



#### **ACPA's 77th Annual Meeting Abstracts**

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### I. Panel Workshop for Parents/Caregivers: Advocacy for School Age Children with Craniofacial Conditions

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Background/Purpose: Our goal is to present to mental health professionals and craniofacial center providers a Panel Workshop on helping parents/caregivers learn how to best advocate for their school age children with craniofacial conditions who experience academic and social challenges. Information about various types of schools, choosing a school, enrollment and school placement, in-school special services, parent–teacher conferences, how to transfer to another school, at home tutoring following surgery, and other related topics will be discussed. In addition, we will address academic stress, developing a realistic schedule, bullying interventions, technology and social media guidelines, and provide families with relevant resources.

Methods/Description: The panelists focused on topics including: academic advocacy, academic testing, neuropsychological and behavioral assessments, IEP's(Individual Educational Plans), optimal classroom placement, academic modifications and accommodations, in-school special services, how to ease the burden of academic stress, and providing information on available services. Social advocacy focused on issues such as how to help children feel comfortable starting school, transitions to grades and new schools, return to school after surgery, and ways to cope with social issues including bullying. Panelists included the craniofacial team psychologist, senior social worker, nurse practitioner, a nonprofit organization's director of family programs, as well as the parent of a school age child, an adult patient, a school administrator, and an educational/advocacy representative from a community organization.

#### 2. Five Centers Look at Perioperative Cleft Palate Care

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Background/Purpose: Cleft and Craniofacial Center's across the country perform cleft palate repairs daily. Each center provides unique care to patients during the pre- and postoperative period to minimize risk and optimize outcomes. Care for patients can vary greatly from

team to team. Five diverse teams will outline and discuss the pre- and postoperative guidelines used to manage patients in the perioperative phase of palate repair in this interactive eye-opener. The panel will discuss the rationale behind differences, such as geographic and demographic differences, and surgeon preferences.

Methods/Description: The perioperative care of patients with cleft palate varies greatly from team to team. Our panel consisting of 5 nurses/coordinators from different teams throughout the United States will discuss aspects and rationale of perioperative cleft palate care. During our interactive eye-opener, each nurse/coordinator will be discussing center demographics including size, patient volume, surgery timing, and surgery technique. We will also discuss pre- and postoperative lab testing, nutrition and feeding, use of restraints, pain medications, wound care, and follow-up. This session will be informative to both new and experienced team members. There will be time allotted for questions and discussion at the end of the panel.

### 3. Professional Competencies for the Perceptual Evaluation of Cleft Palate Speech: A Focus Group

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Background/Purpose: In 2014, a draft set of professional competencies for the perceptual evaluation of cleft palate speech was published from the Speech Assessment Task Force of the 2013 International Cleft Palate and Craniofacial Congress. Additionally, the task force generated a series of general and competency-specific questions that could guide subsequent appraisal and study of these competencies. The publication included a recommendation for further review and revision of these competencies, with scope for expansion of this document. The aim of this focus group will be to seek input and discussion from members of the ACPA on required refinements and amendments to these draft competencies.

Methods/Description: A focus group will be facilitated by a group of experienced Speech-Language Pathologists with knowledge of the original development of the draft competencies. The presentation will include a review of the scope of the perceptual assessment competencies; a review of the existing competencies; and a facilitated discussion around the requirements of each. The application of these competencies to crosslinguistic speech assessment will also be considered. Consensus agreement will be recorded and items requiring further discussion or review will be identified for broader evaluation. Participants will be supplied with a copy of the publication from the Speech Assessment Task Force for review. The new professional

competencies document will be generated from the focus group based upon consensus agreement of the participants. In addition to this, a process for the subsequent dissemination of this document for review by the wider ACPA membership will also be established.

#### 4. Sleep Disordered Breathing in Children with Craniofacial Abnormalities: What You Need to Know

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Background/Purpose: A significant overlap exists between craniofacial disorders and sleep-disordered breathing/obstructive sleep apnea in children. The goal of the session is to fill the knowledge gap in understanding sleep-related breathing disorders in children with complex craniofacial malformations. This session will highlight recent advances in diagnosis and management in a succinct and interactive case-based format with a focus on obstructive sleep apnea across the life span from neonates with Robin sequence to teenagers with facial deformities. Attendees will also learn about state-of-the-art obstructive sleep apnea management including surgical and nonsurgical options for children.

Methods/Description: This session focus on filling the knowledge gap in understanding sleep-related sequelae of common craniofacial disorders seen in pediatric practice that overlap with pediatric sleep-disordered breathing (SDB). The majority of medical professionals do not undergo formal or structured training in sleep medicine during their early careers. Therefore, this proposal will aim to bridge the knowledge gap by (1) creating awareness of the learning gap pertaining to pediatric SDB and (2) sharing information to improve skills to eliminate the gap. Using common pediatric craniofacial conditions as a template, a case-based interactive format will be used for instruction. As a result of attending this session, participants will be better able to integrate diagnosis and management of SDB in the care of their patients.

#### 5. Journal Manuscript Preparation and Submission

Jamie Perry (1), Stephen Conley (2)

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Background/Purpose: This session will be given by members of *The Cleft Palate-Craniofacial Journal*'s editorial board. Editors will discuss what constitutes a good scientific manuscript, what kinds of manuscripts are accepted and what is required by *The Cleft Palate-Craniofacial Journal*. Common problems in manuscript preparation and ways of avoiding them will be addressed.

### 6. Anatomy of the Bilateral Cleft Lip Nasal Deformity and Its Correction

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Background/Purpose: The nasal deformity of the bilateral cleft lip is an intrinsic component of the deformity. Uncoupling of the orbicularis oris sphincter results in lateral displacement of the alar bases and protrusion of the premaxilla and prolabium. Additionally, the medial crura of the lower lateral cartilages are splayed with the fibrofatty tissue of the nasal tip intervening between the tip defining points of the middle crura, resulting in an underprojecting nasal tip and shortened columella. Synchronous repair of the nasal deformity at the time of primary cheiloplasty is essential for treatment of the condition, as

cheloplasty and delayed nasal repair results in secondary stigmata including a wide, underprojecting nasal tip, short or nonexistent columella, and flaring of the nasal ala. The aim of this presentation is to describe the anatomy of the bilateral cleft nasal deformity and to discuss surgical approaches to its correction including the role of presurgical infant orthopedics and strategies for treating asymmetric bilateral clefts.

Methods/Description: Anatomic studies of nasal anatomy in the setting of bilateral cleft lip will be reviewed along with a brief overview of the history of staged repair techniques for bilateral clefts. The authors will present an overview of techniques for synchronous bilateral cleft lip and nasal correction with specific attention given to the Mulliken technique of synchronous bilateral cleft lip repair. Outcome studies of various techniques will be discussed as well as the role of presurgical infant orthopedics in treating bilateral clefts and the role of cleft lip adhesion for optimizing outcomes in an asymmetric bilateral cleft lip. A comprehensive primary surgical repair of the bilateral cleft lip nasal deformity is a safe and achievable goal. Key steps are surgical and/or orthopedic repositioning of the premaxilla, adequate medialization of the alar bases, reconstruction of the orbicularis sphincter, reconstruction of the nasal floor, and unification of the middle crura of the lower lateral cartilages under direction visualization. Presurgical infant orthopedics is an important adjunct treatment for patients with complete clefts as is lip adhesion for patients with asymmetric bilateral clefts.

### 7. Beyond Bottle Selection: Critical Thinking in Management of Complex Feeding Difficulties and Dysphagia in Children with Cleft Lip/Palate

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Background/Purpose: It is well-documented that infants with cleft lip and/or palate (CL/P) often experience feeding difficulties that may negatively impact growth, particularly in the early months of life. Many children with CL/P benefit from compression-based bottle systems and cleft-specific feeding strategies; however, the level of evidence supporting feeding interventions in individuals with isolated orofacial clefts is limited due to small sample size, heterogeneity of the populations, and lack of replication. Moreover, feeding and swallowing difficulties for children with craniofacial anomalies, other congenital anomalies, comorbidities, and/or developmental or cognitive delay are often more complex than for isolated CL/P. Such cases require advanced critical thinking skills for assessment and treatment. The purpose of this session is to explore diagnostic and management challenges in pediatric patients who present with complex feeding and swallowing difficulties beyond those expected with typical CL/P.

Methods/Description: In this interactive workshop, speech-language pathologists from a large medical center will present a variety of cases of infants and young children with cleft and craniofacial conditions who have feeding/swallowing difficulties related to ankyloglossia, laryngo-malacia, and other anatomical, respiratory, gastrointestinal, and oral sensory comorbidities. The presenters will describe the use of patient-focused care to develop a treatment plan for each case. Specifically, referrals to other disciplines, consideration for utensil selection, and other therapeutic strategies will be discussed. Challenges such as slow growth/poor weight gain and swallowing safety will be considered. Attendees will have the opportunity to practice critical thinking regarding complicated feeding and swallowing profiles in this population through participation and collaboration. The more exposure clinicians have a variety of feeding profiles, the more factors they may consider in

assessment and treatment of feeding/swallowing difficulties for future patients with cleft and craniofacial conditions.

# 8. A Common Sense Approach to Patient and Family Centered Craniofacial Team Care: An Honest Community Discussion About Strategies to Optimize Patient and Family Experience Regardless of Your Team's Size or Resources

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Background/Purpose: Interdisciplinary team care is the standard for treating patients with a craniofacial difference and managing the medical complexity of each patient's unique diagnosis. The physical practicalities however of long-term team follow-up and coordination can leave families and patients confused, with lost opportunities for gaining knowledge and support for their child. In this round table discussion, the first author will review best practices for improving the physical experience of team visits for patients and their families, techniques for maximizing the communication and retention of pertinent information to the patient's family and innovative ways to foster community support for patients and their families in the first 20 minutes. The authors will review our 2 teams' practical implementations of these principles and discuss our successes and our ongoing challenges given clinic time and financial limitations in the middle 20 minutes. The last third of the session will be interactive with the audience and a frank, open discussion about how to best leverage clinician and staff time and monetary resources to optimize how patients and families experience team care.

Methods/Description: The 60-minute session will start with a review of best practices around (1) improving the physical experience of team care for patients, (2) maximizing family care givers' information retention, and (3) creating a supportive local community for patients with a craniofacial difference. The first author will then review the practicalities, challenges, and results of their team's yearlong optimization project aimed at improving these 3 areas of their team's care. The senior author will provide their team's alternative approaches to these issues. This part of the hour will be grounded in the qualitative interviews we conducted with families around their experience of team care. Our descriptive data reveals that even long-established patients and their families can be confused about the basics of craniofacial team care and that simple, repetitive interventions can improve information retention. Topics will include fresh (even contrarian) thinking on the logistics of a lengthy interdisciplinary clinic visit, updated formats for sharing information with patients and families including social media, and techniques for garnering resources within your health-care system for patients with a craniofacial difference. This will be a candid discussion and will include cost in staff and volunteer hours and techniques for maximizing patient experience with limited time and money. In the final 20 minutes of the session, we will foster an honest, open discussion among participants to share their successes and ongoing challenges in optimizing how patients and families experience team care.

#### 9. T & A in Patients with Cleft Palate and/or VPI

Pablo Antonio Ysunza (1), Matthew Rontal (1)

(1) Ian Jackson Craniofacial and Cleft Palate Clinic, Royal Oak, MI

Background/Purpose: Performing tonsillectomy and adenoidectomy (T & A) in patients with cleft palate and/or velopharyngeal

insufficiency (VPI) without a cleft has been addressed by several reports in the related scientific literature. However, the issue is still controverted. Adenoid tissue contributes to velopharyngeal closure during speech. In contrast, hypertrophied pharyngeal tonsils can hinder velum movements during speech causing or increasing VPI. When surgery for VPI is being performed, an enlarged adenoid pad can force the surgeon to set a pharyngeal flap or a sphincter pharyngoplasty lower than planned decreasing their effectiveness for restoring velopharyngeal closure. Thus, in selected cases, an adenoidectomy may be a prerequisite for successfully performing surgery for VPI. Another issue to be considered is when to perform adenoidectomy: in the same surgical stage of velopharyngeal surgery? A couple of months before? Several months before? Concerning hypertrophied tonsils in cases of VPI, besides the possible limitation of velar movements tonsils can create a high risk of obstruction when pharyngeal flap or sphincter pharyngoplastsy is performed. The purpose of this article is to address and discuss the questions: When T & A should be performed? How should it be performed? How long should velopharyngeal surgery be delayed after T & A?

Methods/Description: The different roles of tonsils and adenoids in velopharyngeal closure will be demonstrated and discussed using diagrams, videonasopharyngoscopies, and videofluoroscopies. Statistics about how often T & A is indicated in preparation for velopharyngeal closure at the Ian Jackson Craniofacial Clinic of Beaumont Royal Oak Hospital in the last 5 years will be presented and discussed. The controverted indication of adenoidectomy in preparation for velopharyngeal surgery for correcting VPI will be discussed. The risk of obstruction by hypertrophied pharyngeal tonsils when velopharyngeal surgery is performed will be discussed. The possible difficulties of performing tonsillectomy and adenoidectomy after velopharyngeal surgery will be described and discussed. The important issue of timing of T & A in preparation for velopharyngeal surgery will be discussed, and histopathological findings at different times after adenoidectomy will be presented. The surgical techniques for performing T & A in patients with cleft palate and/or VPI without a cleft will be described and their advantages and disadvantages will be presented and discussed.

#### 10. A Discussion and Interpretation of Speech-Language Pathology Assessment Procedures for Allied Team Professionals

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Background/Purpose: There are 3 ways that the speech-language pathologist (SLP) studies speech production, and these are physiologic, acoustic, and perceptual. Each study method is important in understanding normal and disordered speech production, because of the different information that each provides. However, the decisive test for a person with a communication disorder(s) is the perceptual impact of the problem. What is the impression of a person with a communication disorder that other speakers form when engaging with them in verbal communication? This is particularly important for speakers with cleft palate, because they may present with problems that affect the different speech production subsystems of respiration, phonation, resonation, and articulation. The primary problems exhibited by speakers with cleft palate involve the systems of articulation and resonance. Terms such as compensatory errors and hypernasality are used by the SLP in the identification of specific speech production errors and are in many cases unfamiliar to other team members. Often times, other professionals are certainly aware of an individual's

communication deficits, but they are not aware of the specific deficits and their impact. Consequently, there can be confusion in regard to the interpretation of speech assessments and recommendations made by the SLP in the overall management plan of a child who presents with cleft palate. The purpose of this presentation is to discuss in lay terms the different assessment data obtained by the SLP and the interpretation of those data in the treatment of clients with cleft palate.

Methods/Description: The presentation will cover 3 topic areas. (1) Introduction to speech study methods and the speech processes. The 3 different speech production study methods will be discussed along with the speech production processes. The study methods include perceptual study (listening), acoustical study (measuring the parameters of frequency, intensity, and time), and physiologic study (functions of the speech mechanism). The different types of clinical data will be discussed in reference to the speech processes of respiration (breath control for speech), phonation (voice production), resonation (cavity resonance), and articulation (producing speech sounds). (2) Perceptual study and interpretation. The uses of different perceptual measures will be discussed along with the interpretation of the elicited speech production data. Discussion of hypernasality and articulation errors will be presented along with audio and video examples to illustrate the perceptual characteristics. 3. Physiologic/acoustical study and interpretation. Instrumental assessment in the form of physiologic and acoustic measurement is an important adjunct and supplements an SLP's perceptual observations. Physiologic measures such as nasoendoscopy, nasometry, and aerodynamics will be discussed along with the interpretation of the instrumental techniques.

#### II. Attitude Not Answers: Insights from the Patient's Perspective

Jake French (1)

(1) Beaverton, OR

Methods/Description: Jake French helps health-care professionals and family members who care for people with oral cleft and craniofacial conditions gain a deeper understanding of what it feels like to be a patient, so that you can recognize our concerns and meet their needs even better. As a nationally recognized keynote speaker and C6 quadriplegic, Jake's firsthand examples are both heartfelt and humorous. He will show you why it's your attitude and not the answers that can make all the difference in patients. A common misconception is you must do more or know everything. The truth is, we deal with so different people along our medical journey, and we just want to feel like we have a cohesive team that has our back and will always be there to help us find the right answers. Here is your hidden chance to shine. With the right attitude, you can become more effective at collaborating with your fellow professionals as a team and show us how to stick up for ourselves by developing the type of mind-set that helps us solve problems and get the most out of our medical experience. This refreshing speech will help renew a care provider's passion to provide outstanding service, while giving easy to implement tools to increase your connection to those you support.

## 12. 'It Was Meant to Be': Challenges and Strengths of School-Aged Children with Cleft Adopted from China: A Qualitative Study

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Background/Purpose: There is a significant population of children with cleft lip and/or palate (CL/P) who have been adopted from China. While much of the literature has focused on medical outcomes such as receiving corrective surgeries later than noninternational adoptees (Swanson et al., 2014), there has been comparatively less research on the psychosocial outcomes for these children. For example, there is little literature that describes how children adopted from China with CL/P cope with potential challenges related to acculturation, discrimination, or bullying in addition to stressors associated with the medical condition and its sequelae. The present study utilized qualitative methods to richly characterize the experiences of school-aged children with CL/P who have been adopted from China.

Methods/Description: Participants were recruited through a multidisciplinary CL/P medical clinic. Nine child–parent ( $M_{\rm age}=10, 6$  females, 3 males) dyads participated in 1-time semistructured interviews. Interviews were audio recorded, transcribed verbatim, and analyzed according to principles of interpretative phenomenological analysis (Smith, Flowers, & Larkin, 2009).

Results: Preliminary results suggest that these families demonstrate adaptive coping via close family connectedness, quality time, transparent communication, religion/spirituality, and therapies (eg, speech, mental health) when needed. Child participants generally indicated positive self-concept with self-conscious emotions primarily associated with facial differences due to CL/P as opposed to race. Speech and hearing difficulties were also associated with self-conscious emotions. Child participants identified personal strengths readily and endorsed positive emotions regarding their adoption. Parent participants described great meaning associated with the process of adoption in addition to the challenges associated with financial and time burdens. Families typically maintained some material possessions and yearly traditions (ie, Chinese New Year) to commemorate Chinese culture, though child participants were predominantly assimilated to white majority culture.

Conclusions: This study takes a unique, qualitative approach to further understand an understudied population. This study highlights paths of positive family adjustment through family belongingness, connectedness, and openness. Honest dialogue regarding CL/P and adoption appeared to normalize experiences and empower the child participants. Although several child participants presented with a history of emotional or behavioral difficulties (eg, poor social skills, emotional sensitivity), family and community support appeared to be significant protective factors.

### 13. A Competency Assessment Tool for Unilateral Cleft Lip Repair

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Background/Purpose: Objective evaluation of surgical performance is becoming an increasingly important part of how trainees are evaluated and attending surgeons are credentialled. Evaluation tools include global rating scales of performance (eg. Objective Structured Assessment of Technical Skills or OSATS) as well as procedure-specific skills checklists. Because unilateral cleft lip repair is a core procedure for plastic surgery, oral surgery, and otolaryngology training, a procedure-specific checklist would aid in evaluating performance and

determining who is appropriate for independent practice. A challenge however is that numerous repair techniques exist making evaluation challenging. Here, we sought to create a unilateral cleft lip repair evaluation tool that is agnostic to specific repair technique.

Methods/Description: A panel of 4 surgeons was assembled with expertise in the 3 most common repair techniques: Millard rotation-advancement, Mohler modification, and Fisher anatomic subunit. A Delphi process was undertaken in 3 rounds to come to a consensus about what aspects of repair were most important and spanned all techniques: (1) Unguided generation of ideas, followed by stratification of ideas into core domains: marking, performance, and final outcome. (2) Review of aggregate ideas to identify common themes that spanned technique (eg. "Reposition the greater segment inferiorly so peaks of Cupid's bow are symmetric"), followed by drafting of specific data points and discarding ideas supported by only one surgeon. (3) Review of draft data points, followed by revision to clarify goal and discarding points endorsed by only one surgeon.

Results: After consensus points were drafted, the checklist was reviewed with a survey methodologist refining language to minimize bias and develop a 3-pronged scale for assessing performance of each checklist point. Ultimately, a 19-item assessment tool was generated encompassing key areas of cleft lip repair including marking the repair, incision and tissue handling, muscular approximation, and symmetry of the final result. Notably, although all 4 surgeons perform primary rhinoplasty and relevant checklist points were considered, they were not included in the final tool because primary rhinoplasty is not universally performed.

Conclusions: Here, we have demonstrated that despite variation in unilateral cleft lip repair techniques, common themes between them exist that can be used to assess performance and outcome. This has potential implications for how surgical trainees are assessed, the credentialing process, and could even be used to evaluate the qualification of surgeons who wish to participate in surgical missions.

### 14. Accuracy of Dolphin Three-Dimensional Software in Predicting Upper Airway Volumetric Changes After Orthognathic Surgery

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Background/Purpose: Orthodontists and surgeons have been looking for more accurate methods to predict surgical outcomes in patients with skeletal discrepancies. Predicting postsurgical changes in the airway volume may be important for patients with a high risk for sleep-disordered breathing. The purpose of this study was to compare the accuracy of Dolphin 3-dimensional (3D) software in predicting upper airway volumetric changes that would occur after orthognathic surgery.

Methods/Description: The sample consisted of 20 subjects from the surgical clinic of a graduate orthodontic program who have been treated with Le Fort I maxillary movement, BSSO, with or without genioplasty. All subjects had to have preoperative (T0) and at least 6 months postoperative (T1) CBCTs that were imported to Dolphin 3D software. 3D voxel-based superimposition on the cranial base was performed for T0 and T1 to accurately measure the skeletal surgical movements. Virtual orthognathic surgery was performed at T0 to mimic the actual skeletal osteotomies using the treatment simulation tool in Dolphin 3D. A predicted 3D soft tissue image (Tp) was generated based on the Dolphin virtual skeletal planning. The upper

airway was segmented in Dolphin software and exported into Mesh-Mixer software as STL surface files in both T1 and Tp. The measurements of the 3D volume of the airway were calculated in MeshMixer. Means and standard deviations of upper airway volume were compared and correlated using paired *t* test and Pearson correlation coefficient. Interrater reliability analysis for 10 subjects was done using intraclass correlation.

Results: Intraclass correlation showed an excellent interrater reliability (ICC = 0.994, P < .001). No statistically significant differences (P = .07) were found between the upper airway volume of T1 (mean =  $16220 \pm 7545$ ) and Tp (mean =  $14684 \pm 7789$ ). Pearson correlation showed a strong correlation between T1 and Tp (r = 0.897, P < .001), indicating that Dolphin 3D surgical predictions could accurately predict airway volumetric changes that may occur after orthognathic surgery.

Conclusions: Dolphin 3D may be a reliable tool in predicting airway volumetric changes that may occur after orthognathic surgery. It would be advantageous to incorporate this assessment in the 3D virtual planning of orthognathic cases, especially for cases with a high risk for sleep-disordered breathing. Future studies with larger sample sizes are needed to confirm the findings of this study.

#### 15. Acellular Dermal Matrix For Repairing Cleft Palate Fistula

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Background/Purpose: Fistula presence represents a failure of the surgical repair of the cleft palate. Fistula repair is challenging with a high rate of fistula recurrence that increases with each added repair attempt. In the present retrospective study, we review the efficacy of using acellular dermal matrix (ADM) for cleft redo palate fistula closure.

Methods/Description: Twenty consecutive patients were included in the study from 2013 to 2016. Each patient was assigned a Veau designation and a Pittsburgh fistula classification. All patients with palate fistula, underwent fistula repair utilizing ADM as an interposition layer.

Results: There were 11 females and 9 males. Mean patient age was 13.9 years, ranging from 2 to 43 years. The mean length of follow-up was 9.7 months. Complete fistula closure was obtained in 16 patients; 1 patient had asymptomatic recurrent fistula; 2 patients had partial closure with reduction in fistula size and minimal nasal regurgitation; 1 patient developed a recurrent fistula without changes in symptoms (success rate of 85%).

Conclusions: Utilizing ADM for cleft palate fistula repair as an interposition layer is a safe and simple procedure, with satisfactory outcomes that reduces fistula recurrence compared to closures without ADM. A larger, prospective, randomized trial is required for determining efficacy in secondary and tertiary fistula repairs.

## 16. American Indian Alaskan Native (AIAN) Access to Appropriate Cleft Lip and Palate Treatment Using Geographic Information Systems (GIS)

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Background/Purpose: American Indian Alaskan Natives (AIAN) have unmet health-care needs with health facilities being frequently inaccessible and/or medically obsolete. AIAN patients' access to subspecialty, particularly surgical subspecialty care, is even more sparse. AIAN populations have the highest incidence of cleft lip and palate (orofacial clefts [OFCs]) when compared with other ethnic groups. We aim to determine the AIAN population's proximity, availability, and access to American Cleft Palate-Craniofacial Association (ACPA) accredited centers for treatment of OFCs.

Methods/Description: To receive ACPA accreditation, teams must include experienced and qualified professionals from medical, surgical, dental, and allied health disciplines working in an interdisciplinary and coordinated system to provide standard of care to patients with OFCs. An internet search of ACPA-affiliated surgeons was conducted including training type (plastic surgery, otolaryngology, craniofacial fellowship). ACPA centers and density of craniofacial (CF) trained surgeons were mapped using Geographic Information Systems (GIS) and overlaid with 2018 census population data to visually display possible disparities. Tribal Census Tract data were mapped including ACPA centers to assess possible disparities in high-density AIAN populated lands. Total cleft need in high-density AIAN populated lands based on Tribal Census Tract data were calculated by: total population within geographic unit × % AIAN alone or in combination × % children ages 0 to 19 years old × 0.0036 CLP incidence.

Results: The results included 359 surgeons at 146 ACPA centers (including duplicate surgeons staffing multiple sites). This was reduced to 215 fellowship-trained craniofacial surgeons (excluding duplicates) at 123 ACPA centers (excluding duplicates). GIS mapping demonstrates geographical isolation of NA populations from accessing appropriate care in ACPA-recognized centers. The total annual potential pediatric cleft need for AIAN areas in study is 1042 children. Two states with high-density AIAN populated lands (ND and WY) have no ACPA-recognized centers; 47.1% (8/17) of ACPA centers in high AI/AN lands have no CF trained surgeons versus 78.9% (135/171) CF staffed ACPA centers nationally.

Conclusions: AIAN populations are underserved by ACPA-accredited centers, and more drastically, by CF fellowship-trained centers. Not addressing OFCs in a timely and quality manner can lead to worse outcomes and further marginalization of these children. With information on up-to-date disease burden and volume capacity of regional ACPA centers, accessibility from NA lands to ACPA centers (distance, roads, public transportation), state-by-state public aid coverage for all necessary OFC and gaps in coverage (ie, dental, orthodontic, oral surgery), and adherence to OFC treatment timeline and barriers to care, we will be capable of making data-driven, informed decisions to more effectively ensure AIAN access to appropriate surgical care.

#### 17. Anatomy of the KISS Principle for Trainee Cleft Surgeons: The Quest for a Cleft Aesthetic Code of the Philtrum and Median Tubercle in Unilateral Cleft Lip Surgery using Strictly Anatomical Concepts

Bona Lotha (1)

(1) Yemen Smile, Sana, Yemen

Background/Purpose: The function and aesthetics of a seemingly innocuous orbicularis oris muscle cannot be underestimated. A KISS could be defined as the juxtaposition of 2 orbicularis oris muscles in a state of sustained, passive, and active contraction. The KISS principle in cleft lip surgery posits that the way forward for a successful outcome lies in keeping it simple and straightforward. Getting it right the first time would serve as the perfect launching pad for any cleft service

as primary unilateral cleft surgery is the first test of a surgeon's skill at clefts. One of the key strategies in understanding the anatomy of change in unilateral cleft lip surgery, using the KISS principle (keeping it sequential and straightforward) lies in a good knowledge of the orbicularis oris muscle–dermis complex, and a proper sequencing of surgery steps. Our focus is on the perfect simulation of a normal muscle dermis complex and enhancing the lip pout along the median tubercle. Combined with some nuances of lip repair, fine tuning of cleft lip repair is achievable with reasonably good aesthetic functional outcomes.

Methods/Description: Using the KISS principle during cleft lip surgery, a straightforward sequence of muscle approximation is (1) Marginalis muscle sutures with Cupid bow dermal hitch in the superior aspect to cause dimpling and prevent lip notching. (2) Two to 3 fine sutures attach the medial peripheralis to lateral, overlapping muscle and dermis to enhance the philtrum. (3) Above this, a muscle z-plasty is done below the columella base. (4) The subluxated caudal septum is shifted to a more midline position by gentle dissection and internal cinch suture added. (5) A 1 to 2 mm separation of mucosa and horizontal mattress sutures evert the tubercle. Minor salivary glands or fat are included below the median tubercle paying attention to nuances such as the small z-flap of red vermillion, mucosa, above Cupid roll, alar shaping, and botox post-op add to the final improvement.

Results: A successful repair of a cleft lip deformity includes proper alignment of all the labial and alar elements, floor of nose reconstruction when needed, adequate muscle flaps for symmetric closure, and a dynamic philtrum as well as an attractive pout of the median tubercle. Once the finer aspects of lip repair are done, the lip looks a lot better and natural.

Conclusions: Getting it right the first time is an important aspect of cleft lip surgery. Understanding the nuances of a good functional and aesthetic cleft lip surgery requires experience with larger numbers of surgeries, because no 2 cleft lip deformities are the same. There are many good techniques and plastic surgeons have personal preferences, but the most important variable is the surgeon. A successful repair of a cleft lip deformity includes proper alignment of all the labial and alar elements, floor of nose reconstruction when needed, adequate muscle flaps for symmetric closure, and a dynamic philtrum as well as an attractive pout of the median tubercle.

### 18. Augmentative and Alternative Communication (AAC) Use Among Multidisciplinary Cleft and Craniofacial Team Patients

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Background/Purpose: The goal of this research is to establish preliminary data regarding the number of patients who visit multidisciplinary cleft and craniofacial teams that use augmentative and alternative communication (AAC) supports. During visits to a multidisciplinary team, the treating speech-language pathologist is tasked with assessing and addressing needs in the areas of feeding, swallowing, articulation, language, voice, and resonance. Some patients will present with complex communication needs (CCN) severe enough to warrant use of augmentative and alternative communication (AAC). To date, there is neither documented information regarding the prevalence of AAC use among patients in the care of multidisciplinary cleft and craniofacial centers nor is there a standard, accepted protocol on how to best support these patients. This study seeks to establish preliminary data on the prevalence of AAC use among patients in craniofacial team care and draft basic guidelines for providers.

Methods/Description: This IRB-approved retrospective study consisted of chart reviews for all patients who visited the ACPA-approved multidisciplinary cleft palate and craniofacial team at a tertiary, teaching medical center from August 2017 to August 2018. Patients who met inclusion criteria were included in the study. Based on the various data points collected, patients were placed into the AAC-user group or the non-AAC-user group.

Results: A 6.9% (n = 32) of the sample was found to use AAC supports while 93.1% (n = 432) did not. The AAC group had a mean age of 5.1 years (SD: 4.2) and was 68.8% (n = 22) male. The non-AAC group had a mean age of 6.3 (SD 4.9) and was 59.3% (n = 256) male. When comparing the likelihood of having an identified syndromic diagnosis, the AAC group (40.6%, n = 13) was significantly more likely than the non-AAC group (17.6%, n = 76; P = .003) to have one. Conclusions: This is the first study to report the prevalence of AAC use among patients in the care of multidisciplinary cleft and cranio-

use among patients in the care of multidisciplinary cleft and craniofacial teams. This study suggests that a subset of craniofacial team patients may have complex communication disorders that require AAC supports. Craniofacial teams should be aware of resources for these patients, so that the child's communication needs are met in the hospital, school, and community. For neurologically typical verbal communicators who present with pervasive maladaptive articulation patterns that severely reduce speech intelligibility, AAC implementation may hold some short-term value. However, AAC implementation should not be done alternatively to appropriate articulation therapy. Nor should it come at a reduction in the frequency and intensity of articulation therapy.

## 19. Comparison of the Pharyngeal Airway Volume between Patients with Ectodermal Dysplasia and Non-Affected Controls: A CBCT Study

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Background/Purpose: Ectodermal dysplasias (EDs) are a group of inherited congenital disorders characterized by abnormalities of 2 or more ectodermal structures such as the skin, hair, nails, teeth, and sweat glands. ED has an incidence rate of about 1 in every 100 000 live births. Individuals with ED often exhibit maxillary hypoplasia, reduced vertical dimension, and midfacial retrusion. Previous studies using 2D lateral cephalograms found smaller airway dimensions for patients with ED in comparison to controls. The aim of this study is to evaluate the volume and maximum constriction of the pharyngeal airway in patients with ectodermal dysplasia (ED) using cone-beam computed tomography (CBCT) and compare them to controls.

Methods/Description: This retrospective study included 9 patients with ED and 30 controls (CON). We only included controls with class I occlusion and no skeletal discrepancies in any dimension and excluded those with any associated craniofacial conditions. Pretreatment CBCT volumes were imported into Dolphin 3D software, which was used to measure the volume of the nasopharynx (NP), oropharynx (OP), hypopharynx (HP), and area of maximum constriction (MC). For all measurements, the anterior border was set to a plane perpendicular to Frankfort horizontal (FH) passing through PNS, while the lateral and posterior borders were set to the soft tissue contours of the pharyngeal wall. For NP, the upper border was the soft tissue contour of the pharyngeal wall, and the lower border was the plane parallel to FH passing through PNS. For OP, the upper border was plane parallel to FH passing through PNS, and the lower border was the plane parallel to FH plane passing through third cervical vertebra. For HP, the upper border was the plane parallel to FH plane passing through

third cervical vertebra, and the lower border was the plane parallel to FH connecting the base of the epiglottis to the entrance to the esophagus. MC was automatically detected by the software. Independent *t* test was used to compare the volumes and MC. A second investigator remeasured 10 subjects for reliability analysis.

Results: No significant differences were found for the age or gender distributions between ED (mean age =  $12.8 \pm 3.5$ ; 66.7% females) and CON (mean age =  $12.3 \pm 2.1$ ; 63.3% females). All measurements showed normal distribution except HP. Intraclass correlation showed excellent interrater reliability (r > .87, P < .001). Independent t test showed no significiant differences for NP (ED =  $4032 \pm 2305$ , CON =  $3773 \pm 1260$ ; P = .75), OP (ED =  $9592 \pm 4270$ , CON =  $9767 \pm 3584$ ; P = .91), or MC (ED =  $125 \pm 68$ , CON =  $121 \pm 44$ ; P = .85). Mann-Whitney U test showed no significant differences for HP (ED =  $4966 \pm 1872$ , CON =  $4209 \pm 2144$ ; P = .24).

Conclusions: Despite the maxillary hypoplasia and midface retrusion among patients with ectodermal dysplasia, the pharyngeal airway volumes among affected individuals do not seem to be significantly different from those of healthy controls. Future studies with larger sample sizes may be needed to confirm our findings.

### 20. Correlation Between Skeletal and Soft Tissue Asymmetry in Patients with Hemifacial Microsomia: A CBCT Pilot Study

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Background/Purpose: Hemifacial microsomia (HFM) is a congenital condition characterized by hypoplasia of the structures within the first and second branchial arches that can present in various degrees of severity. HFM is characterized by facial asymmetry, with unilateral underdevelopment of the mandible and/or maxilla, orbital asymmetry, ear deformities, soft tissue asymmetry, and nerve involvement. Although it is understood that there is unilateral skeletal and soft tissue discrepancy, the correlation between the 2 has not been evaluated. The purpose of this study was to utilize 3-dimensional (3D) superimposition and color mapping of the soft and hard tissues, to evaluate how the soft tissue discrepancies in various regions of the face may correlate with the skeletal discrepancy.

Methods/Description: This retrospective study analyzed initial CBCTs of 8 patients with HFM. Initial CBCTs were oriented using Dolphin software. The skeletal and soft tissue facial structures were separately exported as STL files. For each of the skeletal and soft tissue surface, the left side of the 3D model was mirrored using midsagittal plane as reference, in Meshmixer software, resulting in a perfectly symmetric skull and face based on 2 left sides (mirrored model). Original and mirrored 3D models were exported to 3D Slicer software and superimposed by a surface best fit method, using the left side as reference. Differences between original right side and mirrored left side were assessed by colormap and were quantified by regional mesh differences on the 3 planes of space, in each group. The 2 planes were combined to produce an absolute difference between the 2 sides. Seven regions of interest were assessed: frontal bone area, endocanthion area, exocanthion area, malar area, maxillary frontal area, mandibular frontal area, and gonion area. The correlations between the amount of skeletal asymmetry and soft tissue asymmetry were evaluated by Pearson correlations.

Results: Hard tissue asymmetry ranged from 1.4 (endocanthion) to 5.5 mm (gonion), while soft tissue asymmetry ranged from 1.5 (endocanthion) to 4.5 mm (gonion). The differences between the amount of skeletal and soft tissue asymmetry ranged between 0.2 (endocanthion) and 1.8 mm (malar region), with soft tissues showing slightly higher levels of asymmetry except for the gonion and frontal regions. Correlation between skeletal and soft tissue asymmetry was highly variable, with the highest correlation at gonion (r = .874, P = .005) and the lowest at exocanthion (r = .206, P = .624).

Conclusions: The results of this study show a high variability for the correlation between skeletal and soft tissue asymmetries among patients with HFM. Open source softwares can be valuable for assessing asymmetries in this cohort of patients. Clinicians should evaluate each component independently for proper diagnosis and treatment planning. Future studies with larger sample sizes are needed to confirm our findings.

### 21. Developing a Sustainable Nasoalveolar Molding Program in Outreach Settings: An Eight-Year Follow-Up

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Background/Purpose: Global Smile Foundation (GSF) is a nonprofit foundation that provides comprehensive cleft care to underserved patients. GSF focuses on long-term follow-up and sustainability of local health-care teams, having engaged in 32 years of follow-up in Ecuador. In 2012, GSF added presurgical nasoalveolar molding (NAM) therapy for their patients in Guayaquil, Ecuador, as part of its sustainability and empowerment initiative. We present longitudinal data on 189 patients treated with NAM and discuss the challenges/barriers to its completion.

Methods/Description: Data were collected from GSF surgical and dental records including patient diagnosis, completion/incompletion, and length of NAM therapy. Surgeon, patient age, peri, intra, and postoperative procedural data were collected for primary cleft lip/nose and palate repair, and any additional surgeries. Follow-up clinical and photographic data were retrieved to document long-term outcomes.

Results: A total of 207 patients were treated with presurgical therapy: 189 patients received NAM therapy, while 18 patients were treated with lip tape and/or nasal elevator. Of the 189 NAM patients who received NAM, long-term follow-up was available for 96 (50.8%) patients, while 84 (44.4%) were lost to follow-up or subsequently seen by another foundation, and 9 (4.8%) are currently undergoing NAM or awaiting primary surgery. Of the 96 patients with long-term followup, 70 (72.9%) had unilateral cleft lip and palate and 26 (27.1%) had bilateral cleft lip and palate; 64 (66.67%) were male and 32 (33.3%) were female. Of those 96 patients, 58 (60.4%) completed NAM therapy, 17 (17.7%) failed to complete it, and 21 (21.8%) had incomplete NAM documentation. The average age at NAM initiation was 36.36  $\pm$  31.39 days (range: 0-157 days) and average length of NAM therapy was 118.98 ± 82.68 days (range: 1-222 days). Patients underwent an average of 2.13  $\pm$  0.93 (range: 1-5) surgeries after NAM initiation, with an average of 0.17  $\pm$  0.43 (0-2) cleft lip/nose revisions, 0.06  $\pm$ 0.28 (0-2) gingivoperioplasty, 0.06  $\pm$  0.28 (0-1) premaxillary setbacks,  $0.07 \pm 0.30$  (0-2) fistula repairs, and  $0.03 \pm 0.17$  (0-1) velopharyngeal insufficiency corrections. Over an 8-year period, 12 NAM providers were trained in Ecuador; 7 provided treatment in Ecuador, and 5 provided treatment internationally, making Ecuador a site for information exchange. Follow-up for NAM patients was an average of  $2.00 \pm 1.77$  (0.22-6.67) years after NAM initiation and  $1.45 \pm 1.77$  (0-6.52) years after their primary cleft lip/nose repair. This includes continued long-term follow-up and comprehensive cleft care in addition to NAM therapy.

Conclusions: With yearly patient follow-up and year-round partnership with local professionals, our model shows successful long-term delivery of NAM therapy as part of a sustainable comprehensive cleft care strategy in outreach settings.

### 22. Developmental and Psychosocial Risks in Children with Pierre Robin Sequence: A Retrospective Chart Review Study

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Background/Purpose: Pierre Robin Sequence (PRS) is characterized by micrognathia, glossoptosis, upper airway obstruction, and frequently, cleft palate. Children with PRS are at risk for feeding and breathing difficulties, including failure to thrive and obstructive sleep apnea. However, few studies have documented developmental and psychosocial risks associated with PRS or receipt of related services. In order to address these gaps, a retrospective chart review study of patients with PRS treated between 2005 and 2018 was conducted at a single pediatric academic medical center's craniofacial center.

Methods/Description: Charts of 108 patients (62% female; mean age at chart review:  $5.97 \pm 4.10$  years) with PRS were reviewed. Demographic (eg, sex, race, age, insurance type, distance to care) and clinical characteristics (eg, associated syndromes or anomalies; type of treatment; history of developmental delays, evaluations, and therapies; learning concerns; behavioral health services and diagnoses) were abstracted. Descriptive statistics and  $\chi^2$  or Fisher exact tests were used to evaluate differences in developmental and psychosocial outcomes by sex, treatment (nonsurgical, mandibular distraction, tongue—lip adhesion), insurance type, and distance to care.

Results: Children were predominantly Caucasian (88%), resided outside of the hospital's county lines (71%), and had Medicaid insurance (63%). Nearly all (96%) had cleft palate; 41% had associated anomalies or syndromes. Regarding treatment, 44% were treated nonsurgically, 32% with mandibular distraction, and 24% with tongue-lip adhesion. History of developmental delay was documented in 74%, most commonly speech/language delays, with no significant differences observed by sex, presence of a syndrome, or treatment type. Of those with developmental delays, 52.5% were referred for a hospitalbased developmental evaluation, 80% received state-sponsored early intervention services, and 81% engaged in hospital-based developmental therapies. There were no significant differences in receipt of services by distance to care or insurance type; however, those who received surgical treatment were significantly (P = .04) more likely to receive developmental therapies than nonsurgically treated children. Across the sample, 24% required an IEP or 504 plan in school. Ten percent were referred for hospital-based behavioral health services, 13% had global developmental delay, 2.8% ADHD, and 0.9% autism.

Conclusions: Developmental delays were common in this sample of children with PRS, with a minority having documented learning or behavioral health concerns. These findings, while descriptive and

limited by use of available data recorded in medical charts, provide a greater understanding of the developmental and psychosocial risks for children with PRS, and can be used to offer anticipatory guidance for clinical care providers and parents. Findings also underscore the need for interdisciplinary care approaches including developmental and psychosocial risk screening.

#### 23. Device Malfunction During Mandibular Distraction for Infants and Young Children with Robin Sequence

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Background/Purpose: Mandibular distraction osteogenesis (MDO) has been increasingly used in infants and young children with severe upper airway obstruction associated with Robin sequence (RS). By enabling gradual mandibular lengthening and soft tissue stretching, this procedure effectively opens the upper airway. The most commonly used devices in MDO have internal and external components that require manual activation, often performed by health-care providers and/or adult family members. These distractors undergo rigorous examination and testing by the various companies, prior to clearance by the Federal Drug Administration. Although complications associated with MDO in infants with RS have been well-documented, there is a paucity of literature precisely describing hardware/device malfunction. The current study reports on the authors' recent experiences with these problems, in an effort to shed light on these complications and identify potential steps to mitigate future issues.

Methods/Description: The authors retrospectively reviewed a prospectively gathered database to identify all young children younger than the age of 3, who underwent MDO using buried internal devices for severe upper airway obstruction associated with severe grade 3 RS from March 2007 to June 2019. We specifically focused on complications attributable to the hardware itself. Discussions were held with the company representatives to better understand these problems, trouble-shoot the issues at hand, and seek avenues for future improvement.

Results: A total of 17 patients met inclusion criteria. The mean age at MDO was  $24.54 \pm 36.63$  (range: 3.14-117.29) weeks. Intraoperative activation of all devices under direct vision resulted in satisfactory distraction. Three (17.6%) patients developed complications directly related to the device. Postoperative hardware malfunction included unilateral device breakdown of component parts (n = 2) and unilateral failure to maintain distraction (n = 1). Two patients required surgical removal and replacement of the device, while the remaining complication occurred during the consolidation phase and did not require intervention.

Conclusions: This report documents a series of device/hardware malfunction in infants and young children undergoing MDO for severe upper airway obstruction associated with RS. Despite all reasonable testing and development, these devices are not infallible, resulting in patient morbidity. Our institution is currently working with device manufacturers to address these complications in effort to improve quality control and minimize risk of future adverse events.

### 24. Do Transverse Problems in Children with Cleft Lip and/or Palate Influence Orthognathic Surgery?

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*Background/Purpose*: The purpose of this study was to determine the effect of transverse problems on future orthognathic surgery and skeletal/dental discrepancies in children with cleft lip and/or palate (CL/P).

Methods/Description: In this retrospective cohort study, we included 126 patients with bilateral and unilateral CL/P (70 males, 56 females; 19 BCLP, 5 BCLA, 71 UCLP, 20 UCLA, 11 CP) who visited Orthodontic Department, Osaka University Dental Hospital between 2002 and 2014, and took at least 2 lateral cephalograms at 7 (T1) and 15 (T2) years old. A total of 27 UCL/P patient among them took computed tomography (CT) before bone graft and was enrolled to examine the width and morphological features of cleft in alveolar. To determine the influence of transverse problem, posterior crossbite, and other cephalometric, independent variables at T1 were examined by logistic regression analysis (P = .05); the width and cross-sectional morphology of alveolar cleft were examined by t test and digital image analysis between favorable/unfavorable outcome groups. As outcomes, clinical decision of the need for orthognathic surgery, skeletal discrepancy (ANB  $\ge 0^{\circ}$  or  $<0^{\circ}$ ), and anterior crossbite (overjet  $\ge 0$  or <0 mm) at T2 were examined.

Results: Posterior crossbite at T1 was determined to be a risk factor for orthognathic surgery and anterior crossbite at T2 (odds ratio: 3.70-6.30). Determined risk factors for orthognathic surgery, skeletal discrepancy, and anterior crossbite at T2 were posterior crossbite, pushback method of palatal closure, smaller protrusion and length of maxillary, smaller S-N length, greater protrusion and length of mandibular, the number of missing teeth, and steeper palatal plane at T1. The width of alveolar cleft before bone graft showed no significant relationship with the prognosis. Cross-sectional morphology of alveolar cleft showed no significant difference between favorable/unfavorable outcome groups.

Conclusions: Posterior crossbite in children with CL/P was proved to be a risk factor for orthognathic surgery and anterior crossbite. Width and morphology of alveolar cleft showed no significant relationship for the prognosis in patients with CL/P.

#### 25. Does Type of Surgical Airway Management Impact Speech Outcomes in Pierre Robin Sequence?

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Background/Purpose: Surgical management of airway obstruction in infants with Pierre Robin sequence (PRS) may include tongue–lip adhesion (TLA) or mandibular distraction osteogenesis (MDO). These surgeries are typically performed in the first months of life, which overlaps with a critical period for speech development. Previous studies have suggested that TLA might temporarily impact early speech production, and encourage certain speech error types; however, little is known about the effects of TLA and MDO on speech outcomes in school-aged children (LeBlanc & Golding Kushner, 1992). This study aims to explore the speech characteristics of school-aged children with PRS who have undergone TLA or MDO and examine group differences in speech outcome.

*Methods/Description*: Nineteen children with repaired cleft palate with PRS (10 female, 9 male; 7 children with syndromes) participated in this study and were divided into 2 groups by surgical history: n=14 MDO, n=5 TLA. All children underwent evaluation by an SLP between ages 5, 0-6, and 11 years as part of standard care. Speech evaluations included standardized articulation testing (Goldman-Fristoe Test of Articulation-3, GFTA-3) and perceptual ratings of

resonance based on a standard speech sample, using published scaling methods (Henningsson et al., 2008, Chapman et al., 2016). Group comparisons were assessed using Wilcoxon rank sum tests,  $\chi^2$ , and Fisher exact tests.

Results: There were no statistically significant group differences in GFTA standard scores (TLA median GFTA SS 87.5 vs MDO median 89.5; P = .73). None of the children in the study exhibited palatalization errors at the time of their school-aged speech evaluation. Only 1 child exhibited backing errors and 2 exhibited compensatory misarticulations. There were no statistically significant differences in articulation performance (P = .23) or hypernasality ratings (P = .11) for syndromic versus nonsyndromic children. For the TLA group, 20% had history of a language delay versus 69\% of the MDO group (P =.12). Fifty percent of TLA patients had a history of speech-language therapy as compared to 89% of children in the MDO group (P = .19). Presence and severity of hypernasality was similar across surgical groups (P = .45). Although the children with TLA underwent palate repair at a significantly later age (TLA = 14.95 months, MDO = 12.07months, P < .03), age at palate repair was not significantly correlated with GFTA scores (P = .79) or hypernasality ratings (P = .55).

Conclusions: Results from this study suggest no group differences in the articulation proficiency or hypernasality ratings of school-aged children with PRS who underwent TLA versus MDO for early airway management. This study provides reassuring information to clinicians and families that there are no differential effects of MDO versus TLA on later speech development. Regardless, children with PRS with cleft palate do exhibit a high overall rate of speech-language disorders and need for speech-language intervention services.

### 26. Early Multidisciplinary Team Management of Infants with Clefts: Number and Types of Interventions

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Background/Purpose: Feeding difficulties are well recognized in infants with orofacial clefts and can lead to poor growth and development, delayed surgical intervention, increased hospitalizations, and increased burden of care. Early multidisciplinary team management and follow-up has been recommended to improve early weight gain and to promote safe and efficient feeding in this population. The purpose of this study is to quantify the number of interventions required for children with CL (cleft lip), CP (cleft palate), and CLP (cleft lip and palate) to maintain adequate nutrition.

Methods/Description: This study was designed as a retrospective chart review of 80 infants with a diagnosis of (CL), (CP), or (CLP), who were evaluated by our Craniofacial Feeding Team. Infants with syndromes and chromosomal abnormalities were included. Any clefting of the alveolus was rated as a cleft palate for the purpose of this study. Core members of the Craniofacial Feeding Team included speech-language pathologists (SLP), advanced practice providers (APP), and a nurse coordinator. Tertiary members included dieticians, lactation consultants, otolaryngology, and a social worker. Infants with clefts born between June 1, 2018, and June 30, 2019, were included. Data were collected from the initial clinic visit until 2 consecutive visits indicating adequate weight gain and efficient feeding, along with consensus from the medical team and family, were obtained. Weights were plotted along the World Health Organization (WHO) growth chart data table for 0 to 2 years. Collected data also included birth weight, gestational age, age at first visit, and cleft type. Interventions included feeding system changes, nipple size or nipple flow changes, changes in the caloric density of milk, referral for instrumental swallowing assessment, and inpatient admission for poor weight gain or dysphagia.

Results: Of the 80 infants in this study, all were evaluated by a cleft team SLP and an APP at each visit. Most initial visits occurred within 10 days of discharge from the birth hospital or NICU; 11 (13.75%) had isolated CL only, 47 (58.75%) had CLP, and 22 (27.5%) had isolated CP only. The most common interventions included a change in the nipple or feeding system, followed by a change in the caloric density of milk. Only 2 (2.5%) infants required inpatient admission for feeding assistance, both of which were later diagnosed with chromosomal abnormalities.

Conclusions: Infants with clefts require numerous interventions and consistent follow-up to maintain general weight gain standards as defined by the WHO and to maintain efficient feeding skills. Failure to meet these standards may set the stage for delayed surgical repairs, increased hospital admissions, and developmental delay. This study described the most common interventions used and the frequency with which they occurred at one cleft/craniofacial center.

## 27. Effectiveness of a 4-Day Speech Therapy Training Program for Cleft Palate Professionals in Abuja, Nigeria

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Background/Purpose: The poster identifies and highlights the effectiveness and challenges in implementing a one-week speech therapy training for health professionals in low- and middle-income countries. Children born with cleft palate need comprehensive care from a cleft team to ensure that they can meet their highest potential and engage in all aspects of life in their communities, including participation in social and educational opportunities. However, in these areas, it is often quite difficult to find health professionals with the capacity to address cleft palate speech characteristics. This means that many children continue to speak with those characteristic cleft palate speech patterns post-palate repair, negatively affecting their overall quality of life. Research has shown that in low- and middle-income countries a partner hospital model such as Smile Train's improves the quantity and quality of cleft care services (Purnell, McGrath, & Gosain, 2015). However, there are varying models as to how to structure these programs once a partner hospital is established. Limited research has been conducted focused on the efficacy of speech therapy trainings intended to provide a lasting, sustainable impact on these communities in a short period of time.

Methods/Description: This poster shares the outcome of a 4-day intensive cleft palate speech therapy training in Abuja, Nigeria, for 20 health-care professionals including 2 trained speech therapists, 5 surgeons, and other related professionals all connected to cleft palate teams from throughout Nigeria. This training, funded by Smile Train, was taught by a cleft palate SLP expert assisted by one graduate SLP student with developing expertise in cleft palate. Authors share data on the participants and effectiveness of the 5-day training in building capacity for cleft palate speech therapy including how the program was structured with both didactic teaching and clinical application with patients. Authors also share how challenges were identified and met including the need for post-training support through Whatsapp, the development of therapy materials in the local languages, and ongoing in-person mentoring. Finally, the authors provide insight on how to enhance the effectiveness of these trainings and to scale this approach using a train-the-trainer model.

## 28. Evaluating and Monitoring Speech Assessment Competencies for Speech-Language Pathologists New to the Evaluation of Cleft Palate Speech

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Background/Purpose: Speech-language pathologists (SLP) new to the perceptual evaluation of cleft palate speech require a coordinated approach to their education, training, clinical supervision and ultimately, the development and establishment of their clinical competence. The monitoring of these competencies also requires a mechanism where both the SLP and their managers can "check-off" a core competencies document and track required competency-based training. There is a need for a computerized solution to this monitoring which can be easily tailored to the individual SLP's needs, shared with the SLP, and also allow managers to evaluate the ever-changing progress of potentially more than one SLP's skills.

Methods/Description: A Microsoft Excel (Microsoft Corp, 2010) workbook was constructed to provide a computerized solution to the tracking and monitoring of SLPs' clinical competencies. The workbook was based on, and populated with, a draft set of clinical competencies for the perceptual assessment of cleft palate speech (Fitzsimons, 2014). Each SLP's worksheet within the workbook includes provision to record the SLP's progress against both standardized and customizable competencies. Information able to be added for each competency includes the specific criteria for each competency, the target and completion dates for achieving that competency, the staff member(s) responsible for facilitating the necessary training and education of that competency, and the status of each competency. The worksheet includes a summary section where the SLP and the manager(s) can see an "at-a-glance" overview of the completed, pending, and overdue competencies which forms the basis of a development plan for each SLP.

Results: This computerized workbook represents a standardized approach to the evaluation and monitoring of professional competencies for SLPs. The document utilizes a widely available computerized format (Microsoft Excel) and as such, the constituent data can be exported and shared in any digital form to facilitate cross-center benchmarking projects. The workbook can be tailored to the needs of both an individual SLP and/or a specific center/hospital and provides a framework to support a new SLP's skill development and professional competence.

Conclusions: A computerized solution to the tracking and monitoring of speech assessment competencies for speech-language pathologists new to the evaluation of cleft palate speech has been developed. The document will be available to ACPA members and feedback on the scope and inclusions of the document will be sought.

### 29. Factors Influencing Timely Preparation of Alveolar Bone Grafting: A Survey of the ACPA

Aleshia Pringle (1), Diana Jodeh (1), Taryl Crsip (1), S. Alex Rottgers (1) (1) Johns Hopkins All Children's Hospital, St. Petersburg, FL

Background/Purpose: Alveolar bone grafting is a vital part of cleft care. Factors most predictive of bone grafts success are the age at grafting and the presurgical orthodontic and dental preparation. In this study, we surveyed team coordinators from ACPA-certified cleft teams to identify factors that are commonly felt to impact a team's

ability to ensure patients are orthodontically and dentally prepared for alveolar bone grafting at the appropriate time.

*Methods/Description*: A 17-question survey was designed to investigate factors associated with team care, treatment protocol preferences, resources available to team coordinators, and access to care barriers. The survey was organized in Qualtrics software, and an e-mail was sent to cleft coordinators from each of the 167 teams identified by the ACPA. Coordinators were asked to complete the survey. Descriptive statistics and bivariate analyses including  $\chi^2$  and Student t tests were used.

Results: Forty-five of the 167 cleft coordinators certified by the ACPA completed the survey (response rate = 27%). Fifty-three percent of coordinators answered that more than 75% of their patients are orthodontically and dentally prepared for bone graft surgery. Most cleft teams' preferred age range for bone grafts is 8 to 13 years of age (48.89%) followed by 6 to 8 years of age (46.67%). Approximately half of the cleft teams have a designated case manager (55.56%), and the vast majority (82.22%) of cleft teams feel having a case manager increases the rate of timely bone grafts. Forty percent of coordinators answered that 50% to 75% of patients have governmental funding. Most common dental coverage model for cleft clinics was a combination of university/hospital employed and private practice dentists (33.33%). One-third of coordinators answered that 25% to 50% of patients seek care from a dentist who participates in the team. Most common orthodontic coverage model for team care was private practice volunteers (33.33%) and 42.22% of coordinators answered 50% to 75% of patients seek care from an orthodontist who participates in the team. Approximately half of the coordinators use the EMR to monitor pending and/or preparing bone graft patients. The presence of a designated case manager and the percentage of patients seeking care from an orthodontist or dentist that participates in the team were not significantly associated with the percentage of patients who are orthodontically and dentally prepared for bone graft surgery (P = .11; P =.42, and P = .32) respectively.

Conclusions: The majority of team coordinators felt that inclusion of a case manager in their team did or would improve compliance and timely preparation of patients for bone grafting. Our survey data demonstrated the employment of a case manager and a higher percentage patients seeking care from an orthodontist or dentist affiliated with the team were not predictive of improved rates of preparation in time for bone grafting. Significant recall bias concerning success of bone graft preparation may affect these results.

# 30. Identifying Three-Dimensional Facial Fluctuating Asymmetry in Normal Pediatric Individuals: A Panel Assessment Outcome Study of Clinicians and Observers

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Background/Purpose: From an ecology and evolutionary biology standpoint, perfect bilateral symmetry is defined as the optimal outcome of the development of bilateral traits in the absence of perturbations. For human facial anthropometrics, perfect bilateral symmetry almost never exists, because random variations in asymmetry, within limits, have been recognized as normal and are called facial fluctuating asymmetry. When facial asymmetry is clinically obvious (called

facial asymmetric deformity, which is more commonly directional rather than fluctuating), surgical or nonsurgical treatment may be required. Since the restoration of symmetry is the main goal of facial asymmetric deformity reconstruction, establishing facial fluctuating asymmetry by specific ethnic and age groups is crucial as it may be adopted as a target of treatment as well as applied as normative data for genetics, orthodontics, and surgical disciplines.

Methods/Description: This study measured 3-dimensional facial fluctuating asymmetry in 600 normal and healthy Taiwanese individuals (6-12 years old) and assessed the perceptions of increasing levels of facial fluctuating asymmetric severity by using a panel composed of 20 clinicians (surgical professionals), as well as 20 adult and 40 preadolescent observers.

Results: On average, this normal cohort presented a facial fluctuating asymmetry of  $0.96 \pm 0.52$  mm, with  $0.52 \pm 0.05$ ,  $0.67 \pm 0.09$ ,  $1.01 \pm 0.10$ , and  $1.71 \pm 0.36$  mm for levels I, II, III, and IV of severity, respectively. For all categories of raters, significant differences in the average symmetry–asymmetry scale values were observed, with level I < level II < level III = level IV (all P < .01, except for level III vs IV comparisons with P > .05). For level I, preadolescent observers presented a significantly (P < .05) higher symmetry–asymmetry scale value than adult observers, with no significant (all P > .05) differences for other comparisons. For overall facial asymmetry and levels II, III, and IV, no significant (all P > .05) differences were observed.

Conclusions: This study reveals that the normal pediatric face is asymmetric, and the panel assessment of facial fluctuating asymmetry was influenced by the level of severity and the category of raters and contributes to the literature by revealing that preadolescent raters present a similar or higher perception of facial asymmetry than adult raters.

#### 31. Interprofessional Education to Advance Interprofessional Practice

Kameron Beaulieu (1), Janet Brockman (2), Hannah Sanford-Keller (2) (1) IDD/Oregon Health & Science University, Portland, OR, (2) Oregon Health & Science University, Portland, OR

Background/Purpose: Interprofessional Education (IPE) is an activity that occurs when two or more professions learn about, from, and with each other to enable effective collaboration and improve outcomes for individuals and families. Similarly, Interprofessional Practice (IPP) occurs when multiple service providers from different professional backgrounds provide comprehensive health care or educational services by working with individuals and their families, caregivers, and communities to deliver the highest quality of care across settings (American Speech and Hearing Association, adapted from the World Health Organization). The parameters for Evaluation and Treatment of Patients with Cleft Lip/Palate or Other Craniofacial Differences states that "It is thus essential that all team members be trained and experienced in the care of patients with craniofacial differences. Each team must take responsibility for assuring that team members not only possess appropriate and current credentials but also have requisite experience in evaluation and treatment of patients with craniofacial differences. Teams should assist members in keeping current with their specialties by supporting and encouraging their participation in continuing education activities and attendance at professional meetings. This presentation will review collaborative efforts to create interprofessional education in cleft palate speech for allied health providers.

Methods/Description: Speech pathologists with advanced training in cleft can support cleft care through interprofessional education,

increasing knowledge of the community allied health provider. To address gaps in the education of allied health providers in cleft palate speech, and to support collaboration and communication between the craniofacial team speech pathologist and allied health providers, multiple pathways for adult learning have been developed across disciplines. This poster presentation will review collaborative efforts to create interprofessional, cleft specific, education, course content, and pathways for interprofessional education.

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#### 32. Intraprofessional Otolaryngology and Craniofacial SLP Collaboration: Improving Quality of Care

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Background/Purpose: The relationship between craniofacial anomalies and complex airways is well documented. There are also high incidences of feeding difficulties, communication impairments, and need for airway management in this population. As many as 20% of children with a craniofacial syndrome require a tracheostomy, which adds increased level of care. Multidisciplinary team management is the growing standard of care for both populations. While each team may include a Speech-Language Pathologist, they may not have a cohesive SLP team that cares for children with both airway and craniofacial anomalies. Given the complexity of patient needs and variability in practice patterns, the need for an integrated SLP team was identified. The purpose of this study was to evaluate the impact of a dedicated craniofacial and ENT SLP team on increasing utilization of therapy services in acute care, increasing use of one-way speaking valve for patients with a tracheostomy, increasing access to infant weight check visits, and improving family and community education.

Methods/Description: In 2017, the ENT and Craniofacial Speech-Language Pathology group was formed to address observed lapses in care for children with airway and craniofacial anomalies. Responsibilities, best practice procedures, clinic structure, and team rounding were established during the initial phase. A line of communication was developed between acute care and outpatient speech pathology teams to address needs upon admission through discharge to the specialty clinics. System-wide education was provided to all outpatient speech therapists to establish a continuum of care with standard practices. A retrospective review of 80 infants with cleft or craniofacial diagnoses and 92 new tracheostomy patients since 2017 were reviewed. Number of cleft and craniofacial patients referred for airway evaluation were documented, as well as time to clinic appointment from referral date, overlapping diagnoses of tracheostomy and/or airway diagnosis and craniofacial diagnosis, and number of patients with a language/feeding evaluation order during acute care stays.

Results: Through the initial 2 years of this program, we have seen an increase in therapy services during the acute care phase increased from 66% to 90%, one-way speaking valve use increased from 36% assessed for readiness to 100% assessed, improved staff and family

education, increased number of infant weight check clinic visits (increased by 36%), and greater collaboration within the community. Patients were often able to obtain same day airway or craniofacial evaluations if concerns arose during an ENT or craniofacial speech clinic visit.

Conclusions: A dedicated SLP team for children with airway and craniofacial anomalies positively impacts continuity of care, access to therapy, feeding outcomes, and staff education.

#### 33. Morphological Variations of the Levator Veli Palatini Muscle in Children With 22q11.2DS

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Background/Purpose: 22q11.2 deletion syndrome (22q11.2DS) is the most common genetic cause of velopharyngeal dysfunction (VPD) with a population prevalence of 1:2000 people. The primary muscle responsible for velar elevation during speech activities is the levator veli palatini (levator) muscle. MRI investigations on the velopharyngeal mechanism so far in the 22q population have focused on 2-dimensional measures and have not included information on 3-dimensional muscle shape and structure. No studies to date have the assessed muscle morphology, shape, and structure of the levator muscle in children with 22q11.2DS. The purpose of this study was to examine morphological variations of the levator muscle in children with 22q11.2DS in comparison to those in a nonsyndromic, noncleft cohort using MRI and advanced 3-dimensional computer technology.

Methods/Description: A total of 20 participants, 10 with a confirmed diagnosis of 22q11.2DS and 10 with normal velopharyngeal anatomy (age range: 4-12 years) completed the study. Children in the 22q11.2DS group were excluded if they had a history of prior palatal or velopharyngeal surgery. The control group was sex and age matched to the 22q11.2DS group to control for the effects of growth and sex on the variables. Participants were imaged using a high-resolution 3D anatomical scan called SPACE. All magnetic resonance images were transferred into Amira 6 Visualization Volume Modeling software. Quantitative and qualitative measures related to muscle volume and area were obtained. T tests were conducted to assess differences between the 2 groups. A P value of < .05 was considered to be statistically significant.

Results: Preliminary results indicated significant variations among the 22q11.2DS group compared to the matched controls. The total volume of the levator muscle was found to be significantly smaller in the 22q11.2DS group compared to the control group. The levator muscle area was also found to be significantly smaller in the 22q11.2DS group compared to the control group. Significant differences were also noted for major and minor axes measures in the halfway point between origin and midline insertion within the velum. The 22q11.2DS group had greater variability across the measures, as demonstrated by larger standard deviations.

Conclusions: This study aimed to use a morphologic approach to provide 3-dimensional data on the levator muscle in children with and without 22q11.2DS. Preliminary results from this study support our hypothesis that children with 22q11.2DS present with significant differences in levator muscle morphology. Given that the levator muscle is the primary muscle responsible for velar elevation during speech, specific anatomical parameters of this muscle that shed insight on structural and functional features would be beneficial in understanding the complex underlying pathogenesis of VPD in 22q11.2DS.

### 34. Novel Retinoic Acid—GATA3 Signaling Pathway Controls Development of Primitive Choanae

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Background/Purpose: In the previous study, we investigated the roles of retinoic acid (RA) signaling in the etiology of craniofacial defects including choanal atresia. From these results, we already identified number of factors that strongly correlate with RA signals in craniofacial development, Gata3 was one of the gene which exhibit significant reduction of expression in RA deficient mouse. Gata3 is well-known to play critical roles during embryogenesis. Indeed, Gata3 mutation in human also results in hypoparathyroidism, sensorineural deafness, renal anomaly (HDR) syndrome which associate with craniofacial anomalies. However, its detailed role for embryonic craniofacial development is still elusive and requires further investigation.

Methods/Description: In order to investigate the expression pattern of Gata3 in embryonic craniofacial development, we performed in situ hybridization and immunohistochemistry using an antisense probe and antibody which detects Gata3 during craniofacial development. Additionally, tamoxifen inducible Gata3 knock out mice was installed for functional assessment of Gata3 during craniofacial development. Morphological analysis was performed using both fluorescent nuclear imaging and micro CT. Histological sections were produced to analyze cellular morphology as well as behavior such as apoptosis and cell proliferation.

Results: Bioinformatic analysis using the dataset of RNAseq from Rdh10 mutant identified Gata3 as a strong etiological factor of choanal atresia. For this reason, we first assessed the expression pattern of Gata3 during craniofacial development. Strong expression of Gata3 could be observed in developing frontonasal process especially around the primitive choanae. Similar expression pattern could be observed by immunohistochemistry. The expression of Gata3 in craniofacial region was substantially reduced in Rdh10 mutant. Interestingly, when Gata3 was eliminated in the early embryonic stage (E9.5) using the conditional knock out mouse, deformation of choanal development could be observed. Furthermore, significant reduction of cell proliferation in nasal epithelium was detected in Gata3 mutant which could explain cytological mechanism of choanal deformation.

Conclusions: These results indicate important molecular crosstalk between RA and Gata3 signaling which is critical for normal cranio-facial development including important structures like primitive choanae.

## 35. Objective and Visual Evaluation of Speech for Patients With a Cleft Using the Noble Speech Visualization System

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(1) Kagoshima University Graduate School of Medical and Dental Science, Kagoshima, Japan, (2) Kumamoto University Graduate School of Science and Technology, Kumamoto, Japan Background/Purpose: There are various ways to evaluate speech after cleft palate (CP) repair, and the feedback system among them is said to be useful. The purpose of this study is to visualize the degree of hypernasality using the speech visualization system, and to visualize changes in articulation using the neural network system (NN) in patients with cleft palate.

Methods/Description: For hypernasality; 15 Japanese subjects were enrolled in this study, and they included 5 noncleft subjects, 5 CP patients with good velopharyngeal closure function (VPF), 5 CP patients with velopharyngeal closure dysfunction (VPD). All subjects pronounced Japanese vowel, /a/ /i/ /u/ /e/, and /o/. Their voices were converted into digital data and saved. For visualization, the first, second, and third formant-frequencies were converted into 3 primary color signals; red, green, and blue, and the phoneme characteristics of the vowel sounds were demonstrated as special colors. For Articulation; 8 patients who underwent closure of the fistula using the hinged-flap method were included. The articulation was analyzed using NN. This is a speech feature estimation system, which can objectively show the place and manner of articulation. Over time, we analyzed changes in articulation after surgery, and compared these with healthy children.

Results: For hypernasality; /a/, /i/, /u/, /e/, and /o/ were visualized in different colors. Noncleft subjects and CP patients with good VPF demonstrated almost the same color patterns for the 5 vowels. However, CP patients with VPD showed a different color pattern, shifting to more bluish. The second formant of CP patients with VPD was different compared to the other subjects. For articulation, there were 7 patients who exhibited backed to velar; however, after training this disappeared. In the analysis utilizing NN, the place of articulation returned to its normal place after training.

Conclusions: From analysis of the vowel's color patterns and changes in articulation, speech visualization system and NN has been suggested to be useful as an evaluation method of the cleft palate speech. These systems made it possible to perform a more objective evaluation, its were more effective in confirming the benefit of the training delivered by the therapist and it also provided visual feedback to the patient.

### 36. Outcomes of Cranioplasty Strategies for High-Risk Complex Cranial Defects: A 10-year Review

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Background/Purpose: A variety of studies have independently investigated primary calvarial reconstruction, but few have examined high risk complex cranial defects. Up to 33% of patients who undergo reconstruction have hostile preexisting conditions with coexisting soft tissue and osseous defects due to history of radiation, prior failed cranioplasty, or concurrent infections. Though studies have evaluated autologous reconstruction for hostile craniums, the use of alloplastic reconstruction alongside autologous reconstruction for hostile defects has yet to be evaluated. Our objective was to retrospectively evaluate the use of alloplastic reconstruction alongside autologous reconstruction for hostile cranial defects.

Methods/Description: A retrospective review of patients who underwent cranioplasty of a hostile site at the University of Alabama—Birmingham between January 2008 and December 2018 was performed. The patients were stratified into 3 groups based on the type of implant used: autogenous (bone), alloplastic (Osteomatch-PEEK, Titanium, PMMA), or mixed (combination of both types of graft).

There were 43 total cases in this time period; 15 autogenous, 23 alloplastic, and 5 mixed. The primary outcome metric was a complication in the year following cranioplasty, identified by flap or bone graft failure, necrosis, or infection. Statistical analysis included t tests and  $\chi^2$  tests where appropriate using SPSS.

Results: The most common type of hostile cranial site resulted from prior or concurrent infection of the scalp (35%). Between the groups, there was no significant difference in history of prior failed cranioplasty, open wound, radiation, infection, or CSF leak. The purely autogenous group had the highest complication rate (85%) and the alloplastic group had the lowest complication rate (38%). When stratified by specific material used for reconstruction (15 bone, 14 PEEK, 10 titanium, and 5 PMMA), overall complication rate was statistically significant (P = .009;  $\chi^2$  test) with PEEK implants having the lowest complication rate (21%). The complications ranged from hematoma and infection, hardware malfunction, CSF leak, or partial flap necrosis (16.3%). The analysis documented an overall complication rate that was statistically different between the 3 groups (P = .012). No difference was seen in the rate of complication between left right or bihemispheric cranioplasties, the hostile setting, the etiology of craniectomy, the size of the defect, comorbid conditions, or harvest location. Multivariate analysis showed that the only factor independently associated with a risk of complication following cranioplasty of a hostile defect was the type of material used in cranioplasty (P = .021).

Conclusions: This analysis interestingly found that in the setting of hostile cranial defects, cranioplasties would benefit from the use of prosthetic implants instead of autologous bone grafts, not only for avoidance of donor-site morbidity but also for decrease in overall complications.

### 37. Outcomes of Modified Nasoalveolar Molding in Patients With Complete Unilateral Cleft Lip and Palate

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Background/Purpose: Nasoalveolar molding (NAM) is a presurgical treatment that is clinically used to lessen the severity of the cleft deformity in order to facilitate primary surgical repair. Denture adhesive has been utilized to secure and improve retention of the NAM appliance. However, it could contribute to potential adverse effects including cytotoxicity, irritation, and infection. This prospective cohort study was conducted to evaluate the outcomes in patients with complete unilateral cleft lip and palate (UCLP) after employing modified NAM without denture adhesive.

Methods/Description: Eight patients with UCLP who received modified NAM at Queen Sirikit National Institute of Child Health, Bangkok, Thailand were included in this study. Standard basilar view photographs were taken and dental models were created. The measurements of nostril height, nostril width, columella angle, nasal base angle, cleft width, palatal arch length, and palatal arch width were obtained. Statistical analyses were used to compare the differences between pre- and posttherapy measurements.

Results: After the modified NAM therapy, there were statistically significant increases in nostril height and columella angle with the differences of pre- and posttherapy measurements of 2.38 mm and 36.30 degree (P < .05), respectively. Furthermore, there were statistically significant decreases in nostril width, cleft width, and palatal arch length with the differences between pre- and posttherapy measurements of 3.27 mm, 7.74 mm, and 1.73 mm (P < .05), respectively.

Conclusions: It is promising that the modified NAM without denture adhesive method could be performed effectively as favorable outcomes of this modified technique were achieved in our study.

#### 38. Palatal Fascia Reconstruction Using Buccinator Myomucosal Flap (BMMF) for Palatal Lengthening: A New Concept in Primary Cleft Palate Repair

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Background/Purpose: Cleft palate is one of the common congenital anomalies affecting 1/1000 live birth whether isolated or associated with cleft lip. There are various techniques developed along medical history for the treatment of cleft palate. All of the operations aimed for restoration of the anatomical and functional reconstruction of the palatal muscles. In this work, we present our nouvelle addition of reforming the palatal fascia (tensor palati fascia) using buccinator myomucosal flap as an addition for the primary cleft palate repair.

Methods/Description: Sixty patients with unilateral complete cleft lip and palate and isolated cleft palate were operated upon by single surgeon during the period May 2016 to May 2018. Age at time repair ranged from 8 to 13 months. The technique used was modified Furlow without lateral release incision. The muscles were reoriented and posteriorly placed in post half of the palate. The buccinator myomucosal flap was raised on the left side and added to the oral layer closure. Preoperative and immediate postoperative measurements of palatal length was taken from the bony edge of the palate till the base of the uvula. The patients were followed up for 12 months postoperatively. Results: Postoperative palatal length was increased by 20% to 45%

Results: Postoperative palatal length was increased by 20% to 45% from preoperative measurement with mean of 33%. We had one case of dehiscence due to severe postoperative infection and 4 cases of early postoperative fistulae two of them healed spontaneously and needed no further management.

Conclusions: Palatal fascia reconstruction is a new concept for restoring cleft palate length and function as it provides not only physical lengthening of the palatal musculature but also a soft pliable attachment to the hard palate which aloe free mobility later on. The addition of the flap diminishes the raw areas that might result from classic Furlow; furthermore, it alleviates the need for lateral release incision which might affect growth. This is a preliminary result of the technique which need further assessment in the years for palatal mobility, velopharyngeal function, and midface growth.

### 39. Postoperative Velopharyngeal Function After Newly Modified Velopharyngoplasty

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Background/Purpose: Our department, Oral-Facial Disorders, is responsible for pharyngeal flap operations to improve velopharyngeal function and also to evaluate pre- and postoperative speech function. We have newly adopted a unified velopharyngoplasty combined with Furlow's method for soft palate treatment since 2008. The purpose of this study was to examine the postoperative results on velopharyngeal function after utilizing our new operation methods.

Methods/Description: Forty-three speakers with cleft palate underwent newly modified velopharyngoplasty in our department between 2008 and 2018. Two subjects who would not receive the evaluation

were excluded. Pre- and postvelopharyngeal motion during speech and blowing were evaluated using a nasopharyngoscope and classified into 3 states: incompetence, borderline incompetence, and competence. Nasometry was also performed to evaluate the nasalance score of a standard sentence, a low-pressure sentence, and a high-pressure sentence pre- and postoperation. We compared velopharyngeal motion and nasalance score between pre- and postoperation.

Results: On preoperative endoscopic findings, 75% of subjects showed velopharyngeal incompetence in all tasks, and 25% of subjects demonstrated incompetence in speech but borderline incompetence or competence in blowing. On postoperative findings, 73% of subjects showed velopharyngeal competence in all tasks. The remaining subjects did not demonstrate incompetence but borderline closure with fair speech intelligibility. The averages of the pre- and postoperative nasalance score were 47.7% and 17.0% for the standard sentence, 40.9% and 17.1% for the low-pressure sentence, and 52.6% and 18.4% for the high-pressure sentence, respectively. There was a significant difference between the pre- and postoperative nasalance scores for all 3 sentences (Mann-Whitney U test, P < .001).

Conclusions: These results indicated that our new operation method, unified velopharyngoplasty combined with Furlow's method, could achieve fair to excellent velopharyngeal function postoperatively.

## 40. Pre-Surgical Orthodontics in Individuals With CLP Submitted to Orthognathic Surgery. How Long Does It Take? Preliminary Results

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Background/Purpose: Surgical orthodontic treatment time is expected to be more extensive for individuals with CLP. Those individuals have inherently different structures that may require a strategic approach compared with noncleft maxillary deficiency with Class III malocclusion. The global time spent for noncleft individuals submitted to surgical orthodontic treatment is between 23 and 29 months, and the median duration for the presurgical treatment is 17 months. The length of orthodontic treatment for individuals with CLP were assessed by Semb (2005), and the mean length ranged from 3.3 to 8.5 years. There was no data about the presurgical treatment for individuals with CLP. This work aimed to assess the time spent for orthodontic preparation for orthognathic surgery in individuals with CLP as a primary end point. Secondarily, the study aimed to provide a better understanding of factors as the presence of alveolar bone graft, cleft affecting the alveolar ridge (classification, type), and the geographical factors between individuals from other states and local individuals.

Methods/Description: A sample of 169 individuals was collected, based on data from the orthodontic records of patients who had undergone orthognathic surgery from 2013 to 2018. Statistical analyses were used to evaluate the time of preparation for orthognathic surgery and the influence of alveolar graft, type of cleft, and the distance between the city of origin of patients and the center where the treatment was conducted, on the time spent for presurgical treatment.

Results: A mean value of 9.34 years was found for the presurgical orthodontic treatment; the time spent from treatment onset until surgery was 9.73 years; and 3.14 years between the day of surgery request and the day of surgery. The alveolar ridge was affected in 86.04% of individuals. Inferential statistics were applied to evaluate the time spent for the presurgical orthodontic treatment in a distribution of

individuals who had undergone alveolar graft before surgery, and individuals who did not have an alveolar graft. No statistically significant differences were found between these groups. Thus, it is important to describe a clinically significant difference of 1.36 years faster for the group of individuals who had done the alveolar graft before surgery. In this sample, 44.58% of individuals needed to travel from other states with longer distances than local individuals, yet no statistically significant differences were found between these groups for the time of treatment.

Conclusions: The time spent for the presurgical orthodontic treatment is more extensive for individuals with CLP. Even though no statistically significant differences were found between the presurgical time and the presence of alveolar bone graft, more research is necessary to better understand this variable. More studies might be developed to better understand the global factors and contextual characteristics that could influence the time spent for orthodontic preparation for orthognathic surgery.

### 41. Prenatal Team Visit for Cleft Lip/Palate: Results of a Parent Survey

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Background/Purpose: At the 20 week ultrasound, parents may learn of the prenatal diagnosis of cleft lip and possible cleft palate. For those with no family history of a cleft, this can cause distress and confusion, diminish the joy of the pregnancy, and raise concerns of other birth anomalies. Worried parents go to the Internet for information, which adds to their unease. An "in person" prenatal consultation with members of the cleft team aims to support parents faced with this new diagnosis by providing valuable information about having a baby with cleft lip and palate. The prenatal consult sets the stage for parents to have a strong connection with the cleft team, which is needed for continuity of care as the child grows. A short 5 question survey was administered at the beginning and end of the prenatal consult to evaluate the effectiveness of the visit to prepare parents prior to the birth of their baby.

Methods/Description: As part of the prenatal visit, parents met with the cleft surgeon, speech pathologist/feeding specialist, and the team social worker. A short 5 question survey asking parent comfort with feeding, postnatal care, surgical plan, cleft team involvement, and anxiety was administered at the beginning and again at the end of the consult. Survey results before and after the consult were compared. The prenatal visit included a discussion of feeding difficulties and demonstration of a specialty feeding bottle, meeting team members and learning their roles, describing the treatment plan, and introducing the ACPA's information for families.

Results: During a 6-month period, 15 sets of parents who came to a prenatal visit with the cleft team participated in a 5 question pre and postsurvey. Preliminary findings indicated that the prenatal cleft team visit was an effective means to prepare parents and eased early feeding concerns, connected family with the cleft team, and helped lower anxiety as they understood that a team of professionals was available to answer their questions. Survey results showed that scores increased from presurvey to postsurvey, indicating a higher level of confidence that they could meet the needs of their child born with a cleft lip and palate.

Conclusions: The prenatal visit reassures parents that there will be a team of professionals to answer their questions and ensure that their baby has healthy growth and development. The results of this qualitative survey reinforced the commitment of the cleft team regarding

the format in place for prenatal consultation and education. Follow-up over time showed that the majority of parents continued their care with the cleft team following the child's birth. Parents were more familiar with the feeding concerns and understood milestones in the first year of care. They were proactive in advocating for their child's care, even if the birth took place at a hospital that was distant from the cleft team location.

# 42. Proposal of References Points to Determine if the Position of the Jaws Generates Harmful Forces in the Temporomandibular Joint in an Orthognathic Surgery Patient

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Background/Purpose: For several years, digital tools have been a way of making diagnosis and planning in patients with an indication of orthognathic surgery. However, most of these programs have high costs are difficult to use and do not predict postsurgical damage to the temporomandibular joint (TMJ) as a result of the new mandibular positions.

Methods/Description: We randomly selected 120 patients with an indication of orthognathic surgery (63 men and 57 women, with an average age of 23.8 years) who were taken lateral radiographs of the skull and a cone-beam scanner of the TMJ. It is worth mentioning that this is a representative sample of patients from the Santiago of Chile MR. On the lateral radiographs of the skull, a vector was drawn from the most superior point of the mandibular condylar to the mandibular angle, called "vector a." The second vector was drawn from the mandibular angle to the most posterior dental contact point, called "vector b" and then, these 2 vectors were added geometrically generating the vector we call "vector c." For convenience, the vector "module c" and the vector "module b" were divided, and the result is called the "coefficient v." At the same time, with the information of the cone beam scanner of the TMJ were classified according the morphological characteristics of the condyles proposed by Koyama (2007), assigning a category to each patient, to associate the "coefficient v" at different stages of joint damage. The data obtained are carried out the Wilcox statistical test for dependent variables with the "R" software.

Results: The results delivered by the Wilcoxon statistical analysis method showed a consistent association between the "v" coefficient and the different levels of joint damage. Therefore, for each type of Koyama classifications, it groups similar values of "coefficient v." This would relate a mechanical condition (consequences of maxillary position) with levels of TMJ damage.

Conclusions: Our study demonstrates that the calculation of the "v coefficient" is not only an easier, fast, and more cheaper tool than current software's, but also allows screening early biomechanical condition and prevent damage to the TMJ after orthognathic surgery.

#### 43. Qualitative Analysis of Early Literacy Engagement Among Latina Mothers of a Child With a Cleft

Alexis Johns (1), Cindy Ola (2), Erin Wallace (2), Jenny Thach (2), Emily Gallagher (2), Yvonne Gutierrez (1), Brent Collett (2)

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Background/Purpose: Children born with a cleft are at risk for difficulties in their language-based skills, including reading. Several factors are posited to impact their literacy, ranging from variability in hearing and speech to differences in brain development. Sociodemographics are linked to reading outcomes and can further increase the risk for children with a cleft. Early familial engagement in language and literacy activities may reduce the risk for reading deficits. A greater understanding of early family literacy environments, particularly among families with socioeconomic risk factors, can assist in designing strategies to promote early literacy for children with a cleft. Methods/Description: Two structured focus groups and an interview (mean 64 minutes) were held in English or Spanish with 7 mothers (mean age 31 years) of children ages 13 to 42 months with a nonsyndromic cleft palate only (CP) or cleft lip and palate (CLP). To target underrepresented higher-risk families, having public insurance and/or being Spanish-speaking were also inclusion criteria. Children were 33 months old on average with CLP (57%) or CP (43%). Spanish was the first language for all but one mother, 57% immigrated to the US a mean of 13 years ago, and 57% had less than a high school education. Focus groups topics for this presentation included the value of early literacy, parental childhood experiences with reading, home reading practices, and challenges in promoting reading. Transcribed and translated content was analyzed in an inductive thematic approach. Themes included high parental value placed on reading and motivation to improve upon their own childhood reading experiences, which were described as minimal until starting school. They discussed goals of bilingualism with challenges in balancing both languages in the home. Mothers conceptualized reading activities broadly, including parentchild reading and library visits as well as shared reading of online content and reading with siblings. Mothers expressed interest in learning more strategies to promote child literacy. Challenges included managing children's frustration/inattention, finding Spanish/bilingual books, low parental literacy, limited parental time, uncertainty of how to teach reading, and difficulty accessing intervention services. Parents also discussed the larger context of having a child with a cleft, including health-care burdens and social concerns. In conclusion, mothers were motivated and placed high value on improving child literacy. They described a range of reading activities and challenges. We are using these data to inform caregiver-focused literacy interventions that can be introduced during craniofacial team visits. Strategies may include building child motivation, interactive reading, behavioral strategies, bilingual teaching, advocacy for services, using online resources, and establishing family reading habits.

## 44. Reliability of Caregiver Reporting and Agreement on a New Measure of Infant Cleft Observer Outcomes (ICOO)

Salene Jones (1), Meredith Albert (2), Cassandra Aspinall (3), Claudia Crilly Bellucci (4), Carrie Heike (3), Kathy Kapp-Simon (2), Brian Leroux (5), Donald Patrick (5), Janine Rosenberg (6), Laura Stueckle (3), Todd Edwards (5)

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Background/Purpose: The goal of this study (NIDCR #R01-DE024986) is to develop and validate an outcomes instrument that assesses observations (eg, of clinical signs) that can be reliably reported by caregivers. This article examined the test–retest and inter-rater agreement between caregivers of infants with cleft lip with

or without cleft palate on a new measure, the Infant Cleft Observer Outcomes (ICOO) instrument.

Methods/Description: Caregivers and their infants (n = 118) were enrolled prior to primary lip repair. Caregivers completed a 69-item observer-reported outcome measure daily for 7 consecutive days. Caregivers reported on the following 10 domains for their infants: breathing; behavioral/emotional; communication; comfort; ear problems; feeding; hearing; facial skin and mouth; sleep; and vocalization. Items were also categorized as problems such as having dry skin or as positive signs if content reflected a desirable behavior like cooing. All domains were represented in the problems category except communication and vocalization. All domains were represented in the positive signs except breathing and ear problems. A third category of 10 items reflected general parental impressions in each domain. When a second caregiver was available and willing to participate, data were collected from both family caregivers (n = 47) to assess caregiver agreement before surgery, 2 days after surgery, and 2 months after surgery. To assess test-retest reliability (consistency of a caregiver with themselves), a subset of primary caregivers (n = 41) and secondary caregivers (n = 19) who completed surveys 2 weeks apart. Test–retest and inter-rater reliability were estimated using the intraclass correlation coefficient (ICC). Group level ICCs are recommended to exceed 0.70 to indicate satisfactory reliability.

Results: Test–retest reliability was high for most items (ICC > 0.7 for 59 of 69 items for primary caregiver and for 53 items for secondary caregivers). However, inter-rater reliability was only low to moderate for most items (15/69 item-level ICCs over 0.70 before surgery, 24/69 ICCs over 0.70 2 days after surgery, and 31/69 ICCs over 0.70 2 months after surgery). Agreement between caregivers was higher for items assessing problems (mean ICC = 0.58) and general impressions (mean ICC = 0.55) than for items assessing presence of positive signs (mean ICC = 0.39). Agreement between caregivers was consistently low for hearing (mean ICC = 0.26) and behavior/emotion (mean ICC = 0.43) and consistently high for feeding (mean ICC = 0.68). Across domains, which caregiver reported more problems or positive signs varied.

Conclusions: Caregivers were consistent over time in assessing their infants but disagreed with one another for most items. Amount of time spent with the infant as well as rater bias might explain these results. Health-care providers should be aware of differing perspectives between caregivers. When caregivers disagree, health-care providers may need to investigate reasons for the discrepancy or query the same caregiver over time.

### 45. Risk Factors Associated With Developmental Risk on the Ages and Stages-3 for Infants With Cleft Lip With or Without Cleft Palate

Meredith Albert (1), Todd Edwards (2), Janine Rosenberg (3), Laura Stueckle (4), Claudia Crilly Bellucci (5), Brian Leroux (2), Salene Jones (6), Carrie Heike (4), Kathy Kapp-Simon (1)

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*Background/Purpose*: The Ages and Stages Questionnaire (ASQ-3) is a parent-report screening measure used to assess infant development; however, there is little data on use of the ASQ-3 with infants with CL+P. This presentation will discuss risk factors associated with low ASQ-3 scores.

Methods/Description: Families (n = 94) of infants born with CL±P (65 male, 69 CLP) were recruited from 1 of 3 national craniofacial clinics or through online groups for parents. The ASQ-3 was completed prior to lip repair surgery as part of a larger study on caregiver perceptions of infant health and well-being. The ASQ-3 is a reliable and valid parent-report measure that screens infant development across 5 domains: Communication, Gross Motor, Fine Motor, Problem Solving, and Personal-Social. An ASQ-3 score 2 SD below the normative mean is considered "below the cutoff" and suggests need for additional assessment and possible intervention. Information on risk factors were obtained through interview and included: diagnosis, prematurity, additional major medical concerns (eg, cardiac), race/ethnicity, SES, and family history.

Results: Mean infant age at assessment was 5.3 months (SD = 2.5, range = 1 to 14 months.); 12% were premature; 33% spent time in the ICU; 13% had additional medical risks; 12% had an immediate family member with clefting; 64% were white, not Hispanic; 6% of caregivers had less than a high school education, and 17% had a household income under \$35 000. Rates of individual domain scores below the cutoff were as follows: Communication, 19%; Gross Motor, 21%; Fine Motor, 25%; Problem Solving, 20%; and Personal Social, 20%. Forty-nine percent of the infants had at least one ASQ-3 domain score below the cutoff; 26% had two or more scores below the cutoff. Of these 26%, 40% were between 3 and 5.5 months of age, while 24% were over 7 months of age. Infants with CLP were more likely to score below the cutoff (P = .036) in two or more domains when compared to infants with CLO. Males were more likely than females to score below the cutoff in all domains. No other risk factors were associated with developmental risk individually or cumulatively.

Conclusions: A significant number of infants with  $CL \pm P$  scored in the clinical range on the ASQ-3. Males and infants with CLP were at greater risk. Prematurity, presence of additional medical risks, family SES, and ethnicity were not associated with increased risk. Infants screened at risk require close monitoring, strategies for home interventions by caregivers across identified risk areas, and possible referral to an infant intervention program.

### 46. The Development of Standard American English and Spanish Phrases for Assessing Speech in Preschoolers With Cleft Lip and Palate

Kelly Cordero (1), Chelsea Sommer (2), Caitlin Cummings (3), Kathy Chapman (2), Judith Trost-Cardamone (4), Adriane Baylis (3), Angela Dixon (5), Kristina Wilson (6), Cindy Dobbelsteyn (7)

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Background/Purpose: When assessing the speech of children with cleft lip and palate (CLP), it is important to use stimuli that are in their native language and that are age-appropriate. Currently in the United States (US), many craniofacial teams use the standard American English sentences from the Cleft Audit Protocol for Speech-Augmented-Americleft Modification (CAPS-A-AM; Chapman et al., 2016). These sentences, created by Trost-Cardamone (2012) follow recommended phonological guidelines that allow speech-language pathologists (SLPs) to accurately sample and differentially diagnose articulation and resonance disorders (Brondsted et al., 1994; Henningsson et al., 2008). There is also a Spanish sentence set which was created following these same

parameters. This set originated from Colombia and is used in a variety of treatment areas with Spanish speaking patients (Cleves et al., 2009). These sentences were designed for school age children and can be difficult to use with preschool age children. Therefore, adaptations of the CAPS-A-AM English sentences and Spanish sentences were needed in order to improve applicability to younger children with CLP in the United States. This poster will discuss the process of adapting the sentences and creating new shorter phrases in English and Spanish. Presenters will also discuss the clinical implications of conducting and interpreting findings from cross-linguistic articulation and resonance assessments.

Methods/Description: This poster will explain the process of developing the preschool English and Spanish standard phrases for use with the CAPS-A-AM assessment protocol. We will describe how we utilized the "universal parameters" set forth by Henningsson et al. (2008) for creating these new stimuli. These parameters incorporate including high and low vowels, using only one target sound per sentence, and no nasal sounds in sentences that target high pressure sounds. The parameters also indicate that these sentences should contain monosyllabic words with consonant-vowel or consonantvowel-consonant structure. The Spanish phrases were adapted and created by CLP-trained, bilingual English and Spanish-speaking SLPs. This poster will address the uses of the English and Spanish phrases with bilingual patients both clinically and in a study of early speech outcomes for children with CLP funded by the NIH. This poster will include a comparison of shared and unshared phonemes across the languages for a more accurate description of skill set compared to a single language assessment.

#### 47. Two Cases of Oral Synechiae With Cleft Palate

Yuumi Lee (1), Koichi Ueda (1)

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*Background/Purpose*: Oral synechiae is a fibrous or soft tissue adhesion between maxilla and the mandible. The presence of oral synechia along with cleft palate is a rare. We report 2 cases of oral synechia with cleft palate and describe pathological results.

*Methods/Description*: Our cases restricted mouth opening due to the presence of band adhesion; 37-day-old girl forced feeding problem and 8-day-old boy was obstructed to intubate. Therefore, we operated surgical excision with local anesthesia or no anesthesia.

Results: Case 1 had restricted mouth opening due to the presence of 2 band adhesion between the floor of the mouth and the free margin of the cleft palate. The infant faced feeding problems due to bad general condition, and therefore a surgical excision of the synechia was decided on. After the surgical excision, the distance of mouth opening was improved in 15 mm from 10 mm and allowed normal feeding. Case 2 had restricted mouth opening due to the presence of a band adhesion between the floor of mouth on the right side and the free margin of the cleft palate. At 8 days after birth, the infant had an operation of pulmonary stenosis and atrial septal defect. We expected that limited mouth opening made intubation difficult, therefore operated surgical excision. Mouth opening improved in 16 mm from 12 mm and intubation was operated safety.

Conclusions: Oral synechiae with cleft palate is a rare anomaly. Limited mouth opening is serious complications for airway management and feeding and may lead to subsequent growth abnormalities.

#### 48. Validation of a Novel Grading System for Unilateral and Bilateral Cleft Lip Outcomes

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Background/Purpose: International cleft initiatives require a mechanism for assessing whether a surgeon is performing successful operations. When determining whether a cleft lip repair outcome is acceptable, the severity of a patient's cleft at initial presentation must be considered. At present, there is not a widely agreed upon scale for grading the preoperative severity or postoperative surgical outcome for patients with unilateral and/or bilateral cleft lip. This study presents the Hubli grading system, which uses 5 categories to generate separate pre- and postoperative ratings, and then combines these ratings to produce one scaled measure of overall cleft repair acceptability. The aim of this study is to validate the Hubli grading system as a reliable and reproducible system for characterizing both unilateral and bilateral cleft lip repair outcomes.

Methods/Description: Nine craniofacial surgeons from a variety of countries independently evaluated 2489 patients undergoing both unilateral and bilateral cleft lip repair by Smile Train partner surgeons from April 2004 to December 2018. Preoperative severity and post-operative surgical result were assessed, and these numbers were then multiplied to determine whether a surgical outcome was acceptable based on initial cleft severity. Intra-rater and inter-rater reliability were calculated as percentages of agreement.

Results: Intra-rater and inter-rater acceptability scores had an agreement of 94.50% and 87.04%, respectively. These results indicate that using the Hubli grading system, raters are able to reliably and reproducibly grade the acceptability of a cleft lip repair based on initial presentation of cleft severity and postsurgical result. Notably, 94% of surgical outcomes were rated as "acceptable," suggesting that the vast majority of cleft lip repairs performed by Smile Train partner surgeons have a successful surgical outcome.

Conclusions: This study validates the use of the Hubli grading system to evaluate the acceptability of both unilateral and bilateral cleft lip repair surgeries. To date, this study is the largest of its kind to evaluate cleft lip severity and repair outcomes, and the novel Hubli grading system is the only scale to account for the acceptability of surgical outcomes based on initial cleft severity. This system is best designed to identify surgeons with suboptimal outcomes in order to implement timely, individualized training on a global scale.

## 49. The Double Opposing Z Plasty +/- Buccal Flap Repair of Cleft Palate: A 29-Year Review of Complications

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Background/Purpose: The Double Opposing Z- plasty +/- Buccal Flap Approach (DOZP+/-BFA) is a proven alternative to traditional palate repairs. A published article has demonstrated that using the DOZP+/-BFA achieves excellent speech outcomes regardless of the patients cleft width or classification. However some surgeons are slow to change from traditional approaches fearing the possibility that severe complications will frequently occur when using a more anatomic reconstruction where outside tissue is added to the surgical plan.

The purpose of this presentation is to review the complications seen during the first 29 years using the DOZP+/-BFA.

*Methods/Description*: This study was a retrospective review of complications and care management for 505 patients over a 29 year time frame. Measures include but were not limited to Veau classification, postoperative trauma, syndrome status, Pierre Robin, fistula type, and additional surgeries required.

Results: Of the 505 patient cohort there were 32 complications and 30 fistulas. For nonsyndromic, non-Pierre Robin patients (N = 333) there were 16 fistulas. The overall fistula rate was 4.8%, however fistulas > 2 mm comprised only 1.8%. There was 1.2% postoperative trauma rate. The number of fistulas requiring more than one surgery was zero. The number of fistulas requiring outside flaps was also zero. The number of nightmare complications was zero. One syndromic patient required 2 closures. All fistulas were closed with local tissue rearrangement. No external flaps required for repairs. Secondary speech surgeries were required more often for patients with fistulas than for patients without fistulas. However, the final speech resonance scores for patients with fistulas were similar to the scores achieved in patients without fistulas.

Conclusion: The DOZP+/-BFA is a safe approach with few complications. The types of fistulas seen over 29 years indicates that the majority of the fistulas seen are of the small nuisance variety and are easily managed with low impact on the patients. The fact that zero fistulas occurred requiring outside flaps demonstrates that DOZP+/-BFA patients are at low risk of experiencing the morbidity of multiple surgeries. Additionally, all fistulas had limited effect on long term speech outcomes.

#### 50. Withdrawn

### 51. Caregiver Report of Sleep and Breathing in a Multinational Cohort of Infants and Children With Craniofacial Microsomia

Kelly Evans (1), Amelia Drake (2), Milagros Duenas-Roque (3), Carrie Heike (1), Paula Hurtado-Villa (4), Alexis Johns (5), Leanne Magee (6), Harry Pachajoa (7), Liliana Porras (8), Zarante Ignacio (9), Daniela Luquetti (1)

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Background/Purpose: The presence of craniofacial anomalies is a well-recognized risk factor for obstructive sleep apnea (OSA). Untreated pediatric OSA is associated with neurodevelopmental sequelae, including learning and behavioral problems. Parent reported snoring is common among children and adolescents with craniofacial microsomia (CFM); however, studies have been limited by small samples and insufficient phenotypic characterization. While CFM is a condition with a wide range of severity and abnormal airway morphology, the association between specific CFM characteristics and the degree of breathing problems remains unclear. We hypothesized that sleep and breathing problems are prevalent in children with CFM and specific phenotypic subgroups within CFM have a heightened risk for OSA.

Methods/Description: Participants with CFM (isolated microtia included) were prospectively recruited from 9 craniofacial centers through the CAUSE study, an active NIH-funded study. Data

collected through caregiver interviews and medical record review included phenotypic characteristics, medical diagnoses, surgical interventions, academic, and demographic data. Sleep was assessed using the parent reported Pediatric Sleep Questionnaire (PSQ) for children 1 year or older. A new Infant Sleep Questionnaire (ISQ), modified from the PSQ, was administered to infants less than 1 year. PSQ and ISQ data were summarized. Higher sleep-related breathing disorder (SRBD) scores equate to a higher risk for a sleep-related breathing disorder. SRBD scores were evaluated by clinical subgroups hypothesized to have a higher risk of airway obstruction. Wilcoxon rank-sum tests were used for all comparisons.

Results: The cohort consisted of 170 participants recruited in the United States (49%) and South America (51%). The average age at ascertainment was 7 years (SD: 6 years) and 60% were male. Complete sleep questionnaire data was obtained from 84% of the study population (ISQ n = 24, PSQ n = 119). Among children <1 year of age, 13% snored more than 50% of the time, 8% had heavy or loud breathing, and 8% turned blue during a feed. Among children 1 year and older, 16% snored more than 50% of the time and 23% had heavy or loud breathing. Hyperactivity and inattentiveness were present in up to 33%. The average SRBD scores were overall normal (mean: 0.2, SD: 0.2), however varied by clinical subgroups. SRBD scores were significantly higher among those with unilateral or bilateral mandibular hypoplasia (P = 0.04), cleft palate (P = 0.04), and among those who had undergone prior surgical airway intervention (P = 0.03). SRBD scores were not associated with academic performance.

Conclusions: Although the overall risk for OSA is low in this cohort, evidence of sleep disordered breathing was associated with mandible and cleft involvement and persists after surgical airway treatments. Further investigations of who is at highest risk for OSA and may benefit from overnight sleep studies and interventions to reduce sleep-related sequelae are needed.

# 52. Comparison of Speech Outcomes and Velopharyngeal Insufficiency Following Maxillary Distraction and Conventional Le Fort I Maxillary Advancement in Patients With Cleft Lip and Palate

Amanda Gerlach (1), Matthew Ford (2), Jesse Goldstein (3), Cleo Yi (4), Bernard Costello (2), Ian Chow (5), Lindsay Schuster (1)

(1) Children's Hospital of Pittsburgh, PA, (2) Children's Hospital of Pittsburgh of UPMC, PA, (3) University of Pittsburgh School of Medicine, PA, (4) University of Pittsburgh, School of Dental Medicine, Orthodontic Residency Program, PA, (5) University of Pittsburgh, PA

Background/Purpose: This investigation evaluates velopharyngeal function and articulation differences in patients with clefts who have undergone LeFort 1 advancement. Patients with cleft lip and palate often have midface hypoplasia with malocclusion which may result in articulation changes obligatory to malocclusion. Surgical maxillary advancement may worsen velopharyngeal function, with previous reports citing rates of VPI of 0% to 84%. Obligatory articulation changes have been observed to resolve or improve following correction of malocclusion. The data presented will aid in informed consent discussions surrounding orthognathic surgery and speech changes.

Methods/Description: This retrospective review evaluates speech parameters in patients with a history of complete cleft lip and palate or palatal cleft who had conventional LeFort 1 maxillary advancement or maxillary distraction osteogenic advancement. Pre- and postoperative speech measures include the Pittsburgh Weighted Speech Score (PWSS) to assess for symptoms of velopharyngeal incompetence (VPI) and formalized speech evaluation with clinical notation of

articulation patterns. Articulation was categorized as: (1) normal, (2) obligatory distortions secondary to malocclusion, or (3) retained speech distortions or misarticulation following surgery. Fifty-six patients were included in the study group (23 female, 33 male). Cleft categories were 20 BLCLP, 9 right ULCLP, 25 left ULCLP, 2 complete palatal cleft. Average age at time of surgery was 16.95 with postoperative speech assessment occurring an average of 6.8 months after intervention.

Results: PWSS averaged 2.68 before surgery and 4.41 after surgery (P < .001). Two patients had improved PWSS; 13 patients (23%) had worsening of PWSS of clinical significance. Of 10 patients with VP competence (PWSS = 0): 2 developed VPI (PWSS > 7), 5 developed borderline VP incompetence (PWSS = 3-5). Two patients changed from borderline competence to borderline incompetence; 4 patients changed from borderline VP function to overt VPI; 49/56 (87.5%) patients had presurgical obligatory speech distortions attributed to malocclusion; 30/49 (61%) had normal articulation postoperatively, 15/49 (30.6%) had retained distortions, correctable through speech therapy.

Conclusions: 87.5% of patients requiring LeFort 1 osteotomy had articulation differences attributed to malocclusion; most resolved with correction of malocclusion or could resolve through speech therapy for retained distortions. On average, symptoms of velopharyngeal dysfunction increased in patients undergoing LeFort 1 advancement (23% of patients experienced clinically significant change).

Main Objectives: Develop basic knowledge of velopharyngeal function/its relationship to maxillary advancement surgery and the relationship of the anterior dental relationship and articulation. Gain knowledge surrounding informed consent considerations of speech changes in surgical maxillary advancement.

#### 53. TGF- $\beta$ Signaling Regulates Characterized Periostin and Tenascin C in Soft Palate Development

Takayoshi Sakai (1), Shirabe Ohki (2), Kayoko Ogata (2), Yang Chai (3), Kyoko Oka (2)

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 University of Southern California, Los Angeles, CA

Background/Purpose: We focused on characterized expression of extracellular matrix during soft palate development, including Periostin (Postn) and Tenascin C (TnC). The immunohistochemical expression of Postn and TnC appeared to be wider and more clearly defined in posterior palatal mesenchyme before/after palatal fusion. However, there is little understanding regarding the role of the characteristic expression pattern of extracellular matrix protein through anterior—posterior palatal shelves.

Methods/Description: Using immunohistochemical analysis, the expression of Postn and TnC was examined in Wnt1-cre;Tgfbr2fl/fl, K14-cre;Tgfbr2fl/fl, and Tgfb3-/- mice. Quantitative real-time PCR was performed to investigate Postn and TnC expression in mouse cranial neural crest cell line (O9 -1) and primary embryonic palatal mesenchymal cells (MEPM). Secreted protein in culture medium was also quantitated by ELISA.

Results: In immunohistological expression of Postn and TnC in TGF-β signaling deficient mice, Wnt1-cre;Tgfbr2fl/fl, K14-cre;Tgfbr2fl/fl, and Tgfb3-/-, Postn expression showed no changes in all categories of mice. Interestingly, TnC expression was diminished in K14-cre;Tgfbr2fl/fl and Tgfb3-/- mice exhibiting soft palate cleft, but not in Wnt1-cre;Tgfbr2fl/fl mice. These results indicate that TnC expression in soft palate mesenchyme during palatogenesis is regulated by

TGF- $\beta$  signaling of palatal epithelium. In O9 -1, Fibronectin, Postn, and TnC mRNA expressions were stimulated by TGF- $\beta$ 3, but not in MEPM. The O9 -1 stimulated by TGF- $\beta$ 3 exhibited a fibroblast cell shape. ELISA confirmed that secreted TnC protein in cultured medium of O9 -1 was also increased. Those responses were attenuated by the existence of inhibitor intercellular signaling of TGF- $\beta$ , Smad, and p38.

Conclusions: These results indicate that Postn and TnC is an important extracellular matrix protein in soft palate development. It is suggested that TnC expression in palatal mesenchyme is paracrine regulated by TGF- $\beta$  in the palatal epithelium, and plays a role in the differentiation of fibroblasts in immature palatal mesenchymal cells.

# 54. First Long Term Outcome Study of Metopic Craniosynostosis Reconstructions Utilizing 3 Different Surgical Techniques: Endoscopic Suture Release vs. Cranial Distraction vs. Open Cranial Vault Remodeling—A 5-Year Analysis

Eric Stelnicki (1), Dean Hertler (2)

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Background/Purpose: The purpose of this study was to analyze the long-term results of metopic craniosynostosis repair via 3 different techniques performed in the first year of life. In this study, we compare and contrast our 10 years results with the endoscopic expanded strip craniectomy versus the endoscopic cranial distraction versus the open approach. We hypothesize that the choice of surgical technique may change if one looks at 5-year outcomes and contrast them to immediate and 1-year results.

Methods/Description: This is a retrospective review of 10 consecutive patients treated with each of the metopic cranial vault reconstruction methods. A total of 3 groups of patients were examined: Group 1 (n =10) underwent endoscopic strip cranial vault remodeling following by the 3 to 6 month use of a postoperative cranial molding orthosis (CMO) prior to 5 months of age. Group 2 (n = 10) underwent endoscopic cranial distraction, removal of distractors, followed by a 3 to 6 months used of a postoperative CMO prior to 5 months of age. Group 3 underwent open cranial vault remodeling at 8 to 10 months of age. Morbidity and mortality, need for additional surgery, type of additional surgery, degree of scaring, need for blood transfusion, and overall patient satisfaction was examined in each group All patients in all groups had successful initial cranial vault remodeling in each of the 3 groups. At 3 months postoperative, each group showed correction of the trigonocephaly, improvement in the temporal narrowing, and expansion of the skull with absence of craniosynostosis. There was no mortality or postoperative infection in any group. Blood transfusions were given in 30% of Group 1, 40% of group 2, and 60% of Group 3. There were no transfusion reactions or postoperative complication from transfusion. Hundred percent of Group 2 patients required a second operation to remove the distractors between postoperative day 30 to 45. At 1 year post operatively, none of the groups had signs of recurrent trigonocephaly and all had equal developmental measures for skull growth. Five years post operatively, the groups differed significantly. Severe temporal hollowing was noted in 80% Group 1, 50% of Group 2, and 10% of Group 3 (P = 0.01). Open skull defects were noted in 10% of Group 1, 30% of Group 2, and 10% of Group 3. Reoperation was recommended for 80% of Group 1, 40% of Group 2, and 10% of Group 3. Patient satisfaction was highest in Group 5 years postoperatively.

## 55. Single Versus Two-Stage Secondary Alveolar Bone Grafting (SABG) in Patients With Repaired Bilateral Cleft Lip and Palate

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Background/Purpose: Repair of bilateral alveolar defects in patients with previously repaired bilateral cleft lip and palate is a challenging surgical scenario with previously reported increased complications and need for revision. Multiple approaches to improve outcomes have been suggested, including operating at an earlier age and correcting the bilateral clefts in separate procedures. Herein, we describe our institution's experience performing SABG in patients with complete bilateral cleft lip and palate.

Methods/Description: This is a retrospective cohort study of patients with a history of repaired bilateral cleft lip and palate who underwent maxillary SABG between January 1, 2012 and March 9, 2019 and had at least 6 months of follow-up in the cleft team outpatient clinic. Patients with syndromic features or immunodeficiency were excluded. Medical records of all patients were reviewed for demographic information, pre-, peri-, and postoperative course, and imaging. Discrepancies in records were resolved by reevaluation by an orthodontist. Three-dimensional cone beam computed tomography (3DCBCT) was used to assess postoperative graft taken at the 6-month postoperative visit. The orthodontic cleft team classes take as excellent, adequate, none, or forming a continuous labial bridge. "Adequate" is defined as clinically sufficient for dental implant, while "excellent" is defined as take beyond the alveolus to the hard palate.

Results: Three-hundred sixteen patients were identified as having undergone SABG, of which 57 had repaired complete bilateral cleft lip and palate with 6 months follow-up and 3DCBCT imaging. This represents 103 cleft sites; 11 patients had unilateral repair without repair of the 2nd cleft site. Graft quality was poorer in our bilateral cleft cohort compared to our age-matched unilateral cleft cohort (P < .05). There was a significant (P < .05) difference in age at operation between patients with at least adequate take at one cleft site (8.28+/-1.36) and those without (9.05+/-1.93). Orthodontic preparation, surgical history, and concurrent procedures did not have a significant impact on the quality of graft take. Nine patients had 2-stage unilateral cleft repairs, with an average of 286+/-187 days between procedures. An additional 11 patients had single unilateral repairs. There was no significant difference in take between patients who underwent unilateral versus bilateral repair. Need for reoperation between unilateral and bilateral repair (30% vs 40.5%) was not found to be significantly different; complication rate was higher in patients who received single-stage repair (16.7% vs 0%).

Conclusions: Improved outcomes in bilateral cleft repair in our cohort was shown to be most likely if operation occurred at an earlier age. Repair of bilateral alveolar clefts in separate procedures may reduce complication rate and reoperation. Physicians should take this into account when considering performing bilateral SABG in patients with an expected complicated postoperative course.

## 56. Parent Led, Therapist Supervised, Articulation Therapy (PLAT)—A Trial of Parent Delivered Intervention Supported by Connected Health

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Background/Purpose: Fifty percent of children with cleft palate have persistent speech problems at 5 years of age (Britton et al., 2014) requiring speech and language therapy. However, there is a lack of access to regular therapy in many countries. Although parent training/involvement has positive outcomes in early communication skills in cleft palate and noncleft speech disorders, little is known about parents undertaking intervention for cleft speech disorders. Connected Health programs have been used to provide speech therapy services to those who would not otherwise receive therapy. The aim of the study was to evaluate an alternative method of facilitating improvements in speech by engaging parents.

Methods/Description: Forty-six children, aged 2.9 to 7.5 years, were included in this 2-phase, two-center, noninferiority randomized controlled trial (RCT). In the parent-led arm, parents attended a 2-day training course were given a speech program and conducted homebased therapy for 12 weeks. Parents were supported by a Cleft Speech and Language Therapist (CSLT) using connected health (Facetime) and a one-to-one face to face session. In the control arm, the group received 6 therapy sessions with a research Speech and Language Therapist comparable to that provided by local services. Speech recordings were undertaken pre- and postintervention. Percent consonant correct (PCC) was the primary outcome measure. Activity and Participation were analyzed using the Focus on Outcomes for Children Under Six (FOCUS) and Intelligibility in Context Scale (ICS). Postintervention, parent experience was evaluated. Nineteen parents (83%) participated in telephone interviews or focus groups conducted by an independent Psychologist/Speech and Language Therapist.

Results: Both groups showed a statistically significant improvement in PCC (mean change = 21%; P < .001; 95% CI = 12.27-19.86), Focus (mean change = 18.87; P < .001; 95% CI = 10.87-25.73), and ICS (mean change = 2.27 P < .001; 95% CI = 1.41-3.13) scores. There was no statistically significant difference between the groups. Parents praised the content, flexibility, and pace of the intervention program devised by the therapist. They commended FaceTime, particularly when tasks were not working. Challenges included how they felt "somewhat daunted when taking on the course" but retrospectively "were very happy to have participated." They reported that at times they were overwhelmed by the materials. They expressed concern at being viewed by their child as a teacher. Making PLAT a routine was difficult especially for working parents. Parents provided excellent critical feedback on how to improve training and fine-tune the therapy programs.

Conclusions: Results indicated that parent led articulation therapy when supported by a CSLT using connected health achieves comparable outcomes to standard speech therapy for children with cleft palate speech disorders and is acceptable to parents. These results will inform future development of the service model.

### 57. Nasopharyngoscopy: Methods for Obtaining A Successful Examination With Preschool Children and Interpretation

Ann Kummer (1)

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Background/Purpose: Nasopharyngoscopy allows direct visualization of the velopharyngeal valve during speech. Therefore, it is commonly used by craniofacial professionals to evaluate velopharyngeal function and dysfunction. Nasopharyngoscopy can show the size, shape, location, and cause of a velopharyngeal opening. This information is

valuable in determining the best surgical procedure to achieve the most successful outcome for the patient. If either residual hypernasality or nasal emission is noted after secondary surgery, or if there is evidence of airway obstruction, nasopharyngoscopy is particularly useful in determining the type of treatment or revision surgery that is needed for further correction. Although nasopharyngoscopy is an excellent diagnostic procedure, it can be challenging to perform on young children. The purpose of this session is to provide methods, tips, and tricks for obtaining a successful nasopharyngoscopy evaluation in children as young as age 3, while causing minimal distress to the child (and the parent). In addition, this session will focus on interpretation and use the nasopharyngoscopy findings to determine the surgical procedure that has the best chance of success for each individual patient.

Methods/Description: In this study session, the presenter will discuss the basic techniques of nasopharyngoscopy and also describe some tips and tricks to elicit necessary cooperation from very young children. The presenter will then explain how nasopharyngoscopy can be used to determine the size, shape, location, and cause of the velopharyngeal opening. Numerous short videos of nasopharyngoscopy examinations will be presented for participants to evaluate and discuss. The presenter will describe how the nasopharyngoscopy findings can be used to determine which surgical procedure has the best chance of a successful outcome for the patient. Finally, the presenter will discuss how nasopharyngoscopy can be used to evaluate secondary surgery for velopharyngeal insufficiency in order to develop appropriate strategies for revision, when necessary.

### 58. Identification of a Core Outcome Set for Reporting Outcomes of Management of Velopharyngeal Dysfunction; the COS-VPD Initiative

Catherine de Blacam (1), Adriane Baylis (2), Richard Kirschner (2), Susan Smith (3), Debbie Sell (4), Kathleen Sie (5), Helen Harris (6), David Orr (1) (1) Dublin Cleft Centre, Dublin, Ireland, (2) Nationwide Children's Hospital, Columbus, OH, (3) Royal College of Surgeons in Ireland, Dublin, Ireland, (4) Great Ormond Street Hospital NHS Trust, London, United Kingdom, (5) Seattle Children's Hospital, Seattle, WA, (6) London, United Kingdom

Background/Purpose: Velopharyngeal dysfunction (VPD) is present in up to 40% of patients following cleft palate repair (Britton et al., 2014). A child with VPD, unable to easily produce oral consonants, may develop compensatory articulation, as well as present with hypernasality and nasal air emission/turbulence. The overall result is decreased intelligibility and acceptability of speech, and functional and social impairments. There are several surgical approaches for the management of children with VPD that can be broadly described as palatal, pharyngeal, and palatopharyngeal procedures. However, standard treatment protocols for VPD have not been welldefined. In a systematic review, the presenters of this session found mainly retrospective case series describing results of surgical interventions for VPD using diverse parameters, particularly with regard to perceptual speech assessment (de Blacam et al., 2018). There is a need for a core outcome set to reduce both outcome reporting bias and heterogeneity across studies, thus allowing meaningful collation and comparison of results across different etiologies, surgical protocols, and institutions. The COS-VPD initiative is an international effort to establish a core outcome set for the reporting of VPD management outcomes.

*Methods/Description*: In this session, an international group of surgeons and SLPs will describe the COS-VPD Initiative's development. A summary of the systematic review from which a preliminary list of

core outcomes was derived will be reviewed (de Blacam et al., 2018), as well as a summary of feedback obtained from 15 surgeons and 50 SLPs who attended an international VPD meeting and participated in a COS-VPD feedback session. The methodology for the development of a core outcome set will be introduced including steering group formation, protocol development, categorization of outcomes, and the Delphi process. Potential core outcomes will be presented and audience feedback gathered. Participants will have the opportunity to share their opinion on the outcomes to be included in the core outcome set during this interactive session. Future directions for clinical and research applications will be discussed.

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#### 59. Meet-and-Greet for Multidisciplinary, Multi-Site Cleft Collaboratives—With Lightning Talks

Alexander Allori (1)

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Background/Purpose: Since the early days of CSAG, Eurocleft, and Americleft, the number of multidisciplinary, multisite cleft collaboratives has grown. Each group is intent on focusing on "metrics that matter"—but what does that mean? What is each group doing? Who are the people involved in each group? Is there opportunity for strategic alignment between projects? The purpose of this study session is for anybody involved in (or interested in participating in) a multisite cleft collaborative to meet other like-minded individuals, share their works-in-progress, receive some feedback, and possibly identify opportunities for collaboration.

Methods/Description: Anybody and everybody who has participated is presently participating, or would like to participate in a multisite cleft collaborative is invited to attend this study session. Each participant will have the opportunity to present a summary of his/her project. We encourage a "Lightning Talk" format, whereby each participant may give a brief presentation consisting of maximum 7 slides; however, no slides are required for participation. The moderator will facilitate discussion, but the focus is on open exchange in an inclusive and respectful environment. There are 4 main goals of this workshop: (1) Participants should have the opportunity to "get their project known," even if it is a work-in-progress that is not ready for abstract presentation or manuscript publication; (2) Participants should receive constructive feedback on their projects that may help them improve their projects; (3) Participants should gain a better appreciation for the landscape of existing (and planned) multisite collaborative projects; (4) Participants will get to know each other! In theory, all of this may foster better synergy and/or complementarity among projects and may encourage closer collaboration between groups. After this Study Session, we would like to set up an online forum where all groups will have representation and may continue their interaction.

### 60. A 70-Year History of Unilateral Cleft Lip Repair: A Simulator-Based Symposium

Court Cutting (1), Roberto Flores (1), Qisi Wang (2), Yutian Tao (2), Eftychios Sifakis (2)

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Background/Purpose: The evolution of primary unilateral cleft lip repair represents a series of incremental modifications pioneered by a distinct group of master surgeons. It is through understanding the purpose of each evolutionary step, the limits and compromises of these steps, and the subsequent modifications which followed, can a greater understanding of the art of cleft lip repair be realized. This course will trace the conceptual development of unilateral cleft lip repair over the past 70 years using a novel, real-time computer-based cleft lip simulator. A first order accurate biophysics implementation within the simulator will be used to demonstrate the cleft lip repair techniques described to reveal the strengths and weaknesses of each stage of unilateral lip repair development. The course will begin with the Tennison Randall lower triangular lip repair, as it is still in common use today. This will be followed by Skoog, Wynn, and Mustarde adding an upper triangle to the lip repair. The various stages in the progression of the Millard repair will then be carefully traced along with the biophysics which are likely responsible for why Millard altered his original design. The modifications of the Millard design by other surgeons, and the reasons for them, will then be carefully traced. The modifications covered will be those of Noordhoff, Mohler, Cutting, Fisher, and others. The interaction between lip repair technique and primary correction of the cleft-lip nasal deformity will be discussed in detail. Simulator-based demonstrations will be augmented with patient examples from the senior author's clinical practice which illustrate the conceptual difficulties encountered at various stages in the historical development of primary unilateral cleft lip and nose repair.

Methods/Description: The principal method used in this course will be real-time computer-based surgical simulation. A unilateral cleft lipnose model involving skin, mucosa, muscle, bone, cartilage, and teeth was derived from an MR scan of an adolescent with an unrepaired unilateral cleft. Alterations in the model are illustrated with first-order accurate biophysics using a new software base called projective dynamics. Surgical tools provided are scalpel, suture, hook, and undermine of both skin/mucosa and bone/cartilage. Surgical "'history" files are used to step through a succession of cleft lip repairs in the surgical eclectic. The presentation will be augmented with photographs from the senior author's long career further illustrating why successive alterations in technique were made.

## 61. Evidence-Based Assessment of the Internationally Adopted Child With Cleft Lip and Palate: Practice Strategies for the Team Speech-Language Pathologist

Caitlin Cummings (1), Katie Garcia (1)

(1) Nationwide Children's Hospital, Columbus, OH

Background/Purpose: Cleft lip and palate (CLP) teams must be prepared to provide culturally and linguistically appropriate care for patients of diverse backgrounds, including children who are internationally adopted (IA) (Swanson et al., 2014; U.S. Department of State, 2017). Speech-language pathologists (SLPs) who work with IA children with CLP are presented with unique challenges. In addition to the known negative impact of CLP on articulation and expressive language (Chapman & Willadsen, 2011; Chapman et a., 2003), IA children often have histories of institutionalization and delayed cleft surgical management (Glennen, 2007; Kaye et al., 2019). In addition,

IA children experience an abrupt shift from their native language to a new language environment—a process known as second first language acquisition—which is not equivalent to monolingual or bilingual development. Finally, IA children have heterogeneous origins and can display variable speech and language profiles. Although research on markers of speech and language disorders in IA children with CLP is emerging (Larsson et al., 2017; Morgan et al., 2017, 2018; Scherer et al., 2018), evidence to guide SLP practice is still limited. The purpose of this study session is to review the most current literature on the speech and language development of IA children with CLP and describe clinical best practices for evaluating communication skills. This innovative session extends traditional discussions by focusing on the role of the SLP and the type, timing, and interpretation of clinical assessments to ensure comprehensive and evidence-based care for IA children and their families.

Methods/Description: Two CLP-team SLPs will summarize current literature and discuss their experiences working with IA children with CLP in the interdisciplinary clinic and outpatient therapy settings. Language milestones of IA children will be reviewed. Strategies for teams working with International Adoption Clinics will also be discussed. Attendees will be provided with suggested time points and clinical materials for evaluating IA children with CLP, including how to determine therapy recommendations, if intervention is warranted. Case studies, with audience participation, will be incorporated to illustrate how SLPs should conduct an evidence-based assessment and triage different therapy and/or surgical needs while collaborating with other members of the team, family, and community SLP providers. Finally, the session will discuss other current initiatives to improve evidence-based care and awareness of the communication needs of IA children within the team setting.

#### 62. Models of 22q Team Care in the United States

Oksana Jackson (1), Yvonne Gutierrez (2), Alexis Johns (2), Daniela Schweitzer (3), Marina Clarke (3), Jessica Kianmahd (3), Donna McDonald-McGinn (4), Daniel McGinn (1), Amanda Smith (5), Scott Hickey (5), Emily Gallagher (6), Kaylee Paulsgrove (6), Susan Hughes (7), Jill Arganbright (7) (1) Children's Hospital of Philadelphia, PA, (2) Children's Hospital Los Angeles, CA, (3) University of California Los Angeles, CA, (4) Children's Hospital—Clinical Genetics Center, Philadelphia, PA, (5) Nationwide Children's Hospital, Columbus, OH, (6) Seattle Children's Hospital, Seattle, WA, (7) Children's Mercy Kansas City, Kansas City, MO

Background/Purpose: Patients with 22q11.2DS present to many cleft/ craniofacial teams in the Unites States for assessment and management of speech and language disorders including velopharyngeal dysfunction. Because patients with 22q11.2DS are at risk for multisystem involvement and developmental deficits during childhood, they require coordinated care from a multidisciplinary team. Unlike the well-established team care standards for cleft and craniofacial care, management models for patients with 22q vary widely. While some core team members are consistent across regions of the United States, there is marked variability in both the composition and structure of team care for 22q patients, even among larger teams. Also, 22q teams face unique challenges, often related to geographic distribution of patients and available resources. As a result, management approaches vary in ways that may impact clinical outcomes of patients with 22q. The goal of this presentation is to highlight various models of 22q team care across several institutions in the United States. This will serve as a useful resource for individuals and groups that are interested in starting a 22q team or continuing to grow their current 22q team. Methods/Description: ACPA's 22q Special Interest Group offers this

Methods/Description: ACPA's 22q Special Interest Group offers this session which will follow a "show and tell" approach with

contributions from five 22q teams across the Unites States. The aim is to show common characteristics between teams and those that are variable. The presentation will allow 22q teams that are in their infancy or trying to get started, to see different ways of setting up a team as well as answering questions that may be helpful as they build their team. Each 22q team representative will present specifics on their hospital type and location, the structure and format of their 22q team including specialist composition, care coordination strategies, funding, team conferencing, as well discussing current challenges the teams are facing and insight into how they are overcoming these issues. Teams will also share their approaches to patient education and outreach by highlighting ongoing local and national 22q patient/ family activities. The aim of this will be to increase provider awareness of these events and inspire 22q teams to encourage and foster these types of opportunities for their 22q patients and families. Lastly, audience participation will be encouraged through a question and answer session.

### 63. Strategies for Successful Implementation of a Standardized Speech Outcome Protocol in the Cleft Palate Clinic

Kristina Wilson (1), Adriane Baylis (2), Kelly Cordero (3), Angela Dixon (4), Kathy Chapman (5), Cindy Dobbelsteyn (6), Anna Barigayomwe (7), Judith Trost-Cardamone (8)

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Background/Purpose: In addition to clinical responsibilities of evaluating speech production skills and velopharyngeal function, speech-language pathologists on cleft palate teams also collect data that can be used to monitor and report speech outcomes for research and internal quality improvement initiatives. This session focuses on the logistics involved with implementing standardized speech outcomes assessment procedures based on the Americleft speech protocol, which has been utilized in numerous clinics across the United States. The goal of this session is to share data collection strategies and techniques that can be integrated into clinical practice to improve patient care and allow for possible research application, while also complying with institutional regulations.

Methods/Description: This session will focus on practical strategies for incorporating the Americleft speech protocol into clinical visits. Suggestions for video and audio recording equipment will be provided. Standardized speech samples for English-speaking and Spanish-speaking preschool children through adults will be shared (Trost-Cardamone, 2012). The CAPS-A-AM rating scale (Chapman et al., 2016) as well as a clinical adaptation of the tool (Chapman et al., 2017) will be reviewed. Strategies for storing, transmitting, and evaluating this data to complete quality improvement projects and conduct research while complying with HIPAA and IRB requirements will be discussed.

#### 64. Advanced Practice Providers in Cleft Care: Where Do We Fit In

Katherine Shedd (1), Alison Kaye (1), Meghan Tracy (1), Clare Gargaro (2), Payton Leonhardt (3)

(1) Children's Mercy Kansas City, Kansas City, MO, (2) Vanderbilt Children's Hospital, Nashville, TN, (3) Phoenix's Children's Hospital, Phoenix, AZ

Background/Purpose: While the roles of the plastic surgeon, speech therapist, and orthodontist in the care of children with orofacial cleft conditions are well established, the incorporation of advanced practice providers (APP) specializing in this patient population has been more recent. As a result, the roles and responsibilities of APPs, including nurse practitioners (NP) and physician assistants (PA), in cleft and craniofacial patient care varies substantially across the nation. Knowing that the roles vary, underutilization of the APP becomes more likely. Our goal is to better understand the roles and responsibilities among APPs to ensure that they are utilized to their full scope of practice within the cleft team.

Methods/Description: An IRB approved survey study of APPs practicing in Cleft Multidisciplinary Clinics approved by the American Cleft Palate Association (ACPA). Written consent was obtained from all study participants. This presentation will provide an opportunity to review the varying roles and responsibilities reported by APPs in the survey. A panel of APPs will be present to provide insight and lead discussion. In addition, we will identify roles and responsibilities that an APP may hold that may benefit their cleft team and allow the APP to practice at the top of their scope.

#### 65. Robin Sequence and Mandibular Distraction— The Multidisciplinary Management From Infancy to Adolescence

Michael Lypka (1), Zarmina Ehsan (1), Dan Jensen (1), Heather Hendricks (1)

(1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: In many institutions, mandibular distraction has become the primary management of the neonate with a hypoplastic mandible and respiratory compromise. A multidisciplinary team approach is necessary to identify neonates who might benefit from the procedure and manage the patient after the procedure. Many of the parameters for patient selection and follow-up are controversial, with varying imaging, airway, and sleep evaluations. The purpose of this course is to discuss the comprehensive multidisciplinary approach to mandibular distraction in patients with Robin Sequence, including patient selection and work-up, technical aspects of the operation, management of complications, and long-term follow-up.

Methods/Description: With a panel consisting of a craniofacial surgeon, otolaryngologist, sleep physician, and orthodontist, the course will explore a series of cases of Robin Sequence, with audience participation encouraged. Work-up, including airway and sleep evaluation, surgical technique, and long-term follow up, including orthodontic considerations, will be discussed.

#### 66. Neonatal Ear Molding: A Practical Hands-On Instructional Course

Elizabeth Wetz (1), Jessica Grant (1), Laura Hanna (1), Savannah Brown (1), Sarah Wiggins (1), Kaylyn Gerth (1), Kimly Nguyen (1), Rami Hallac (2), Christopher Derderian (3), James Seaward (3), Alex Kane (4)

(1) Children's Health, Dallas, TX, (2) Analytical Imaging and Modeling Center, Children's Health, Dallas, TX, (3) University of Texas Southwestern Medical Center, Dallas, TX, (4) UT Southwestern School of Medicine, Dallas, TX

Background/Purpose: The true incidence of neonatal auricular deformities is unclear, with reported rates from 0.1% to 45%. Ear molding has been growing in popularity over the last 20 years as a nonsurgical treatment option and several techniques have been described. These techniques vary widely in complexity, compliance, and cost, yet few

published large series of ear molding outcomes are available in the literature. We have developed an inexpensive and effective technique for neonatal molding that demonstrates predictable and successful outcomes. We propose a practical hands-on instructional course using flexible 3D printed models for providers interested in developing an ear molding practice.

*Methods/Description*: This course will familiarize participants with our ear molding technique and provide hands-on instruction and practice utilizing flexible 3D printed models.

### 67. Clinical Research in Craniofacial Microsomia: Progress, Pitfalls, and Future Goals for International Engagement

Carrie Heike (1), Maarten Koudstaal (2), Amelia Drake (3), Daniela Luquetti (4), Alexis Johns (5), Nicola Stock (6), Canice Crerand (7)

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Background/Purpose: Children with craniofacial microsomia (CFM) are at risk for significant differences in facial appearance due to asymmetry and significant functional problems such as respiratory compromise, feeding difficulties, and hearing loss. Children with CFM are also at increased risk of having extracraniofacial anomalies that warrant screening. Significant advances in the field of cleft lip and palate have been made in recent years, with several international consortiums working to standardize outcome measurement, large-scale cohort studies being established in the United States, Europe, and Australasia, and an increased understanding of health-care needs and psychosocial impacts for patients and families. In contrast, fewer efforts have been made to progress knowledge and health-care delivery for individuals with CFM. The specific goals of this study session are to review recent results from recently published, large, multicenter studies and active clinical research projects in CFM, to identify gaps in current knowledge, and to discuss future goals for the field.

Methods/Description: In this 90-minute session, participants will hear from 6 panelists from a variety of disciplines with experience in conducting large, multicenter studies in CFM. Panelists will describe past, current, and planned projects to collect standardized data in CFM. Speakers will summarize studies on the following topics: health-care services, craniofacial surgeries, airway anomalies and obstructive sleep apnea, genetics, psychosocial outcomes, social determinants of health and at-risk screening. The last portion of the session will include an interactive component with opportunities for discussion about the material presented and future aims of the study group, which include international collaborations and greater involvement of patient and caregivers as stakeholders in study design and execution.

## 68. Comparison of Speech Outcomes Using Superiorly Based Pharyngeal Flaps Versus Buccal Palatal Flaps for Velopharyngeal Insufficiency

Andrea Biaggi-Ondina (1), Brady Anderson (1), John Teichgraeber (1), Matthew Greives (1), Phuong Nguyen (1)

(I) McGovern Medical School at the University of Texas Health Science Center at Houston (UTHealth), Houston, TX

Background/Purpose: Cleft palate repair may be complicated by velopharyngeal insufficiency (VPI) which occurs in 20% to 30% of

patients. VPI is characterized by insufficient closure of the velopharyngeal valve. Two common procedures to address VPI include superiorly based posterior pharyngeal flaps (PPF) and buccal myomucosal flaps for palatal lengthening, with or without a revision Furlow palatoplasty. Though both procedures have distinct advantages and drawbacks, there has not yet been a direct comparison in speech outcomes and effectiveness.

Methods/Description: A retrospective chart review was performed of all patients with cleft palate who underwent a procedure for VPI from January 2014 to April 2019. Inclusion criteria were patients who underwent secondary surgery with PPF or buccal flap for documented VPI. Demographic information was abstracted along with surgical findings and measurement from preoperative imaging studies, including nasoendoscopy and lateral palatal motion studies. Pre- and postoperative speech assessments were performed by a trained speech-language therapist (SLP) using the University Parameters for Reporting (UPR) for Speech.

Results: Thirty-six patients underwent PPF and 10 patients underwent buccal flaps with adequate follow-up. Preoperative velopharyngeal gap sizes did not differ significantly between patients undergoing buccal flaps (5.3 mm  $\pm$  4.8) versus those with PPF (6.0 mm  $\pm$  4.0; P=0.75). URP hypernasality scores improved for both buccal flaps (2.1 to 0.4; P<0.1) and PPF (2.0 to 0.6; P<0.1), though there was no significant difference between the 2 cohorts (P=0.61). Patients with PPF were more likely to develop complications (63.9% vs 0%) than those undergoing buccal flaps. The most common complications due to PPF included snoring (33.3%), obstructive sleep apnea (8.3%), midface retrusion (8.3%), and persistent VPI (5.6%). However, given the pedicled flap, all (100%) patients in the buccal flap group underwent flap pedicle division versus 13.9% of patients in the PPF group. When excluding pedicle take down, the revision rates were 0% for buccal flaps versus 13.9% for the PPF group.

Conclusions: Both buccal flaps and superior based pharyngeal flaps are effective methods for treating VPI with significant improvement in hypernasality speech scores. There was not a significant difference in speech improvement between pharyngeal flaps and buccal flaps. There were lower rates of sleep apnea in the buccal flap group, though they necessitated a second surgery for pedicle division.

### 69. Using Artificial Intelligence to Analyze Facial Action Units Following Facial Reanimation Surgery

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Background/Purpose: Facial expressions can be analyzed systematically using the Facial Action Coding system which links discrete facial action units to specific emotions. This study evaluates the use of a machine learning technology to directly measure facial action unit and emotional expression both before and after facial reanimation surgery.

Methods/Description: Fifteen study subjects with facial palsy were evaluated both before and after undergoing cross facial nerve graft and free gracilis muscle transfer. Video footage was obtained of subjects with their face both in repose and with a posed, closed-lip smile. The video data was then analyzed using the Noldus FaceReader™ software application to measure the 28 action units and the happy and sadness emotion detected within each clip.

*Results*: During smile, activation of the action units corresponding to happy emotions increased from average intensity of 1.5/4 pre to 2.71/4

postoperatively and 1.0/4 pre to 1.83/4 postoperatively in the lip corner puller and cheek raiser AUs, respectively. This corresponds to an increase in overall happy emotion detected from 13% to 42% (P < .0001). Conversely, the "lip corner depressor" action unit, associated with sad emotions, decreased from average of 2/4 pre to 0/4 postoperative during smile. The sad emotion detected decreased from 15% to 9% (P = 0.092). The other 2 action units associated with sad emotions ("inner brow raiser" and "brow lowerer") did not show any change pre- and postoperatively value of 1/4.

*Conclusions*: This study provides the first proof of concept for the use of a machine learning software application to objectively detect facial units changes and quantify facial expression before and after surgical reanimation.

# 70. Workflow and Operational Strategies for a Longitudinal 3D Craniofacial Morphologic Study: Three-Year Practical Model Experience from the Healthy Taiwanese Chinese Elementary School Students-Derived Normative Database

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Background/Purpose: Longitudinal studies are considered to be a gold standard for understanding the normal craniofacial morphologic development, but the operational management for implementation of such studies is not often addressed in detail. This study describes the strategies used to recruit and maintain high levels of participation in a longitudinal study involving annual 3D craniofacial soft-tissue images from healthy Taiwanese Chinese elementary school students aged 6 to 12 years.

Methods/Description: The key aspects for project delineation, implementation, and initial 3-year practical experience were portrayed in an integrated multistep workflow: ethics- and grant-related issues; contact, approval, and engagement from school stakeholders (dean, director, and teacher) and parents; didactic approach to recruit the students; staff training with task design; tripod station-based data collection day with educative (oral hygiene and psychosocial interaction stations) and craniofacial imaging (3D surface and anthropometric information) measures; reinforcement tactics to sustain the longitudinal annual participation after the first enrollment; and definition of 3D data processing and analysis. Randomly selected students and teachers answered an experience satisfaction questionnaire with 5-point Likert scales.

Results: Six of 7 contacted schools accepted to be part of project. All parents who attended the explanatory meetings accepted to join this project. A cohort of 550 students participated at baseline enrollment, with a follow-up rate of 80% in the second data collection. The average questionnaire-related scores were of  $4.2\pm0.7$  and  $4.4\pm0.6$  for teachers and students, respectively.

Conclusions: This 3D craniofacial norms would benefit multidisciplinary teams facing a myriad of cleft-craniofacial deformities in globally ethnic Chinese population, particularly useful for characterization of craniofacial phenotypic variation, for making quantitative morphologic comparisons, and in therapeutic planning and outcome assessment. The described strategies would assist other groups to start the planning of data collection for establishment of their own age, gender-, and ethnic-specific normative database.

# 71. Trans-Palatal Approach for Resection of a Pediatric Clival Retropharyngeal Sarcoma: Implications for Expanded Application of the Craniofacial Surgical Armamentarium

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Background/Purpose: Rhabdomyosarcoma (RMS) is the most common pediatric soft tissue tumor accounting for about 3% to 5% of all childhood malignancies. Approximately 40% of all RMS occur in the head and neck region. Retropharyngeal RMS are rarely reported in the literature. Their location poses a unique set of challenges for surgical resection, namely that they are obscured by the presence of the palate and their close association with the retropharyngeal carotid artery. Given the familiarity with this region, the craniofacial surgeon is ideally poised to perform surgical extirpation of tumors within the retropharyngeal region. The purpose of this case presentation is to demonstrate the ability of the craniofacial surgeon to assist in surgical extirpation of tumors arising in the retropharyngeal space.

Methods/Description: This case presentation will discuss the nature and pathophysiology of pediatric head and neck RMS with an emphasis on the surgical approach utilized to remove the tumor. We will detail in depth the planning of extirpation, the technique employed, and the events that lead to this patient being seen and ultimately treated by the craniofacial team. A discussion will be made regarding the potential role that the craniofacial surgeon can play in the management of head and neck tumors in the pediatric setting and why the craniofacial surgeon is well positioned to deal with these scenarios.

Results: The transpalatal approach for resection of the retropharyngeal rhabdomyosarcoma permitted for clearance of the T2 signal on iMRI, R1 histopathologic resection with microscopic disease on the carotid sheath, and rapid convalescence allowing for resumption of adjuvant chemotherapy and proton radiotherapy.

Conclusions: This case highlights the ability of the craniofacial surgeon to utilize and apply ones familiarity and surgical skills in this forum to pediatric tumors of the anterior skull base.

## 72. Does the Twist or FGFR3 Mutation in Bicoronal Synostosis Affect Morphometrics Two Years After Fronto-Orbital Advancement: A Comparison Study

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Background/Purpose: Saethre-Chotzen (SC) and Muenke (Mu) syndromes are 2 of the most common syndromes associated with bilateral coronal synostosis (BCS). Their respective mutations (TWIST and FGFR3) have been shown to affect osteogenesis in vitro, but it is unclear whether these mutations affect clinical outcome after fronto-orbital advancement (FOA) compared to nonsyndromic BCS (nsBC) cases. Our aim was to analyze CT scans of SC and Mu syndrome cases who underwent FOA for BCS, and compare the morphology before surgery (t0), immediately after surgery (t1), and 2 years after surgery (t2) to nsBC who underwent FOA, and to normal controls.

*Methods/Description*: This was a retrospective study design. Inclusion criteria were all BCS cases who underwent FOA from 2005 to 2017. Age-matched normal controls had CT scans for noncranial deforming reasons. We made the following measures using 3D Slicer software:

cephalic (CI) and turricephaly index (TI), horizontal and vertical bossing ratio (HBR, VBR), sella-frontal (SF) and midsagittal (MS) vector analysis, supraorbital retrusion (SOR), cranial fossa area, and vault volume. Data was compared between groups and to controls at each time point, and between time points within each group using ANOVA with Tukey's HSD correction (significance P < .05).

Results: Seven SC, 6 Mu, 11 nsBC, and 10 controls were included. At to all BCS cases had significantly greater CI, TI, frontal bossing (HBR, VBR), and SOR compared to controls. Bandeau analysis revealed significant anterior restriction (SF0) and biparietal widening (SF90), while midsagittal analysis revealed significantly greater height and posterior restriction in BCS. Volume was significantly restricted in the posterior vault, but was greater in the anterior and middle vaults. Lower forehead was over-advanced with FOA at t1 with improvements to anterior fossa restriction and SOR, but it was normalized at t2 after growth of the normal controls. Turricephaly measures remained higher in BCS but decreased between t1 and t2. FOA did not address posterior skull base restriction in BCS, which persisted at t2. There were no statistical differences between the SC, Mu, and nsBC groups at any time point, with few exceptions: Mu had a significantly larger middle fossa area and volume than SC and nsBC at t0 and t2 and total volume at t2 due to greater biparietal width and vault height.

Conclusions: FOA adequately treats lower forehead position in BCS 2 years after, but turricephaly and decreased skull base length persist. With few exceptions, the syndromic vs nonsyndromic BCS morphology was not significantly different at any time point.

## 73. A Novel Method for Fabricating Naso-Alveolar Molding Appliances Using 3-Dimensional Workflow and Clear Aligners

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Background/Purpose: Presurgical infant orthopedics have been utilized for over 50 years as an adjunctive therapy for the correction of cleft lip and palate (CLP). Nasoalveolar Molding (NAM) appliances are traditionally made on dental stone models, and fabricated with an acrylic molding plate. NAM therapy often requires weekly visits for adjustments or fabrication of a new appliances, requiring an extensive chair-side time at each visit. Weekly visits may extend for 10 to 20 weeks, and may impose a significant burden of care. In the current study, we propose a new approach to utilizing a digital workflow and 3D printing to fabricate clear aligner NAM devices that minimize the chairside visit time and may minimize the frequency of visits.

Methods/Description: We present 3 patients with unilateral CLP, with defect sizes ranging from 5 mm to 16 mm, that we treated with digitally designed clear aligner NAM appliances. A traditional impression using Siltec putty material is acquired then poured and the stone model is scanned with an intraoral scanner. The STL file was imported into MotionView software. The alveolar segments are digitally segmented and moved to the desired final position. The total distance moved is divided into a sequence of 1 to 2 mm increments, allowing us to create a staged number of digital models. The models are 3D printed along with button templates to allow free form positioning of the button on each model. Buttons are added directly to the model and attached using Triad gel. Once the button is placed, a vacuform machine is used to fabricate an 0.040' aligner for each stage. The button is retained in the aligner and easily separates form the model. The nose piece is added at the appropriate stage with a .030' orthodontic wire and Triad for nasal support. Triad and Assure plus are used to adhere

the wire to the aligner, with retention holes cut out for increased retention

Results: Appointments for the NAM adjustments have transitioned to primarily counseling and discussion and less adjusting of the appliance. The initial appointment takes approximately 30 minutes in order to maximize vestibular extension and consult the parents. Try in and fit verification takes less than 10 minutes. The placement of the nose-piece may require additional  $\sim\!30$  minutes as they are adjusted chair-side to the particular child. Benefits of the aligner are improved fit at each stage, visual appreciation of blanching tissue, more precise increments of activation, reduced chair-side time for patients and providers, and minimizing the number of visits as it is possible to deliver multiple stages at once.

Conclusions: NAM aligners may save on average 20 to 30 minutes per appointment and can reduce the number of total visits needed in half or more depending on compliance. The appointments become more focused on our feeding team and counseling the family, instead of the labor-intensive adjustment of the traditional appliances. NAM aligners may provide similar benefits to the traditional approach while reducing the burden of care.

## 74. The Impact of the Buccal Myomucosal Flap on Speech and Surgical Outcomes in Cleft Palate: A Systematic Review

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Background/Purpose: A systematic review was conducted on the buccal myomucosal flap surgical approach for primary palatoplasty and secondary surgery for velopharyngeal insufficiency in individuals with cleft palate. This is the first systematic review of the buccal myomucosal flap approach and its impact on speech and surgical outcomes pertaining to velopharyngeal anatomy and function.

Methods/Description: A systematic review was carried out according to PRISMA guidelines. A search of the literature in EMBASE, Pubmed, Scopus, CINAHL, and Cochane was performed. Articles were included if the patients received the surgical technique for primary cleft palate repair or secondary surgery for VPI and if the study provided adequate details on the methods of the perceptual speech assessment or visualization of postoperative velopharyngeal anatomy.

Results: A total of 11 studies were included in the review with a total of 1013 patients. Variability in perceptual speech assessment scales and methods for assessing velopharyngeal competence were noted in the studies. Following surgery, normal resonance was achieved in 77.4% of patients and no nasal air emission was reported in 54.7% of patients. Improvements in velopharyngeal closure was reported in 81.8% of patients. Negative surgical outcomes were reported in 10.8% of patients and 9.9% required further surgery.

Conclusions: This systematic review suggests the buccal myomucosal flap results in an improvement in overall speech intelligibility, resonance, nasal air emission, and velopharyngeal closure. Improvements in intelligibility, resonance, and nasal air emission are similar to those of other secondary surgeries for VPI. Higher incidences of surgical complications were noted for traditional primary palatoplasty repairs than the buccal myomucosal flap technique. Lower surgical complication rates were noted for traditional methods for secondary surgery for VPD than the buccal myomucosal flap. To ensure accurate comparisons between surgical techniques, the development of an international, standardized method for assessing and reporting speech outcomes and velopharyngeal competency is recommended.

#### 75. Speech at Home—An Innovative Approach to Developing Therapy Programmes

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Background/Purpose: The Parent Led Articulation Therapy (PLAT) RCT showed that parents are effective in delivering therapy following in-depth training, based on a child-specific program designed by the cleft speech pathologist and supported using connected health. Parentled intervention for cleft palate articulation therapy could be an alternative model of service delivery. One aspect of this intervention package was the development of speech therapy programs with their resources, all labor-intensive activities for busy therapists. Furthermore, parents reported materials were overwhelming and difficult to access during the trial. An online resource, named Speech at Home, is nearing completion and aims to address this problem. The aim of this presentation is to demonstrate one aspect of this resource—the speech program builder.

Methods/Description: The speech program builder is built on a framework of overall goals, aims based on listening and production and 142 different therapy activities. The activities are focused on target consonants contrasted with error sounds, listening activities, and establishing speech through syllables, words, phrases, sentences, and stories. Resources include a video archive on how sounds are made, cleft speech sound errors and how to elicit targets, and pictorial material covering sound concepts, consonants and vowels, error sounds, and worksheets for working at the different levels of the speech hierarchy. At word level, pictures have been selected and ordered addressing the unique challenges of therapy in cleft speech. When an activity is selected, the required resources are automatically linked to it. Therapists require some training in the application of the speech program builder.

*Results*: Aspects of the speech program builder will be demonstrated. *Conclusions*: The speech program builder is an innovative approach to developing speech programs, with the potential to facilitate this task for therapists and help parents access their programs and required resources more efficiently.

### 76. Which Dysmorphologies of Unilateral Lambdoid Synostosis Are Normalized by Current Surgical Technique?

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Background/Purpose: Unilateral lambdoid synostosis (ULS) is a rare form of craniosynostosis and is considered a primarily posterior deformity. Surgery is therefore largely focused on expansion of the posterior cranium. More recently, attention has been focused on ULS asymmetries of the skullbase and middle and anterior cranial fossa. The purpose of this study was to use morphometric analysis to identify differences in the cranium and skullbase in a large series of ULS cases before and after posterior cranial vault expansion. The goal was to determine which dysmorphologies are normalized by current surgical technique.

Methods/Description: We performed morphometric digital analysis of 21 cases diagnosed with ULS. Fifty-six individual landmark points were placed in 3D Slicer software on preoperative (t0), immediate

postoperative (t1), and 2-year postoperative (t2) CT scans. We calculated multiple linear and volumetric measurements using automated programs and compared the measures against age-matched controls. Comparison was done using regression analysis (significance P < .05).

Results: ULS anterior fossa area was larger than normal at all 3 time points. Middle fossa area was similar to normals at t0, increased at t1, and was the same as normals at t2 (P < .05). Although mean posterior fossa was smaller than normal at t0, was larger at t1, and comparable to normals at t2, none of these trends were significant. Ear position, middle cranial fossa, posterior fossa, and cranial base angulation were significantly different in ULS compared to normal and were not affected by surgery. On axial analysis, the posterior affected side was expanded after surgery, but the unaffected side remained larger at t1 and t2.

Conclusions: ULS is not an isolated posterior fossa condition. Anterior fossa and skullbase asymmetry measures were not affected by surgery and remained different. Surgery had more effect on the middle cranial fossa than the posterior fossa. Surgery successfully expanded the affected axial measures, but did not adequately decrease the unaffected side. Modifications in technique can work to address these residual deformities.

## 77. Aesthetic Outcomes of Patients With Unilateral Cleft Lip and Palate Following NasoAlveolar Molding Therapy in an Outreach Setting

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Background/Purpose: Global Smile Foundation (GSF) is a not for profit foundation whose founders and volunteers have been providing cleft care to underserved communities around the world for 32 years. In 2012, GSF incorporated nasoalveolar molding (NAM) into its treatment model in Guayaquil, Ecuador. We present an evaluation of nasolabial aesthetic outcomes and scarring in patients treated with NAM prior to primary cleft lip repair versus patients who were not, in similar outreach settings.

Methods/Description: The Cleft Aesthetic Rating Scale (CARS) used frontal photographs taken at least 1-year post primary cleft lip repair to assess the nose: tip, nostrils (symmetry, size, flaring), and upper lip (vermillion symmetry and continuity and length of the philtrum), ranging from 1 (very good) to 5 (very poor). Photos were standardized to reveal only the nasolabial area and excluded any time points after any additional surgical revision to the nasolabial area. Each NAM time point was matched to a control time point based on their age at primary cleft lip repair (maximum of 3 months) and their time postoperative from primary cleft lift repair (maximum of 6 months). All included NAM and control patients had unilateral cleft lip and palate, and were from Ecuador. Patients with congenital syndromes other than cleft lip and palate (CLCP) affecting facial appearance were excluded. Twelve independent raters including 3 surgeons, 3 orthodontist, 3 pediatric dentists, and 3 medical students rated the photographs. As a modification to CARS, raters were also asked to assess scar quality utilizing a previously developed scar subtype scoring system.

Results: Of the 189 patients treated with NAM in Guayaquil since 2012, 96 patients had long-term follow-up, and 27 patients with 34 photographic time points qualified for inclusion, 15 (55.56%) male, 12

(44.44%) female. The average time post primary cleft lip repair was 2.19  $\pm$  1.65 months (range: 0.98-7.37 months). Matched to the patients who had received NAM were 31 control patients with 34 photographic time points, 26 (83.87%) male, 5 (16.13%) female with an average time post primary cleft lip repair of 2.19  $\pm$  1.63 months (range: 0.93-6.98 months) The average rating of Nasal aesthetics was significantly lower in patients who had received NAM compared to nasal controls (2.60  $\pm$  1.05 vs 2.82  $\pm$  1.12;  $P\!\!<$  .01).The average rating for lip aesthetics showed similar differences between patients who had received NAM and matched controls (2.23  $\pm$  0.96 vs 2.56  $\pm$  1.07; P< .01). Similarly, the average rating of scar quality was significantly lower among patients who had received NAM (1.82  $\pm$  0.93 vs 2.03  $\pm$  0.89; P< .01).

Conclusions: Using the CARS and a modified scar rating scale, patients who had received NAM were found to have superior nose and lip aesthetic outcomes in comparison to non-NAM controls. This suggests that NAM is not only feasible in an outreach setting, but it is also associated with improved lip, nose, and scar outcomes.

#### 78. Cranial Vault Distraction as a Treatment for Refractory Hydrocephalus

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Background/Purpose: Hydrocephalus is an increased volume of cerebrospinal fluid in the central nervous system due to disturbance in the flow, absorption, or formation of CSF. It affects 3 in 1000 live births in the US. The mainstay of treatment is ventricular shunting. While this has increased survival, shunts can fail or lead to conditions such as slit ventricle syndrome or post-shunt craniosynostosis. If shunting fails, treatment options are limited. Patients may experience headaches, papilledema, and developmental delays. The same principles of cranial vault expansion that are used in treatment of craniosynostosis have been implemented in the treatment of patients with refractory hydrocephalus. However, this technique is invasive and can be limited by the amount of soft tissue coverage available. Cranial vault distraction osteogenesis allows for similar expansion but with less dissection of dura and more controlled expansion of soft tissues. This has not yet been described for treatment of refractory hydrocephalus. We predict that cranial vault distraction is a safe and effective treatment.

Methods/Description: This is a case series of consecutive patients at our institution who underwent posterior cranial vault distraction for hydrocephalus refractory to shunting. Objective outcome measures included lumbar puncture opening pressure, papilledema, head circumference, and emergency department visits or hospital admissions for shunt-related issues. Subjective outcome measures included patient- and parent-reported headaches and improvement in developmental milestones.

Results: Three patients were identified. All patients were 2 or 3 years old at the time of distraction. All patients presented with headaches. Two had elevated LP opening pressures and the third had not been tested. None had papilledema. They all had 3 to 5 ED visits/hospital admissions for headaches or shunt-related problems. One patient had severe developmental delays and was nonverbal. All 3 patients underwent successful craniotomy with placement of 2 or 3 uniplanar distractors. Distraction was completed and devices were subsequently removed 3-months later. All have had resolution of their headaches. The nonverbal patient has had drastic improvements in communication. One patient was readmitted for a pin site infection and one had wound breakdown treated with outpatient wound care. There have been no other ED visits or hospital admissions.

Conclusions: In our series, cranial vault distraction has preliminarily proven to be a safe and effective treatment for refractory and symptomatic hydrocephalus. It uses principles of cranial vault expansion but with a more favorable safety profile in terms of dissection and soft tissue management. Though it does require a second operation for device removal, we feel it is a beneficial technique and plan to continue this series.

### 79. Use of Patient-Specific Biodegradable Implants in Pediatric Craniofacial Surgery

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Background/Purpose: The premature fusion of calvarial sutures often leads to severe deformities of the skull that affect the fronto-orbital region. We present for the first time a 3D planning tool that allows the manufacture of a corresponding osteotomy and drill guide, as well as a resorbable implant for the fronto-orbital advancement procedure (FOA). The implant, optimized for minimal material usage, guarantees an anatomically correct reconstruction of supraorbital margins and correction of the forehead, by taking overcorrection into consideration.

Methods/Description: A 7-month-old female presented with premature right unilateral synostosis of the coronal suture and corresponding deformity of the skull. Preoperative MRI data was imported into iPlan software (version 3.0.5; Brainlab, Feldkirchen, Germany) and further processed. 3D osteotomies for a standard FOA procedure were performed. A resorbable patient specific implant (PSI) was built by the KLS Martin Group (Tuttlingen, Germany) consisting of poly(D, L-lactic acid) (PDLLA). FOA was performed and osteotomy lines and drill holes were transferred according to the cutting guide with a piezoelectric device and a burr. Each bony piece was then reconfigured in the appropriate place within the resorbable PSI.

Results: The above-described technique for reconfiguring a deformed skull using preplanning information in conjunction with patient-specific osteotomy and a drill guide, as well as a PSI with predrilled fixation points, allows for computer-assisted surgery and quality control in FOA procedure. The targeted reconfiguration in our case was time-saving. After surgery, the patient experienced no infection, inflammatory response, or implant-related problems, such as instability or deformity. During the 12-month follow-up, stereophotogrammetry showed further growth of the cranium and normal development of the child; in particular, the forehead was smooth.

Conclusions: FOA including reconfiguration of the forehead in craniosynostosis can be performed with patient-specific resorbable implants, based on computer-assisted preoperative planning tools. The combination of techniques described above potentially represent a paradigm shift. While conventional techniques are limited to reshaping the skull into a harmonious shape using fewer, but larger, bony fragments, the technique presented here allows for preplanned reconfiguration of the skull using multiple fragments covered by a resorbable PSI, with adequate stability and no aesthetic compromise.

#### 80. Unique Techniques Utilizing Rib Grafts for Mandibular Reconstruction in the Pediatric Population

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Background/Purpose: Mandibular reconstruction requires a grafted segment of sufficient height and stability to support subsequent dental implant placement. Double barrel graft techniques and distraction osteogenesis after mandibular reconstruction are 2 methods used to achieve this. However, as reported in the literature, these methods have only been seen in cases using fibular grafts. Rib grafts are reported to have not only a lower donor-site morbidity when compared to fibular grafts but they also provide adequate defect coverage in children. As such, we propose their use with a double barrel technique and with distraction techniques.

Methods/Description: Four pediatric patients underwent mandibular mass resection and reconstruction using rib grafts. The first patient had a single rib graft placement only and is included for comparison with the remaining 3 patients. Of these 3, one patient underwent single-rib graft placement with subsequent vertical rib distraction while in the remaining 2 patients, we utilized a double-barreled rib technique stacked horizontally in one patient and vertically in the other.

Results: From March 2018 to May 2019, 3 patients with the average age of 11 presented with mandibular tumors or tumor-like lesions. The lesions were resected and all patients underwent immediate mandibular reconstruction with rib graft. Due to postoperative wound complications, the rib graft was completely removed in one patient. One of the 3 patients has since had dental implants placed which have fully osseointegrated and is awaiting final dental prosthesis. The remaining patient is currently in the healing phase; once full bony union is complete, dental implants will be planned.

Conclusions: Rib grafts are often thought to be too thin and short to completely reconstruct mandibular defects. However, autogenous rib graft has proved to be an excellent source of bone in the pediatric population. Along with having the benefit of lower donor-site morbidity compared to a free fibula or iliac crest, rib is also extremely versatile. Mandibular distraction osteogenesis (MDO) is an available and viable treatment option when the resultant height of rib graft is insufficient. Utilization of MDO has the unique advantage of stretching the soft tissue envelope while allowing formation of new bone, facilitating the placement of implants and oral rehabilitation in pediatric patients.

# 81. Combining Virtual Surgical Planning and Innovation to Treat Unilateral Lambdoid Craniosynostosis: The Sand-Dollar and Staves Technique

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Background/Purpose: Occurring once in every 40 000 live births, unilateral lambdoid synostosis (ULS) is the rarest form of craniosynostosis, representing 1% to 4% of all craniosynostosis cases. Due to the associated cranio-caudal shift seen in ULS, surgical correction is technically challenging from a morphological standpoint. While numerous techniques have been proposed that attempt to provide optimal postoperative morphology, surgical correction is often unable to correct every aspect of asymmetry and often leads to unsatisfactory postoperative results. Herein, we present a novel "Sand-Dollar and Staves" technique, aided by virtual surgical planning, for the repair of ULS.

Methods/Description: A retrospective chart review was performed identifying all patients diagnosed with ULS at Texas Children's Hospital. A zigzag coronal incision is performed, and an anteriorly based

pericranial flaps are elevated, exposing the calvarium. Prefabricated cutting guides are placed and the calvarium is marked. To treat the occipital flattening on the side with the synostosis, a wedged suturectomy is performed with additional barrel staves to allow for compensatory vault expansion. A large circle centered over the bulging of the occipitoparietal region on the contralateral side is cut out above the open lambdoid suture leaving the open sutures intact. This piece is then barrel staved in a radial fashion, leaving only the center intact and creating a "sand-dollar" appearance. This circular disk is flattened and trimmed while keeping the entire construct in one piece. The modified sand-dollar is then fixed back onto the calvarium using an absorbable plating system. While gentle pressure is applied to the sand-dollar piece as it is being secured, the flattened ipsilateral side with the barrel staves demonstrates compensatory filling. Postoperative results were evaluated by 6 board-certified craniofacial surgeons using the Whitaker Classification.

Results: Four patients underwent surgical correction with the Sand-Dollar & Staves technique. The procedure was performed at mean age of 11.7 months. The mean operative time was 2.5 hours. Intraoperative blood loss ranged from 50 to 100 mL with no blood transfusions required. Total hospitalization time was 2 to 3 days. No postoperative complications were encountered. When the cranial shape was assessed subjectively by the parents, an acceptable aesthetic outcome was achieved in all cases. Mean Whitaker Classification scores ranged from 1 to 1.5. Patients were followed for a mean of 10 months.

Conclusions: The Sand-Dollar & Staves procedure is a novel, single-stage approach for the management of ULS with decreased operative time, blood loss, and hospital stay with satisfactory aesthetic outcomes. This approach offers a valuable alternative that meets the functional and aesthetic goals for cranial correction of lambdoid synostosis with improved safety profile in comparison to previously utilized open techniques.

#### 82. Revisiting the Use of Iliac Chondro-Osseous Grafts for the Multi-Operated Cleft Nose

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Background/Purpose: The cleft nasal deformity is one of the more challenging aspects of cleft care, made so because of the complex intrinsic and extrinsic forces that act to not only create the deformity but also to maintain it. Inherently the cleft nose is marked by a deficiency and deformation of local tissues, which is even more so pronounced in the multioperated nose. A number of surgical techniques and approaches have been developed and utilized for correction of the cleft nasal deformity which utilize both local and regional autogenous tissues. However, in situations where there is a paucity of both local and regional tissues, as might be the case in a multioperated cleft nose, the reconstructive effort becomes complicated. The iliac chondro-osseous graft is an ideal graft for predictable framework correction in patients who lack or have pathologic local tissue to work with.

Methods/Description: We will present our technique for harvesting and utilizing split chondro-osseous iliac crest grafts for restoring nasal projection and support. Pre and postoperative evaluation and images will be presented to highlight the utility and fidelity of this technique. A discussion of outcomes including possible complications will also be presented.

Results: Select patient cases will demonstrate the reliability and utility of the iliac crest chondro-osseous graft. Pre- and Postoperative photographs will highlight the effect of this graft. Complications, although rare, may occur and will be discussed.

Conclusions: The use of the iliac crest chondro-osseous graft for cleft rhinoplasty provides reliable, durable, and predictable results especially in the multioperated nose. This is a simple technique which can be applied across the spectrum of cleft rhinoplasty and can be adopted and added to the armamentarium of any cleft surgeon.

#### 83. Deep Learning: Detection of Craniofacial Abnormalities Using Standard Clinical Photography

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Background/Purpose: Development of an objective algorithm to diagnose and assess craniofacial conditions has the potential to facilitate early diagnosis, especially for care providers with limited craniofacial expertise. Deep learning, a branch of artificial intelligence, is capable of producing algorithms which automatically analyze and categorize disease without human assistance. Convolutional neural networks (CNN) have excelled in medical image analysis to automatically classify disease. In this study, we developed CNN models to detect and classify nonsyndromic craniosynostosis (CS) and cleft lip (CL) conditions using 2D images alone.

Methods/Description: We created an annotated data set of labeled CS (normal, metopic, sagittal, and unicoronal) and CL (normal, unilateral, bilateral) conditions using standard clinical photography from the image repository at our center. We extended this data set by adding photographic images of children with craniofacial conditions from the Internet. The web image diagnoses were validated by 2 craniofacial surgeons. A total of 1077 images were used in this study. We divided the sets into 67% training and 33% testing groups and processed the images for consistency in view, size, and quality. We developed a CNN model using a pretrained ResNet strategy to classify the data as normal, metopic, sagittal, or unicoronal, and a pretrained AlexNet strategy to classify bilateral cleft or unilateral cleft. Once trained, we tested the model for accuracy.

Results: For CL, the validation and testing accuracies were 92.2%. Case detection for CL was 67.2% for bilateral and 88% for unilateral. For all CS types, overall testing accuracy was 90.6%. The sensitivity and precision were 100% and 60% for normal, 100% and 100% for metopic, 93% and 100% for sagittal, and 70% and 100% for unicoronal, respectively. Misclassified images included those with features proving challenging for the algorithm, such as imperfect views or obstructive characteristics.

Conclusions: Overall, the CNN model performed with promising accuracy. These results support the idea that deep learning has a role in diagnosis of craniofacial conditions. Using standard 2D clinical photography, such systems can provide automated screening, detection, and diagnostic confirmation of these conditions, minimizing radiation and financial cost to patients and families. In the future, ML may be applied to prediction and assessment of surgical outcomes, or as an open-source remote diagnostic resource.

### 84. Primary Vermillion Border Correction in the Incomplete Cleft Lip Repair

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Background/Purpose: Vermillion border irregularity can be found after primary unilateral cheiloplasty. Vermillion border excess is especially found after incomplete cheiloplasty. Since 2017, primary vermillion border correction in the incomplete cleft lip was performed. This retrospective study analyzed the anthropometric outcomes of Chang Gung modification rotation advancement cheiloplasty for both the complete and incomplete cleft lip groups and compare the vermillion symmetry.

Methods/Description: From 2014 to 2018, there are totally 203 patients who underwent unilateral cheiloplasty. One hundred eighty-seven patients (100 incomplete vs 87 complete) have at least 1 year postoperative adequate medical and 1:1 photographic records. These 2 groups were measured as to vermillion area, vermillion height, vermillion width, and Lateral lip length, using the ratio of the cleft to noncleft side. The incomplete groups were also divided as to whether the vermillion border correction or not.

Results: The vermillion height were found higher in these 3 groups (complete vs incomplete vs incomplete with primary correction), but much higher (P < .05) in the incomplete cleft lip group without primary correction. The vermillion width, and lateral lip length were found shorter in the incomplete cleft lip whether or not correction. The vermillion area is almost equal in groups of complete and incomplete lip without correction. The vermillion area of the incomplete lip with primary correction is a bit smaller than other groups.

Conclusions: The cleft side vermillion border excess is often found after the incomplete cleft lip repair. Primary vermillion border correction is suggested in unilateral incomplete cheiloplasty for better symmetry.

### 85. A Pilot Study to Determine the Utility of Genetic Consultation in Multidisciplinary Cleft and Craniofacial Clinic

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Background/Purpose: The role of genetic consultation includes counseling, offering genetic testing, reviewing overall pediatric care, and identifying referrals as indicated. Patients with cleft lip/palate (CLP) may especially benefit from such evaluation, but these potential advantages have not been well quantified. The purpose of this study was to document the beneficial effects of genetic consultation as a standard evaluation for all patients seen at the Cleft and Craniofacial Center (CCC) at our institution.

Methods/Description: The authors retrospectively reviewed 4 random days of CCC team evaluations at our institution between October 2018 and December 2018. A total of 20 patients were reviewed. All patients were provided with a genetic consultation. There were 9 cases of likely isolated CLP (45%), 8 cases of probable syndromic CLP (40%), 2 cases of Robin sequence (RS) with cleft palate (CP; 10%), and 1 case of craniofacial microsomia (5%).

Results: All of the patients were seen by a board-certified geneticist and received counseling regarding likely etiology and recurrence risk. The 2 patients with RS had normal development and were offered Stickler syndrome testing. All the syndromic patients were offered genetic testing, other laboratory screening considered, or referral recommended. Examples included: (1) Patient with Down syndrome who missed several years of proper screening. Standard laboratory testing was ordered, and he was referred for additional therapies. (2) Patient with CLP, global delay, arthrogryposis, and bony abnormalities. Laboratory screening, including renal function test and a kidney

sonogram showed a pelvic kidney. Genetics recommended urology consultation, which ultimately ruled out bladder pathology and led to a treatment plan. In addition, she began physical therapy for worsening arthrogryposis of her hands. (3) Patient with CP and short stature was discovered to have levo-scoliosis and an unusual gait, and was referred to orthopedic surgeon. An MRI of the spine has now been scheduled, and surgical options may be considered.

Conclusions: Based on the high frequency of complicated and syndromic cases (40%) seen in our CCC, and the positive contribution of the genetic evaluation on medical management, this study supports the inclusion of a genetic evaluation as part of the standard of care for patients with cleft and craniofacial anomalies. We believe that genetic evaluation as part of a CCC captures patients who may otherwise be lost to follow-up provides an opportunity to reinforce genetic counseling and optimize medical care for complex and syndromic patients.

### 86. A Qualitative Assessment of Bone Mineral Density in Individuals With Hemifacial Microsomia; A CBCT Study

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Background/Purpose: Hemifacial microsomia (HFM) is a craniofacial condition characterized by a disturbance in the development of the structures of the first and second branchial arches. It can be manifested in different degrees of severity. HFM is the second most common congenital craniofacial condition after cleft lip and palate with prevalence of 1 of every 4000 to 5600 live births. Facial asymmetry of the mandible alone or in conjunction with the maxilla is the main characteristic feature of HFM. While it is understood that the quantity of the bone on the affected side of the mandible is compromised, few studies have evaluated the quality of the bone among this cohort of patients. The aim of this study is to utilize cone-beam computed tomography (CBCT) to compare the bone mineral density on the affected versus nonaffected side, among individuals with HFM.

Methods/Description: This retrospective study included 9 individual with HFM (2 females and 7 males, with mean age of 11 years and 10.8 years respectively). Pretreatment CBCT volumes were imported into Invivo5 software, which was used to measure the bone density through Hounsfield Units (HU) in 3 regions of the mandible; inferior to the lower lateral incisors, inferior to the first molar, and at the ramus inferior to the sigmoid notch. All measurements were done at 10 mm superior to the inferior border of the mandible. Each region was measured at the buccal cortical bone, lingual cortical bone, and cancellous bone, yielding 9 areas of measurements on each side. The densities on the right and left sides were compared by Wilcoxon signed-rank test.

Results: Overall, the bone mineral density on the affected side tended to show slightly lower values when compared to the nonaffected side. Differences ranged from 44 HU at the cancellous bone of the first molar region to 234 HU at the buccal cortical plate of the ramus region. Differences were only statistically significant at the ramus region for the buccal (P=.008) and lingual (P=.008) cortical plates and at the lower incisor region at the buccal cortical plate (P=.018) and cancellous bone (P=.038). The differences, however, did not seem to be clinically significant.

Conclusions: Among patients with HFM, bone mineral density on the affected side seems to be slightly lower than that on the unaffected side. The differences are emphasized at the ramus and anterior body of the mandible. Findings from the current study show that the quality of

bone on the affected side may be slightly reduced, but not to clinically significant levels. Future studies with larger sample sizes are needed for better understanding of the quality of bone among patients with HFM.

### 87. Administration of Steroids in Patients Undergoing Orthognathic Surgery Is Associated With 90-Day-All Cause Readmissions

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Background/Purpose: Perioperative systemic steroids are broadly used in orthognathic surgery to prevent postoperative complications, but it is unclear whether this practice is beneficial and concerns about potential side effects have been raised. The purpose of this study is to examine the association of steroid use during orthognathic surgery and postoperative outcomes including major complications and 90-day all-cause readmission.

Methods/Description: A multicenter retrospective cohort study was implemented utilizing the Pediatric Health Information System (PHIS) database, a collaborative of 45 hospitals. The study population was composed of patients 13 to 21 years of age at the time of their orthognathic surgery, identified by ICD-9 procedure codes 76.62-76.68, between January 1, 2004, and September 30, 2014. Steroid exposure was defined as having been billed for the generic drug code for Dexamethasone (154035) at any time for up to 7 days from the surgery date. The outcome variables included major complications (bone debridement and hardware removal) identified by ICD-9 codes 76.01, 76.97, and 78.69 and 90-day all-cause readmission. Unadjusted and adjusted random-intercept logistic regression models were utilized to assess the association between steroid exposure and these outcomes.

Results: The sample included 5405 patients, with a mean age of 17.1  $\pm$  1.68 years and 61% were female. Seventy percent were of white ethnicity. The most common procedures were Le Fort I alone (39%), followed by bimaxillary surgery (23%), then mandibular osteotomy alone (20%). Fifty-three percent of patients were exposed to steroids, with 19% exposed only on the day of surgery, 27% on the day of surgery and after, and 7% after the day of surgery. Major complications occurred in 8.6% of patients and 90-day all-cause readmission was 11.2%. In models adjusting for age, sex, race, procedure, length of stay, and hospital variation, the odds-ratio of steroid exposure was 1.23 (95% CI = 0.96-1.59) for major complications and 1.50 (95% CI = 1.20-1.88) for 90-day all-cause readmission.

Conclusions: The administration of steroids in patients undergoing orthognathic surgery is significantly associated with increased odds of 90-day-all cause readmission. Association between steroid use and complications and readmission may represent a causative relationship or a surrogate marker of medical conditions predisposing patients to complications. Large, administrative databases such as the PHIS database do not allow determination of this, but future prospective study is warranted.

### 88. Advanced Genioplasty for Airway Constriction in the Upper Respiratory

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Background/Purpose: Corrective jaw surgery often required as patient grow even to improve sleep apnea after maxillofacial surgery. We

would like to introduce 5 cases who undertook advanced genioplasty to improve the airway condition.

Methods/Description: The objects are 2 patients with Treacher-Collins syndrome, 2 patients with Crouzon syndrome, 1 patient with Goldenhar syndrome. There were 3 males and 2 females; their ages ranged from 9 to 23 years (mean: 16.2 years). In the 5 cases, we compared the preoperative condition with postoperative one by the apnea index, airway width by CT and cephalogram, and subjective symptoms.

Case Reports: Case 1: A 23-year-old female diagnosed with sleep apnea with micrognathia due to Treacher-Collins syndrome. An interposition genioplasty and segmental osteotomy of the mandible were performed. Case 2: A 23-year-old male diagnosed with severe sleep apnea due to Crouzon syndrome. The patient had previously undergone Le Fort III osteotomy and distraction (twice), tongue reduction, and sliding genioplasty. The airway constriction had improved after the previous genioplasty. However, it recurred with increase of bodyweight, and so nighttime CPAP management was resumed. Hence, distracting genioplasty was performed to obtain enough anterior movement. Case 3: A 9-year-old female with micrognathia due to Goldenhar syndrome. The patient underwent tracheostomy to correct airway obstruction at 2 months of age, and had recently undergone bilateral mandibular body distraction and reconstruction of the right temporomandibular joint by a rib and costochondral complex graft. Interposition genioplasty was then performed by adding a rib bone graft to create further airway expansion.

*Results*: Airway constriction and subjective symptoms were improved postoperatively in all cases. 4 cases were improved apnea index. The patient of case 3 no longer had respiratory distress on attaching the voice prostheses.

Conclusions: The method of genioplasty with geniohyoid muscle advancement as a treatment for sleep apnea was first reported by Riley et al. in 1989. We confirmed the usefulness of this method not only for cases of upper airway constriction without maxillary hypoplasia, but also for sleep apnea syndrome that had recurred after achievement of dental articulation. Furthermore, distraction genioplasty and rib bone graft will help to obtain great effect for severe airway constriction in micrognathia.

## 89. Analysis of the Cleft Lip and Palate Section of the Plastic Surgery In-Service Training Exam: A Strategic Approach to Guide Resident Studying

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*Background/Purpose*: Our purpose was to analyze the cleft lip and palate related questions on the Plastic Surgery In-Service Training Exam over a 10-year time span (2010-2019) to better guide a more systematic approach to studying for this section of the exam.

Methods/Description: Using the American Council of Academic Plastic Surgeons In-Service training exam website, we systematically went through the listed cleft lip and palate questions, breaking them down by subject matter tested and question taxonomy. We then analyzed the most commonly tested subject matter along with question taxonomy in order to provide insight into how this section of the exam was tested.

Results: Depending on the year, up to 4% of the 250 questions may be directly cleft lip and palate related. The most commonly tested topic in this section was velopharyngeal insufficiency (26%), followed by anatomy and classification of cleft lip and palate (18%). In regard to the various topics, treatment knowledge was tested most often (44%) followed by knowledge of the condition (30%). The majority of

questions (74%) strictly involved memorization or recall of the topics tested, whereas 26% required a higher-level of thinking such as making a diagnosis or providing the next best step in management from an unknown diagnosis.

Conclusions: Evaluation and analysis of the cleft lip and palate portion of the Plastic Surgery In-Service Training Exam will more systematically guide resident preparation and comprehension of the tested material.

#### 90. Cartilage Grafting Outcomes in Intermediate and Definitive Cleft Rhinoplasty

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Background/Purpose: In subjects with cleft lip and palate, the ideal surgical techniques and timing to correct nose deformity remain controversial. Our study compares the number of revisions and cartilage grafts required to achieve satisfactory outcomes in intermediate and definitive rhinoplasty age groups.

Methods/Description: A retrospective chart review of a single surgeon's experience was conducted. Subjects were stratified into 2 groups: "intermediate rhinoplasty" = cleft rhinoplasty performed during the ages of mixed dentition (5-13 years) and "definitive rhinoplasty" = cleft rhinoplasty performed after age 13. Data collected included number of surgeries, cartilage grafts, and complications. Statistical analyses were performed using  $\chi^2$  and student t tests in GraphPad Prism 8. A P value of < .05 was used as a threshold for determining statistical significance.

Results: A total of 46 subjects with a cleft nose deformity underwent 65 rhinoplasty procedures. The intermediate group included 18 males (85.7%) and 3 females (14.3%). The definitive group included 16 males (64.0%) and 9 females (36.0%). Twenty-one subjects in the intermediate group had 34 total operations, and 25 subjects in the definitive group had 31 total operations. In the intermediate group, 6 subjects (28.6%) underwent a total of 6 rhinoplasty procedures that required cartilage grafting, whereas 15 subjects (71.4%) underwent a total of 26 rhinoplasty procedures that did not require cartilage grafting. In the definitive group, 18 subjects (72%) underwent a total of 21 rhinoplasty procedures with cartilage grafting, while 7 subjects (28%) underwent a total of 9 rhinoplasty procedures where cartilage grafting was not required. The difference in the number of cartilage grafts performed between the 2 groups was statistically significant (P = .008). Regarding cartilage donor sites, there were 14 conchal cartilage, 6 septal cartilage, 2 lower lateral cartilage cephalic trim, 2 nasal dorsal hump cartilage grafts, and 1 from a previous malpositioned graft. In addition, 1 subject who also underwent simultaneous alveolar bone grafting utilized iliac cartilage cap as the cartilage donor site. There were no complications observed with the cartilage donor sites utilized.

Conclusions: Cartilage grafting is a useful and valuable tool in both intermediate and definitive cleft rhinoplasty. In our experience, conchal cartilage is the preferred donor site with equivalent outcomes among all cartilage graft options. Definitive cleft rhinoplasty required more cartilage grafting procedures to achieve satisfactory outcomes, which may relate to the severity of their deformity. Furthermore, we observed that intermediate rhinoplasty is effective, has a low risk of complications, and occasionally prevents the need for further rhinoplasties during the definitive age period.

#### 91. Characteristics and Academic Productivity Among Pediatric Plastic Surgeons in the United States

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Background/Purpose: The characteristics that predispose plastic surgeons to a career in pediatric plastic surgery remains unclear. Therefore, the aim of this study is to analyze the characteristics of current pediatric plastic surgeons and to determine their academic productivity.

Methods/Description: Pediatric plastic surgeons were identified through an Internet search of all academic children's hospitals affiliated with an Accreditation Council for Graduate Medical Education (ACGME) accredited integrated or independent plastic surgery program. Demographics, training background, institutional and leadership positions, and academic productivity were determined.

Results: Initial comparisons of surgeon productivity were completed using Wilcoxon Rank-Sum Analyses. Measurements of correlation for age, residency graduation year, program size, NIH funding, total publications, H-index, and total citations were made using the Pearson Correlation Coefficient with P value measuring a null hypothesis of zero correlation. Determination of H-Index, total publications, and total citations by years of post-residency experience was made using the frequency of these publication measurements per-year. A total of 304 pediatric plastic surgeons were identified. The average age is 48.2, with 57.9% (n = 176) of the cohort completing residency before 2009. The majority of pediatric plastic surgeons were white (n = 217,71.8%) males (n = 235, 77.6%). Clinical fellowships were completed by 86.8% (n = 263) of the cohort, with craniofacial surgery (n = 181, 59.7%) being the most common followed by hand surgery (n = 54, 17.8%). Among the cohort, 41.1% had clinical fellowship training at 10 top institutions, with the top 7 most represented programs being University of Pennsylvania (n = 19, 6.2%), University of California-Los Angeles (n = 16, 5.3%), Harvard University (n = 15, 4.9%), University of Toronto (n = 14, 4.6%), New York University (n = 14, 4.6%), University of Washington (n = 12, 3.9%), and University of Pittsburgh (n = 10, 3.3%). Among the cohort, 25.7% (n = 78) held leadership positions within their institutions (fellowship or residency directors, and chiefs/chairs). A significant higher academic productivity was found among research fellowship-trained surgeons, chiefs of pediatric plastic surgery, fellowship directors, and members of departments of plastic surgery. Those who completed an independent residency had a significant higher H-index and number of citations.

Conclusions: Pediatric plastic surgery is represented by surgeons of diverse training background, highlighting that there are alternative pathways to becoming a pediatric plastic surgeon. An elite cohort of programs has trained the most pediatric plastic surgeons and produced the most chiefs. Lastly, high academic productivity was found to be correlated to certain demographic and leadership variables highlighting its impact on career advancement.

#### 92. Craniosynostosis: Risk Factors for Delayed Primary Surgery and Predictors of Early Operation

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Background/Purpose: Patients with craniosynostosis often undergo delayed primary surgery, defined as primary operations performed after 12 months of age, which places them at a higher risk of complications compared to patients who are treated earlier. Past studies have investigated risk factors related to delayed presentation for craniosynostosis management. Given the wide variability between presentation and surgery, however, patient age at time of surgery would be a better metric for assessing these risk factors for delay. The purpose of this study is to elucidate risk factors for delayed surgical correction of craniosynostosis and to identify factors associated with younger patient ages at the time of operation.

Methods/Description: Retrospective chart review was conducted from November 2011 to September 2018 on patients with a documented diagnosis of craniosynostosis presenting for primary surgical management. We analyzed 19 risk factors potentially associated with delayed primary surgical intervention. A Wilcoxon rank sum test was used to determine P values for comparisons between patients in different age cohorts at time of surgery. Logistic regression was used to model the relationship between potential risk factors and patient age at surgery. Odds ratios (ORs) and 95% confidence intervals (CIs) were generated for each variable. A P value of < .05 was considered statistically significant.

Results: Of the 208 patients evaluated for craniosynostosis management, 123 (59.1%) met final inclusion criteria. The majority of patients were male (68.3%). We found that a higher percentage of white patients received surgery before 12 months of age compared to nonwhite patients (58.5% vs 41.5%, P = .046). Significant differences in patient age at time of surgery were also noted based on type of craniosynostosis (P = .004); patients with sagittal craniosynostosis were more likely to receive surgery before 12 months of age, whereas patients with unilateral coronal and multisuture craniosynostosis were more likely to receive surgery after 12 months of age. Patients with syndromic craniosynostosis and congenital anomalies were significantly more likely to receive surgery after 12 months of age (P =.019; P = .007). Logistic regression confirmed that syndromic status and type of craniosynostosis were highly correlated with age at surgical intervention (OR = 0.11, CI = 0.01-0.94, P = .04; OR = 0.20, CI = 0.08-0.50, P < .01).

Conclusions: While previous studies have focused on risk factors for delayed presentation of patients with craniosynostosis, we aimed to explore the risk factors for delayed surgical intervention. We found that nonwhite patients with syndromic, unilateral coronal, or multisuture craniosynostosis and comorbidities were more likely to undergo surgical intervention after 1 year of age. Understanding these risk factors can drive evidence-based interventions designed to promote earlier presentation conducive to safer, less invasive surgical treatments.

## 93. Does Size Actually Matter? A Comparison of Velopharyngeal Gap Size from Imaging and Universal Parameters for Reporting Speech Scores

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Background/Purpose: Patients with cleft palate can present with disordered resonance due to velopharyngeal insufficiency (VPI). Speechlanguage pathologists (SLPs) can rate hypernasal resonance using the Universal Parameters for Reporting (UPR) speech outcomes, with a 4point scale for hypernasality. Structurally, velopharyngeal gap size can be quantitatively measured using nasopharyngoscopy or videofluoroscopic palatal motion studies (VF-PMS). We aimed to evaluate the correlation of UPR versus quantitative imaging findings.

Methods/Description: A retrospective review was conducted of all patients with cleft palate at a single institution who were diagnosed with VPI from 1999 to 2014. All patients underwent perceptual speech evaluations using the UPR by a trained SLP and velopharyngeal imaging by either nasopharyngoscopy, VF-PMS, or both to evaluate velopharyngeal function and velopharyngeal gap size. The speech sample was customized to elicit maximal velopharyngeal function at various levels, including isolation, word, and phrase level as deemed appropriate by the SLP. Speech sound errors consisting of compensatory misarticulations were avoided during imaging. During VF-PMS, multiview imaging measured the distance between the velum to the posterior pharyngeal wall during sustained /i/ and /s/. The radiologist recorded images obtained at maximum velar movement to analyze maximum closure and documented millimeter measurement of velopharyngeal gap findings. During nasopharyngoscopy, the SLP obtained quantitative measures based on en view findings. Correlation was assessed using Spearman rank correlation coefficient.

Results: Of 113 patients who underwent speech evaluation by an SLP, 98 (87%) had a VF-PMS, 54 (48%) had nasopharyngoscopy, and 39 (35%) had both. Correlation of UPR hypernasality scores with gap size during sustained /I/ and /s/ during VF-PMS was 0.39 and 0.46, respectively. Correlation of UPR hypernasality scores with gap ratio from nasopharyngoscopy was 0.39. Correlation of UPR speech understandability and gap size during sustained /I/ and /s/ was 0.24 and 0.32, respectively. Correlation of UPR speech understandability and gap ratio from nasopharyngoscopy 0.34.

Conclusions: Diagnosis of hypernasal resonance is dependent upon perceptual evaluation such as the UPR, but our study demonstrated low correlation with the physical gap size seen on VPI imaging. Given the ease of use and limited impact to patients, it remains a screening tool to identify patients who may require further evaluation of resonance to determine candidates for secondary speech surgery.

#### 94. Early Mandibular Distraction in Pruzansky III Craniofacial Microsomia: Matthews Device to Avoid Bone Grafting

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Background/Purpose: Early mandibular distraction is the preferred treatment of Pruzansky III craniofacial microsomia with clinically significant airway obstruction. While the primary goals of distraction include resolution of airway obstruction and avoidance of tracheostomy, secondary considerations include protection of the temporomandibular joint (TMJ) and preservation of future reconstructive options. Current standard approaches of early mandibular reconstruction with a free bone flap followed by distraction are often complicated by ankylosis and limit future reconstruction. As an alternative to early reconstruction, we present the case of a 4-year-old patient with Pruzansky III hemifacial microsomia secondary to Goldenhar syndrome whose severe airway obstruction was successfully managed with the unilateral placement of a Matthews device coupled with bilateral mandibular distraction.

Methods/Description: A 4-year-old female with a history of Goldenhar syndrome with associated right-sided Pruzansky III craniofacial

microsomia of the mandible presented with progressively worsening, severe obstructive sleep apnea and increasing nighttime oxygen requirements. Her apnea hypoxia index (AHI) at presentation measured 42. Surgical management was indicated in an attempt to avoid tracheostomy. Following multidisciplinary discussion, it was decided to forego traditional early mandibular reconstruction in favor of a novel approach. Bilateral mandibular distractors were placed, and to avoid driving the atrophic condyle into the TMJ, a Matthews device was secured to the right cranium and mandible.

Results: The patient tolerated distractor placement, distraction, and subsequent hardware removal without complication. Tracheostomy was completely avoided, the TMJ was successfully protected by the Matthews device, and no ankylosis was observed. Postdistraction, the patient's AHI decreased to 8 and her nighttime breathing was noted to be markedly improved. The patient is currently doing well 13 months postdistraction. We anticipate that she is unlikely to require further surgical intervention until achieving skeletal maturity, when she will be able to undergo custom joint and ramus implant placement.

Conclusions: Our case describes a novel approach to early mandibular distraction in children with Pruzansky III craniofacial microsomia complicated by severe airway obstruction. This approach offers the advantage of tracheostomy avoidance without the increased comorbidities associated with free bone grafting. Our findings suggest that simultaneous MDO coupled with placement of a unilateral Matthews device is an effective and safe alternative to address mandibular-based airway obstruction in the setting of Pruzansky III craniofacial microsomia.

## 95. Effects of Lexical Characteristics of Sentence on Intelligibility Scores of Children With Velopharyngeal Insufficiency

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Background/Purpose: One of the factors affecting intelligibility of speech aside from the acoustic signal is speech stimuli characteristics like length and complexity of speech stimuli, and also type of words in the stimuli. If different type of words and sentence complexity result in comparable speech judgements, then the choice of speech material when evaluating cleft palate speech could be decided on which tasks are the easiest for both the clinician/researcher and the child.

Methods/Description: Ten children with velopharyngeal insufficiency (VPI) were recorded while repeating randomly selected sentences from the Hearing in Noise Test (HINT). These audio recordings were then utilized by 70 naïve listeners in a transcription task to obtain speech intelligibility scores. The stimulus material was further analyzed to provide information regarding certain sentence characteristics that could potentially influence speech intelligibility scores (ie, total number of words per sentence and ratio of function words to content words).

Results: Intelligibility scores were calculated as the number of words correctly identified (75.2%-98.45%). Multiple regression analysis revealed that the model predicted the outcome significantly  $F_{2,1397} = 9.44$ , P < .001. Increased number of words per sentence was significantly associated with increased intelligibility score (B = .025, t = 2.97, P = .003) but the ratio of function/content words did not significantly contribute to variance.

Conclusions: The findings suggest that in speakers with VPI with high intelligibility scores, the length of sentence plays an important role in

the degree of understandability. The longer the sentence, the probability of being understood is higher.

### 96. Evidence-Based Cleft Care in Lower Resource Settings

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Background/Purpose: Most literature constituting the evidence base for cleft practices originates from centers in higher resource countries. However, implementation of care protocols from higher resource settings (HRS) to lower resource settings (LRS) is not always feasible or appropriate given unique social, cultural, economic, and logistic demands. The purpose of this study was to systematically review evidence for cleft management protocols appropriate to LRS.

Methods/Description: Systematic review of the literature utilizing PubMed and the Cochrane Database was conducted following PRISMA guidelines. The initial search yielded 4031 studies. After abstract review, 44 studies underwent full text review. Inclusion criteria were defined as relevance to cleft lip and cleft palate surgery, low- or middle-income country setting, and presence of a specific recommendation.

Results: From a preoperative planning standpoint, in regions and practice settings with reduced compliance, performing cleft palate repair first was found to increase compliance to return for cleft lip repair, compared to performing cleft lip repair first and anticipating return for cleft palate repair (Level II, Grade B). From an operative standpoint, combined cleft lip and palate surgery is safe and effective in patients with limited access to care or limited follow-up (Level I, Grade B). Moreover, simultaneous closure of hard palate and cleft lip decreases the incidence of oronasal fistulas (Level II, Grade B). Ambulatory surgery is safe for children without cardiac or respiratory comorbidities undergoing primary lip repair and should be considered in the cases of uncomplicated lip reconstruction (Level II, Grade B). Closure of the nasal floor at the time of cleft lip repair reduces frequency of symptomatic nasolabial fistula and revision lip surgery (Level IV. Grade C). From a postoperative standpoint, antibiotics do not decrease wound complications after cleft lip repair; on the other hand, for cleft palate repair, 5 days of postoperative antibiotics are helpful in reducing postoperative complications (Level I, Grade A). In areas where multidisciplinary care for comprehensive cleft lip and palate care is unavailable, descriptive studies suggest that telemedicine can be effective for preoperative evaluation, as well as for improving postoperative care and speech therapy (Level III, Grade B).

Conclusions: This review revealed practices fundamentally different in LRS, practices with demonstrable cost-savings, and innovative practices originating in LRS. Cumulative consideration of these findings begins to define evidence-based recommendations for cleft care applicable in LRS.

### 97. Genomic Research Testing in a Population With Congenital Craniofacial Malformations

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Background/Purpose: Many craniofacial anomalies have an underlying genetic etiology. The evolving standard of care for many craniofacial disorders includes genetic evaluation and testing, which often

impacts management and prognosis. However, current genetic diagnostic technologies cannot always provide a diagnosis. Not having a specific diagnosis can cause emotional stress and anxiety for families. In addition, newer genetic testing technologies are expensive and often not covered by insurance. Due to these factors, families look for other opportunities that may aid them in their pursuit for an answer. Participation in research can be an option for families. Using next generation sequencing techniques, we query genomes of affected individuals and their family members to identify gene variants that cause craniofacial malformations and use these genetic data to better understand craniofacial conditions.

Methods/Description: Over 500 individuals (patients and family members) have been enrolled in our study. Data reviewed from our study population includes demographics, comprehensive phenotypic information, and samples collected. Samples include blood, saliva, and/or tissue, which are processed for genetic studies and the potential for immortalized cell lines. We reviewed the time and effort involved in sample collection and results disclosure, and ensured that research is performed in a CLIA-approved laboratory for results disclosure. Genes of interest are further studied, including development of animal models.

Results: In a population of over 263 probands who have been offered participation in our study, 173 have been enrolled. Previous reasons for nonconsent or nonapproach include time commitment, lack of interest in research, loss to follow-up, and previous clinical findings that support a diagnosis. Ninety of the probands are male (52%) and most identify as white (87.3%). Probands have a wide variety of disorders including cleft lip, cleft palate, craniosynostosis, dysmorphic craniofacial features, Pierre Robin sequence, and brain malformations. On average, families that enroll in the study have 2.04 individuals who submit samples in addition to the proband, providing additional data that is useful for genomic interpretation. Family members include parents, siblings, aunts, uncles, and grandparents. Close to 100 probands have been sequenced and several articles have been published to share genomic data and findings with the medical and scientific community.

Conclusions: Research genetic testing in a craniofacial population is a reasonable diagnostic option when other testing has been negative. In our experience, families are pleased when we are able to make a diagnosis and understanding when no diagnosis is made. Additionally, research testing generates clinical and scientific questions. This leads to further basic science studies which, hopefully, paves the way for better treatments and expands knowledge of craniofacial disorders.

### 98. Graduate Students' Clinical Self-Efficacy: Impact of an Intensive Cleft Palate Clinical Practicum

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Background/Purpose: It is expected that speech-language pathologists (SLPs) acquire a specific and distinct skill set needed to assess and treat individuals with cleft lip and palate (CLP) in graduate school education and clinical training. However, studies indicate that the majority of graduate programs offer less than a full required course on cleft palate and most graduate students do not have clinical experience with this population. As a result, SLPs may feel insufficiently prepared to provide speech services to individuals with CLP (Bedwinek et al., 2010; Vallino et al., 2008). Expanding clinical practicum opportunities in the area of CLP during graduate studies may increase

graduate clinicians' confidence or self-efficacy and help cultivate competent SLPs in this area. This study aimed to examine self-efficacy as graduate students gained clinical skills during an intensive international CLP clinical practicum and to determine the relationship between graduate students' self-efficacy ratings on skills related to their experience and their performance as judged by clinical supervisors' feedback.

Methods/Description: Eighteen graduate students in the Communication Sciences and Disorders program at Teachers College, Columbia University, participated in the study. Graduate students participated in the CLP clinical practicum for 5 days with healing the children and completed a self-efficacy survey that measured their confidence in evaluation, treatment, and professional behavior at the beginning and end of their CLP clinical practicum. To compare students' self-efficacy ratings with clinical supervisors' evaluation of performance, clinical supervisors answered questions regarding graduate students' performance at the end of their CLP clinical practicum.

Results: A Wilcoxon signed-rank test was utilized to examine pre-to-post practicum changes. Results from the supervisors' questionnaire were analyzed descriptively. A significant pre-to-post practicum group effect (P < .05 and P < .001) was found for all dependent variables under the self-efficacy evaluation and treatment categories. No significant pre-to-post practicum changes were observed for the variables pertaining to professional behavior, except for 2 questions related to the students' ability to display initiative in individual conferences and convey professional information to colleagues. Supervisors' perceptions of students' initial performance and progression were overall consistent with the students' ratings of self-efficacy.

Conclusions: This intensive practicum program for CLP shows significant and positive changes in graduate students' self-efficacy in specialty areas, such as assessment and treatment in CLP. This may be an effective way to increase graduate students' experiences in working with children with CLP.

# 99. Impact of Velopharyngeal Dysfunction on Intelligibility of Speakers With Cleft and/or Craniofacial Disorders Measured by Inexperienced Listeners

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Background/Purpose: Improvement in intelligibility is a primary aim in remediation of velopharyngeal dysfunction. Despite the large influence of intelligibility measurement in cleft and craniofacial care, there is no standard measure of intelligibility that explains the kinds of difficulties that listeners who are inexperienced with resonance disordered speech may encounter. While a correlation can be drawn between intelligibility and the overall presence of velopharyngeal dysfunction, further investigation into how severity of resonance disorders contributes to intelligibility would provide a more informative approach to intervention. Some speech error types including maladaptive articulations and phonological error patterns are known to negatively impact intelligibility more so than other errors. However, exploring the impact of differing severity levels of hypernasality on intelligibility has not yet been explored. This study aims to investigate the effect of disordered resonance on intelligibility of individuals with velopharyngeal dysfunction measured by inexperienced listeners.

Methods/Description: Fifteen individuals with a history of cleft and/or craniofacial disorders and velopharyngeal dysfunction will serve as speakers. The speakers are classified into 5 groups (a. Normal

resonance, b. Mild hypernasality, c. Moderate hypernasality, d. Severe hypernasality, e. Mixed Hypernasality and Hyponasality). Each group speaker group is made up of 3 individuals. Each speaker previously recorded 10 speaking tasks including single words and short phrases that will be analyzed by inexperienced listeners (1. Papa, 2. Popped, 3. Up, 4. Hi how are you, 5. Mama made, 6. Lemon jam, 7. Buy, 8. Baby, 9. A Bib, 10. Hamburger). Listeners will be recruited through the crowdsourcing platform Amazon Mechanical Turk (AMT) and through public online invitations in order to investigate the type of difficulties listeners in the general population might encounter. A web-based platform compatible with AMT, Intelli-turk©, will be used to test the effect of resonance disordered words and phrases on the listeners' ability to understand. Listeners will be required to complete (1) word recognition task by typing "the real word or phrase you think you hear into the text box provided" and (2) direct magnitude estimation (DME) rating of intelligibility by describing "how easy-difficult was this word or phrase to understand." Intelligibility scores will be calculated by an average of at least 25 different listeners' scores for every speaker recording. It is hypothesized that listeners inexperienced with speech disorders can rate speech samples using DME and reflect different levels of intelligibility in agreement with severity of hypernasality and word production accuracy measures.

#### 100. In Search of the Optimal Pain Management Strategy for Children Undergoing Cleft Lip and Palate Repair: A Systematic Review and Meta-Analysis

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Background/Purpose: Adequate postoperative analgesia yields improved surgical outcomes while decreasing length of stay. As no guidelines presently exist, we sought to identify the most effective and safe perioperative pain management strategies for children undergoing primary cleft lip and palate repair. Our outcomes of interest included the requirement for, and time to rescue analgesia in the postoperative period.

Methods/Description: A systematic search of MEDLINE, Embase, Cochrane library, Scopus, and Web of Science databases was conducted. Two hundred thirty unique titles were then assessed by 2 independent reviewers. Pooled analysis of variables was conducted, and data pertaining to common approaches in decreasing postoperative analgesia were compared.

Results: A total of 42 studies involving 583 and 1584 patients undergoing cleft lip and palate repair, respectively, met inclusion for comprehensive analysis and data abstraction. In children undergoing cleft palate repair, palatine block demonstrated the greatest latency to first analgesia ( $F_{8,325}=210,\,P<.0001$ ), but was not associated with a decrease in total opioid consumption relative to alternative pain strategies. In cleft lip, bilateral infraorbital nerve blocks resulted in the greatest increase in latency to first analgesia (215.76 minutes; 95% CI: 83.26 to 448.26, P<.005) and demonstrated a mean decrease in morphine consumption of 0.2 mg/kg/d (95% CI = -0.20 to -0.20, P<.00001). No significant intervention-related complications were identified.

Conclusions: A variety of effective methods exist to decrease postoperative pain. Palatine nerve block show the greatest effectiveness in palate repair, while bilateral infraorbital nerve block demonstrates an opioid sparing effect and increased the latency to first analgesia. All studied interventions demonstrated safety in this pediatric cohort.

### 101. Initial Consonant Cluster Production in Children With Repaired Cleft palate

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Background/Purpose: In the United States, 1 in 1574 infants is born with cleft palate (hereafter CP; Mai et al., 2014). Research conducted regarding speech characteristics in children with CP has primarily focused on resonance errors. Currently, there is limited research in speech development in this population. In particular, research evaluating consonant cluster production in children with CP is extremely rare. Locke (1983) reported that one-third of all English monosyllabic words begin with a consonant cluster and the majority of monosyllable words end with a consonant cluster. Thus, it is important to understand how children with CP produce consonant clusters. The purpose of this study was to investigate characteristics of singleton consonant and consonant cluster production in school-aged children with CP as compared to normally developing children in order to understand whether children with CP possess age equivalent consonant production skills.

Methods/Description: A total of 20 English-speaking children aged between 4 to 7 years old were recruited for this study. Ten children had a history of repaired cleft palate (mean age = 6;0) while the other 10 normally developing children were age matched (M = 5;9). Six children with CP reported a history of unilateral, with 4 reporting the current presence of a fistula. All children with CP had received speech therapy previously. A standardized articulation test and additional words were administered in order to obtain overall articulation skills as well as consonant cluster productions.

Results: The current study found that children with CP produced all elicited singleton consonants as well as consonant clusters with significantly less accuracy compared to the normally developing group. While standard scores of all normally developing children were within normal limit, standard scores of the half of children with CP were within 2 standard deviations below the mean for his chronological age. Production accuracies of all subtypes of consonant clusters (eg, twin, blue, tree, star, spring) in children with CP were significantly lower than those of children with normal development. Finally, children with CP produced different error patterns from normally developing children. Most errors were considered compensatory errors due to structural abnormalities, difficulties controlling resonance, and/or learned misarticulated patterns.

Conclusions: The findings of the study revealed that children with cleft palate do not produce consonant clusters as accurately as their normally developing peers although all had received speech therapy previously and half of the children currently receive speech intervention at school. Even though consonant like twin, that is, an early developing consonant that 90% of normally developing 3-year-old children produce correctly, was produced with significantly lower accuracy by children with CP compared to normally developing children.

### 102. Intensive Outcome After Speech Therapy and Pharyngeal Bulb Prosthesis

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Background/Purpose: Compare the speech outcome of patients with cleft lip and palate before and after an intensive speech therapy program (ISTP) associated to the use of a pharyngeal bulb prosthesis.

Methods/Description: Twenty operated cleft lip and palate subjects (8 males and 12 females) presenting with velopharyngeal insufficiency and hypodynamic velopharynx were selected to this study. All of them had hypernasality and/or compensatory articulations (CAs). Due to hypodynamic velopharynx, a pharyngeal bulb (in steady of secondary surgery) combined with an ISTP was indicated. The ISTP had 45 sessions of therapy (3 sessions a day) during 3 weeks. All subjects had their speech evaluated before the ISTP and immediately after the ISTP. Perceptual-auditory evaluation of the occurrence of hypernasality and CAs was performed by 3 experienced speech pathologists, upon the recordings of 12 sentences with recurrent high-pressure consonants, 3 sentences with recurrent low-pressure consonants (Brasil-Cleft protocol), counting of 1 to 20, and spontaneous speech; b) nasometry during the reading of a short text, consisting only of oral sounds (Oral Text), and 15 sentences with recurrent high and low pressure consonants (BrasilCleft protocol).

Results: The results of perceptual evaluation and nasometry showed that most subjects improved their speech after ISTP.

Conclusions: A structured ISTP, combined with the use of a pharyngeal bulb, is a fast and efficient method for correcting speech disorders of patients with cleft lip and palate/velopharyngeal dysfunction.

### 103. Is Telepractice for Cleft Speech Clinically Applicable in Japanese Language?

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Background/Purpose: Unlike USA, telepractice (TP) for speech therapy has not been yet approved by the Japanese government and also Japanese Association of Speech-Language-Hearing Therapists as a certain tool of speech therapy in Japan. The number of Speech-Language-Hearing Therapists (ST) in cleft field has recently decreased in Japan, and the postoperative speech and VPF management may not be sufficiently performed in some cases. Especially patients living in a remote area have a difficulty to access to the cleft team. Not only patients in a remote area but also patients with some family background cannot have frequent speech therapies. Due to the lack of therapy options in Japan, besides speech therapy in-person, there are some children who are left behind and grow up without appropriate treatments because of the geographical and family background factors. There are few reports of TP for cleft speech in Japan. This study aimed at clarifying whether TP could provide auditory accuracy to ST in Japanese language, and whether it was beneficial for patients in Japan.

Methods/Description: We assessed speech of enrolled patients with cleft speech and/or speech sound disorder (SSD) and without SSD, in both in-person and TP. The apps used were Skype, LINE, and Face-Time. Patient families chose which app to use. The device used for TP by patients was not specified in particular. Through TP, the change of VPF and articulation were assessed over time. To see if there was the positive effect from TP, patients were routinely assessed in-person and by TP. Inclusion: Patients aged 0 to 18 years with cleft lip/palate, SMCP, VPD, SSD without cleft; patients whose families could video-chat at home using the app via high-speed Internet; patients who were able to visit the hospital for regular assessment in person;

patients spoke Japanese as the first language. Exclusion: Hearing loss requiring hearing aids.

Results: Twenty-five children were enrolled in this study; 19 patients with cleft speech and/or SSD improved their speech; 3 patients without SSD at the first assessment stayed with good speech. Other 3 patients who had been very young at the first assessment did not develop any SSD. There were 2 patients who were detected as decreasing VPF through TP. They were sent to the assessment in person and performed the naso-endoscopy. In these assessments in person, the decrease of their VPF was confirmed.

Conclusions: From the results of this study, it was found that TP could improve patients' speech sufficiently in Japanese language. In addition, the auditory assessment by ST through a speaker was the same as one in-person. In other words, this study result may indicate that TP can be operated in speech therapy as well as in Japan.

### 104. Kenny-Caffey Syndrome Type 2: A Unique Presentation and Craniofacial Analysis

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Background/Purpose: Kenny-Caffey Syndrome Type 2 (KCS2) is characterized by short stature, skeletal dysplasia, primary hypoparathyroidism, and delayed closure of the anterior fontanelle; it has also been associated with additional rare anomalies. While the genetic basis and endocrine dysfunction of KCS2 have been studied thoroughly, an exhaustive craniofacial profile has not yet been compiled. As a result, patients with more subtle presentations may experience delayed diagnosis. We present the first-encountered case of a patient with KCS2 and concurrent craniosynostosis and provide a comprehensive, systematic characterization of all reported KCS-associated craniofacial features.

Methods/Description: A 5-month-old infant was referred to our institution for multiple congenital anomalies and increasing complexity of care. His physical features and persistent hypocalcemia were suspicious for KCS2 and prompted comprehensive genetic testing. A pathogenic variant in the FAM111A gene was detected, confirming the diagnosis of KCS2. Our patient was found to have brachycephaly/ plagiocephaly, a large forehead with frontal bossing, a wide anterior fontanelle, small eyes, hypertelorism, a hypoplastic midface, and a V-shaped mandible. At 5 months of age, he developed hydrocephalus and subsequent imaging showed a Chiari I malformation and bilateral craniosynostosis. Bilateral orbital edema was detected, and he underwent endoscopic third ventriculostomy, septostomy, and choroid plexus cauterization without complication. Given that the patient's remaining sutures were widely patent, and the fact that his thin calvarium would not withstand operation, surgical management of his craniosynostosis was delayed.

Results: We characterized KCS2-associated craniofacial features across the literature by dividing the head into upper, middle, and lower thirds. Of the patients identified with KCS2 to date, the majority presented with frontal bossing, absence of the diploic space, microphthalmia, hypertelorism, and micrognathia with a triangular-shaped face. Common features in the upper third were consistently reported between all patients; however, less commonly reported features in this region included depression of the sella turcica and microcephaly. The middle third had the greatest variation, and rare features included

strabismus, congenital glaucoma, absent frontal sinuses, nasal bridge depression, beaked nose, and a long philtrum. Less commonly reported features of the lower third were limited to a V-shaped mandible.

Conclusions: An absent diploic space in the upper third, microphthalmia and hypertelorism in the middle third, and micrognathia in the lower third with a triangular shaped face are key craniofacial features best used for diagnostic confirmation of KCS2. Awareness of uncommon presentations and maintenance of a high degree of suspicion for KCS2-associated anomalies are critical for early detection and intervention planning.

#### 105. Model for Tissue Engineered Vascularized Flaps for Patient-Specific Temporomandibular Joint Reconstruction: A Pilot Study

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Background/Purpose: Hemifacial macrosomia (HFM) is the most common congenital deformity affecting the temporomandibular joint (TMJ) and the second-most common of all craniofacial anomalies. Loss of TMJ (congenital or developmental) during childhood presents a clinical dilemma for which there is no viable regenerative solution. In children, the presently available autologous and alloplastic reconstruction options are not ideal. We propose to develop a patient specific, 3-Dimentional (3D) printed biologic pedicled flap approach to TMJ reconstruction.

Methods/Description: In the pilot study, a patient-specific mandibular condyle porous scaffold was designed directly from 6-month-old Yucatan minipig computed tomography (CT) scans. A Schwartz P triply periodic minimal surface (TPMS) of 40% volume fraction was used to create a porous mandibular condyle scaffold (Figure 2A). An integrated collar with screw holes was designed to fix the scaffold to the ramus following condylectomy (Figure 2A). The scaffold was 3D printed from polycaprolactone (PCL) using a laser sintering process. After ethylene oxide sterilization, mandibular condyle scaffolds were soaked in a solution of 1-mg BMP2 then implanted in the temporalis muscle as a flap (Figure 3A). After 6 weeks in situ in the muscle, a condylectomy was performed and the muscle flap with scaffold was rotated into place. The scaffold was attached through the collar fixation to the ramus with titanium screws (Figure 3B). CT scans were obtained 6 months after flap rotation and condyle reconstruction.

Results: Reconstructed condyles had 78% (Figure 4A) and 68% (Figure 4B) height of contralateral condyles when measured from the same point. From microCT scans, reconstructed condyles had 67% (Figure 4A) and 36% (Figure 4B) of total volume and 107% (Figure 4A) and 105% (Figure 4B) of the bone volume fraction of the contralateral condyles.

Conclusions: In this pilot project, we demonstrated that in the Yucatan minipig model, a patient-specific, 3D printed biologic scaffold adsorbed with 1 mg BMP2 implanted in the temporalis muscle will attain sufficient bone regeneration.

### 106. Natural History of Facial Growth in Unoperated Cleft Defects: A Systematic Review and Meta-Analysis

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(1) Children's Hospital of Los Angeles, Los Angeles, CA, (2) Keck School of Medicine of USC, Los Angeles, CA, (3) University of Southern California, Los Angeles, CA Background/Purpose: Orofacial cleft deformities are prevalent congenital defects which can affect approximately 7.75 neonates out of every 10, 000 live births. After surgical repair of the orofacial cleft, high rates of midface hypoplasia requiring orthognathic repair have been reported in the literature. The etiology of this midface hypoplasia is incompletely understood. Popular belief suggests that the growth retardation of the maxilla is secondary to scar tissue formation and disruption of the natural growth plates during the surgical repair of the cleft. In various countries, patients with unoperated clefts have been examined to gauge the underlying growth potential of the maxilla untouched by surgical intervention. As a result of small patient populations in each individual study, no conclusions can be drawn. We describe a meta-analysis of maxillary cephalometric measurements in unoperated cleft patients to gain a more complete understanding of the cleft palate's underlying growth potential.

Methods/Description: A systematic literature review and metaanalysis was performed according to PRISMA guidelines in the PubMed database. Studies were included for their focus on background information, cephalometric measurements in unoperated cleft patients, or maxillary growth mechanics. Means and standard deviations of SNA, SNB, and ANB angles along with measurements of maxillary anterior—posterior distance were collected from patients who possessed unoperated unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), and isolated cleft palate groups (ICP). Statistical analysis was run with independent parametric t tests. Results: A total of 33 peer-reviewed articles were included in the

Results: A total of 33 peer-reviewed articles were included in the study. From these, 10 articles included the appropriate cephalometric measurements for comparison. A total number of 468 patients were included in the meta-analysis. This contained 289 patients with UCLP, 36 patients with BCLP, and 143 patients with ICP. SNA angles of the UCLP and BCLP groups demonstrated significant increases from the control group (84.5°  $\pm$  4.0° vs 82.3°  $\pm$  3.5°,  $P \le$  .001) and (85.8°  $\pm$  2.8° vs 82.3°  $\pm$  3.5°  $P \le$  .001), respectively. The ICP demonstrated a decrease in SNA angle (79.2°  $\pm$  4.2° vs 82.3°  $\pm$  3.5°  $P \le$  .001). The anterior–posterior length of the maxilla was decreased in UCLP and ICP groups (52.32  $\pm$  4.05 mm vs 53.98  $\pm$  3.25 mm  $P \le$  .015 and 39.29  $\pm$  3.61 mm vs. 46.54  $\pm$  3.03 mm  $P \le$  .001, respectively).

Conclusions: In patient with unoperated cleft lip and palate, the maxilla does not exhibit the degree of midface hypoplasia typically seen after operative repair. The data presented suggests that the midface hypoplasia could be attributable to extrinsic factors on the palate. More studies are required to delineate the etiology of midface hypoplasia typically seen after operative repair of the orofacial cleft.

#### 107. Periodontal Health of Individuals With Non-Syndromic Cleft Lip and/or Palate: A Meta-Analysis

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Background/Purpose: Periodontal disease is an inflammatory polymicrobial condition involving a complex interplay between the pathogenic bacteria and the host. The authors conducted a systematic review and meta-analysis to assess the periodontal health status of children, adolescents, and adults with nonsyndromic cleft lip and/or palate (CL/P).

Methods/Description: The authors conducted a search of indexed databases (PubMed, Cochrane, Web of Science, Embase, Science direct, and LILACS) without language restriction up to and including August 1, 2019, and used cross-referencing to further identify articles. The eligibility criteria were as follows: (a) observational studies with

original data that statistically compared the periodontal health status of (b) individuals with CL/P without syndromes to (c) those without CL/P. The authors performed meta-analysis using Cochrane Community Review Manager 5 software and used standardized mean difference (SMD) with 95% confidence intervals (CI) as a measure of the effect size between CL/P and risk of periodontal disease. Random effects model using the inverse variance estimator was performed in order to obtain an overall summary estimate of the mean difference of plaque index (PI), gingival index (GI), periodontal probing depth (PPD), and clinical attachment loss (CAL) between CL/P and non CL/P groups. An assessment of the methodological study quality was performed according to the modified Newcastle-Ottawa Scale.

Results: The database search generated 650 records, and 30 full-text articles were reviewed. Ten studies comprising 1127 individuals (510 with CL/P and 617 without CL/P), with age ranging between 2 and 28 years, fulfilled our selection criteria and were included in the meta-analysis. The meta-analysis revealed significant difference in PI scores (SMD = 0.41, 95% CI = 0.27-0.55) and GI scores (SMD = 0.55, 95% CI = 0.30-0.79) between individuals with and without nonsyndromic CL/P. Additionally, individuals with nonsyndromic CL/P had higher periodontal probing depth and attachment loss overall compared to those without CL/P; however, this finding was not statistically significant.

Conclusions: Individuals with CL/P showed an increased risk for plaque accumulation and gingival inflammation, and may have poorer oral hygiene compared with individuals without CL/P; whether this translates into periodontal disease remains an open-ended question. More awareness among clinicians, patients, and caregivers would promote better periodontal preventive management for CL/P patients.

## 108. Preliminary Report on the Development of a Language Questionnaire for Children With Cleft Lip and Palate from Diverse Linguistic Backgrounds

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Background/Purpose: In the United States, approximately 60 million people speak a language other than English in the home (U.S. Census Bureau, 2015), and many cleft lip and palate (CLP) teams have reported an increase in the linguistic diversity of their patient caseloads (Edwards & Bonilla, 2004). Yet, only about 6% of speechlanguage pathologists (SLPs) are bilingual (ASHA, 2018). Many SLPs have limited knowledge about multilingualism (Hammer et a., 2004), how to work with families that speak different languages (Bedore et al., 2011), or how to identify a language difference from a disorder when a child is from a non-English speaking or bilingual background (Kritikos, 2003). As a result, SLPs on CLP-teams may conduct only a cursory review of the child's linguistic background, which can lead to biased speech and language evaluations, incorrect diagnoses, and inappropriate interventions. One initial step toward addressing this problem is for SLPs to base their practices on the available evidence regarding multilingual and cross-linguistic assessments, which dictates that the child be assessed in their primary language(s). In addition, by obtaining information from parents about the child's language exposure and use, such as the percentage of input, percentage output, and proficiency of each language, the full extent of their linguistic proficiency can be applied to clinical decision-making (Goldstein et al., 2010; Restrepo, 1998). There is currently no set of standard practices for SLPs to implement when assessing the linguistic background and language use of diverse populations of children within the CLP-team clinical setting. To address that need, this poster will present findings from a scoping literature review and consensus of a working-group of monolingual and bilingual team SLPs regarding the development of a novel language questionnaire (LQ) to use with children with CLP from diverse linguistic backgrounds from 1 to 36 months of age.

Methods/Description: This poster will describe the process of developing an English/Spanish version of the LQ, currently being trialed in the clinical setting and as part of a multisite, NIH-funded investigation of speech and surgical outcomes in children with CLP. Best practice recommendations for how language background and the spectrum of bilingualism in linguistically diverse young children with CLP should be assessed and interpreted via questionnaires will be discussed. Descriptions of the LQ forms and structure will also be provided, along with data regarding its feasibility, challenges encountered, and opportunities for improvement identified during trial use.

### 109. Reduction of Unrepaired Cleft in a Northern Nigerian Community

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Background/Purpose: According to guidelines of surgical cleft repair, cleft lips are repaired at about 10 weeks after birth followed by associated cleft palate repair within 12 to 18 months of life. Persistence of the condition well beyond the expected time of primary surgical repair is known as unrepaired cleft; with the age limit defined as being above 2 years, 6 years or even 12 years by various authors. This management guideline is a far-cry from what is obtained in developing countries (especially Sub-Saharan Africa) where substantial number of cases of unrepaired clefts are still reported. This has been ascribed to the poverty, lack of adequate awareness, dilapidated health-care facilities, and remoteness of standardized surgical care for cleft anomalies. The study aims to determine the prevalence of unrepaired clefts in Minna, a community in Northern Nigeria during 2 free surgical outreaches in 2011 and 2017 and to determine principal factors influencing the outcome.

*Methods/Description*: A comparative study involving patients with cleft lip and palate anomalies at 2 free surgical outreaches held in Minna in 2011 and 2017. From case notes of cleft patients attending the surgical outreach, data were collected and analyzed, independent t test was used to compare the mean ages in both group of patients while statistical associations between categorical variables were determined using the Pearson  $\chi^2$  test. For all statistical tests and comparisons used, probability values <.05 implied statistical significance and rejection of the null hypotheses.

Results: A total of 127 patients were encountered at both surgical outreach programs. Mean age of patients in 2011 and 2017 was 14.3  $\pm$  14.98 and 6.4  $\pm$  9.93 years, respectively. In 2011, most patients (27.1%) received information about the free surgical program via friends and relatives while radio/media broadcast represented the most common source of information (26.1%) for participants in 2017. There was a 29.6% drop in prevalence of unrepaired cleft from 75.3% in 2011 to 45.7% in 2017 at the free surgical outreach programs. Similarly, adult cleft prevalence dropped from 32.1% to 13.0% within the study period. Mean age of patients was reduced from 14.3  $\pm$  14.98 to 6.4  $\pm$  9.93 years in 2011 and 2017, respectively.

Conclusions: A tripartite approach involving an indigenous surgical foundation (CFDF, Abuja, Nigeria), an international funding organization (Smile Train, New York) and governmental support, in creating awareness through cleft awareness campaigns and patient education, and providing free surgical care in the management of cleft lip and palate anomalies as in a significant way created a harvesting phenomenon and subsequent surgical repair of unrepaired clefts in the region and has encouraged younger age presentations at local hospitals from where they are referred to cleft centers within the region for definitive surgical management.

## I I 0. Remote Ascertainment of Families With a History of Orofacial Clefts Through Social Media and Clinician Partnerships

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Background/Purpose: Orofacial clefts (OFCs) are the most common craniofacial birth defect in humans, affecting approximately 1 in 700 individuals. Large cohorts are increasingly required in biomedical research yet recruiting such a cohort of OFC cases is hampered with traditional recruitment methods. Previous studies have been primarily site-based, requiring scheduling with busy craniofacial clinic visits and have been limited to the patient population at any given clinic-posing significant barriers to sample size and diversity. In a pilot study, we established a social-media based recruitment strategy coupled with online informed consent, questionnaires, and mail-in saliva collection kits.

Methods/Description: Here, we report the results of this study to assess the feasibility and efficacy of a social-media based remote recruitment for individuals and families with either nonsyndromic OFCs or a rare OFC syndrome (Van der Woude syndrome). In a test of these recruitment mechanisms, we initiated a one-time social media campaign targeting 3 active cleft support groups on Facebook (FB).

Results: From this initial outreach, we were able to enroll 139 participants from 111 families located across 34 US states and 6 countries. Eighty-six percent completed the online survey portion of the study. Saliva Sample kits were then mailed to participants and their family members, with a 6-month return rate of 55%, resulting in 163 total samples collected. Though most (88%) of our families were nonsyndromic, we also recruited syndromic families, including 9 with Van der Woude syndrome. The majority of OFC individuals who participated were minors (75%) with most under the age of 6 (61%), who were enrolled by their parents.

Conclusions: Using social media support groups to target-specific patient populations presented an advantage over site-based recruiting methods, allowing for increased awareness and accessibility to a wider range of participants. By advertising our study directly on online platforms and allowing interested participants to self-enroll, we were able to greatly reduce recruitment costs and streamline the participation process. In the future, on top of social media outreach, we plan to actively collaborate with clinicians who can provide referral information to patients using provided recruitment materials, minimizing the time commitment for clinicians and their staff. In conclusion, our recruitment design has been shown to successfully expand OFC research recruitment to new populations and increase sample diversity, while minimizing burden on researchers, clinicians, and participants.

## III. Sociodemographic Factors Associated With Establishment of a Dental Home in Patients With Craniofacial Differences

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Background/Purpose: The ACPA and American Academy of Pediatric Dentistry (AAPD) recommend that children establish a dental home by 12 months of age. The ACPA parameters state that the dental needs of patients with craniofacial (CF) differences are best met by a pediatric dental specialist. Results of this study will determine whether patients from our CF team are meeting these recommendations and identify potential barriers (demographic factors, etc) to establishing a dental home at a young age.

Methods/Description: A cross-sectional study was conducted of all active patients 12 months to 10 years of age seen by our center's CF team. Only patients who had their age of first dental visit recorded were included. Data included: CF anomaly type, gender, race/ethnicity, insurance type, age at first CF team visit, age at first dental visit, type of dental home (general or pediatric), county of residence, presence of pediatric dentist in county of residence, dental home in county, distance to hospital from home, and distance to dental home from home

Results: Of the subjects who had a dental home, 16% established care with their dentist by the recommended age of 12 months. By 24 months 48% established a dental home and by 36 months 69% had a dental home. The mean age at first dental visit was 28 months, and ranged from 5 months to 105 months (8.75 years). Eighty-six percent of subjects saw a pediatric dentist for dental care; 67% saw a dentist within their home county, and 58% established care with our hospital's dental center. The only statistically significant variable associated with younger dental home establishment was closer distance to the hospital from home.

Conclusions: Most patients, 84%, are not establishing a dental home by the recommended age of 12 months. However, most, 86%, are having their dental care provided by a pediatric dentist as recommended by the ACPA. Based on the data collected, sociodemographic factors, aside from distance to hospital, do not appear to be associated with establishment of a dental home at a younger age.

### II2. Supporting Mothers Through Providing Breast Milk to Babies With Clefts

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Background/Purpose: According to the AAP and the WHO, breast milk should be provided to babies for the first 6 months of life exclusively, and up to 1 to 2 years or longer. The rate of babies who had received breast milk in the United States was 83.2% in 2015, while comparatively only 67% of patients with clefts receive breast milk. Providing breast milk is proven to have many known benefits for infant and maternal health, including benefits specific to the cleft population, such as lowering risk of otitis media and positive long-term effects on neurodevelopment. Mothers of cleft babies may not be receiving appropriate support in initiating and facilitating breast milk production for their babies. The purpose of this poster presentation is to describe interventions that can be implemented in order to support mothers of babies with cleft lips and palates in providing breast milk for their babies.

Methods/Description: A literature review was conducted in order to ascertain ways the nurse practitioner can support mothers of babies with cleft lips and palates in breast milk production. Barriers to providing breast milk will be identified. Specific interventions will be presented that can be applied when caring for cleft patients and families pre and postnatally in order to achieve higher breast milk provision rates for this patient population. Interventions that are included and that will be expanded upon are prenatal consultations, providing information regarding health benefits specific to this population, providing accurate and correct breast milk and pumping information, offering support throughout pre and postnatal stages, educating caregivers, peer support, engaging partners, and educating health-care providers.

#### 113. The Development of a Customized Trans-Sinus Maxillary Distraction Device: A Preliminary Study

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Background/Purpose: The conventional maxillary distraction osteogenesis is including external and internal approach. Both method has its limitations, such as social inconvenience for the external device and hardly vector control for the internal devices. The newly designed trans-sinus maxillary distraction device provides a precise surgical outcome by customized designing and manufacturing. The purpose of this study is to compare the mechanical properties with conventional devices and test the precision on skull model surgery.

Methods/Description: The design of the customized distraction device was based on a maxillary hypoplasia patient secondary to cleft lip and palate. The design of the fixation part of the distraction device is strictly matching the anterior maxillary sinus bony surface and avoiding teeth roots and suborbital nerve damaging. The distraction rod is placed through the sinus on both sides with same parallelism. The device was manufactured through combination of select laser melting (SLM) printing technology and traditional technology. The mechanical properties were tested compared with the commercial distractors. The skull model surgery was performed to assess the distraction accuracy of the customized distractor compare by pre and post CT measurements.

Results: The customized device can be fixed on the bony surface without prebending the fixation plate. The adjacent anatomic structures such as tooth root can be reconstructed by software which might reduce the intraoperative unexpected damage. The location of plate hole can be planned in the strut area of the maxilla which can provide sufficient mechanical support during distraction procedure. The mechanical tests showed the appropriate properties of customized device compare with conventional device. The skull model surgery showed higher precision and better degree of parallelism of customized trans-sinus distraction device.

Conclusions: The newly designed trans-sinus maxillary distraction device has good mechanic properties which can transfer the surgical plan to the operation with high accuracy. The potential application of this device can improve the clinical efficacy of maxillary distraction osteogenesis on cleft patients.

### I I 4. The Role of Flexible Nasendoscopy for Surgical Decision-Making in Outreach Settings

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Background/Purpose: Management of velopharyngeal dysfunction (VPD) is particularly challenging in outreach settings. While nasendoscopy is a valuable tool for the surgeon in industrialized cleft care settings, there is limited access to the necessary equipment in international outreach settings. To evaluate the role of nasendoscopy in surgical decision-making, a prospective study was conducted to compare surgical treatment recommendations with and without nasendoscopy data. The aim was to compare surgical recommendations for individual VPD cases with and without nasendoscopy data in mission-based care.

Methods/Description: A 2-year prospective cohort study comparing surgical recommendations with and without nasendoscopy data was performed. Participants consisted of 5 craniofacial surgeons participating on an international cleft lip/palate team mission. Each surgeon screened between 7 and 17 patients (n = 52 patients) for velopharyngeal dysfunction (VPD) during a day long screening and triage session prior to a surgical mission in Guayaguil, Ecuador. Each surgeon rated the patient's speech and resonance according to their own guidelines based upon a speech sample and clinical experience and made surgical recommendations. All patients who were identified as having VPD where then evaluated by the team speech pathologist who conducted nasendoscopy evaluations on a portable KayPentax system with video archiving. Each study was then deidentified and presented to the evaluating surgeon who made surgical decisions based upon the nasendoscopy video. Pre and post nasendoscopy results were analyzed using descriptive statistics.

Results: Our preliminary analysis indicates that, surgeons in an outreach setting modified their treatment recommendations for VPD an average of 63.2% of the time when presented with nasendoscopy information. Across the 5 surgeons in this study, the range of recommendation agreement between pre and postnasendoscopy ranged from 33% to 40%. Mean agreement among raters was 36.8% (SD = 4.49). Conclusions: The results of this study suggest the need for flexible nasendoscopy to be an integral component of the cleft surgical decision-making process in outreach settings. We conclude that incorporating nasendoscopy information would improve the delivery of comprehensive cleft care internationally.

#### I 15. Three-Dimensional Evaluation of Secondary Alveolar Bone Grafting in Patients With Cleft Lip and/ or Palate

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Background/Purpose: Secondary autogenous bone grafting (SABG) of the alveolar cleft is essential for the occlusal management of patients with cleft lip and/or palate. The outcome of the bone bridge in SABG has routinely been evaluated on 2-dimensional images using dental radiographs. However, recent recommendations suggest that the bone bridge should be evaluated using 3-dimensional (3D) conebeam computed tomography (CBCT). The purpose of the present study was to assess the usefulness of 3D-CBCT for examining the alveolar cleft and bone bridge in patients who underwent SABG.

Methods/Description: Twenty clefts of 16 patients (4 bilateral cleft lip and alveolar [BCLA], 6 unilateral cleft lip and alveolar [UCLA], 1 bilateral cleft lip and palate [BCLP], and 5 unilateral cleft lip and

palate [UCLP]) with cleft lip or/and palate who underwent SABG in the University of the Ryukyus Hospital were enrolled. The surgical procedures for SABG were performed in accordance with the modified methods of Bergland et al. All patients underwent CBCT (3DX Multi-Image Micro CT/3D; Accuitomo, Morita, Japan). The acquisitions were completed by multiplanar reconstructions (coronalsectional, axial-sectional, and sagittal-sectional 3D-CBCT images) using i-View software (J Morita Corporation, Osaka, Japan). i-View software was used to obtain measurements on the 3D-CBCT reconstructed images in the midline of C1 passing the anterior nasal spine (ANS) point on the axial images, on the line connecting both orbital points on the coronal images, and in the Frankfurt horizontal plane on the sagittal images. The alveolar cleft before SABG on the axial 3D-CBCT images was classified into 3 categories for statistical analysis. In patients who were followed-up for more than 6 months after SABG, the bone bridge was evaluated by measuring the bone width and bone height at the A point plane on axial 3D-CBCT images, and the interalveolar septal height and piriform aperture height on coronal 3D-CBCT images.

Results: The alveolar cleft before SABG was classified as type B (anterior width wider than the posterior width) in 15 of 20 clefts (75.0%). The bone bridge width at the A point plane was 11.3 mm at the mesial site and 9.1 mm at the distal site, which was considered sufficient. In the UCLA/UCLP cases, the bone bridge height at the cleft site was shorter (-3.8 mm) than the alveolar bone height at the normal site, and the lower edge of the piriform aperture at the cleft site was slightly lower (-1.6 mm) than that at the normal site. The bone bridge width after SAGB was 91.0% of the width of the alveolar cleft, which was considered acceptable. The width and height of the bone bridge were slightly poor in cases with a preoperative alveolar cleft width of 10 mm or more, and countermeasures such as improvement of the surgical procedures and preoperative orthodontic treatment were considered necessary.

Conclusions: The use of 3D-CBCT images to measure the alveolar cleft and the bone bridge before and after SABG achieves acceptable outcomes.

#### I 16. Treatment and Long-Term Outcomes of Children With Co-Occurring Childhood Apraxia of Speech and Velopharyngeal Insufficiency

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Background/Purpose: As each are relatively rare, the co-occurrence of childhood apraxia of speech (CAS) and velopharyngeal insufficiency (VPI) is truly exceptional and poses unique challenges to speechlanguage pathologists (SLPs) and otolaryngologists (ENTs) in the treatment of these children. CAS is a deficit of speech motor control and impacts timing of articulatory movements, including those of the velum. Such mistiming may cause children with CAS to exhibit intermittent or inconsistent hypernasality (Baas et al., 2015) and aberrant closure of the velopharyngeal port during speech (Sealey & Giddens, 2010). Co-occurring childhood CAS and VPI may occur in children with genetic phenotypes that include craniofacial anomalies and motor speech disorders. Although speech treatments for CAS and VPI as separate speech disorders are well-described in the literature, very little is known about treatment for children with concomitant CAS and VPI. The purpose of the current project is to report long-term outcomes of children with concomitant CAS and VPI.

Methods/Description: A retrospective review of multidisciplinary VPI clinic records identified 34 records of children who were diagnosed with both CAS and VPI. Inclusion criteria included evaluation by SLP

and ENT, surgical intervention by ENT, and at least one follow-up visit to judge the outcome of the surgical intervention. The records were reviewed to extract communication and medical diagnoses, description of CAS and other communication disorders, velopharyngeal structure and function pre and post intervention, instrumental and perceptional measures of resonance pre and post intervention, type of surgical management, description of speech therapy, and length of follow-up.

Results: All patients had a diagnosis of CAS, with many children exhibiting other communication disorders. Velopharyngeal insufficiency was determined by results of nasendoscopy. All children received surgical treatment for their VPI. Improved velopharyngeal function and/or improved speech and resonance were reported in all cases. Many of the patients received ongoing speech therapy for several years with approximately 1/3 of the patients receiving at least one period of intensive speech therapy. While some patients had resolution of their VPI symptoms, no patients were considered "resolved" of their motor speech disorder.

Conclusions: This retrospective review of patients with co-occurring VPI and CAS identified a subset of children whose speech sound production was meaningfully enhanced by management of their VPI. Moreover, the success of the surgical intervention for VPI may have been facilitated by intensive speech therapy. We conclude that intensive speech therapy and ongoing reassessment of motor speech and articulation skills, as well as systematic reevaluation of velopharyngeal function is needed for children with CAS and VPI to ensure timely modifications to surgical and speech therapy treatment plans.

### 117. Use of Intraoral Scanner in Presurgical Infant Orthopedics for Children With CLP

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Background/Purpose: Recently, intraoral scanners are utilized in many dental treatments. It is known that digital dentistry can provide more accurate treatment and better appliances, and also simplify the classical process of intervention. Moreover, digital dentistry can reduce the treatment time and the burden of patients. We have been performing presurgical infant orthopedics for children with CLP using passive plates. During the fabrication of the plates, alginate impressions have to be taken from infants. Since infants with CLP have high risk of vomiting and accidental swallowing during the alginate impression, it is essential to manage these risks strictly. To overcome these problems, intraoral scanners were used for infants with CLP and the accuracy of scanned images was evaluated.

Methods/Description: Digital impression (STL data) of the maxillary arches was taken from 2 infants (40-day-old infant with cleft lip and alveolus, and 55-day-old infant with cleft lip, alveolus, and palate). The used intraoral scanner was Trophy 3DI Pro (Trophy Radiologie Japan). To improve the quality of the scanned image, clefts of alveolus and palate were partially flattened by the silicone impression material. Alginate impression was also taken from 2 cases and the plaster models were fabricated. By using the desktop scanner, these plaster models were 3-dimensionally compared with the STL data obtained from the intraoral scanner.

Results: The used intraoral scanner didn't have difficulty in taking STL data of 2 infants with CLP. The insertion of the silicone impression material to the clefts helped scanning. The shape difference between the STL data obtained from the intraoral scanner and the plaster models fabricated after the alginate impression was less than

100 µm. Finally, the passive plates which were fabricated from the STL data showed an acceptable fit to the infants.

### 118. Variability and Excess in Opioid Prescribing Patterns After Cleft and Craniosynostosis Surgery

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Background/Purpose: Opioid overprescribing has been frequently reported in pediatric surgery, but has not been well-documented for craniofacial procedures. Perioperative and inpatient opioid use reduction via nerve blocks has been studied in the cleft population, but prescribing practices at hospital discharge and home opioid use relative to prescribed amounts have not been reported. New persistent opioid use after surgery is a major concern of the opioid crisis, and this phenomenon has been demonstrated in patients undergoing cleft-related surgeries. Accidental ingestion and drug diversion are additional risks of pediatric opioid prescriptions. Critical analysis of current prescribing patterns is essential for curbing excess opioid prescriptions and achieving safe prescribing practices.

Methods/Description: We retrospectively reviewed records for all patients undergoing surgical intervention for cleft lip and/or palate or craniosynostosis at a single center over a 12-month period. Variables collected included patient age, procedure type, number of opioid doses prescribed at discharge, and concomitant nonopioid analgesic prescriptions. Prescribed opioid doses were analyzed with nonparametric univariate analysis. Correlations between patient age and number of prescribed doses were analyzed with Spearman rho. Preliminary data from an ongoing prospective analysis of actual home use of prescribed opioids are also presented, based on in-person patient surveys.

Results: Ninety-five patients with cleft lip and/or palate and 44 patients with craniosynostosis underwent a surgical procedure in the study period. Median prescribed opioid doses for the most common procedures were 15 for primary lip/nose repair (range = 5-67, interquartile range [IQR] = 10-33; n = 19), 10 for primary palatoplasty (range = 6-75, IQR = 10-16; n = 27), 11 for secondary palatoplasty (range = 5-38, IQR = 8-19; n = 12), 10 for alveolar bone graft (range =5-28, IQR = 10-17; n = 22), 10 for endoscopic synostosis repair (range = 0-50, IQR = 7-17; n = 29), and 21 for open cranial vault remodeling (range = 0-42, IQR = 9-27; n = 10). Ninety-five percent of patients were also prescribed acetaminophen, and 88% of patients over 6 months of age were prescribed ibuprofen. In patients with cleft lip and/or palate, there was a weak negative correlation between age at surgery and prescribed opioid doses (rs = -0.231, P = .024). Thirteen patients were prospectively surveyed for actual home opioid use. Procedures included 1 primary lip/nose repair, 2 primary palatoplasties, 1 secondary palatoplasty, 6 alveolar bone grafts, 1 rhinoplasty, 1 open cranial vault remodeling, and 1 endoscopic synostosis repair. Fiftyfour percent used no opioids at home, 31% used 1 to 2 doses, 8% used 3 to 5 doses, and 8% used 6 to 10 doses. All used nonopioid analgesics as first-line medications.

Conclusions: Postoperative opioid prescriptions vary widely after common cleft and craniosynostosis procedures, and younger patients with cleft lip and/or palate may be more likely to be prescribed more doses. Actual home opioid use is less than prescribed amounts.

### 119. ERAS, Al, and Neural Network in Cleft and Craniofacial Care

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*Background/Purpose*: Educational Objectives: Cognitive: The attendees will understand what is ERAS and how to develop it psychomotor. The attendees will be able to organize and implement ERAS affective. The attendees will appreciate the importance and the value of AI and neural network in cleft and craniofacial care.

Methods/Description: There is a need to adopt enhanced recovery after surgery, or ERAS, in cleft and craniofacial care. Developed in the 1990s, ERAS was the result of sustained and systematic approach to improve surgical outcomes and to reduce complications in colorectal and cardiothoracic surgery. Due to the tremendous success, both the AMA and ACS have accepted ERAS in recent years. However, to date, very few ERAS protocol or pathways exist in cleft and craniofacial care. The ASCFS established a presidential taskforce during its 2019 annual meeting in Tucson, Arizona, to address this. The proposed panel will provide the audience with information to bridge a critical gap in our knowledge and practice. The ASCFS taskforce has developed ERAS protocols for 5 commonly performed cleft and craniofacial procedures. Lip repair, palatoplasty, alveolar bone graft, Le Fort I advancement, and FOAR. These ERAS protocols will be available to all attendees. As the second decade of this century closes, advances in AI such as machine learning and neural network have drastically transformed how we deliver health care in isolated areas. But the application will spread. The ability to mimic human cognition coupled with "Big Data," NSQIP, HCUP, NCHS to name just a few, will propel cleft and craniofacial care in ways we cannot even imagine at present. It is time that we take the first step.

#### I20. Smile Oregon—Building a Locally Sustainable Non-Profit Community-Based Organization to Improve the Lives of Patients and Families Affected by Cleft or Craniofacial Conditions

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Background/Purpose: There are several teams providing cleft and craniofacial care to the patients and families affected by cleft and craniofacial differences in the state of Oregon. However, many families and patients continue to lack access to care, community, and support through this difficult journey in cleft and craniofacial health care. Therefore, Dr Garfinkle, and several members of the greater Oregon community came together to establish a nonprofit community-based organization to raise awareness, resources, and support for these families through SMILE OREGON. This organization has grown in members, events, resources raised and given out to families and in its own organizational structure over the years, and we felt it would be an important message to share with others in the cleft community nationally to share ideas of how this organization was created and has helped many families.

Methods/Description: Smile Oregon was conceived 12 years ago by Dr Judah Garfinkle who wanted to create a sustainable resource for families affected by cleft and craniofacial conditions who may need

support, education, or other resources. It has grown from 2 founding members to 12 board members and it currently serves any and all children or families in need of resources in their cleft/craniofacial journey. SMILE OREGON is the only nonprofit organization in the state of Oregon which is composed of community-based volunteers with the sole purpose of raising awareness, advocacy, education, and resources for families affected by cleft and craniofacial conditions. We have built a large community resource across an urban and rural territory through social events, fundraisers, online, and printed media. We have raised and awarded thousands of dollars to patients and families affected by cleft/craniofacial conditions, we have enjoyed recruiting several dozen community partners to sponsor events, and have connected 100's of families and other affected by cleft/CF conditions to build a much stronger sense of community in our state of OR.

Conclusions: Navigating the journey of a cleft and craniofacial condition alone is difficult and challenging. This journey can be improved for families and patients by local teams, volunteers, and families to band together to organize and help create resources, ease of access, and improved sense of belonging/community. SMILE OREGON is an example of such a way we are attempting to help those families and patients on this journey. We would be excited to share our experiences, growth, and those of patient's/families' who have benefitted from SMILE OREGON's presence.

#### 121. A Preliminary Study of Anatomical Changes Following the Use of a Pedicled Buccal Fat Pad Graft During Primary Palatoplasty

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Background/Purpose: The pedicled buccal fat pad graft (BFP) is a well-documented technique for closure of oral defects. When utilized during primary palatoplasty, it has been hypothesized that this technique results in increased vascularized tissue at the posterior hard palate, preventing wound contracture, lengthening the velum, optimizing maxillary growth, and maintaining a favorable depth:length ratio (Levi et al., 2009). However, these hypotheses have not been systematically examined using a comparison group. MRI studies have demonstrated potential clinical utility to improve postsurgical speech outcomes by analysis of velopharyngeal (VP) variables. The purpose of this study was to use MRI to examine the surgical impact of the BFP on VP anatomy after primary palatoplasty. We hypothesized that participants with cleft palate treated with the BFP would exhibit more favorable anatomical dimensions that are more similar to the noncleft study group when compared to participants with cleft palate treated with traditional repair techniques.

Methods/Description: 15 English-speaking children aged 3 to 7 years participated in this study as part of 3 groups: noncleft, traditional palatoplasty, and palatoplasty with BFP. The 5 children with BFP were operated on by the same surgeon. Participants across all groups presented with normal resonance at the time of the MRI study. The MRI protocol, processing methods, and analysis were consistent with previous investigations of the velopharynx using MRI. All analyses were designed to quantify differences in anatomical variables across

the 3 groups. Using an intraclass correlation, reliability was completed on 80% of participants 4 weeks after initial data analysis.

Results: Nonparametric statistical analyses were utilized for comparing measures between the participant groups due to the small sample size and nonnormal distribution of data. All assumptions were adequately met for the Kruskal-Wallis H test. Median values were significantly different among groups for velar length (P=.042), effective velar length (P=.048), effective VP ratio (P=.046), levator length (P=.021), extravelar levator length (P=.009), and levator origin-origin distance (P=.030). Post hoc analysis revealed a statistically significant difference between the BFP and traditional repair groups for effective VP ratio (P=.040), extravelar levator length (P=.033), and levator length (P=.022). Inter-rater reliability was r=.813 for effective velar length and r=.762 for sagittal angle. Intrarater reliability was r=.951 and .995, respectively.

Conclusions: Upon further investigation, this technique may create a more advantageous system for speech production over traditional surgical techniques. Additional research employing a larger sample size should be completed to evaluate cases with VPI as well as cross-sectional data across the life span. Larger sample sizes will also allow for integration of speech data in the statistical analyses while controlling for age, race, and sex.

# I 22. Patient- and Family-Reported Outcome Measures Included in the ICHOM Standard Set for Cleft Care: An Examination of Their Psychometric Performance in an International, Multicenter Sample

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Background/Purpose: The International Consortium for Health Outcomes Measurement (ICHOM) has proposed a standard set of outcome measures for the comprehensive appraisal of cleft care. It stresses the importance of the perspectives of patients and their parents or carers by incorporating several diverse outcome measures covering multiple domains: the patient-reported CLEFT-Q (9 subscales), the NOSE scale for nasal breathing, the COHIP-OSS for oral health, and the family-reported Intelligibility in Context Scale (ICS) on overall speech. While each of these instruments has undergone some degree of reliability testing in their original development, they have not yet undergone robust psychometric evaluation when used in clinical practice with point-of-care collection of outcome metrics. Given that these outcome measures are intended to facilitate future comparative effectiveness research and quality-improvement projects, it is critical that the Standard Set be appraised for robustness and clinical significance of the instruments that it includes. Therefore, the aim of this study is to evaluate the psychometric performance of the patient- and familyreported outcome measures included in the ICHOM Standard Set for cleft care after implementation.

Methods/Description: Outcomes data were collected from patients with cleft lip and/or palate, aged 8, 12, and 22 years at the Erasmus MC (NL), Boston Children's Hospital (USA), Duke Children's Hospital (USA), and McMaster University (CA) between November 2015 and April 2019. Deidentified data were examined with Rasch Measurement Theory (RMT). This analysis is based on the probability that

individuals will answer a set of items in a certain manner based on their abilities, which are assumed to be indicative of cleft-related outcome. Thus, Rasch analysis can provide insight in the strengths and limitations of each individual scale to determine which ones are most useful in cleft outcome studies.

Results: Data were collected from 748 unique patients. The RMT analysis provided support for reliability and validity of the CLEFT-Q subscales and the ICS. The Person Separation Index values were above 0.83, except for the CLEFT-Q Eating and Drinking subscale (0.54), the CLEFT-Q Speech Distress subscale (0.66), the NOSE scale (0.38), and the COHIP-OSS (0.56). In the complete Standard Set, 6 out of 102 items were significantly outside the fit criteria and 14 out of 102 items had disordered thresholds; of these, the COHIP-OSS showed no ordering of thresholds at all.

Conclusions: This study illustrates that the majority of the patient- and family-reported components of the ICHOM Standard Set for cleft care (ie, the CLEFT-Q subscales and the ICS) are reliable tools for assessing domain-specific cleft outcomes. However, the NOSE and COHIP-OSS scales may not be particularly useful for accurately stratifying cleft-related outcome. This is likely because they seem to work like a symptom checklist rather than a measurement scale in the assessment of 8- and 12-year-old patients with cleft.

### 123. Trends in Utilization of Computer-Aided Surgical Planning in Pediatric Craniofacial Surgery

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Background/Purpose: While the use of computer-aided surgical planning (CASP) has been well-described in the adult craniofacial literature, there has been little written about pediatric uses or trends. The purpose of this study is to evaluate the evolving utilization of CASP for pediatric craniofacial procedures.

Methods/Description: Our prospective, IRB-approved craniofacial registry was queried for index craniofacial procedures from January 2011 through December 2018. Data were collected regarding utilization of traditional surgical planning versus CASP, as well as the extent of CASP's influence on the operative procedure. These data were analyzed for trends over time and compared using Pearson  $\chi^2$  test.

Results: During the study period, a total of 1131 index craniofacial cases were performed, of which 160 cases (14.1%) utilized CASP. Utilization of CASP collectively increased over time, from 2.0% in 2011 to 18.6% in 2018 (P < .0001). Utilization rates of CASP varied among procedures from 67% of osteocutaneous free tissue transfers to 0% of craniosynostosis cases (P < .0001). The most profound contributor to increase in CASP utilization was orthognathic surgery, utilized in 0% of orthognathic procedures in 2011 and 68.3% of orthognathic procedures in 2018. Moreover, utilization of components of the CASP system demonstrated unique footprints for each procedure. Free fibula transfer cases utilized all CASP components for essentially every procedure during which the CASP workflow was engaged; for example, 100% of those cases utilized computer preoperative planning, 92% of cases utilized stereolithographic models, 100% of cases utilized intraoperative cutting guides, and 69% utilized prefabricated implants. On the other hand, almost all orthognathic cases utilizing CASP enlisted the support of computer preoperative planning and intraoperative splints, but models, guides, and prefabricated implants were rarely utilized. For orthognathic procedures, we noticed a significant difference in the percent utilization of stereolithographic models, intraoperative guides, and prefabricated implants when compared over the years (P < .0001). Most other procedures showed stable utilization of all components over the study period.

Conclusions: Utilization of computer-aided surgical planning for pediatric craniofacial procedures is increasing, especially for complex orthognathic procedures and osteocutaneous free tissue transfers. Utilization patterns of individual components within the CASP system demonstrate unique footprints across the spectrum of craniofacial procedures, which reinforces the specific and variable benefits of this workflow for treating pediatric craniofacial disorders.

### 124. Long-Term Speech Outcomes in Patients With Robin Sequence After Tongue-Lip Adhesion

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Background/Purpose: Tongue-lip adhesion (TLA) is commonly used as a surgical treatment for upper airway obstruction (UAO) in patients with Robin sequence (RS). The effect on speech outcomes after TLA and subsequently cleft palate (CP) repair in patients with RS is insufficiently investigated. The aim of this study is to assess the effect of TLA on long-term speech outcomes in patients with RS.

Methods/Description: All consecutive patients with RS (with or without TLA, 1993-2014) who underwent CP repair according the von Langenbeck technique with intravelar veloplasty at the Amsterdam UMC were retrospectively reviewed and compared to patients with isolated cleft palate (ICP). RS was defined as micrognathia, glossoptosis, and UAO, and in patients with "isolated," either in isolated RS or ICP, there was a clear documentation of no other anomalies. Variables analyzed were sex, CP-type, age at TLA, associated syndromes with RS, and age at CP repair. Speech was assessed as binary outcomes by the senior craniofacial speech pathologist of multidisciplinary cleft team and included all assessments between the age of 3 and 6 years. These outcomes included: secondary speech operation, velopharyngeal insufficiency (VPI), hypernasality, and articulation errors by assessment of cleft speech characteristics (CSC), including 4 categories: 1. Passive, 2. Nonoral, 3. Anterior oral, and 4. Posterior oral. Results: Forty-one patients with RS (56% syndromic, 44% isolated RS) and 61 patients with ICP underwent CP-repair (at median ages of 9.3 and 9.4 months, P = .207, respectively) with sufficient speech follow-up. Of the patients with RS, 23 (56%) underwent a TLA at a median age of 12 days (range: 2-100 days). CP characteristics were different: soft palate only 24% RS versus 59% ICP, soft + hard palate 76% RS versus 41% ICP, P = .001. Median age at time of all speech evaluations was 4.5 years (range: 3.0-6.3) for RS versus 4.6 years (range: 3.3-6.3) for ICP, P = .581. VPI was diagnosed in 73% of the RS-group versus 62% of the ICP-group, P = .253. Rates of hypernasality and secondary speech operations to resolve VPI were higher in RS compared to ICP, both P = .003. In RS, the posterior oral CSC demonstrated higher rates compared to ICP, P = .038. When comparing RS+TLA versus RS-TLA, no significant differences were observed in variables CP-type and underlying syndromes between the 2 groups. Patients with RS+TLA did not demonstrate differences in VPI, hypernasality, secondary speech operations, or any of the CSC's

(all P > .05) compared to patients with RS without TLA, except for the anterior oral CSC's (74% RS+TLA vs. 28% RS-TLA, P = .003).

Conclusions: Our data demonstrates that patients with RS have worse speech outcomes compared to patients with ICP. In patients with RS, a TLA was not associated with worse speech outcomes compared to RS patients without TLA. However, patients with RS+TLA demonstrated higher rates of anterior oral CSC, that might be related to a different positioning of the tongue after TLA.

## 125. Promising Information—Cognitive Function and Stigma in Patients With Single Suture Craniosynostosis

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Background/Purpose: Children with craniofacial anomalies are often stigmatized and at greater risk for psychosocial disturbances and cognitive impairment than their peers. Studies have shown that children with craniosynostosis have lower appearance ratings and modest differences in neurodevelopment and intelligence compared to healthy children. To date, there is no reliable, convenient, and validated method to measure psychosocial parameters in children with nonsyndromic single suture craniosynostosis (SSC). The purpose of our study is to use the Patient Reported Outcomes Measurement Information System (PROMIS) to evaluate the perception of stigma and cognitive function in nonsyndromic SSC patients.

Methods/Description: Stigma and cognitive function were measured in 42 consecutive patients, 5 years and older, presenting to clinic with repaired nonsyndromic SSC using the National Institute of Health's PROMIS questionnaire with transformed scores normalized for the general population (mean  $\pm$  standard deviation,  $50 \pm 10$ ). Questionnaires were administered as part of clinical care via Research Electronic Data Capture (REDCap) from July 2018 to May 2019. Scores were automatically transferred to electronic medical records. Parents were asked to fill out the Parent Proxy Cognitive Function Questionnaire if the child was younger than 8 years (n = 21). The child's responses were entered for the stigma questionnaire regardless of age. Computerized-adaptive testing was utilized to reduce survey burden and improve sensitivity. As a result, the number of questions answered by each participant varied. Statistical analysis was performed using SPSS version 25.

Results: Forty-two patients (50% male, 39 white patients, 2 African-American patients, and 1 Asian patient) were treated for nonsyndromic SSC and participated in this study (18 sagittal, 12 unicoronal, 11 metopic, and 1 frontosphenoidal). The average age at follow-up was 8.4 years (range = 5-18 years). Our cohort had equivalent cognitive function scores (mean = 51.8  $\pm$  10.2, 95% CI = 48.7-55.0, P < .001) when compared to children without SSC. Children with repaired SSC perceived less stigma (41.7  $\pm$  6.8, 95% CI = 39.6-43.8, P < .001) compared to healthy children. There was a significant negative correlation between the 2 scales (Spearman rho = -.629, P < .001).

Conclusions: Based on patient-reported outcomes, children with repaired SSC have equivalent cognitive function and feel less stigmatized than healthy controls. Lower perceptions of stigma were associated with higher cognitive function scores. These results do not align with previously reported in-depth assessments in patients with repaired SSC which found small but significant deficits in intelligence and small but significant elevations in perceived stigma. Future research is needed to explain the discrepancy. Nevertheless, PROMIS

questionnaires offer a convenient, validated method of measuring psychosocial parameters in children with SSC which might otherwise be difficult to obtain during standard follow-up visits.

## 126. Is Poverty Associated With Orofacial Clefts? An Analysis of Orofacial Cleft Risk and Socioeconomic Status Utilizing the United States Birth Data

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Background/Purpose: Increasing evidence suggests that lower socioeconomic status (SES) is associated with delayed cleft care; however, the relationship between poverty and the incidence of cleft lip/palate remains elusive. We aim to investigate the association between SES and cleft lip with or without cleft palate (CLP) and cleft palate only (CPO) in the United States (US) after controlling for demographic and environmental risk factors.

Methods/Description: United States 2016 and 2017 birth data (n=7820~866) were abstracted from the National Bureau of Economic Research's Vital Statistics Natality Birth Data. We excluded births with missing data for CLP or CPO (n=11~765), with both diagnoses of CLP and CPO (n=268), or with both cleft and another congenital or suspected chromosomal disorder (n=479). Individual-level proxies for SES included maternal education, use of the Special Supplemental Nutrition Program for Women, Infants, and Children (WIC), and payment source for delivery. Multiple logistic regression was used to control maternal and paternal demographics, prenatal care, maternal health risk factors, and infant sex and weight.

Results: 6 250 956 live births with complete data were included in the multiple regression analysis; 3547 (0.06%) patients with CLP and 1418 (0.02%) with CPO were identified. Maternal education level of bachelors' degree or higher and prenatal care started in the first trimester were protective against CLP (AOR = 0.73 [0.62-0.85], P <.001 and AOR = 0.65 [0.48-0.87], P = .004, respectively) but not CPO. Positive WIC status was associated with an increased risk of CPO (AOR = 1.24 [1.06-1.45], P = .009) but not CLP. Source of payment for delivery was not associated with the risk of either CLP or CPO. Other risk factors for CLP included male sex (AOR = 1.67 [1.55-1.81], P < .001), first-trimester tobacco smoking (AOR = 1.02 [1.00-1.03], P = 0.03), maternal prepregnancy and gestational diabetes (AOR = 1.47 [1.06-2.03], P = .02 and AOR = 1.20 [1.04-1.39], P = .01), and birth weight less than 1500 g (AOR = 1.61 [1.25-2.041, P < .001). Other risk factors for CPO included female sex (AOR = 1.30 [1.15-1.47], P < .001), prepregnancy tobacco smoking (AOR = 1.02 [1.01-1.03], P = .005), maternal infections during pregnancy (AOR = 1.72 [1.23-2.40], P = .002), and increasing paternal age (AOR = 1.01 [1.00-1.03], P = .029).

Conclusions: Orofacial clefting was associated with multiple indicators of lower SES. Interestingly, different indicators of lower SES (level of maternal education, WIC status) were associated with different cleft phenotypes (CLP and CPO, respectively). Given that other demographic and environmental factors were controlled in this analysis, these findings suggest that poverty may be the latent factor to the observable associations of maternal education and WIC status with cleft incidence. Our model also confirmed known risk-modifying factors for CLP that are environmentally mediated but associated with lower SES, such as first-trimester tobacco smoking.

### 127. Long-Term Speech Outcomes Following Midface Distraction in Syndromic Craniosynostosis

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*Background/Purpose*: The aim of this study was to examine the effect of midface distraction on speech in patients with syndromic craniosynostosis using the Pittsburgh Weighted Speech Scale (PWSS).

Methods/Description: A retrospective review was completed of patients with syndromic craniosynostosis who underwent midface distraction at the Lefort III level or higher from 2003 to 2019 at a single academic hospital and had complete pre and postoperative speech records. Demographic information, type of procedure, and PWSS were recorded. Subgroup analyses were also performed based on syndrome type and level of advancement.

Results: Seventy-six syndromic patients underwent midface distraction during the study period, but only 19 patients met all inclusion criteria. Patients had the following syndromes: Apert (n = 8, 42%), Crouzon (n = 7, 37%), Pfeiffer (n = 4, 21%). Median age at time of surgery was 6.7 years (range = 4.6-16.3 years), and median length of follow-up was 57 months (range = 7-129 months). Overall, the highest postoperative PWSS score was significantly higher than the preoperative PWSS (.52 vs 2.4, P = .0073) thus indicating a higher degree of VPI postoperatively. Specifically, the PWSS components nasal emission and nasality were significantly higher postoperatively than preoperatively (nasal emission: .053 vs 1.16, P = .0204; nasality: .053 vs .68, P = .035). Mean length of time from distractor removal to highest PWSS was 20.3 months (SD  $\pm$  26.1). There was no significant difference between preoperative PWSS and either the earliest postoperative PWSS (P = .090) or the latest postoperative PWSS (P= .31). On subgroup analysis, 10 patients (52.6%) had preoperative scores of 0 that increased to a non-zero PWSS after surgery (range: 1-6), and decreased at later follow-up. Eighty percent of these reduced scores returned to 0 while 20% returned to a lower, non-zero number (range = 4-5). Four patients (21%) had a PWSS of 0 at all measured points indicating velopharyngeal competency throughout. Three of these 4 patients (75%) had Crouzon syndrome and all underwent a LeFort III advancement.

Conclusions: Midface distraction in syndromic craniosynostosis is associated with a transient decrease in velopharyngeal competence that appears to resolve over time to a large degree in most patients. Future research with a larger sample size would be helpful in determining risk factors for VPI after midface distraction.

#### 128. Clinical Course of Temporomandibular Joint Ankylosis in Pediatric Patients With Craniofacial Anomalies

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Background/Purpose: Temporomandibular joint (TMJ) ankylosis is an uncommon but debilitating condition which can affect feeding, speech, dental health, facial growth, and quality of life. We present an institutional experience treating congenital and acquired TMJ ankylosis, detailing outcomes and potential risk factors of recurrence.

Methods/Description: Patients with ankylosis of the TMJ were identified through retrospective chart review (1976-2019). Clinical records, operative reports, and imaging studies were reviewed for demographics, surgical operations, and ankylosis including mean interincisal opening (MIO) and reankylosis.

Results: Forty-four TMJs with bony ankylosis were identified in 28 patients, 27(96.4%) of whom had syndromes. Mean age at any initial mandibular surgery was  $3.7 \pm 3.6$  (range: 0-14 years). Follow-up was  $13.7 \pm 5.9$  years. Sixteen (57.1%) patients had bilateral ankylosis. Nine cases of ankylosis were congenital, 16 were introgenic (4.5  $\pm$ 3.7 years from initial distraction osteogenesis or autologous mandibular reconstruction) referred from outside institutions in 6 cases, and 3 were postinfectious. Patients having their first mandibular operation at a younger age had more frequent reoperations for recurrent TMJ ankylosis, although this did not reach statistical significance. Improvement in MIO was 21.4 ± 7.3 mm. Ankylosis recurred in 21(75%) cases, 11 of which were iatrogenic, requiring an average of 2 reoperations (range: 1-8). Five patients with congenital TMJ ankylosis required gastrostomy and remained at least partially dependent. Five patients had tracheostomy at the time of TMJ ankylosis surgery: 2 were eventually decannulated and 3 required repeat tracheostomy after ankylosis recurrence and remained tracheostomy-dependent.

Conclusions: Craniofacial anomalies, younger age at mandibular surgery, and number of operations portend to increased risk of TMJ ankylosis as well as tracheostomy and gastrostomy dependence. Despite initial improvement in postoperative MIO, pediatric TMJ ankylosis is associated with high recurrence and multiple reoperations.

### 129. Family-Centered Cleft Palate Speech Therapy: A Model for Improved Speech Outcomes

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Background/Purpose: Team care has been the gold standard for complex patient care, improving decision-making and treatment outcomes. Many patient populations including craniofacial, cystic fibrosis, cancer, and so on have demonstrated superior results using the team model. Assembly line care (vs highly coordinated care) is fraught with problems. In fact, coordinated team care is ACPA's baseline standard for approved team status. In more recent years, children and their families have been included as integral members of the craniofacial team with family education, networking, and inclusion in treatment decisions becoming more standard. Thus, it is ironic that cleft palate speech therapy typically does not embrace this effective and desired paradigm of care. Instead, children attend speech therapy several days per week and often show little or no progress for months or sometimes even years. Cleft trained SLPs often spend considerable time consulting with community speech-language pathologists, often with limited success, and without providing similar capacity-building for the family. This paradigm creates a disconnect between the therapy room and the rest of the child's "talking hours." Cleft speech therapy is based on motor learning therapy which requires significant practice to relearn patterns of sound production. Collaborating with families to ensure they know how to implement the therapy goals in the child's environment and understand their crucial role in cleft speech therapy. With this model, the child receives infinitely more opportunities to practice correct productions once s/he leaves the treatment room. This will increase the likelihood that the child will reach those goals and show carryover more quickly and efficiently than the traditional therapist-patient model with contact once or twice per week. The

purpose of this session is to propose a workable model for family-centered speech therapy.

Methods/Description: Review of disciplines that routinely incorporate home-based components. Present neurological principles of learning contrast model for intense parent training and outcomes of community SLP training in Columbia, South America. Outline model of free cleft palate speech therapy in NYC that includes families and community SLPs. Propose workable craniofacial center-based family focused speech therapy program.

#### 130. The World Cleft Coalition: Team Reflections

Serena Kassam (I), Jamie Perry (2), Ruben Ayala (3), Erin Stieber (4), Gareth Davies (5), Usama Hamdan (I)

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Background/Purpose: The World Cleft Coalition (WCC) is an alliance of international nongovernmental organizations (NGO's) that work in the area of cleft lip and palate and engage in long-term local capacitybuilding partnerships. The WCC was initiated at the 13th International Congress of Cleft Lip and Palate and Related Craniofacial Anomalies in Chennai, India in 2017. The objective set forth by Congress delegates at the end of the first NGO day "NGO's: Think, Treat, Teach," was to encourage international collaboration in the development of an agreed set of globally recognized minimum core practice and best practice guidelines for safe and comprehensive cleft care. In addition to creating these guidelines, it was the collaborative process between the founding NGO's that was the unexpected strength and success of this initiative. It is an example of diverse organizations collaborating in the enhancement of cleft care. The purpose of this panel discussion is to present results of joint efforts to establish best practice guidelines and to solicit and engage attendees to discuss their experiences in building and supporting worldwide cleft care.

Methods/Description: Through collaboration among participating organizations (American Cleft Palate Craniofacial Association, European Cleft Organization, Global Smile Foundation, Operation Smile, Smile Train, and Transforming Faces) and consultations with outside constituents and partnerships, the WCC created an "International Treatment Program Standards." From February 2017 to Present (August 2019), the team has met monthly/bimonthly via videoconferencing. This exchange was built on sharing experiences between the founding NGO's on what is of primary importance for ensuring the delivery of high-quality cleft care in outreach settings. The group was able to present the core standards at the European Cleft and Craniofacial Equality Conference in Nis, Serbia Sept 2018. This was followed by the official launch of the Program Standards at the 76th ACPA Conference in Arizona, April 2019, and then a presentation at the European Cleft and Craniofacial Meeting in Utrecht, Netherland, June 2019. The goal was to compile standards for safe, comprehensive, and sustainable cleft care. The starting point was a review of existing guidelines (internal and external) to create globally recognized international treatment program standards. Focus was kept on ethical, safe, accessible, and patient-centered care, not on technique and timing. Attention to wording was focused on being inclusive and encouraging, avoiding mandatory and prescriptive language. These national and international presentations also allowed significant feedback from various participants. Several organizations have voiced their support. Statements of official support, versus endorsement, are being considered. A website was constructed to enhance awareness and dissemination of the proposed standards. This is an example of diverse organizations collaborating in the enhancement of cleft care globally.

#### 131. Nutritional Challenges in Oral Clefting: A Multidisciplinary Approach to Support Growth and Avoid Malnutrition in the Neonatal and Perioperative Periods

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Background/Purpose: Children born with oral clefting face a variety of challenges related to feeding and weight gain in the first year of life. These children are at increased risk for growth disturbance, malnutrition, and even delays in surgical care or complications as a result of slow growth or poor weight maintenance. In the neonatal period, families often struggle with learning or applying appropriate feeding techniques to maximize their infant's nutritional intake. Neonatal feeding challenges are compounded by eventual transition to solids, cup training, and perioperative dietary restrictions that can unnecessarily increase family stress during the first year. Nutritionally, there is also concern for children losing weight after surgical interventions as a result of pain, dietary restrictions, and increased nutritional requirements. Multidisciplinary management of feeding and growth challenges throughout childhood is crucial to support best outcomes for these children and their families. In our experience, comprehensive team-centered nutritional care should include the cooperation of a committed group of feeding specialists that evaluate and manage patients in both inpatient and ambulatory settings with an organized and multifaceted approach as the child ages. Our primary aims are to avoid malnutrition and support healthy growth and development while easing the stresses associated with feeding challenges in this special population.

Methods/Description: This multidisciplinary presentation will identify the role of each feeding-related specialty involved in our cleft nutritional management approach and discuss nutritional needs, pearls, and pitfalls at each stage of infant and child development. The importance of early, frequent, and supportive interactions with families will be stressed and we will detail the appropriate assessments and interventions necessary to help these children and their families meet age-appropriate nutrition and lactation goals throughout the first year of life. We will highlight ways in which different team compositions can maximize their newborn feeding support, teach food and cup transitions, and provide comprehensive growth and feeding recommendations. We will also discuss our strategies for perioperative nutritional interventions in the inpatient and ambulatory settings to support families, respect required dietary restrictions, and identify appropriate high-calorie nutrition sources to help prevent significant postoperative weight loss and help ensure the children will recover and heal appropriately after lip and palate surgery. The target audience for this presentation will be anyone involved with nutrition, feeding, or lactation education and assessment of patients with a cleft or craniofacial condition.

### 132. CAPS-A-AM Speech Ratings: Training, Practice, Calibration

Angela Dixon (1), Adriane Baylis (2), Kathy Chapman (3), Anna Barigayomwe (4), Kelly Cordero (5), Kristina Wilson (6), Cindy Dobbelsteyn (7), Judith Trost-Cardamone (8)

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TX, (7) Dalhousie University, Halifax, ND, (8) California State University– Northridge, Northridge, CA

Background/Purpose: Resonance and nasal air emission are commonly accepted speech parameters rated by speech-language pathologists (SLPs) on cleft palate and velopharyngeal dysfunction (VPD) teams. These parameters can be considered complex to newer SLPs or to other disciplines within the team. This can cause confusion for patients and families as terms are used interchangeably or incorrectly by different disciplines. Additionally, experienced SLPs may have specialty training in this area, but barriers such as distance, time, patient privacy, and so on make it difficult to calibrate subjective clinic judgments with other specialty trained SLPs. This study session seeks to define the speech parameters of hypernasality, hyponasality, and audible nasal emission as defined and utilized with the Cleft Audit Protocol for Speech-Augmented-Americleft Modification (CAPS-A-AM, Chapman et al., 2016). Additionally, this study session will provide opportunities for the novice rater to practice listening and rating speech samples, as well as an opportunity for the experienced SLP to calibrate judgments.

Methods/Description: This study session will be interactive in format. Time initially will be spent discussing and defining the speech parameters and rating scale points and definitions. Speech samples will be utilized to illustrate each parameter discussed. Attendees will be given the opportunity to rate different samples on each parameter, allowing time for discussion. To conclude 1 to 3 case studies will be presented and attendees will have the opportunity to rate on a multitude of parameters at one time, consistent with the SLP's role in the clinic setting.

### 133. The Black Hole: Prevention and Correction of Fistulas Following Cleft Palate Repair

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Background/Purpose: Fistula following cleft palate repair is one of the most significant long-term complications of cleft palate care, as persistent nasal air-escape impairs speech and persistent oronasal regurgitation impacts dental, Eustachian, and nasal hygiene. Reoperation to close a fistula has considerable burden on children and the additional scar limits velar movements and impairs facial growth. The chances of successful palatal closure diminish with each additional surgical attempt and, as such, successful primary cleft palate repair or reconstruction of an existing fistula is of paramount importance to long-term outcome and quality of life.

Methods/Description: This session will involve a combination of presentations, case examples, and panel discussion of difficult cases. Prevention: We will start with detailed surgical anatomy and its application to surgical dissection. We will describe technical maneuvers that can assist with closure and highlight potential pitfalls that can result in complications. We will discuss potential "lifeboats" that can be used during repair. We will emphasize technical nuances that may help to avoid complications. Correction: Fistulas can occur in various locations and in various magnitudes. We will describe surgical options and approaches to closure of a fistula if one has occurred. We will use cases that illustrate potential scenarios. Case discussion: A significant portion of this session will be dedicated to presentation of cases and detailed interdisciplinary discussion. Participation by all will be greatly encouraged.

#### 134. Mandibular Distraction Osteogenesis for Neonatal Airway Obstruction: Optimizing Results With Evidence-Based Patient Selection and Use of Virtual Surgical Planning

Raj Vyas (1), Melissa Kanack (1), Irfan Ahmad (2), Neal Nakra (2) (1) University of California, Irvine, Orange, CA, (2) Children's Hospital Orange County, Orange, CA

Background/Purpose: The objective of this study session is to discuss recent interdisciplinary advances in the management of neonatal airway obstruction. Existing surgical/nonsurgical treatment choices and patient selection algorithms are reviewed. We then present new polysomnography data (including previously unknown normative data on neonates in the first weeks of life) in order to highlight evolving considerations in identifying candidates for mandibular distraction. We discuss the promise of light-based volumetric probes currently being studied at our institution that offer a more objective assessment of the degree and location of neonatal airway obstruction. We will review the advantages and step-by-step technique of incorporating computer-assisted modeling/design to enhance surgical outcomes and minimize complications from mandibular distraction. Finally, perioperative considerations and complications are reviewed.

Methods/Description: This presentation is evidence-based and systematic in its review of existing practice patterns and evolving techniques and technologies. The presentation will focus on the contributions of multiple specialties to evidence-based optimal patient selection. New polysomnography data on normative infants in this age range is presented and used to evaluate, for the first time, existing algorithms of care. Additional contributions from ENT, neonatology, pulmonology, anesthesia, genetics, GI, therapists, and many others are discussed to emphasize the importance of a multidisciplinary/interdisciplinary approach to patient selection and management. Illustrations, photographs, video, and 3-dimensional models are used to highlight the planning and execution of computer-assisted surgery. Audience members will have the opportunity to handle and assess 3-dimensional models, cutting guides, and distractor devices during the presentation and after. The author will engage audience members to share current practice patterns and areas of improvement while providing an overview of his own learning curve with these approaches as well as offering practical solutions and perspectives.

# 135. Genetic Testing in the Craniofacial Clinic: On the Development of Algorithms for Testing in Patients With Orofacial Clefting, Craniosynostosis, and Branchial Arch Anomalies

Anne Hing (1), Emily Gallagher (1), Yvonne Gutierrez (2), Glenn Rosenbluth (3), Ophir Klein (3), Daniela Schweitzer (2), Hazel Perry (3), Jessica Kianmahd (4), Michael Cunningham (5)

(1) Seattle Children's Hospital, WA, (2) Children's Hospital Los Angeles, CA, (3) University of California San Francisco, CA (4) University of California Los Angeles, CA, (5) University of Washington, Seattle, WA

Background/Purpose: The majority of patients cared for by multidisciplinary craniofacial teams have conditions with orofacial clefting, craniosynostosis, or branchial arch anomalies. Team providers must determine whether the patient's findings represent an isolated condition or possible underlying genetic syndrome. For each patient, the provider must make recommendations for additional assessments ranging from laboratory tests, imaging, and subspecialty consultation to assure that the child is safe and ready for surgical procedures and to guide medical management. This includes need for and timing of

genetics consultation and testing. The goals of this workshop are to: (1) provide a framework for providers regarding indications for genetic assessment of children with orofacial clefting, craniosynostosis, and branchial arch anomalies, and impact of the genetic diagnoses on medical and surgical management, and (2) determine utility of proposed genetic testing algorithms for children with orofacial clefting, craniosynostosis, and branchial arch anomalies.

Methods/Description: A panel of experts in craniofacial medicine (pediatricians and geneticists) will discuss their approach to the genetic assessment of patients with orofacial clefting, craniosynostosis, and branchial arch anomalies. Presenters will provide workshop participants with (1) a summary of frequency of underlying genetic causes, (2) potential impact of genetic testing on patient care, and (3) an algorithm for genetic assessment and testing for each major category. Workshop participants, assisted by experts will have the opportunity to apply the algorithms in the assessment of provided case scenarios.

### 136. Working Differently Together: Building Team Strength Through Our Differences

Richard Kirschner (1)

(1) Nationwide Children's Hospital, Columbus, OH

Background/Purpose: A unified vision is critical to successful team care, but team members inevitably bring different personality types and leadership styles to the teams that they serve. Although different temperaments may be a source of conflict in team relationships, they can also be a source of great synergy and strength. In order to best care for patients, communication between team members must be transparent, frequent, and authentic. In this highly interactive session, we will share ways to understand and capitalize on our differences in order to provide effective team care to our patients through both everyday challenges and periods of tumultuous change.

Methods/Description: This session includes a look at different personality types and leadership styles. Through storytelling and examples from the corporate world, we will share tips, tools, and takeaways to help team members forge a positive relationships with one another and to successfully work differently together. A high-level introduction of the Myers-Briggs personality types and Kiersey temperaments will be used to stimulate a discussion of how to best work across type and temperament. Small-group role-play and discussion will apply this knowledge to illustrative scenarios. A question and answer period will follow.

### 137. Speech and Language Disorders in 22q11.2 Deletion Syndrome: Management Guidelines

Cynthia Solot (1), Adriane Baylis (2), Debbie Sell (3), Anne Mayne (4), Christina Persson (5), Oksana Jackson (1), Donna McDonald-McGinn (1)

(1) Children's Hospital of Philadelphia, PA, (2) Nationwide Children's Hospital, Columbus, OH, (3) Great Ormond Street Hospital NHS Trust, London, United Kingdom, (4) NHS, Essex, London, (5) Institute of Neuroscience and Physiology, Speech and Language Pathology Unit, Gothenburg, Sweden

Background/Purpose: Speech and language disorders are hallmark features of 22q11.2 deletion syndrome (22qDS). Learning disabilities, cognitive deficits, palate abnormalities, velopharyngeal dysfunction, behavioral differences, and various medical and psychiatric conditions are also major features of this syndrome. The complexity and variability in the phenotypic presentation of 22qDS poses significant management challenges to craniofacial teams and speech-language

pathologists (SLPs). While general medical care guidelines for management of 22qDS have been available for several years (Bassett et al., 2011), specific suggestions for speech-language pathologists, and others engaged in the management of velopharyngeal dysfunction and communication disorders in 22qDS, were still needed. The purpose of this session is to summarize the state of the art of current clinical and scientific knowledge regarding speech-language disorders in 22qDS and provide recommendations for best practices for clinical management.

Methods/Description: An expert international group of SLPs and health-care providers with extensive clinical and research experience in 22qDS was formed, and over a period of several years developed a consensus set of principles and practical suggestions for management of communication disorders in 22qDS (Solot et al., 2019). In this session, a general overview of the phenotype and care needs of individuals with 22qDS will be presented. 22qDS best practices for diagnosis and management of speech-language disorders in 22qDS will be discussed. In this session, members of the expert consensus group will present and discuss the best practices, including diagnostic procedures, treatment protocols and associated management recommendations for SLPs, surgeons and other practitioners caring for this population across the life span. Approaches to implementing 22qDS best practices will be discussed, as well as strategies to optimize outcomes and minimize long-term communication impairments.

### 138. Sequential Care for the Patient With Hemifacial Microsomia

John Girotto (1), John Polley (1), Ann Schwentker (2), Kongkrit Chaiyasasate (3), Sharon Aronovich (4)

(1) Helen DeVos Children's Hospital, Grand Rapids, MI, (2) Cincinnati Children's Hospital Medical Center, Cincinnati, OH, (3) William Beaumont Hospital, Royal Oak, MI, (4) University of Michigan, Ann Arbor. MI

Background/Purpose: The most challenging reconstructions in cranio-maxillofacial surgery are hypoplastic asymmetries. Hemifacial microsomia (HFM) is the most common. HFM is a variable and heterogeneous malformation involving the lower two-thirds of the face and ear. This represents embryologic hypoplasia of the structures arising from the 1st and 2nd brachial arches. The challenges of reconstructing severe grades of HFM are well recognized. Numerous protocols for autologous reconstruction have been advanced. Unfortunately, these have not been substantiated in patient series with consistently acceptable results. Coordinated care often proceeds through puberty and can involve orthodontics, soft tissue restoration, skeletal orthognathic surgery, TMJ reconstruction, and auricular correction or prosthetics. To improve the outcomes for children with HFM and discuss the multiple stages of surgical/orthodontic interventions. New customizable techniques will also be presented.

Methods/Description: This upper level course will start with a brief discussion of HFM and its etiology. We will then review our extensive experience with distraction osteogenesis. Emphasis will be placed on long-term follow-up and the significant relapse that is experienced with these unique patients. For patients with class IIB and III HFM, we will present our techniques for customized TMJ reconstruction. We will also present customized TMJ options for other conditions. The role of orthodontic coordination is emphasized. The soft tissue envelope will be addressed next. Comparisons between microsurgical interventions and fat grafting will be presented. Finally, our preferred method for autologous ear reconstruction and the unique technical challenges of the patient with HFM are reviewed.

## 139. The Multidisciplinary Management of Obstructive Sleep Apnea in Children With Jaw Deformities, Dental Problems, and Craniofacial Conditions

Zarmina Ehsan (1), Michael Lypka (1), Heather Hendricks (1), Dan Jensen (1)

(1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: Obstructive sleep apnea (OSA) is prevalent in up to 5% of children and if left untreated can lead to morbidity across multiple health-related outcomes, including neurocognitive, cardiovascular, and metabolic sequelae. Although adenotonsillectomy has historically considered curative, we now know that in a significant proportion of children (up to 40% in a recent meta-analysis) AT does not result in complete resolution of OSA. The prevalence of persistent OSA (defined as OSA that persists despite first-line therapy with AT) is particularly high in those with severe OSA at baseline, obesity, and underlying medical complexities (particularly craniofacial malformations). Limitations of the second-line treatment option, continuous positive airway pressure (CPAP), are that adherence is low and it may be less effective for children with craniofacial malformations, and good adherence may contribute to abnormal facial growth in this population. Using a case-based, interactive format, this session will review a practical, evidence-based and systematic approach to evaluate and manage this growing cohort of children with obstructive sleep apnea in the context of craniofacial malformations focusing on individualized, patient-centered care.

Methods/Description: This presentation will be structured as follows: (1) Introduction: High impact, brief overview of pediatric obstructive sleep apnea focusing on the burden of disease and available management options (Zarmina Ehsan, MD, Pulmonary and Sleep Medicine [5 minutes]); (2) Understanding sleep disordered breathing and obstructive sleep apnea in children with craniofacial abnormalities, jaw deformities, and dental problems (Zarmina Ehsan, MD, Pulmonary and Sleep Medicine [20 minutes]); (3) Surgical treatment of obstructive sleep apnea in children: beyond adenotonsillectomy (Daniel Jensen, MD, Otolaryngology [25 minutes]); (4) Surgical management of obstructive sleep apnea in children with jaw deformities, dental problems, and craniofacial conditions (Michael Lypka, DMD, MD [25 minutes]); (5) The role of orthodontics in managing obstructive sleep apnea in children (Heather Hendricks, DDS [25 minutes]); (6) Wrapup and concluding remarks (Zarmina Ehsan, MD [5 minutes]); and (7) Question and answer session with entire panel (15 minutes). A 2-hour advanced course is the best format for this presentation as it will allow the abovementioned experts to review what is known, present new and emerging research data, and provide practical recommendations using a case-based interactive format. The question and answer session at the conclusion of this symposium will allow a unique opportunity for ACPA 2020 attendees to interact with the experts and discuss challenging cases.

### I 40. Bilateral Alveolar Cleft Grafting FromConventional to Complex: Double Your Pleasure?

Michael Lypka (1)

(1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: Most literature and educational content on alveolar bone grafting in the patient with cleft lip and palate is focused on the unilateral situation. The bilateral case, while occasionally straightforward, is unarguably more complicated from a technical perspective and results are typically less favorable than in the unilateral case. Bilateral cases are often complicated by a malpositioned, often diminutive, premaxilla, large anterior palatal fistulae, or

multiple previous failed attempts at grafting. Surgical access for proper soft tissue closure and maintenance of blood supply to the premaxilla can be a challenge. The purpose of this presentation is to discuss the management of alveolar clefts in patients with bilateral cleft lip and palate, starting with the conventional and moving to the more complex.

Methods/Description: The basic technique of bilateral alveolar cleft grafting will be described. Splint fabrication will be discussed. More complex cases will be highlighted, including advanced methods for closure, such as premaxillary osteotomy, segmental osteotomies, distraction osteogenesis, and soft tissue flaps. The presentation will be case based with photographic and video descriptions, with audience participation welcomed.

## 141. Innovation in Cleft Palate Reconstruction: How the Anatomic Palate Restoration Philosophy Can Guide the Use of the Buccal Flap

Robert Mann (1)

(1) Spectrum Health Medical Group, Grand Rapids, MI

Background/Purpose: The challenge for the cleft palate reconstructive surgeon is to successfully repair every type and anatomical variation of cleft palate. Presently, the vast majority of surgeons use single pattern repairs which are highly inflexible and may only work well on certain anatomic presentations. Many surgeons revert to older, more growth restricting techniques on complete or wide clefts. The Double Opposing Z- pasty +/- Buccal Flap Approach (DOZP+/-BFA) fits within the Anatomic Palate Restoration Concept and has demonstrated excellent speech results regardless of cleft width or classification. In addition to excellent speech results, excellent growth results have also been presented. Nonanatomic secondary VPD surgeries such as Pharyngeal Flaps and Sphincteroplasties are fraught with problems. Whereas, the Buccal Flap Palate Lengthen Procedures have proven successful and leave patients with more normal anatomy. The purpose of this workshop is to teach the Anatomic Palate Restoration Philosophy. This workshop will also demonstrate how to use the buccal flap and other useful flaps to more effectively reconstruct the cleft palate, thus improving outcomes in both primary and secondary palate repair.

Methods/Description: The buccal myomucosal flap has proven its great applicability in cleft palate repair. The use of this flap and others will be taught via video, slides, didactic presentation, and audience interaction. The senior presenter will relate his experiences with over 1500 buccal flaps surgeries used in various cleft palate applications. The Anatomic Palate Restoration Philosophy (APRP) will be described. Using the APRP has led to the development of multiple reconstructive procedures that leave the patients with more normal anatomy.

#### 142. Clinical Considerations for Safe and Positive Feeding Advancement for Infants With Cleft Palate or Cleft Lip and Palate From Birth to 13 Months

Allyson Goodwyn Craine (1)

(1) Kaiser Permanente, Portland, OR

*Background/Purpose*: Infants with cleft palate and cleft lip and palate present with feeding challenges. The purpose of this session is to review their unique swallow physiology and clinical considerations to safety advance feeding skills from birth to 13 months of age.

Methods/Description: Newborns with cleft palate present with specific feeding needs based upon unique anatomy and swallow

physiology. Key clinical considerations for safe and positive feeding experiences and progression in the first 13 months of life of infants with cleft palate (CP) and cleft lip and palate (CLP) will be explored. Feeding considerations for infants with cleft lip or cleft of the soft palate only will not be addressed in this presentation. In this session we will review: swallow physiology unique to infants with CP/CP. Explore how compensatory muscle movements and poor or absent inner oral pressure impact the oral, pharyngeal, and esophageal phases of the swallow. Eight clinical considerations for feeding the newborn with CP/CLP including hypoglycemia intervention considerations, swallow safety, support at breast, initial feeding, endurance concerns, positioning, the importance of burping, and tips of clearing nasal congestion while feeding without the use of suction. Bottle selection and developmental feeding strategies for infants with CP/CLP. Specific clinical considerations when selecting common bottles designed for infants with cleft palate and their progressive use before and after cheilorrhaphy and as the infant matures. Specific positioning, handling, and feeding techniques are also reviewed. Successful advancement of solids for infants with CP/CLP provides strategies to safely advance the infant's capacity to safely eat solids and optimize oral motor development from 4 to 6 months through palatoplasty between 9 and 12 months of age. Review of pre and postoperative feeding follow-up after discharge proven to provide strong outcomes with intentional continuity of care.

## 143. Comprehensive Management of the Patient Undergoing Distraction Osteogenesis—A Primer for Nurses/Coordinators

Patricia Chibbaro (1)

(1) Wyss Department of Plastic Surgery, NYU Langone Health, New York, NY

Background/Purpose: Distraction osteogenesis is a well-established standard of care for the patient requiring craniofacial reconstruction. It was first introduced in 1989 as an alternative to the traditional approach to lengthening of the human mandible. Over the course of 30 years, distraction has evolved into a treatment option for reconstruction of the entire craniofacial skeleton, beginning as early as the neonatal period. This intervention, as with cleft/craniofacial care in general, requires a comprehensive team approach to support the patient and family successfully through the treatment journey. The nurse/coordinator is often at the center of this process, which can be extremely challenging for both the patient and family, serving as the liaison between them and the other members of the team. This role/ responsibility can be especially overwhelming to the nurse/coordinator who is new to this position. Thus, the purpose of this foundational course is to provide a framework for comprehensive management of the patient/family undergoing distraction osteogenesis to the novice nurse/coordinator on a cleft/craniofacial team.

Methods/Description: Utilizing multiple case examples and drawing on 30 years of experience as a cleft/craniofacial nurse practitioner caring for patients undergoing distraction osteogenesis, the presenter will provide a comprehensive overview of this treatment option, with specific emphasis on the roles/responsibilities of the team nurse/coordinator. This will include identifying the types of distraction available (cranial, midface, maxillary, mandibular), determining who is a an appropriate candidate (both patient and family), reviewing the necessary preoperative workup for each procedure, discussing how to optimally prepare the patient and family (taking into consideration the age of the patient), reviewing postoperative care protocols and anticipating possible postoperative complications.

### **144. Prosthetic Management of Velopharyngeal Dysfunction**

Kaylee Paulsgrove (1), Kathryn Preston (1), Barbara Sheller (1), Randall Bly (1)

(1) Seattle Children's Hospital, Seattle, WA

Background/Purpose: Surgery is often the first-line approach in the management of velopharyngeal dysfunction (VPD) for individuals with cleft and craniofacial conditions. When surgical options have been exhausted or in cases where surgery is contraindicated, prosthetic management can be an effective alternative. With advancements in surgical techniques, use of prosthetics for the management of VPD has been eliminated from many centers; however, not all patients with VPD are optimal surgical candidates. Contraindications for surgical VPD management include obstructive sleep apnea, inadequate tissue due to previous surgeries, or serious medical conditions that contraindicate anesthesia or surgery (ie, certain cardiac disorders). Overall, the decision to pursue surgical versus prosthetic management of VPD should be determined by the multidisciplinary team. At our craniofacial center, the multidisciplinary team including otolaryngologists, plastic surgeons, speech-language pathologists (SLP), pediatricians, and orthodontists determine the patient's surgical versus prosthetic candidacy. Once an individual is referred for prosthetic management, the otolaryngologist performs ongoing instrumental evaluation in order to guide management. The orthodontist fabricates and modifies the speech appliances as needed. The SLP works in conjunction with the orthodontist to adjust the appliance to meet the specific VPD needs of the patient. The goals of this foundational course are to (1) discuss rationale for use of speech prosthesis in lieu of surgical management (2) facilitate multidisciplinary discussion about the decision-making process, and (3) provide an overview of the process involved in prosthetic management of VPD, from the initial evaluation to the final stages of prosthetic modification.

Methods/Description: This foundational course will include perspectives from a pediatric dentist/orthodontist, speech-language pathologist, and otolaryngologist with experience treating children with complex craniofacial conditions. Patient populations will be described, processes will be defined, and protocols will be discussed. Cases will be used to highlight key points. The presentation will provide an overview of the working relationship between the team members that are integral to the success of the speech prosthesis as well as specific information about holistic patient assessment, appliance selection and fabrication, and medical coding. The information presented in this session is meant to be of interest to SLPs, surgeons, pediatric dentists, orthodontists, prosthodontists, and any others involved in treating VPD in children with complex medical and/or craniofacial conditions.

### 145. Genetics 101: A Primer in Cleft and Craniofacial Genetics for the Non-Genetics Provider

Susan Hughes (1), Lauren Grote (1), Alison Kaye (1)

(1) Children's Mercy Kansas City, Kansas City, MO

Background/Purpose: It is well understood that orofacial clefting and craniofacial disorders can be syndromic or isolated, resulting from either a single genetic variation or a combination of multiple genetic and environmental factors, respectively. We commonly treat both isolated and syndromic anomalies within a multidisciplinary team setting. A genetic specialist may not always be readily available at the time of initial presentation. Therefore, it is imperative that the evaluating clinicians be able to recognize certain constellations of features associated with known cleft and craniofacial syndromes.

Having a basic understanding of the etiology will allow for appropriate navigation of the numerous genetic tests and interpretation of the results. This can allow for a more streamlined process and utilization of resources. Ultimately, understanding the genetic contribution to the clinical presentation will help clinicians provide better medical management for each individual patient.

Methods/Description: This lecture-based foundational course is designed to help explain the basics of genetic and environmental causation of common cleft and craniofacial conditions. This will serve as a primer on topics like inheritance patterns, genetic family trees, basic genetic testing strategies, and interpretation of results. We will discuss the role of a genetic counselor versus a clinical geneticist and what a formal genetic consult entails. Attention will be given to different specific syndromic and nonsyndromic diagnoses and describe how a clinician might work to recognize a child at risk for a formal genetic diagnosis. As a part of this education, we will explain how different genetic tests may or may not help identify a suspected problem, when and how certain tests should be ordered, and how those results are interpreted. Lastly, we will touch on how genetics researchers are working to identify new candidate genes which may have a role in cleft and craniofacial conditions.

### 146. Introduction of Ultrasound Technology in Speech Intervention for Children With Cleft Palate

Sue Ann Lee (1)

(1) Texas Tech University Health Sciences Center, Lubbock, TX

Background/Purpose: Visual feedback has been used in speech interventions for children with speech sound disorders in order to help the children visualize articulatory gestures and movements for various speech sounds. During the past decade, ultrasound became a new tool to provide visual feedback during speech intervention. Using this innovative technology, patients are able to see their lingual image on the monitor to identify correct or incorrect gestures. The majority of studies utilizing ultrasound focused on persisting errors of articulation within typically developing children (Byun et al., 2014; Preston et al., 2016; Preston et al., 2013). Research using ultrasound therapy was conducted with several special populations including patients with cochlear implants and childhood apraxia of speech (Adler-Bock et al., 2007; Preston et al., 2013; Gibson et al., 2017). However, very limited evidence is currently available for children with cleft palate. To our knowledge, only one study examined children with submucous clefts using ultrasound (Roxburgh et al., 2015). Furthermore, ultrasound has mainly used in research, but not in clinical practice. Most speech pathology practitioners who work with children with cleft palate are not familiar with such technology. The purpose of this foundation course (nonresearch) is to introduce how ultrasound technology can be used in speech intervention. Ultrasound will be a powerful tool to remediate children with cleft palate because it gives strong visual feedback on placement errors. It is well-known that children with cleft palate produces distinctive speech errors including glottal stop/fricative, pharyngeal stop/fricative, and mid-dorsum palatal stop. These sounds replace oral stop and fricatives resulting in placement errors. Using ultrasound, patients are able to identify lingual contour and gestures on the ultrasound monitor. Visual and verbal feedback to change lingual placement will help children with cleft palate improve their speech production accuracy.

Methods/Description: During presentation (120 min), we will start with discussion about fundamentals of ultrasound technology including definition of ultrasound and types of probes etc. Next, we will review English consonant system and show ultrasound images of each sound produced by normal speakers without any speech issues

appears. Then, we will show ultrasound images of children with cleft palate and video clips of ultrasound intervention sessions for children with cleft palate. Finally, we will provide participants with a hands-on practice session using ultrasound. We will teach verbal technique and tips.

## 147. Education of Medical Professionals to Improve Successful Feeding and Weight Gain in Infants With Cleft Lip/Palate

Helen Huff (1), Kristi Thaete (1), Brenda Sitzmann (1), Alison Kaye (1) (1) Children's Mercy Kansas City, MO

Background/Purpose: Children who are born with a cleft lip/palate face a variety of challenges related to feeding and weight gain in the neonatal period. Families often struggle with learning and using feeding techniques to help maximize their infant's nutritional intake. Our approach to comprehensive team-centered care for these patients includes assessment, treatment, and education provided by feeding specialists from Nutrition, Occupational Therapy, and Lactation. Education and resources are provided to the family beginning with the prenatal appointment. Despite this multidisciplinary, comprehensive, and coordinated care, we discovered a trend of newborns failing to meet weight gain goals. Contributing factors include varying degrees of knowledge regarding feeding and growth expectations from nurses and clinicians located outside our hospital at referring hospitals and physician offices. To address these challenges, our team developed an educational program for medical professionals who may care for these newborns outside of the cleft team setting. The target audience for this program are medical professionals who care for parents and newborns with cleft lip/palate and those in a position to help educate noncleft specialists on appropriate feeding goals and techniques. Our aims are to increase successful infant feedings demonstrated by improved growth and decreased admissions for failure to thrive in the cleft lip/palate population.

Methods/Description: This presentation provides an overview of our multidisciplinary educational program for medical professionals which focuses on cleft care for the newborn and why these infants have difficulty with breast and bottle feeding from a standard bottle nipple. We will describe our hands-on approach for providing this valuable cleft feeding education and support for primary care and bedside providers. Additionally, we will discuss how we have targeted our different learning cohorts for future replication of similar education programs elsewhere.

## 148. Standardization for Reporting of Nasopharyngoscopy and Multiview Videofluoroscopy: Where Are We Now, What Have We Learned?

Thomas Watterson (1), David Jones (2), Lynn Grames (3)

(1) University of Nevada, Reno School of Medicine, Reno, NV, (2) University of Wyoming, Laramie, WY, (3) St. Louis Children's Hospital, St. Louis, MO

Background/Purpose: ASHA Special Interest Group 5, Craniofacial and Velopharyngeal Disorders, offers this eye opener session of case presentations to ACPA attendees. In 1988, an international, multidisciplinary working group of craniofacial professionals convened to devise standard methods for reporting findings from velopharyngeal imaging studies. The intent was to provide seamless communication from one center to another, and to facilitate consistency in reporting research and treatment outcomes. The recommendations were published in 1990 with a call for validation studies. As time has passed, newer professionals have entered the field without awareness of the

standards, and validation studies are scarce, yielding concern for our evidence base. The intent of this eye opener is to review these standards, demonstrate their use, and discuss recent validity research in detail.

Methods/Description: The rationale for developing the standardized reporting approach will be reviewed, and the basic method will be presented. Video examples of nasopharyngoscopy and multiview videofluoroscopy will illustrate the methods of reporting, as well as the advantages and limitations of using the standardized approach. The results of a multicenter study that investigated intra-rater and inter-rater reliability in the evaluation of video clips of VP endoscopy for speech will be presented. In that study, 5 raters at 3 different centers rated video clips using metrics recommended by the International Working Group: the size of VP opening, location of opening in the VP port, and the pattern of VP closure. There were a total of 37 speakers in the study: 21 were normal speakers and 16 were patients being evaluated for VPD. Intra-rater reliability, measured by percentages of exact agreement, was high for each rater and for all metrics. However, inter-rater reliability varied from slight-to-moderate depending on the metric. Future direction for use of the standardized approach will be discussed.

#### 149. Strategies for Optimizing Audiologic Assessments and Minimizing Sedation for Young Children With Cleft Lip/Palate and Craniofacial Conditions

Krista Roper (1), Carlee Jones (1), Daniel King (1), Christine Holmes (1), Karma Tockman (1), Alexander Allori (1), Jeffrey Marcus (2), Ana Maria Fernandez (1), Jeff Cheng (3), Eileen Raynor (3)

(1) Duke University, Durham, NC, (2) Duke University Division of Plastic, Maxillofacial & Oral Surgery, Durham, NC, (3) Duke Health System, Durham, NC

Background/Purpose: Hearing loss has a significant impact across the life span, particularly during the school-age years when there may be considerable effect on education and psychosocial well-being. Early identification of hearing loss in young children has the potential to change the trajectory of hearing functionality, speech and language development, quality of life, and other outcomes. Given the importance of early diagnosis and management of hearing loss, it is critical to be able to evaluate infants and young children as early as possible. Unfortunately, this age range presents particular challenges for behavioral and objective testing, and often sedated auditory brainstem response testing (ABR) is warranted. In recent years, interprofessional guidelines have emerged calling for restraint in the use of general anesthetics in infants and young children due to potential deleterious effects on neurocognitive development. The purpose of this presentation is to describe alternative behavioral and objective audiometric techniques that may be used to avoid unnecessary sedation (or at least minimize the duration of sedation). The benefits and challenges of each method will be discussed, with particular focus on the appropriate age ranges to which these techniques apply in the cleft and craniofacial population.

Methods/Description: A pediatric audiologist with extended experience working as part of a multidisciplinary cleft and craniofacial team will address the overarching implications of temporary and permanent hearing loss in infants and children, particularly the impact on those with cleft and craniofacial conditions. We will review methods of behavioral and electrophysiological testing, including Visual Reinforcement Audiometry (VRA), Conditioned Play Audiometry (CPA),

unconventional techniques, two-person testing, Otoacoustic Emission Testing (OAEs), sedated or nonsedated Auditory Brainstem Response Testing (ABR), and Auditory Steady State Response Testing (ASSR). Test methods that do not require sedation will be discussed in depth, including the benefits and challenges of each modality. In addition, videorecorded examples of each method will help to demonstrate the testing techniques, assessments, and the subtle responses provided by children. The aim of this discussion is to broaden the cleft and craniofacial team's "toolkit." We hope this will encourage reflection of your team's current audiologic practices, spur consideration of alternative/adjunctive strategies in these challenging situations, and improve overall care for children with cleft and craniofacial conditions.

## 150. Advancing Genetic Technology: How Application of Genetic Diagnostic Testing Impacts the Management of Craniofacial Disorders

Patricia Bender (1), Farrah Mahan (1), Howard Saal (1) (1) Cincinnati Children's Hospital Medical Center, OH

Background/Purpose: Many craniofacial disorders have an underlying genetic etiology. Over 50% of children born with cleft palate and 30% of children born with cleft lip with or without cleft palate will have genetic syndromes. Many of these disorders are rare and, in the past, our ability to make diagnoses was limited. However, genetic and genomic testing has advanced tremendously over the past decade with the development of next-generation sequencing. This technology has provided the opportunity to make diagnoses of rare and unique genetic disorders. There still remains a need to perform tests which have been in existence for decades, including chromosome analysis and fluorescence in situ hybridization (FISH). The purpose of this study session is to educate participants about genetic testing, how and when it is done, and how the results of these tests directly impact the management of children with craniofacial disorders and their families.

Methods/Description: This study session will offer a comprehensive review of genetic and genomic testing. The presenters will review the different types of genetic tests available: (1) chromosome analysis, (2) fluorescence in situ hybridization (FISH), (3) Sanger sequencing, (4) chromosomal microarray, and (5) next-generation, whole-exome and genome sequencing and the application of appropriate tests to specific clinical presentations. Limitations of genetic testing and ethical issues which have arisen as a result of extensive genetic testing, whole-exome and genome sequence, will be explained. Multiple clinical examples and scenarios will be used to illustrate the principles of genetic testing and to encourage audience participation.

#### 151. Nuances in Cleft Lip Revisional Surgery: The Importance of Deformity Analysis, Surgical Planning, Timing of the Revision, and Long-Term Followup

Jeffrey Goldstein (1)

(1) Children's Mercy Hospital-Kansas City, MO

*Background/Purpose*: Secondary cleft lip deformities can be challenging to correct, sometimes more challenging than a primary cleft. They can result from the congenital deformity or be iatrogenic in nature. Scar, growth restriction, or complications from the primary

repair can further compromise the aesthetics of the lip. The purpose of this eye-opener will be to address secondary unilateral and bilateral cleft lip deformities. Great attention will be placed on analyzing the various defects and then planning and executing surgery based on the underlying problem. The effects of malocclusion and facial growth restrictions on lip aesthetics will also be addressed. Appropriate timing for different types of reconstruction will be reviewed. Long-term follow-ups will be presented with an emphasis on the effects of facial growth on cleft lip revisional surgery. Grafting techniques (autologous fat and mucosa) will also be discussed. Attendees are encouraged to bring or submit in advance to the primary author difficult revisional cases to discuss in the last 15 minutes.

## I 52. Understanding Speech/Resonance Disorders and Velopharyngeal Dysfunction Using a "Science Experiment" Approach

Ann Kummer (1)

(1) Cincinnati Children's Hospital Medical Center, OH

Background/Purpose: Many craniofacial professionals do not fully understand and appreciate the complexities of speech production. As such, they are not aware of how structure and the laws of physics can impact both resonance and speech production. Normal speech production is, in fact, very complex and involves the coordination of the subsystems of respiration, phonation, velopharyngeal function, and speech articulation. Because these subsystems must work together (like a team), an anomaly or dysfunction in one of the subsystems can cause dysfunction of the other subsystems. (For example, a velopharyngeal opening during oral speech production can have an effect on respiration for speech, resonance, and oral articulation. In some cases, it can even effect phonation.) The first purpose of this session is to describe the structure, function, and physics required for normal voice, speech sound production, resonance, and airflow using a "science experiment" approach for demonstration. The second purpose is to provide learners with a better understanding of how abnormal structure and function in the vocal tract can affect resonance and speech sound production.

Methods/Description: The presenter will describe how the various subsystems contribute to both the production of individual consonants and how we alter oral resonance for production of vowels. These descriptions will be augmented by simple demonstrations, analogies, and "science experiments" to enhance the learners' understanding. The presenter will then describe how variations in structure can cause normal variation in resonance and "voice" among individuals and how variations can cause abnormal speech and/or resonance. With this background, the presenter will explain how the perceptual features of a speech/resonance disorder due to velopharyngeal insufficiency can be used to predict the size of the velopharyngeal opening. Finally, there will be a discussion of how surgical alteration of structure in the vocal tract can affect the physics of sound and airflow in a positive (or sometimes negative) way. Many short video clips will be used for illustration. This lecture is appropriate for speech-language pathologists, but most importantly, it is appropriate for all other craniofacial professionals who want to have a better understanding of speech production and speech disorders.

#### 153. Cleft Lip Repair: Measure Twice Cut Once

Raymond Tse (1)

(1) Seattle Children's Hospital, WA

Background/Purpose: "The first time is the best time." Favorable initial cleft lip and nose repair is critical in providing patients with optimal long-term form and function. Current "cut as you go" techniques of cleft lip repair rely heavily on the abilities and experience of surgeons, and the myriad of variations reflects the free-form nature of those repairs. The lack of more defined methods can make cleft lip repair intimidating for new surgeons and can result in variability of outcomes for more experienced surgeons. The "Anatomic Subunit Approximation Approach" to cleft lip repair relies upon anatomic landmarks and anatomic subunits in the design of a cleft lip repair. It involves careful measurement and includes ways to check and recheck the design to help surgeons achieve favorable form. Studies have demonstrated optimal results without the learning curve that other techniques require. Because the approach is based on landmarks, it inherently adapts to all presentations. Although the technique has been widely adopted, the many landmarks and measurements make it seem complex. The purpose of this session is to describe the Anatomic Subunit Approximation approach to cleft lip repair and demonstrate how it adapts to various presentations to produce favorable results.

Methods/Description: We will use step-by-step explanations to illustrate and clarify the approach. We will use case examples that span the spectrum of presentations from complete to microform clefts. We will describe ways to verify and adapt the repair for final tailoring so that there is some flexibility in final repair. We will use a combination of photos, videos, illustrations, and discussions to ensure that attendees benefit from the session. Even if more experienced surgeons do not adopt the technique, an understanding of the approach can help with the nonovert landmarking that occurs with cut-as-you-go repairs. Careful planning makes for accurate and efficient repair and that ultimately benefits our patients!

### 154. Commission on Approval of Teams: ACPA Team Approval Process

Celia Heppner (1)

(1) Children's Medical Center, Dallas, TX

Background/Purpose: An overview of the team approval process and review by the Commission on Approval of Teams will be presented.

Methods/Description: This session will help to demystify the ACPA Team Approval process by review of the application standard by standard. Helpful hints on how to confidently prepare the application will be provided. Any specific questions team members might have about the application process are welcome, making for an interactive presentation.

## 155. Management of Feeding and Swallowing in Infants With Pierre Robin Sequence and Airway Obstruction

Loredana Cuglietta (1)

(1) Stollery Children's Hospital, Edmonton, Alberta, Canada

Background: Infants who have Pierre Robin sequence and cleft palate often face challenges with feeding and swallowing as a result of airway obstruction. Airway obstruction can be managed in multiple ways including continuous positive airway pressure (CPAP), sleep positioning, mandibular distraction osteogenesis (MDO), tracheostomy, and tongue—lip adhesion (TLA). Both clinical and instrumental feeding and swallowing assessments can provide the medical

team with the ability to make informed decisions regarding airway management. Therefore, collaborative care is essential when managing infants with airway compromise, while also supporting oral feeding.

*Purpose*: To discuss involvement of feeding and swallowing specialists to optimize airway management in the Pierre Robin sequence and cleft palate population.

Methods/Description: Case studies of infants followed at the Stollery Children's Hospital with Pierre Robin sequence and airway obstruction will be used to illustrate the following: (1) clinical and instrumental feeding and swallowing assessment options, (2) strategies to optimize oral feeding, and (3) collaborative team care, in essence feeding and swallowing outcomes that inform both nonsurgical and surgical airway management. This presentation is intended for anyone involved in the management of feeding and swallowing of patients with Pierre Robin and airway complications.

### 156. My Changing Face: Preparing Teens for Orthognathic Surgery

Amanda Johnston (1), Kristi Thaete (1)

(1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: Adolescents with various craniofacial diagnoses may develop jaw anomalies or altered jaw growth patterns that require orthognathic surgery. Jaw surgery can significantly alter a patient's appearance and improve their breathing, speech, and ability to chew. The majority of these surgeries are done once a child has completed their skeletal growth in their later teenage years. Often, these are some of the final and most complex surgeries cleft patients may experience. Nurses play a pivotal role in providing preoperative teaching for adolescents about surgical sequelae, common complications, period of recovery, changing facial aesthetics, and the expected course of rehabilitation after undergoing orthognathic surgery. By providing a thorough education in how to effectively manage their diet and activity restrictions, pain, swelling, and potential postoperative complications, teens will be better equipped to anticipate the major changes in jaw function and facial aesthetics as well as the abrupt changes in lifestyle that occur for the first 4 to 6 weeks following the operation. Thorough preoperative education that sets realistic postsurgical expectations can help adolescents and their families better manage their recovery course, reducing potential complications and complaints that can lead to emergency department visits or impact their postoperative healing.

Methods/Description: This PowerPoint presentation provides an indepth overview detailing the presurgical education provided to patients and their parents prior to undergoing orthognathic surgery to help manage expectations, improve compliance, and set realistic expectations of their postoperative course. Following liquid diet compliance is crucial to appropriate healing and an in-depth review of longer term liquid diet management and ideas is provided by a registered dietitian. Postsurgical pain control, oral care, diet and activity restrictions, and management of potential postsurgical complications both in the hospital and at home after discharge will be discussed. This presentation will also discuss suicide screening in the adolescent patient and focus on many of the unique care needs facing the adolescent patients and their family as well as preparing them for the psychosocial challenges

they may experience with a complex surgery that will alter their facial aesthetics.

## 157. 3-D Morphologic Characterization to Optimize Mandibular Distraction in Patients With Pierre Robin Sequence

Matthew Davis (1), Angela Volk (2), Amit Narawane (1), Robert Dempsey (1), Elton Lambert (2), John Wirthlin (2), Edward Buchanan (3)

(1) Baylor College of Medicine, Houston, TX, (2) Texas Children's Hospital, Houston, TX, (3) Baylor College of Medicine and Texas Children's Hospital, Houston, TX

Background/Purpose: In the management of airway obstruction in patients with Pierre Robin sequence (PRS), optimal patient outcomes are achieved when mandibular distraction osteogenesis (MDO) is performed as the primary surgical intervention. A number of previous studies have attempted to characterize the mandibular morphology of patients with PRS. At present, however, quantitative metrics of mandibular morphology cannot be easily translated to guide surgical planning and distractor vector selection. The aim of this study is to compare the mandibles of infants with nonsyndromic PRS to controls to characterize their morphological variance in a manner relevant to distraction. This study also examines whether morphologic measurements can predict which patients will require MDO and shows how airway view grades vary among patients requiring MDO.

Methods/Description: From October 2010 to March 2019, patients under 2 months old with nonsyndromic PRS were identified and age-and sex-matched to control patients. Demographic and perioperative data were recorded, including Cormack-Lehane airway view grades. Computed tomography (CT) scans from these patients were used to generate 3-dimensional (3-D) mandibular models. The following anthropometric landmarks were independently identified by 2 reviewers: bilateral condylions, bilateral gonions, and the menton. A high degree of inter-rater reliability was observed between reviewers. Linear and angular measurements were made. Wilcoxon rank sum and 2-sample t tests were performed. A P value of <.05 was considered statistically significant.

Results: A total of 24 patients (mean age: 0.64 months) with a diagnosis of nonsyndromic PRS and 24 control patients were included in this study. Of the patients with PRS, 17 required MDO, 5 were managed conservatively, and 2 required other management methods. Patients with PRS were found to have shorter ramus heights (16.7 vs 17.3 mm; P=.346) and shorter mandibular body lengths (35.3 vs 39.3 mm; P<.001) compared to controls. Gonial angles were more acute (125.3 vs 131.3°; P<.001) and intergonial angles were more obtuse (94.2 vs 80.4°; p<0.001) in patients with PRS compared to controls. No significant differences in mandibular measurements were found among patients requiring MDO versus conservative management, nor among patients requiring MDO with high versus low airway grades.

Conclusions: Our study examines both the largest and youngest PRS patient population to date, making it the most relevant regarding management of early airway obstruction with MDO. The observed statistically significant differences in mandibular body length and intergonial angle suggest that univector distraction of the mandibular body should allow for normalization of mandibular morphology in patients with nonsyndromic PRS. Our findings also indicate that determining the optimal treatment modality for airway obstruction in patients with PRS remains a clinical decision.

## 158. 5-Year Maintenance of the Pedicled Buccal Fat Pad Graft During Primary Palatoplasty: An MRI Case Study

Katelyn Kotlarek (1), Michael Jaskolka (2), Brad Sutton (3), Silvia Blemker (4), Bruce Horswell (5), Paul Kloostra (5), Jamie Perry (6)

(1) University of Wyoming, Laramie, WY, (2) New Hanover Regional Medical Center, Wilmington, NC, (3) Beckman Institute for Advanced Science and Technology, University of Illinois at Urbana—Champaign, IL, (4) University of Virginia, Charlottesville, VA, (5) Charleston Area Medical Center, WV, (6) East Carolina University, Greenville, NC

Background/Purpose: When used during primary palatoplasty, the pedicled buccal fat pad graft (BFP) is a well-documented surgical technique. Reports have hypothesized that this technique results in increased vascularized tissue within an otherwise denuded space at the posterior hard palate that is maintained after surgical healing. In such, it was expected that the increase in volume and vascularity provided by the BFP would prevent wound contracture, thus maintaining a longer velum and optimizing maxillary growth (Levi et al., 2009; Pappachan and Vasant, 2008). However, these outcomes to date have only been assessed via intraoral inspection and patient observation. Currently, MRI is the only technology that enables 3-D analysis of soft palate tissue structures in living individuals. The purpose of this case study was to (1) examine if the BFP is maintained within the velum at a 5-year postoperative time point and (2) quantify the structural changes in the velum as a result of BFP placement.

Methods/Description: In accordance with the institutional review board, a single participant was recruited as pilot data for a larger study. The participant underwent primary repair of the cleft palate with an intravelar veloplasty and pedicled BFP graft placement before 12 months of age. The MRI was performed using a Siemens 3 Tesla MRI scanner and head coil, which were used to scan the participant nonsedated while lying in the supine position. The MRI protocol, processing methods, and analysis are consistent with that used in previous literature. Both T1-weighted and T2-weighted images were obtained at rest.

Results: Clear borders of the BFP were visualized on the MR image. The BFP appeared as a bright white mass on the T1 image, indicating likelihood of fat content in that region. From the midsagittal slice, the length and height of the BFP were 11.77 and 3.23 mm, respectively. The participant's velar length (28.60 mm) and thickness (7.80 mm) were within the normative range for children of the same age and race (Perry et al., 2018). The effective velar length was 17.37 mm. At midline, the percentage of fat and muscle tissue were 14.02% and 18.56%, respectively.

Conclusions: This is the first case study to investigate preservation of the pedicled BFP graft in vivo after placement during primary palatoplasty. In this single case study, we demonstrate that the BFP is maintained within the velum up to 5 years postoperatively. To a certain extent, there is evidence that the BFP retains its adipose tissue properties and substantial mass within its terminal location in the velum. Employing a larger sample size with comparison groups will allow for evaluation of postsurgical anatomy and short- and long-term outcomes, which may contribute to a better understanding of the surgical impact on velopharyngeal morphology.

### 159. Addressing Ongoing Healthcare Needs in an Adult Cleft Clinic

Jeyhan Wood (1), Wesley Stepp (1), Michael Canfarotta (2), David Zajac (2), Luiz André Pimenta (3), Margot Stein (2), Renie Daniel (1), Joseph Clark (1), William Shockley (1), Amelia Drake (1)

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Background/Purpose: Cleft lip and/or palate (CL/P) anomalies are common congenital defects in the United States (1:900 live births). Pediatric and adult patients with these diagnoses may have many associated medical and physical challenges including speech impairments, difficulty hearing, issues with social integration, and challenges with self-worth and appearance. While some data exist on access barriers to primary craniofacial care, the specific medical and social needs of the adult CL/P population could be better elucidated to provide optimal care. This study aids in the identification of these needs and promotes the development of an interdisciplinary clinic of medical and dental providers to the adult cleft population.

Methods/Description: Qualitative, semistructured interviews were conducted with 30 adult patients, aged 18 to 60, with CL/P. Interviews were focused on social issues related to self-identity, life experience living with an orofacial cleft, socioeconomic access barriers to health care, and needs and desires of each patient with respect to medical and dental management. Additionally, each patient underwent a physical examination to address reported concerns.

Results: Results are presented. Lack of access to practitioners with experience treating CL/P patients, generalized concerns about appearance to individuals beyond close friends and family, and lack of knowledge about additional surgical options were highlighted as key factors important among the sampled population. Of the patients analyzed, 3 required and received additional surgical care (repair of residual palatal fistula, septorhinoplasty), 5 requested additional psychological support, 1 required speech services, and 4 had ongoing dental needs that were addressed.

Conclusions: Adult patients with cleft anomalies routinely experience both medical and social barriers to receiving care. Among our patient population, we have devised several high-need specialties to be integrated into a multidisciplinary adult cleft clinic, including facial plastic surgery, specialized dental care, and psychosocial support. Addressing residual surgical and dental needs appears to be a realistic goal or outcome for adults who present with clefts. Integrating these services into a recurring clinic appears to have a positive impact on the health and wellness of the adult CL/P population.

### 160. An Algorithmic Approach to the Management of Pediatric Isolated, Condylar Fractures

Joseph Lopez (1), Isabel Lake (2), Nima Khavanin (1), Bartlomiej Kachniarz (2), Richard Redett (3), Paul Manson (1), Amir Dorafshar (4)

(1) Johns Hopkins University School of Medicine, Baltimore, MD, (2) Johns Hopkins Hospital, Baltimore, MD, (3) Johns Hopkins Department of Plastics and Reconstructive Surgery, Baltimore, MD, (4) Rush University Medical Center, Chicago, IL

Background/Purpose: The purpose of this study was to examine injury patterns in pediatric mandibular condylar fractures and to evaluate the validity of our institutional treatment algorithm for such fractures based on postmanagement complications.

Methods/Description: A retrospective chart review was conducted on pediatric patients who presented to our institution with isolated mandibular condylar fractures between 1990 and 2016. Patients were categorized by dentition, and information regarding demographics, injury characteristics, management, and complications was compiled.

Results: Forty-three patients with 50 mandibular condylar fractures were identified. Twelve (27.9%) patients had deciduous dentition, 15 (34.9%) had mixed dentition, and 16 (37.2%) had permanent dentition. The most common fracture pattern in all groups was diacapitular

[n = 30 (60%)]; however, older groups showed higher rates of condylar base fractures and bilateral fractures (P=.049 and .009, respectively). Thirty-one patients (72.1%) were treated with nonoperative management, 10 (23.2%) with closed treatment and MMF, and 2 (4.7%) with open treatment and MMF; nonoperative treatment was more common in younger patients (P=.022). Management for 10 (23.2%) patients was nonadherent to the treatment algorithm. Seven patients had complications (16.3%). Common complications included TMJ ankylosis (n = 3) and malocclusion (n = 2). Though complications were seen in all management groups, they were more common in patients whose care was nonadherent to the algorithm (P=.032).

Conclusions: Nonoperative management is preferred in deciduous children. Children in permanent/mixed dentition may undergo closed treatment and MMF if they have malocclusion/open bite, significant condylar dislocation, and ramus height loss >2 mm.

#### 161. Association of MTHFR C677 T Polymorphism With Cleft Lip and/or Palate in Mongolia

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Background/Purpose: Nonsyndromic cleft lip and/or palate (CL/P) are the most common and easily recognizable major congenital malformations. The pathogenesis of CL/P is complex, presumably implicating the interaction of several genetic and environmental factors. MTHFR (5,10-methylenetetrahydrofolate reductase) gene C677 T functional mutation and insufficient folic acid intake during pregnancy were proposed to increase the risk of CL/P. The present study is aimed to explore the association between nonsyndromic cleft lip and/or palate and genetic polymorphism of MTHFR C677 T in Mongolian population.

Methods/Description: The study population consisted of 307 unrelated nonsyndromic CL/P patients and their parents. Recruitment of 67% of participants took place at the Children's Department of Oral and Maxillofacial Surgery of the MCHRC Hospital. The 220 control individuals (120 males and 100 females) were without any congenital anomaly, unrelated to each other and chosen as the CL/P cases at the MCHRC Hospital. They ranged in age between 2 and 20, with 70% as adolescents aged 15 to 19. All participants were Mongolians residing in Ulaanbaatar and did not differ ethnically/racially from the CL/P cases. Genotyping for the MTHFR mutation was performed. Statistical analysis was carried out using  $\chi^2$  tests to compare the composite distribution of the MTHFR CC, CT, and TT genotypes between CL/P patients, their mothers and fathers groups, and the controls. The data were examined for statistical significance at a probability level of .05.

Results: No indication of significant differences for the proportions of CC, CT, and TT genotypes was observed between total patients (CC 54.7%; CT 36.8%; TT 8.5%) and control groups (CC 54.5%; CT 37.7%; TT 7.7%) for either their parents or the controls. Nevertheless, when patients group was divided by cleft types and gender, a difference tendency (P = .09) was found between female CP patients (CC 53.0%; CT 23.5%; TT 23.5%) and female control groups (CC 53.0%; CT 37.0%; TT 10.0%), respectively. Further significant differences were found between male CL and female CL (P = .01), as well as male CP and female CP (P = .04) when cases were compared by their subtypes of oral clefts.

Conclusions: The discrepancy in the distribution of MTHFR genotype among types of oral clefts reflects some heterogeneity in the development mechanism of nonsyndromic CL/P. The TT genotypes might contribute to the pathogenesis of cleft palate in Mongolian females.

### 162. Atlantoaxial Rotatory Subluxation: A Rare Complication of Craniofacial Surgery

Viren Patel (1), Robin Yang (2), Sameer Shakir (3), Scott Bartlett (4) (1) University of Pennsylvania School of Medicine, Philadelphia, PA, (2) Johns Hopkins Department of Plastics and Reconstructive Surgery, Baltimore, MD, (3) University of Pennsylvania, Philadelphia, PA, (4) Children's Hospital of Philadelphia, Philadelphia, PA

Background/Purpose: Neck pain is common in the postoperative period after craniofacial procedures. If patients present with neck pain and torticollis, it could be a manifestation of atlantoaxial rotatory subluxlation (AARS), which describes a rare condition in which there is lateral displacement of C1 relative to C2. When this occurs in the postoperative patient, it is termed Grisel syndrome. In this case series, we report on 3 patients diagnosed with Grisel syndrome after a craniofacial procedure.

*Methods/Description*: A retrospective chart review of a single craniofacial surgeon at a pediatric hospital was conducted over the last 3 decades. Demographics, procedures performed, and management strategies of AARS were included for review.

Results: Three patients were identified who were diagnosed with Grisel syndrome after a craniofacial procedure and required inpatient cervical traction. All patients presented with torticollis within 1 week of their operation. Conservative management was ineffective, and all 3 patients were admitted for inpatient cervical traction, for an average of 13 days followed by an average of 47 days of outpatient therapy. No patients showed any signs of recurrence after removal of outpatient traction device.

Conclusions: Grisel syndrome is a rare, but serious complication of craniofacial procedures. Physicians caring for these patients must have a high degree of suspicion if a patient with a recent craniofacial procedure presents with torticollis. Delaying the initiation of therapy has been shown to lead to higher rates of recurrence and increases the likelihood that patients will require surgical intervention.

### 163. Barriers to Accessing Timely Surgical Cleft Treatment in Low- and Middle-Income Countries

Siddhi Upadhyaya (1), James Gatherwright (2), Rebecca Knackstedt (1) (1) Cleveland Clinic, OH, (2) MetroHealth, Cleveland, OH

Background/Purpose: Orofacial cleft deformities affect 1 in every 500 to 750 births worldwide, resulting in complications ranging from mild to severe when left untreated. In low- and middle-income countries, timely access to surgical repair of cleft lip and palate is a significant challenge contributing to substantial risk of infant mortality. Though this challenge is widely acknowledged, having served as the driving force behind the robust expansion of foundation-based cleft care, the challenges unique to each region vary widely and have not been well defined for each country. The purpose of this study is to examine the region-specific barriers to accessing timely surgical cleft repair.

Methods/Description: A systematic review of peer-reviewed literature was conducted utilizing PubMed, MEDLINE, Scopus, and Cochrane databases according to guidelines specified by the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA). Journal articles from January 1989 to July 2019 were included, and the level of

evidence of each study was recorded. Data analysis was performed using SPSS.

Results: A total of 73 articles were found based on our search. Based on our inclusion criteria, a total of 22 articles were included in our analysis. Six articles were determined to produce level I evidence. Barriers to cleft care most strongly represented included mistrust of health-care professionals, travel costs, lack of financial support, and poor health. Travel costs were the most commonly identified barrier. Subset analysis will be presented, showing specific barriers varied largely by geography and cultural factors.

Conclusions: With the continuing expansion of foundation-based cleft care globally, though many public health models address access to health care, the need exists to enhance models exploring access specifically to surgical intervention in vulnerable patient populations. Our review shows barriers to timely access to cleft lip and palate repair were found to vary primarily by geography and cultural factors, with travel costs serving as the greatest barrier. In relation to these factors, it is necessary for discussion of barriers to care specific to each country to be included in the international dialogue when addressing solutions for disparities in cleft lip and palate care.

### 164. Barriers to Care Among Patients With a Cleft Lip and Palate in Southwest Virginia

Jose Rodriguez (1), Lamve Le (1), Christopher Liao (1), James Thompson (2) (1) Virginia Tech Carilion, Roanoke, VA, (2) Carilion Clinic, Roanoke, VA

Background/Purpose: Southwest Virginia is a mostly rural and underserved area of the country where patients have to travel several hours to get specialized care. Most studies have looked at barriers and disparities in the pediatric population as a whole. There are few studies looking at barriers specific to the care of children with a cleft lip or palate in the United States and no studies looking at these barriers in southwest Virginia. The purpose of this study was to elicit the barriers to care in southwest Virginia to better serve our community. We know that these patients will need multiple operations and multidisciplinary care, so ensuring access throughout their childhood and into adulthood is essential in optimizing their clinical outcomes.

Methods/Description: Eighty caregivers of children with cleft lip and palate were surveyed to assess perceived barriers using the validated Barriers to Care questionnaire. The survey was modified to 25 questions during clinic visits between May 2018 and May 2019. Bivariate analyses using Fisher exact test was used.

Results: Our population was white (74.4%), black (9%), Hispanic (6.4%), Asian (6.4%). The most common type of health insurance was public health insurance (64.4%) and 34.2% had private insurance. Increased travel distance was associated with less access to medical resources (P=.047). There was no correlation between travel distance and household income. Cost of travel increased the chance of forgoing a clinic visit (P=.001). Similarly, the lack of medical resources in a community also increases the chance of forgoing a clinic visit (P=.006); 12.3% of respondents occasionally will miss clinic visits due to cost or distance; 10.4% view financial difficulties as a barrier to care; 8.7% perceived a problematic care process; and 95% were satisfied or very satisfied with the services provided.

Conclusions: Barriers to care were associated with the cost of travel and lack of medical resources in the community. Unfortunately, access to quality health care continues to be unequal in the United States and this is apparent in southwest Virginia. To reduce these barriers to care, we would recommend reducing or offsetting the cost of travel. Having a lack of medical resources in a community could be mitigated by

working with care managers to ensure that patients are maximizing the resources available in and out of the community.

#### 165. Cardiovascular Anomalies in Patients With Non-Syndromic Orofacial Clefts: A Prospective Case Controlled Study

Olutayo James (1), Ogochukwu Sokunbi (1), Olushola Abogidi (2), Adeola Adekunle (2), Olawale Adamson (1), Ajoke Ogunlewe (2), Ekanem Ekure (1), Wasiu Adeyemo (1), Akin Ladeinde (1), Mobolanle Ogunlewe (1)

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Background/Purpose: Cardiovascular anomaly is always a concern in children with orofacial clefts. This anomaly may have an impact on the timing of treatment and mandate special precautionary measures perioperatively. The incidence of cardiac anomalies associated with orofacial cleft in our environment is unknown. This study ascertains the cardiovascular anomalies in a population of patients with orofacial clefts and compare with those of age- and sex-matched control subjects.

Main Objectives of Presentation: To alert clinicians on the need to take precautionary measures perioperatively while treating patients with orofacial clefts.

Methods/Description: This was a prospective case-controlled study carried out at the Oral and Maxillofacial Surgery Cleft Clinic, Pediatric and Community Health Outpatient Clinics of the Lagos University Teaching Hospital (LUTH) Lagos, Nigeria. Participants were all consecutive orofacial cleft subjects aged 2 months and above and age- and sex-matched control subjects who are without orofacial cleft. All eligible subjects (cases and controls) had a full clinical examination, electrocardiography (ECG), and echocardiography tests done. Ethical approval for this study was obtained from the HREC of Lagos University Teaching Hospital.

Results: A total of 120 subjects who satisfied the inclusion criteria participated in the study with 60 subjects in each group. There were 63 (52.5%) males and 57 (47.5%) females with a male to female ratio of 1.1:1. Of the 60 subjects in the orofacial cleft group, 14 (23.3%) had bilateral cleft lip and palate, 12 (20%) had unilateral cleft lip and palate, and 13 (21.7%) had isolated cleft of the palate. Four (6.67%) of the control subjects was diagnosed with congenital heart defects, while 17 (28.3%) of the orofacial cleft group had congenital heart defect (P = .001).

Conclusions: This study shows a statistically significant higher incidence of cardiovascular anomalies in subjects with nonsyndromic orofacial clefts than age- and sex-matched control subjects. The detected cardiovascular anomalies are however of none severe variety. Tessier clefts seem to predispose to cardiovascular anomaly as the 2 cases in this study present with cardiovascular anomalies.

### 166. Cholesteatoma of the External Ear Canal in Goldenhar Syndrome

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(1) University of North Carolina, Chapel Hill, NC, (2) University of North Carolina, School of Medicine, Chapel Hill, NC

Background/Purpose: Oculoauricular vertebral dysplasia, also known as Goldenhar syndrome, includes abnormalities of the first and second branchial arches. Ninety-two percent of patients with Goldenhar syndrome have ear abnormalities, usually consisting of microtia and

conductive hearing loss. Newborn hearing screening results in early diagnosis and management of patients with this condition.

Methods/Description: Descriptions of ear canal cholesteatoma are scant, in reviewing the literature. Despite this, the clinical condition is well known to otolaryngologists and they may recommend radiologic studies, such as CT scanning, for diagnosis and surgical planning. A case series of patients are presented with acquired cholesteatoma of the ear with microtia in patients with Goldenhar syndrome. We attempt to better delineate an uncommon complication in a craniofacial condition and to recommend surveillance for it.

Results: This case series study describes 3 patients with Goldenhar syndrome who presented with the findings and diagnosis of external auditory canal cholesteatoma, all requiring operative removal. In all 3 patients, the ear canal was only partly patent, allowing development of buildup of squamous material and acquired cholesteatoma. Clinical and radiological presentation is reviewed and surgical approach discussed.

Conclusions: This relevant disease process, ear canal cholesteatoma, has significant clinical implications for health as well as hearing. We postulate a mechanism for this complication to occur in patients with microtia and canal stenosis, as opposed to congenital cholesteatoma, which would be more likely to occur with total ear canal atresia. As either of these conditions may occur in patients with Goldenhar syndrome, awareness of this complication makes timely diagnosis and treatment possible in the care of these craniofacial patients.

### 167. Cleft-Palate Speech Therapy: Telesupport as a Home Program Supplement

Naomi Lowe (1), Alison Scheer-Cohen (1)

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Background/Purpose: International humanitarian efforts for patients with cleft palate often lack long-term support for speech deficits. When speech therapy is included, it typically consists of one session initiating a parent-implemented home program, with little follow-up. This research investigated the effectiveness of using telesupport internationally to compliment the traditional parent-implemented home program.

Methods/Description: Ten participants with cleft palate, ages 3 to 12 years, were recruited from a hospital in Tijuana, Mexico. All participants were primarily Spanish-speaking, had cleft-palate-related speech errors, and were not receiving speech therapy at the time of the study. Participants were randomly assigned to one of 2 groups: a home speech program or a home speech program with additional weekly telesupport. Speech production was measured before an 8week home treatment program and after treatment. Measures of speech included word and sentence-level production, as well as a language sample. Targeted treatment phonemes were assessed in preselected treatment words and probe words prior to and following treatment. Errors in phoneme production were evaluated by 2 additional raters who were blind to treatment group, with Cronbach  $\alpha$ achieving acceptable inter-rater reliability (650 items,  $\alpha = 0.73$ ). Additionally, a quality-of-life survey was given to parents before and after participation.

Results: Age was significantly related to amount of time practiced, r(6) = 0.814, P = .014), with older participants practicing more at home. Practice time was marginally increased for older students provided telesupport, as evidenced by a partial correlation, r(6) = 0.725, P = .065. A Wilcoxon signed rank test revealed no significant differences in error rates of targeted phonemes in the targeted words, probe words, or sentence-level production. Additionally, a Mann-Whitney U

failed to show a difference between the 2 treatment groups (P > .05). Despite lack of phoneme improvement at the word level, parent report on implementing treatment after receiving telesupport was positive. Parents in the telesupport group reported being more comfortable with understanding therapy and with home implementation. A Mann-Whitney U showed that although the changes in quality of life were in the predicted direction, none of the differences were significant.

Conclusions: Possible reasons for lack of phonemic outcomes are explored, as well as the potential that telesupport holds for treating speech in children with cleft palate who have limited access to resources, such as trained speech-language pathologists. Additional modifications of home programs are considered based on the result that younger participants were less likely to practice articulation.

### 168. Complex of Octacalcium Phosphate and Atelocollagen to Treat Alveolar Clefts

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Background/Purpose: For eruption of permanent teeth in patients with alveolar bone defects accompanying cleft lip and palate, autologous bone transplantation is generally performed. The purpose of this study was to evaluate postoperative bone formation after implantation of octacalcium phosphate granule and atelocollagen complex (OCP/Col) to treat bone defects in patients with an alveolar cleft accompanying unilateral cleft lip and palate.

Methods/Description: This was a phase III, prospective, multicenter, single-arm clinical trial of OCP/Col for guided bone regeneration, which was registered with the University Hospital Medical Information Network in Japan (registration number: JPRN-UMIN000018192). This clinical trial was approved by the institutional review board of the Pharmaceuticals and Medical Devices Agency in Japan (reference number: OCTC-14001). Research and development on bone regeneration of OCP/Col was proceeded by Tohoku University. Toyobo Co. Ltd sponsored this clinical trial. The clinical trial was conducted at 9 hospitals in Japan. OCP/Col transplantation was performed to treat alveolar cleft in 4 patients with unilateral cleft lip and palate at the Department of Oral and Maxillofacial Surgery, Kagoshima University. The timing and method of surgery were the same as those for autologous bone transplantation. X-rays were obtained preoperatively and after 1, 2, 3, 6, and over 30 postoperative months. The volume of the bone defect was measured preoperatively and after 6 postoperative months via computed tomography. The image analysis software OsiriX (v.4.1.2 Mac OS) was used. The bone bridge was evaluated on an X-ray scan after 6 postoperative months with the Bergland scale.

Results: In all 4 patients who underwent OCP/Col transplantation, permanent teeth erupted at the site of transplantation after 6 post-operative months. On the Bergland scale, all cases were evaluated as grade 1. On computed tomography, bone bridging sufficient for orthodontic movement was observed in all cases. All patients and their families were satisfied, as there were no pain or gait disturbances in the autologous bone collection site following OCP/Col transplantation.

Conclusions: In this clinical trial, all 4 cases of OCP/Col transplantation showed almost the same results as autologous bone grafting for the formation of bone bridge and eruption of permanent teeth. Therefore, OCP/Col was considered as a suitable material, performing the same functions as autologous bone.

### **169.** Controversies in Pediatric Mandibular Tumor Reconstruction

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Background/Purpose: Although the reconstructive paradigm of the adult mandible has been well-documented, defining guidelines for pediatric mandibular reconstruction (PMR) has been more controversial. Fibula and iliac crest bony reconstruction have traditionally been utilized with great success in adults. In children, the reconstructive options are not as straightforward as the pediatric patient is still growing, has mixed dentition, and traditionally cannot have dental implant rehabilitation. One needs to create an optimal functional and aesthetic reconstruction while keeping these considerations in mind. Preserving future mandibular reconstructive options is key as these patients will likely require multiple reconstructions over the course of their lifetime. Guidelines are not established for the optimal time to utilize the various reconstructive options. The purpose of this study is to review the currently available literature regarding PMR and describe our experience.

Methods/Description: A systematic review of the current literature surrounding PMR was completed according to PRISMA guidelines. IRB-approved retrospective chart review of patients who underwent PMR for tumor resections between 2005 and 2019 was performed. Our experience was then combined with the literature review to provide a proposed PMR algorithm.

Results: A systematic literature search was conducted on the PubMed database for peer-reviewed literature resulting in 516 results. Further abstract and full-text review for relevance of these articles yielded a total of 36 studies for the evaluation of reconstructive techniques in PMR. Retrospective chart review yielded 13 patients undergoing PMR, mean age 7.9 (1-16) years. The etiologies for their PMR included ameloblastoma, aggressive fibromatosis, osteoblastoma, fibromatosis, giant cell tumor, myofibroma, odontogenic myxoma, osteosarcoma. PMR methods included cadaveric tissue, corticocancellous iliac crest, rib, custom fabricated endoprosthesis, and custom crib. The mean mandibular defect size for vascularized bone grafts (VBG) and non-VBG was  $12.4 \pm 6.2$ cm and  $7.8 \pm 3.0$  cm, respectively. In total, the mean mandibular defect size was 8.9 ± 4.1 cm. Fifty-four percent of patients had complications associated with repair, the majority being dehiscence and surgical site infection; 46.2% of patients had successful dental implant placement.

Conclusions: Pediatric mandibular tumors resection can be reconstructed with high rates of success and minimal morbidity. Smaller mandibular defects may be restored with less invasive methods allowing for the preservation of VBGs for later reconstruction. Based on our institution's experience, the traditional 6 cm limit on utilizing non-VBG should only be a guideline and may be expanded for different indications. One may want to preserve the gold standard, free fibula, for potential need for future surgery as the patient grows. Regardless of the reconstructive method, dental implants can be placed with a high degree of success, as well, in this population.

#### 170. Does Virtual Surgical Planning Accurately Predict Post-Operative Intracranial Volumes in Patients Who Undergo Distraction Osteogenesis for Craniosynostosis?

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Background/Purpose: Virtual surgical planning (VSP) in cranial vault reconstructive has been shown to decrease preoperative planning time, decrease intraoperative time, and provide superior aesthetic results. In patients with craniosynostosis, the primary outcome metric is change in intracranial volumes (ICV), and in patients who undergo distraction osteogenesis (DO), this is dependent on the length of distraction. In spite of the advantages of VSP use in the literature, there is no study to date that has analyzed the accuracy of VSP in predicting anticipated ICV changes. The purpose of this study is to analyze the actual versus predicted intracranial volumes in patients who undergo distraction osteogenesis for coronal craniosynostosis.

Methods/Description: After institutional review board approval was obtained, a retrospective review of all patients who underwent distraction osteogenesis for unicoronal craniosynostosis and bicoronal craniosynostosis at UC San Diego/Rady's Children Hospital from August 2013 to April 2019 was conducted. Inclusion criteria included a diagnosis of craniosynostosis and patients who have undergone distraction osteogenesis, have documented preoperative and postdistraction osteogenesis computed tomography scans, and have VSP planning with predicted postdistraction intracranial volumes. Preoperative and postoperative computed tomography scans were used to quantify ICV. Measured ICV and VSP-estimated ICV were adjusted in proportion for age-related ICV growth based on our previously reported normative data. The primary outcome measure was calculated as age-adjusted percentage volume change attributable to distraction osteogenesis. Outcome measures were analyzed using paired Wilcoxon signed rank tests. All statistical analysis was performed using IBM SPSS.

Results: A total of 23 patients underwent distraction osteogenesis for cranial vault remodeling for craniosynostosis, 60.9% were female and 39.1% were male patients; 60.9% of patients had a diagnosis of unicoronal craniosynostosis, 8.7% of patients had bicoronal craniosynostosis, 8.7% of patients had lambdoid synostosis, and 21.7% of patients had multisuture synostosis; 8.7% of patients had a diagnosis of Pfeiffer syndrome, 4.3% of patient had a diagnosis of Crouzon syndrome, and 4.3% of patient had a diagnosis of Apert syndrome. The average calculated age-adjusted percentage increase in intracranial volume per millimeter of distraction was 0.38 as compared to the VSP age-adjusted predicted increase in intracranial volume per millimeter of distraction of 0.49. VSP age-adjusted predicted volumes for distraction were higher and this was statically significant (P = .001).

Conclusions: The predicted VSP post DO volume overestimates the change in ICV attributable to distraction in patients with craniosynostosis. These data suggest that VSP data may overestimate expected postoperative ICV and this should be taken into consideration in the preoperative planning.

## 171. Early Cleft Lip Repair as a Viable Alternative to Nasoalveolar Molding for Treatment of Wide Unilateral Cleft Lip

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Background/Purpose: Early cleft lip repair (ECLR) can be performed safely and effectively. One persistent question is whether ECLR may be offered to wide unilateral complete cleft patients who historically would have received nasoalveolar molding (NAM). This study aims to compare the preoperative cleft severity of ECLR patients to those whom underwent NAM pretreatment and compare postoperative outcomes.

Methods/Description: Unilateral CL patients (01, 01, 2005 to 09, 11, 2018) were retrospectively reviewed and divided into 2 groups: ECLR (age <3 months) and presurgical NAM with CL repair (ages 3-6 months). Pretreatment CL severity was assessed using an AI computer engineered system that calculated cleft width ratios (CWR, pretreatment cleft width divided by commissure width). Postoperative symmetry was compared between the patients undergoing ECLR and patients undergoing NAM by measuring bilateral lip length, frontal nasal breath, commissure length, nostril breadth, nostril width, and nasal angle to generate symmetry ratios for each patient.

Results: A total of 27 complete ECLR patients and 25 NAM patients (average age at repair: 32.24 days and 117.56 days, respectively) met inclusion criteria. Mean CWR was 0.520 (SD: 0.121) for ECLR patients and 0.501 (SD: 0.122) for NAM patients (P = 0.566). The average lip length, frontal nasal breadth, and commissure length symmetry ratios, measured on frontal 2-D photos, for the 27 ECLR patients was 0.88 (0.63-1.14; SD: 0.12), 1.05 (0.72-1.36; SD: 0.15), and 0.92 (0.65-1.38; SD: 0.16), respectively. The symmetry ratios for the same measurements in the NAM group were 0.93 (0.68-1.12; SD: 0.09), 1.08 (0.78-1.46; SD: 0.20), and 0.89 (0.65-1.05; SD: 0.10) with no statistically significant difference between these mean anthropomorphic symmetry ratios (P = .181, P = .526, P = .378, respectively). The average nostril breadth, nostril width, and nasal angle ratios among the ECLR patients measured from the worm's eve view were 1.09 (0.90-1.39, SD: 0.13), 1.17 (0.91-1.47, SD: 0.17), and 1.12 (0.87-1.36, SD: 0.14) respectively. The symmetry ratios for the same measurements in the NAM group were 1.12 (0.93-1.38, SD: 0.13), 1.19 (0.83-1.57, SD: 0.18), and 1.14 (0.88-1.55, SD: 0.18) with no significant difference between the groups (P =.480, P = .613, P = .640, respectively). The intra-rater reliability was determined for each anthropomorphic measure with resulting intraclass correlation coefficients (ICC) of 0.913, 0.906, and 0.721 for frontal measures of lip length, frontal nasal breath, and commissure length, respectively. The ICCs for basal view measures of nostril breadth, nostril width, and nasal angle were 0.950, 0.927, and 0.853, respectively. All ICCs indicated good correlations (>0.7).

Conclusions: ECLR provides patients with severe cleft lips an alternative option to NAM with at least equivalent results. With increased experience, long-term data, and increased awareness, we feel that ECLR has the potential to be a paradigm shift in the treatment of the cleft lip/nasal deformity.

## 172. Effects of Pre-Operative Administration of Erythropoietin in Pediatric Patients Undergoing Cranial Vault Remodeling for Craniosynostosis

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Background/Purpose: Over 90% of patients undergoing cranial vault remodeling for craniosynostosis receive blood transfusions to compensate for intraoperative blood loss. However, transfusions are not without risk and can lead to allergic and immune transfusion reactions as well as rare cases of infectious transmissions. Preoperative use of erythropoietin in cranial vault remodeling has been demonstrated to be safe and to reduce transfusion requirements. We set out to determine the impacts that a protocol of preoperative erythropoietin and elemental iron for 4 weeks prior to surgery would have on patient outcomes.

Methods/Description: After obtaining IRB approval, a chart review was performed on patients who underwent cranial vault remodeling between 2005 and 2019 in a single institution. Two cohorts were identified: in the first cohort, preoperative erythropoietin was administered under protocol, and in the second cohort, preoperative erythropoietin was not administered. The cohorts were compared with respect to age, perioperative Hgb levels, estimated blood loss during surgery, packed red blood cell transfusion volume, length of hospital stay, and length of surgery.

Results: Patients who received preoperative erythropoietin by our protocol demonstrated statistically significantly increased preoperative Hgb levels, as well as decreased estimated blood loss, volume of packed red blood cells transfused, length of hospital stay, and length of surgery. Postoperative Hgb levels were not found to be statistically significant, nor was age.

Conclusions: In this study, the administration of preoperative erythropoietin with elemental iron resulted in decreased transfusion requirements among other salutary effects in patients undergoing cranial vault remodeling for craniosynostosis. Further analysis may include the effects of preoperative iron alone. Limitations of this study include retrospective design and nonstandardized criteria used by anesthesiologists as start and end points of transfusion.

## 173. Examining Utilization of Surgical Treatment for Sleep Apnea: A Study of Demographic Disparities Using the National Inpatient Sample Database (NIS)

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Background/Purpose: Sleep apnea is a source of serious morbidity, for which surgical intervention is an effective management option. Barriers to surgical treatment for sleep apnea remain understudied, although demographic factors such as race, gender, and insurance may impact utilization of surgery. In this study, we sought to evaluate demographic disparities in the surgical management of sleep apnea.

Methods/Description: The Healthcare Cost and Utilization Project National Inpatient Sample (NIS) database was analyzed for 2014. Patients aged 18 or older with a primary or secondary diagnosis of sleep apnea were selected and compared against those who had this diagnosis, but also received related surgeries, such as orthognathic surgery and tonsillectomy. Age, race, gender, region, insurance, and procedure type were analyzed between groups using SPSS.

Results: A total of 47 585 patients with primary or secondary diagnosis of sleep apnea were identified, 1600 (3.4%) of whom received surgical intervention. Patients in the surgical cohort were younger, with a mean age of 44.8 versus 55.9 years (P < .001), male (72.8% vs 57.8%; P < .001), and Hispanic (15.3% vs 7.5%; P < .001) compared with the total group. The most common surgical procedures were palate surgery (39.1%) and tonsillectomy (11.9%). Those receiving surgery were less likely to use Medicare (12.2% vs 36.3%) and more likely to use private insurance (67.5% vs 46.7%) than the total group. The largest percentage of patients were in the Midwest (33.0%), but surgical management was the least prevalent in this region (19.7%).

Conclusions: This study identified multiple demographic discrepancies in the utilization of surgical management of sleep apnea. Patients utilizing surgery were more likely to be younger, male, and Hispanic compared to the total group with sleep apnea. Predictably, private insurance was more commonly utilized by those who received surgical intervention, whereas Medicare was more common among the total population with sleep apnea. Furthermore, the largest proportion of patients with sleep apnea were in the Midwest, although surgical management was used the least in this region when compared to other regions of the United States. Using these data, a predictive patient profile can be established to identify patients more likely to utilize surgical management for sleep apnea. Further research must be performed to identify causative factors for the disparities identified.

#### 174. Human Umbilical Cord Mesenchymal Stem Cells Are Useful Bioresource for Experimental Alveolar Cleft Regeneration

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Background/Purpose: Alveolar clefts cause the morphological and functional problems. In our dental hospital, gingivoperiosteoplasty (GPP) is performed with cheiloplasty for infants with alveolar clefts. However, our recent study showed that the bone bridge formation between alveolar clefts was insufficient in some cases. Regenerative medicine using mesenchymal stem cells (MSCs) is expected to improve the bone bridge formation. In human umbilical cords, it is known that immature and pluripotency MSCs are included. Moreover, obtaining human umbilical cord MSCs is invasive to mothers and children. In this study, we examined the bone formation by human umbilical cordderived MSCs using an experimental rat alveolar cleft model.

Methods/Description: Human umbilical cords were digested by enzymatic procedure and isolated cells (UC-EZ) were collected. Using magnetic-activated cell sorting, CD146-positive cells (UC-MACS) were isolated from UC-EZ. To evaluate bone formation in vivo, UC-MACS were transplanted with hydroxyapatite and collagen (HA + Col) into alveolar cleft model.

Results: Both type of cells showed MSC gene/protein expression and multipotency, in vitro. Observation of micro computed tomography and histological staining showed that UC-MACS induced more abundant bone formation than HA + Col implantation solely. Cells immunopositive for osteopontin were accumulated and embedded in newly formed bone. Cells immunopositive for human-specific mitochondria were seen in both mineralized and nonmineralized tissues.

Conclusions: These findings indicate that UC-MACS are responsible for new bone formation. The observation that human-specific mitochondria-positive cells were in mineralized tissues suggests that UC-MACS directly induced bone formation in the transplanted tissues. As conclusion, UC-MACS are useful bioresource for alveolar cleft regeneration.

### 175. Infant Anesthesia in Early Cleft Lip Repair: Assessing Short-Term Safety and Efficacy

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Background/Purpose: Cleft lip represents the most common craniofacial birth defect, affecting 1 in 700 live births worldwide. Dogma established by Millard in 1976 regarding the ideal timing of elective surgery in infants centered on the "rule of 10s" (>10 weeks of age, >10 pounds, and has >10 g hemoglobin and <10 white cell count). This rule of 10's was largely based on outdated and anecdotal evidence. Over the last 2 decades, the risks associated with anesthesia have drastically decreased. Currently, the FDA recommends that anesthesia times in children less than 3 years of age be limited to less than 3 hours in duration to avoid developmental complications in the growing brain. Two large studies, one prospective (GAS) and one ambidirectional (PANDA), have demonstrated safe short- and long-term outcomes. Our study seeks to evaluate short-term anesthetic safety and efficacy in our early cleft lip repair (ECLR) protocol.

Methods/Description: Patients who underwent ECLR (repair before 2.5 months of age) within the last 4.3 years were identified. Retrospective review of their records included abstracted patient demographics, ASA (American Society of Anesthesiologists), vital signs, intraoperative and postoperative medication dosing, total anesthetic time, major and minor complications, and medication side effects. Total anesthetic time was defined as time from induction to extubation. Major complication was defined as a code event, aborted surgery, or intraoperative death. Minor complication was defined as a sustained alteration in heart rate, apnea event, or prolonged emergence time. Medication side effect was defined as a transient alteration in heart rate or hypopnea.

Results: A total of 101 patients underwent ECLR during our study period. All patients were either ASA class 1 or 2. Mean age at of surgery was 31  $\pm$  13 days and mean anesthetic time was 179  $\pm$  36 minutes. Mean emergence time was 7.6  $\pm$  5.7 minutes. Mean length of stay was 1  $\pm$  0.1 day. There were no episodes of hemodynamic instability. A minor complication rate of 1% was observed. A medication side effect rate of 7.7% was observed. Development of medication side effect was not associated with anesthetic time, age at surgery, emergence time, or length of stay. A stepwise binary logistical regression analysis demonstrated minor medication side effects were associated with increases in the total dexmedetomidine and fentanyl administered (36.7  $\pm$  29.4  $\mu g$  vs 13.0  $\pm$  13.2  $\mu g$ , P= .004; 3.6  $\pm$  2.7  $\mu g/kg$  vs 1.8  $\pm$  1.2  $\mu g/kg$ , P= .012).

Conclusions: ECLR patients demonstrated no major complications and an acceptable minor complication rate associated with anesthesia administration. Millard's rule of 10's governing the ideal timing of elective surgery in infants may not apply to ASA 1 and 2 class patients.

### 176. Influence of Speech Stimuli on Perceptual Ratings of Hypernasality

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Background/Purpose: The chosen type of speech stimuli may affect speech judgment in subjects with cleft lip and palate. Prior selection of

speech stimuli may be effective to increase the agreement among listeners. The goal of the study was to assess the influence of speech stimuli on intra- and inter-rater hypernasality reliability in subjects with unilateral cleft lip and palate who underwent palatoplasty with the Furlow procedure or modified von Langenbeck procedure.

Methods/Description: Four hundred audio-recorded speech samples of 80 individuals with repaired unilateral cleft lip and palate, both genders, aged between 9 and 17 years old (mean =  $12 \pm 7$ ), were selected and edited. Speech stimuli analyzed were 12 high-pressure sentences, 6 fricative/plosive voiced sentences, 6 fricative/plosive unvoiced sentences, and 4 low-pressure sentences and number counting. Three experienced speech-language pathologists rated presence or absence of hypernasality, using their internal criteria, in 80 samples of each stimuli (80 samples  $\times$  5 stimuli) as well as 16 samples for intra-rater agreement analysis.

Results: Comparison of intra-rater agreements showed higher agreements for the 12 high-pressure sentences for all raters. Significant differences were found between fricative/plosive voiced sentences and 3 other stimuli (high-pressure sentences, P=.032, fricative/plosive unvoiced sentences, P=.020; and low-pressure sentences, P=.012) for one rater, with lower intra-rater agreement for voiced sentences. Comparison of inter-rater agreements showed significant differences between fricative/plosive voiced sentences and 3 other stimuli (high-pressure sentences, P=.027; low-pressure sentences, P=.029; and counting, P=.016) for 2 raters (1 and 3), with lower inter-rater agreement for voiced sentences. Significant differences were also found between low-pressure sentences and counting (P=.048) for 2 raters (2 and 3), with lower inter-rater agreement for low-pressure consonants.

Conclusions: Voiced sentences and low-pressure sentences had influence on reliability among raters, resulting in lower inter-rater agreements. Voiced sentences also disfavored intra-rater agreements for one rater. Overall, these findings suggest that one could avoid the exclusive use of voiced pressure sentences or even low-pressure consonant while rating hypernasality. High-pressure sentences combining all plosives and fricatives sounds (voiced and unvoiced) may favor ratings of hypernasality even in the presence of compensatory articulations, a speech error commonly identified in association with hypernasality.

#### 177. Influenza as a Measure of Maternal Immune Activation and Its Effects on the Incidence of Encephalocele and Microtia

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Background/Purpose: The activation of the immune system of a pregnant mother can have significant effects on fetal development. Maternal immune activation (MIA) can cause neurodevelopmental disorders including schizophrenia, autism, and so on. We sought to examine MIA and craniofacial development. Since influenza occurs seasonally, it can be used as a measure of MIA. This study measures the association of the incidence of influenza infection in the United States with the incidence of craniofacial congenital deficits, specifically encephalocele and microtia.

Methods/Description: The National Inpatient Sample Database (NIS) was referenced to identify national estimates of infants born with each disease from 2004 to 2013. The gross monthly disease incidences were adjusted based on the number of newborns each month. The National

Respiratory and Enteric Virus Surveillance System's FluView database from the CDC was used to obtain influenza data from 2003 to 2013. The gross monthly influenza incidences were adjusted based on the total US population from the US Census Bureau. Mixed effect logistic regression analyses were conducted to find the association between influenza occurrence and each disease, specifically an odds ratio. The *P* values were calculated with Bonferroni correction. Poincaré graphs were plotted, and Hurst exponents were calculated.

Results: There were 2858 newborns born with encephalocele and 3371 newborns born with microtia from January 2004 to December 2013. Microtia showed no statistically significant correlation with influenza rates (P > .05). Encephalocele showed a strong correlation with influenza rates during the eighth month of pregnancy (OR = 34.538, P = .047).

Conclusions: This study shows evidence of a correlation between influenza occurrence during the eighth month of pregnancy and encephalocele incidence. This suggests that there is an additional trigger for encephalocele development toward the end of the pregnancy. Results of microtia analysis served as a negative methodologic control. Although there appears to be a connection between MIA and encephalocele, we need more evidence to support this theory.

## 178. Institutional Orthognathic Intervention After Orofacial Cleft Repair: A Systematic Review and Meta-Analysis

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Background/Purpose: Orofacial cleft deformities are prevalent congenital defects which can affect approximately 7.75 neonates out of every 10 000 live births. This is by far the most common congenital malformation of the head and neck seen in the pediatric population. As these patients age, they receive surgical correction of their orofacial clefts. The development of midface hypoplasia has been a commonly reported problem that afflicts these patients. Between institutions, the reported incidence of the surgical correction of this midface hypoplasia varies drastically. The relationship between the need for orthognathic corrective surgery and the institution's preferred surgical protocol and threshold for orthognathic surgery remains unclear. Some institutions report orthognathic surgery rates as high as 70%. An improved understanding of the overall incidence of orthognathic surgery and treatment patterns is important to determine optimal treatment and standard of care for these patients.

Methods/Description: A systematic literature review and metaanalysis was performed according to PRISMA guidelines in the PubMed database. Studies were included for their focus on either background information or reports on the rates of orthognathic surgery at select institutions. Number of orthognathic surgeries, age at surgery, syndromic versus nonsyndromic status, number of surgeries until correction of orofacial cleft, and incidence of reintervention were collected from unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), isolated cleft palate (ICP), and isolated cleft lip (ICL) groups.

Results: A total of 15 peer-reviewed articles were included in this study. The mean age at orthognathic surgery was  $20.1 \pm 1.7$  years. When cleft laterality was compared, 30.0% of UCLP patients, 34.1% of BCLP, and 1.8% of ICP patients required orthognathic surgery for correction of their midface hypoplasia. There were no reported cases

of ICL patients requiring orthognathic surgery. The standard deviations of the rates of orthognathic intervention for UCLP, BLCP, and ICP were 16.4%, 22.9%, and 13.9%, respectively. Between institutions, the average number of surgeries needed per cleft patient was 5.7  $\pm$  1.6 surgeries.

Conclusions: The mechanism of developing midface hypoplasia following orofacial cleft repair is yet to be completely understood. By gaining insight into which of these patients require orthognathic correction of this deformity helps initiate the discussion of technique, timing, institutional bias, and their effects on maxillary growth significant enough to require an additional corrective surgery. The rates of orthognathic surgery vary widely in the literature and further studies delineating the reason for this variation need to be completed to gain an appreciation for determining an optimal treatment protocol.

### 179. Longitudinal TMJ Compressive Stresses and Mandibular Growth in Children

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Background/Purpose: To determine whether (1) temporomandibular joint (TMJ) compressive stresses differ between growing dolichofacial and brachyfacial children and (2) there was a correlation between TMJ compressive stress and growth of the mandibular ramus.

Methods/Description: Three-dimensional anatomical geometries were derived from archived lateral and posteroanterior cephalographs (AAOF Legacy Collection) of 36 dolichofacial and 29 brachyfacial individuals at average ages of 6 (T1), 12 (T2), and 18 (T3) years. The 3D geometries were produced for use in a numerical model to calculate longitudinal changes in TMJ normal loads produced by static biting on the mandibular incisor teeth. Compressive stresses were estimated based on the magnitude of load divided by published data describing age-dependent increases in the anteroposterior and mediolateral dimensions of the mandibular condyles. ANOVA and Tukey HSD post hoc tests were used to evaluate time (T1, T2, T3) and diagnostic group differences in TMJ compressive stresses and Condylion-Gonion (Co-Go) heights (millimeters). Regression analyses were used to test for a correlation between age-dependent compressive stresses and mandibular ramus height (Co-Go) within diagnostic groups.

Results: Significant diagnostic group differences were found for T2 and T3 compressive stresses and Co-Go heights (all P < .01). Regression analyses showed that dolichofacial subjects produced significantly higher stresses which correlated with smaller ramus heights ( $R^2 = 0.48$ ). In comparison, brachyfacial subjects had lower compressive stresses which corresponded with larger Co-Go heights ( $R^2 = 0.68$ ).

Conclusions: TMJ compressive stresses may explain, in part, differences in mandibular condyle growth between dolichofacial and brachyfacial phenotypes.

### 180. Multiview Videofluoroscopy Training for Radiologists: A Self-Study Package

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Background/Purpose: The objective assessment of velopharyngeal function in children with suspected or confirmed velopharyngeal insufficiency (VPI) typically involves a combination of videonasendoscopy and/or multiview videofluoroscopy procedures. Successful multiview videofluoroscopy studies require collaboration between

medical and health professionals and an understanding of the technical aspects and clinical goals of the study for all professionals involved. The aim of this project was to develop a computerized education portal for increasing awareness and understanding of the technical requirements and goals of multiview videofluoroscopy studies for medical imaging staff unfamiliar with procedure.

Methods/Description: A Microsoft PowerPoint presentation file was constructed using a single-view navigation panel allowing medical imaging staff to choose from a series of written information about the videofluoroscopy procedure and view embedded video samples of recorded studies. The scope of the education included an overview of the procedure, information about cleft palate and VPI, technical information about each of the videofluoroscopy views, an overview of the anatomy of the velopharynx, and expected radiologist reporting guidelines and examples of both optimal and suboptimal views. The presentation file utilized PowerPoint's "kiosk" mode, which controlled the navigation of users to reduce any chance of error in accessing the included information. A focus group of 4 staff from both the cleft palate and medical imaging departments then reviewed the final package.

Results: Following review by the focus group, 6 small modifications were made, and an additional "module" looking at the process for recording the studies was added. The focus group also recommended that the self-study package format be duplicated to address a similar problem with medical imaging staff's understanding of micturating cysto-urethrogram studies. The final self-study package now consists of 17 single pages (slides) of information which takes approximately 4 to 5 minutes to review.

Conclusions: This self-study package is a potentially useful addition to the orientation information for staff new to the medical imaging department in our institution. A formal content and face validity study now needs to be conducted looking at the information contained within the package, and a qualitative study needs to ascertain the nature of any improvements in the quality of imaging being performed and the level of understanding of the procedure by medical imaging staff. These studies will commence following feedback from the ACPA Membership regarding the included components of the package.

## 181. National Trends in Hospitalization Charges and Utilization of Otological Procedures for Cleft Lip and Palate Patients

Yida Cai (1), Lesley Summerville (1), Samuel Boas (1), Cristin Coquillard (2), Anand Kumar (1)

(1) Case Western Reserve University School of Medicine, Cleveland, OH, (2) University Hospitals Cleveland Medical Center, OH

Background/Purpose: Otological procedures are key components of cleft lip and palate management. However, the overall otological burden and utilization of these procedures in this patient population remain poorly understood. The purpose of this study was to examine national trends in patient utilization and hospitalization charges associated with otological procedures for cleft lip and palate patients over multiple years.

Methods/Description: The Healthcare Cost and Utilization Project National Inpatient Sample database was analyzed from January 2007 to December 2014. All patients who were diagnosed with cleft lip or palate who underwent otological procedures were included. Variables of interest included demographic data, hospital characteristics, hospitalization data, and total hospital charges. All charges were inflated to May 2019 value with the consumer price index. Univariate

and generalized linear models were used to examine associations between various factors of interest and the final adjusted hospitalization charge as well as the change in these factors over the multiple years included.

Results: A total of 62 471 patients were diagnosed with cleft lip or palate during hospitalization between January 2007 and December 2014. Of these patients, 4178 (6.7%) received an otological procedure, including myringotomy (3479, 83.3%), auditory function testing (447, 10.7%), and tonsillectomy (110, 2.6%) and were included in the study. Regression analysis showed that total procedure volume did not change significantly over the years examined (P = .06). Higher procedure volume (>30 cases/year) was associated with large bed size hospitals (P < .001), nonprofit private hospitals (P < .001), and urban teaching hospitals (P < .001). The mean total hospital charge for these patients was \$28 108.87 (interquartile range: \$12 529.55-\$32,854.57). Hospitalization charges also increased significantly in this time period (P < .001), in conjunction with an average hospital length of stay of 1.91 days that did not change significantly during the study period (P = .149).

Conclusions: Otological procedures for patients with cleft lip and palate appear to be utilized mainly at larger, urban teaching and nonprofit private hospitals. While hospital length of stay and procedure volume appears be stable across the years studied, hospitalization charges associated with the procedures rose significantly. Further studies are necessary to examine other factors that could be driving the increased hospitalization charges to better target strategies for reducing health-care burden and improve barriers to access to these important procedures for cleft lip and palate management.

### 182. National Trends in Hospitalization Charges and Utilization of Services for Cleft Lip and Palate Repair

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Background/Purpose: Understanding factors that influence variation in hospitalization charges associated with surgical procedures and national trends in utilization of services can help target strategies to reduce health-care spending and improve patient outcomes. The purpose of this study was to examine national trends in patient utilization and hospitalization charges associated with cleft lip and palate repair over multiple years.

Methods/Description: The Healthcare Cost and Utilization Project National Inpatient Sample database was analyzed from January 2007 to December 2014. All patients who were diagnosed with cleft lip or palate who underwent correction or repair of cleft lip and palate were included. Variables of interest included demographic data, hospital characteristics, hospitalization data, and total hospital charges. All charges were inflated to May 2019 value with the consumer price index. Univariate and generalized linear models were used to examine associations between various factors of interest and the final adjusted hospitalization charge as well as the change in these factors over the multiple years included.

Results: A total of 62 471 patients were diagnosed with cleft lip or palate during hospitalization between January 2007 and December 2014. Of these patients, 34 200 (54.7%) received repair of the cleft lip and palate during that hospitalization and were included in the study. Regression analysis showed that the overall volume of cleft lip and palate repairs decreased during the years included

(P=.028). High procedure volume (>50 cases/year) was associated with nonprofit private hospitals (P<.001), hospitals with a large bed size (P<.001), and urban hospitals, both teaching and nonteaching (P<.001). The mean total hospital charge for these patients was \$26 879.58 (interquartile range: \$15 464.90-\$32 020.12). Hospitalization charges also increased significantly in this time period (P<.001), in conjunction with an increase in hospital length of stay (P<.001).

Conclusions: Utilization of services for cleft lip and palate repair appears to center around large, urban, nonprofit, private hospitals. The overall demand for the procedures appears to be decreasing during the years analyzed, in conjunction with rising associated with hospitalization charges and increasing hospital length stay. Longer length of stay may be contributing to increasing hospitalization charges and may be a target for strategies to decrease the overall health-care burden and increase access to these procedures.

## 183. Optimal Outcomes Reporting (OOR): A New Value-Based Metric for Outcome Reporting Following Cleft Palate Repair

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Background/Purpose: The goal of cleft palate repair is to separate the nasal and oral cavities and to achieve adequate velar structure and function so that the patient can develop normal speech. The manner in which outcomes are assessed from center to center demonstrates wide variability, and metrics for palatoplasty success are not uniform. A standardized success metric would help provide quality benchmarks for caregivers and practitioners.

Methods/Description: We conducted a retrospective review of one surgeon's primary palatoplasty patients from 2007 to 2013. Using speech outcomes data by 5 years of age, we created a novel conceptual quality metric that we have called "OOR" (Optimal Outcomes Reporting). The OOR is designed to reflect the percentage of cleft palate patients who experience the best outcomes: one operation, normal speech by age 5 years, and no palatal fistula.

Results: The OOR metric was used in this study. Ninety-four patients underwent primary cleft palate repair between 2007 and 2013 and had adequate follow-up for a speech evaluation by 5 years of age. Our OOR was 79.8% (75/94). Eight patients had "suboptimal" outcomes for having undergone more than one operation associated with their cleft palate. At the second stage of evaluation, 11 more patients failed for borderline or incompetent speech at 5 years of age. No additional patients fell out of the algorithm at the third stage because no additional patients demonstrated postoperative fistulae. A significantly higher proportion of nonsyndromic patients demonstrated an "optimal" result compared to those patients with a syndromic codiagnosis (67/80, 83.8% vs 8/14, 57.1%; P = .02). Within the nonsyndromic group, those patients who underwent palatoplasty at 12 months of age or less had a higher aggregate OOR than those whose palatoplasty occurred after 12 months of age, but the differences were not statistically significant (53/61, 86.9% vs 14/19, 73.7%; P = .28). Patients who required more than 1 procedure had significantly more clinic visits (32.6 vs 14.9, respectively, P < .01) and accrued a significantly higher total cost compared to "optimal" outcome patients (\$34 019.88 vs \$15 357.25, respectively, P < .01).

Conclusions: OOR represents a novel quality outcome metric that can provide meaningful information for patients with cleft palate. OOR

utilization can help cleft centers adopt changes that matter to patients and their families. By allowing for cross-institutional comparisons in a clear and objective manner, OOR can promote both competition and innovation in cleft palate care that will ultimately improve cleft palate care value by improving quality and patient outcomes while decreasing cost.

### 184. Parents of Children With Cleft Lip and Palate: Perceptions of the Treatment Experience

Seungeun Jung (1), Seunghee Ha (1)

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Background/Purpose: Individuals with cleft lip and palate (CLP) are now diagnosed before birth and undergo treatment as early as right after they are born. Children with CLP generally undergo a lengthy period of treatment lasting 4 to 5 years. This extended treatment period requires tremendous energy from the child and from their family in terms of time, financial support, medical and surgical therapy, and emotional resources. The treatment for CLP likely affects a family's quality of life and its psychosocial dynamics. Despite the importance of this topic, no research has investigated the psychosocial effects of CLP treatment in Korea. This study aims to investigate the perception of parents who have been involved in their child's treatment for CLP and explores how their perception about CLP itself has changed over time. More specifically, this study examines each parent's (1) perception of the treatment process, (2) perception of CLP according to different time period, (3) estimation of how CLP has affected their life, and (4) report of any support they need. This qualitative research sought to understand the current status of CLP treatment from the parents' perspectives and to identify the needs of individuals with CLP and their family for better CLP treatment.

Methods/Description: This study interviewed 7 parents of children with CLP; all children were older than 10 years and had experienced most of the treatment process for CLP. An official in-depth interview technique was used for individual. The following elements were considered to increase the credibility of the research: (1) truth values, (2) applicability, (3) consistency, and (4) neutrality. In addition, peer briefing, member check, and triangulation were conducted while all data were collected, and analysis processes have been recorded as audit data. An in-depth interview technique was used for data collection; the grounded theory method was used for data analysis. The interview was conducted in an unstructured way, and interview data were analyzed after transcription through open coding and axial coding to draw the range and topic.

Results: The results of this research are currently undergoing analysis. We expect to find approximately 70 concepts, 30 low categories, and 10 categories. The categories derived from this study included the negative perceptions and burdens of parents on a lengthy period of treatment of CLP, the frustrations, the challenges for the cleft palate, and the requests for early support in the psychosocial sector.

Conclusions: This research can increase our understanding of parents' perceptions of CLP in general and can offer insight into how these perceptions change during the course of the CLP treatment process. This research may help us identify the needs of families with an individual undergoing CLP treatment and offer greater psychosocial support and resources to these families. In addition, the results of this research are believed to contribute to more efficient treatment that considers the quality of life.

## 185. Preprofessionals' Attitudes Toward Individuals With Disabilities or Craniofacial Differences: Practice Implications for Multiple Disciplines

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Background/Purpose: Negative attitudes toward people with disabilities are commonly reported barriers to participation and inclusivity, and similar attitudes have been reported toward individuals with craniofacial differences (e.g., Lewis, Roberson, & Foulsham, 2017; Shannon, Schoen, & Tansey, 2009). Attitudes held by preprofessional students (undergraduate and graduate students in higher education) represent an important area of study because such students may eventually become the professionals who educate, treat, or interact with individuals with these conditions. Systematic reviews provide high levels of scientific evidence for medical professionals. To date, a systematic review on this topic has not been published. This study provides a comprehensive review of the published literature on this topic, which summarizes the clinical and practice relevance of the attitudes held by preprofessional students in the United States.

Methods/Description: A systematic search of 6 databases that commonly index publications related to craniofacial differences, psychology, and attitudes was conducted using keywords, inclusion criteria, and inter-rater reliability to identify relevant peer-reviewed, scientific literature. To be included, articles must have (1) been published in a peer-reviewed journal or been an unpublished dissertation or thesis that has not been published as a peer-reviewed journal article, (2) been written in English, (3) been an examination of thoughts, feelings, emotions, perspectives, or attitudes, (4) included undergraduate or graduate student participants, and (5) been published prior to the Americans with Disabilities Act (1990). The initial database search yielded 996 articles. From this set, duplicate articles were removed, and titles and abstracts were examined for keywords. A total of 38 articles that met all of inclusion criteria were selected for the comprehensive review.

Results: Results documented and summarized (1) reported attitudes toward individuals with craniofacial differences or disabilities, (2) how attitudes varied by demographic characteristics (eg, educational background, exposure to disabilities, contact), and (3) related psychosocial issues relevant to clinical practice. Implications for higher education and for practitioners from multiple disciplines are discussed.

Conclusions: Negative attitudes directed toward individuals with disabilities continue to be a concern. Several demographic characteristics are consistently related to attitudes (eg, gender, service learning experiences, relationships with individuals with disabilities). Preprofessional and practitioner attitudes can potentially hinder the application of best practices when serving individuals with disabilities. Raising awareness of this issue can eventually contribute to improving attitudes toward this population.

### 186. Requirement of Hyaluronan Synthase-2 in Craniofacial and Palate Development

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Background/Purpose: Cleft palate is a common major birth defect resulting from disruption of palatal shelf growth, elevation, or fusion during fetal palatogenesis. Whereas the molecular mechanism controlling palatal shelf elevation is not well understood, a prevailing hypothesis is that region-specific accumulation of hyaluronan, a predominant extracellular glycosaminoglycan in developing palatal mesenchyme,

plays a major role in palatal shelf elevation. However, direct genetic evidence for a requirement of hyaluronan in palate development is still lacking.

Methods/Description: We used are-loxp systems and genetic approaches to analyze developmental defects caused by tissue-specific inactivation of Has2 throughout the cranial neural crest lineage or specifically in developing palatal or mandibular mesenchyme, respectively, using Wnt1-Cre, Osr2-Cre, and Hand2-Cre transgenic mice.

Results: Whereas both Has2f/f;Wnt1-Cre and Has2f/f;Osr2-Cre mutant mice exhibit cleft palate at complete penetrance, the Has2f/f;Wnt1-Cre fetuses showed dramatically reduced mandible size and complete failure of palatal shelf elevation, whereas Has2f/f;Osr2-Cre fetuses had normal mandibles and delayed palatal shelf elevation. All Has2f/f;Hand2-Cre pups showed reduced mandible size and about 50% of them had cleft palate with disruption of palatal shelf elevation. Results from explant culture assays indicate that disruption of palatal shelf elevation in Has2f/f;Hand2-Cre mutant fetuses resulted from physical obstruction by the malformed mandible and tongue.

Conclusions: Our data indicate that hyaluronan plays a crucial intrinsic role in palatal shelf expansion and timely reorientation to the horizontal position above the tongue as well as an important role in mandibular morphogenesis that secondarily affects palatal shelf elevation.

### 187. Strength in Collaboration: Advanced Training to Support Speech Outcomes in Rural Areas

Hannah Sanford-Keller (1), Kameron Beaulieu (1), Janet Brockman (1) (1) Oregon Health & Science University, Portland, OR

Background/Purpose: Orofacial clefting is the most prevalent birth defect in the United States, affecting approximately 7090 newborn infants annually. Best practice guidelines recommend involvement of interdisciplinary teams in treating individuals with cleft lip and palate to address multidimensional concerns in this population, with speech-language pathologists playing a key role in the evaluation and treatment of children with craniofacial differences. However, many speech-language pathology graduate programs do not include specific coursework on cleft lip and palate, resulting in a lack of training related to evidence-based treatment techniques for this population. In fact, nearly 50% of speech pathologists in one study indicated that they do not feel comfortable treating a child with a cleft (Bedwinek, Kummer, Rice, and Grames, 2010). Speech pathologists with advanced training in cleft can help demystify cleft care through community outreach and education, contributing to increased knowledge of the community provider and improved care for children with clefts. This program will review outcomes of state-wide collaborative efforts to improve speech pathology services for children with cleft palate through advanced training workshops. Strategies to enhance collaborative efforts between cleft palate craniofacial teams and community providers in rural areas will be discussed.

Methods/Description: In order to address lack of training in cleft palate speech and enhance collaboration and communication between the craniofacial team speech pathologist and treating speech pathologist, training opportunities have been developed for education service districts across one state. This poster presentation will review collaborative efforts to create cleft-specific education and training opportunities. Key points of the presentation will include course content, strategies for supporting adult learners, and participant indicators of increased knowledge. Additional outcomes from the training will be discussed, including policy change related to eligibility criteria for

children ages 0 to 3 years with cleft diagnoses, enhanced collaboration across one cleft center and education service districts, and the creation of subsequent case-based learning opportunities.

## 188. Study on Tooth Movement After the Alveolar Bone Grafting in Patients With Unilateral Cleft Lip and Palate

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(I) Ninth People's Hospital, Shanghai Jiao Tong University School of Medicine, China

*Background/Purpose*: The aim of the study was to explore the effectiveness of the cleft-adjacent teeth moved into the grafted alveolar bone in unilateral cleft lip and palate (UCLP) patients and to evaluate the alveolar bone support of the teeth.

Methods/Description: Twenty unilateral cleft lip and palate patients were recruited in this study. The average age was 12 years and 8 months. Periapical radiographs were taken for the cleft-adjacent teeth 3 months after bone grafting (T1) and after teeth moving into the grafted bone (T2) and for the contralateral teeth (T3) when teeth aligned. Alveolar bone support of the moved teeth and the contralateral teeth was measured using the ratio of bone support height to root length. Paired t test was performed for statistical analysis with SPSS17.0 software package.

Results: All the cleft-adjacent teeth were bodily moved into the grafted area. Average alveolar bone support ratio for the moved teeth was 86.48% (T1), 87.11% (T2), and 90.81% (T3) for the contralateral teeth. There was no statistically significant difference between T1 and T2 (P > 0.05). Although the contralateral teeth had the highest alveolar bone support ratio, significant differences were found between T3 and T2 (P < 0.05).

Conclusions: Cleft-adjacent teeth could be bodily moved into the grafted bone from the iliac crest in UCLP patients. A successful level of alveolar bone support for the moved teeth was achieved. Moved-in teeth could be functionally loaded and may be benefit to the reduction of the resorption of grafted bone.

## 189. Three-Dimensional Photogrammetry to Assess Disease Progression in Acquired Hemifacial Atrophy: A Pilot Study

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(1) Boston Children's Hospital, MA

Background/Purpose: Hemifacial atrophy (HFA), also known as Parry-Romberg syndrome (PRS), involves progressive hemifacial atrophy of subcutaneous tissues such as skin, muscle, cartilage, and, in severe cases, bone and often begins in childhood. The direct pathological cause of this syndrome has yet to be identified, although the disease is thought to be likely rheumatologic in nature. Previous studies have documented common areas of the face that are affected. Three-dimensional photogrammetry is an ideal method to monitor discrete small changes between times; however, comparing 2 time points can be challenging in the midst of baseline facial growth in the interval. The purpose of this pilot study is to use 3-D imaging to track the progression of HFA during growth and development.

Methods/Description: This pilot study reports the preliminary findings of a prospective study on pediatric patients with HFA. Subjects in all phases of active and stable disease were included. Three-dimensional photographs (VECTRA 3D; Canfield Scientific) taken at 2- to

3-month intervals during routine clinical care. For each separate time point, the images were analyzed for asymmetry by mirror imaging the halves of the face and creating an overlay heat map. Serial images between time points were analyzed by measuring the difference in volume and again using heat maps to illustrate areas of progressive disease.

Results: A female and a male patient were identified who had 1 year of follow-up in the study. One patient demonstrated ongoing disease progression during the study time period. The other patient demonstrates the use of 3D photogrammetry to confirm the stabilization of disease on medical management.

Conclusions: Three-dimensional photogrammetry is a helpful adjunct to the clinical exam in determining whether there has been subtle ongoing atrophy over time. Using these 3D photogrammetry techniques, this pilot study reports early findings of HFA facial volumetric analysis in actively growing patients across long time periods. A similar methodology could also aid in diagnosis and treatment planning for other acquired or congenital facial asymmetry.

## 190. Transforming the Degradation Rate of $\beta$ -Tricalcium Phosphate Bone Replacement Using 3D Printers

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(1) New York University Langone Health, NY, (2) Hansjörg Wyss Department of Plastic Surgery, New York University Langone Health, NY, (3) New York University College of Dentistry, NY, (4) New York University School of Medicine, NY, (5) New York University, NY

Background/Purpose: β-Tricalcium phosphate (β-TCP), the most common synthetic bone replacement product, is frequently used in craniofacial reconstruction. Although solid β-TCP can be absorbed over time, the slow degradation rate (1%-3%/year) predisposes this product to exposure, infection, and fracture, limiting its use in the growing face where implants are required to grow and remodel with the patient. Our tissue engineering laboratory has successfully leveraged 3D printers to manufacture 3D-printed bioactive ceramic (3DPBC) scaffolds composed of β-TCP in an architecture which optimizes the needs of rigidity with efficient vascular ingrowth, osteogenesis, and degradation kinetics. The latter qualities are further optimized when the osteogenic agent dipyridamole (DIPY) is used. This long-term animal study reports on the new degradation kinetics profile achievable through this novel manufacturing and tissue engineering protocol.

Methods/Description: Twenty-two 1-month-old (immature) New Zealand white rabbits underwent creation of unilateral 10 mm calvarial defects with ipsilateral 3.5  $\times$  3.5 mm alveolar defects. Each defect was repaired with β-TCP 3DPBC scaffolds coated with 1000 μM DIPY. Rabbits were killed at 8 weeks (n = 6), 6 months (n = 8), and 18 months (n = 8). Bone regeneration and scaffold degradation were calculated using micro-CT images and analyzed in Amira software. Cranial and maxillary suture patency and bone growth were qualitatively analyzed using histologic analysis.

Results: Results are reported as a percentage of volumetric space occupied by either scaffold or bone. When comparing time points 8 weeks, 6 months, and 18 months, scaffolds showed significant decreased defect occupancy in calvaria (23.6%  $\pm$  3.6%, 15.2%  $\pm$  1.7%, 5.1%  $\pm$  3.4%; P < .001) and in alveoli (21.5%  $\pm$  3.9%, 6.7%  $\pm$  2.7%, 0.1%  $\pm$  0.2%; P < .001), with annual degradation rates 55.9% and 94.2%, respectively. Between 8 weeks and 18 months, significantly more bone regenerated in calvarial defects (25.8%  $\pm$ 

6.3% vs  $55.7\% \pm 10.3\%$ , P < .001) and no difference was found in alveolar defects ( $28.4\% \pm 6.8\%$  vs  $32.4\% \pm 8.0\%$ , P = .33). Histology showed vascularized, organized bone without suture fusion.

Conclusions: The degradation kinetics of  $\beta$ -TCP can be altered through 3D printing and addition of an osteogenic agent. Our study demonstrates an acceleration of  $\beta$ -TCP degradation from 1% to 3% a year to 55% to 95% a year. Absorbed  $\beta$ -TCP is replaced by vascularized bone and there is no damage noted to the growing suture. This additive manufacturing and tissue engineering protocol has implication to future reconstruction of the craniofacial skeleton.

### 191. Mental Health Care Follow-Up in Children and Adolescents With Craniofacial Anomalies

Melissa Rafferty (1), Irene Baus (2), Catherine Scherer (3), Anand Kumar (4), Rebecca Hazen (3)

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Background/Purpose: Children with cleft lip and/or palate or other craniofacial anomalies experience increased rates of learning disabilities, mental health problems, and teasing or bullying. Craniofacial team social workers often make referrals for psychosocial resources. However, patients and their families may experience significant barriers to their ability to follow through with those referrals, such as insurance difficulties and social stressors. The purpose of this study is to collect data about rates of and types of psychosocial referrals provided by a social worker integrated within an interdisciplinary craniofacial clinic, as well as rate of follow-through with those referrals and potential barriers to follow-through on care.

Methods/Description: Participants were patients with cleft lip and/or palate or other craniofacial anomalies who attended an interdisciplinary craniofacial clinic at a large metropolitan children's hospital in the Midwest. Patients typically attend the clinic on a yearly basis, with meetings with the team social worker included at those visits. Inclusion criteria included 4 to 26 years of age and contact with social worker at clinic visits. Data extracted from medical charts included whether the patient was already receiving mental health services, whether the social worker provided mental health referrals, type of referral, reason for the referral, and whether they followed through with the referral 1 year following their visit. Information on insurance status (private vs public) and any documented history of mental health problems of the parents was also gathered from charts.

Results: A total of 100 participants' charts were reviewed. As documented by the team social worker, 39% of patients were already receiving mental health services. Referrals were made by the social worker for 27% of patients. The following types of referrals were made: individual therapy, family support, developmental behavioral pediatrics, and psychiatry. Documented reasons for referral included anxiety, depression, ADHD, autism, learning difficulties, family support, obesity, and sleep. Eighteen percent of families who were given a referral followed through on that referral within 1 year. Of those with private health insurance, 50% followed through on referrals. Only 14% of families with a Medicaid policy were receiving the recommended services at follow-up, although the differences in proportions were not statistically significant ( $\chi^2 = 2.86$ , P = .09). Of the 19 families with parental history of mental health problems, 11 were referred for services and only 1 followed through with the referral.

Conclusions: Results suggest low rates of follow-up on mental health referrals and highlight the importance of addressing barriers to care. Additional research using parent-reported measures of beliefs about mental health care and barriers to care is needed to better understand ways to improve follow through on mental health referrals.

#### 192. ASCFS Linton Whitaker Lecture

Ian Munro (1)

(I) Naples, FL

Methods/Description: Dr Ian Munro is a pioneer in the field of craniofacial surgery, having played a key role in the development and establishment of the subspecialty. This lecture will follow Dr. Munro's journey across several countries and multiple occupations and discuss some of the major breakthroughs that have changed, improved, and simplified craniofacial surgery. Dr. Munro will share insights he has gained from over 20 years of practice and offer advice to medical professionals just entering their field.

#### 193. Patterns of Academic Risk and Services Received in Children With Isolated Cleft Lip and/or Palate (iCL/P)

Amy Conrad (1), Meredith Albert (2), Canice Crerand (3), Claudia Crilly Bellucci (4), Celia Heppner (5), Farah Sheikh (6), Suzanne Woodard (7), Kathy Kapp-Simon (2)

(1) University of Iowa Children's Hospital, Iowa City, IA, (2) Shriners Hospitals for Children Chicago, IL, (3) Nationwide Children's Hospital, Columbus, OH, (4) Shriners Hospitals for Children-Chicago and University of Illinois at Chicago, IL, (5) Children's Medical Center, Dallas, TX, (6) The Hospital for Sick Children, Toronto, ON, Canada, (7) Lancaster Cleft Palate Clinic, PA

Background/Purpose: iCL/P research has demonstrated a higher risk of learning concerns, particularly in reading. Extensive work has evaluated biological factors (eg, sex and cleft type) and neuropsychological patterns (eg, phonological awareness, auditory wording memory). However, very little work has addressed the potential relationship to school environment, psychosocial functioning, or level of school support received.

Methods/Description: A total of 363 children, ages 8 to 10 years of age (181 males; 54 iCL, 63 iCP, and 246 iCLP) and their guardians were recruited during a regular clinic visit to complete questionnaires designed to evaluate psychological functioning in relation to cleft-related treatment. Participants were recruited from 6 centers across North America from August 2016 through May 2019. Measures included: Academic Functioning (ABAS-III and CBCL subscores), General School Functioning (Cleft-Q and PedsQL subscores), Global Psychosocial Functioning (PedsQL and CBCL total scores), and diagnoses/services received.

Results: Academic Functioning was predominantly in the average to high average range, although boys had significantly lower functioning than girls,  $F_{1,\ 350}=4.175,\,P=.042$ ). This difference was driven by lower performance by boys with iCP (25% at or below the 16th percentile). Of the total sample, 21.1% fell within the at-risk range for academics. Thirty-five participants (10%) had a formal diagnosis of a learning or language disability; 26% of these children had co-occurring diagnoses. Academic Functioning was significantly correlated with ratings of General School Functioning (r range of .140 to .330). However, Academic Functioning was not consistently correlated with Global Psychosocial Functioning (r range of .072 to .283). Finally, 55% had a history of receiving either or academic or

language services; the most common was speech therapy (51%). Reading (11%), writing (3%), and math (8%) services typically started in preschool and steadily increased in frequency through the second grade. Of participants rated within the at-risk range on Functional Academics (n = 43), 81% were receiving some assistance through an individual education plan/special education. While cleft teams were likely to make referrals for speech therapy, referrals for learning concerns were made less frequently.

Conclusions: Academic functioning for the majority of children with iCL/P is within the average range or higher. Results support previous research that boys, and particularly those with iCP, are at a higher risk for poor performance. Academic functioning was strongly correlated with general school functioning and less so to global measures of psychosocial functioning. Of those at risk for academic issues, the majority are receiving services through the school system, but academic referrals are not consistently provided by cleft teams. Screening of academic performance should continue and guardians should be guided in risk factors and ways to advocate for their children within the school system.

## 194. Patients With a History of Oronasal Fistula Repair Exhibit Lower Oral Health Measured With Patient-Centric Outcomes Measures

Diana Jodeh (1), Alyssa Fritz (1), James Cray (2), S. Alex Rottgers (1) (1) Johns Hopkins All Children's Hospital, St Petersburg, FL, (2) The Ohio State University, Columbus, OH

Background/Purpose: Cleft lip and palate is the most common congenital craniofacial anomaly, with oronasal fistula representing the most frequent complication associated with cleft palate surgery. Oronasal fistulae may affect patients' quality of life by impacting their ability to eat, speak, and maintain oral hygiene. We aimed to quantify the impact of previous oronasal fistula repair on patients' quality of life using patient-specific outcomes psychometric tools.

Methods/Description: A cross-sectional study of 7- to 19-year-old patients with cleft palate with or without cleft lip was completed. Patients who had a cleft team clinic between September 2018 and August 2019 were recruited. Members of the research team did not perform primary cleft palate repairs within this cohort. Participants were divided into 2 groups (no-fistula, prior fistula repair). Scores for CLEFT-Q dental, jaw, speech function, and eating and drinking and Child Oral Health Impact Profile (COHIP) scores were recorded. Differences in the individual CLEFT-Q and COHIP scores between the 2 groups were evaluated using a Mann-Whitney U test controlling for sex.

Results: The identified cohort included 66 patients with a history cleft palate. Forty-six patients (70%) had an associated cleft lip. Thirty-six patients did not have a fistula and 30 patients had undergone a fistula repair. Median age was 11 years (range: 7-19 years of age) and 52% were male. The number of fistula repairs ranged from 1 to 5. Scores for CLEFT-Q dental (no-fistula 61.36 vs prior fistula repair 55.93), jaw (no fistula 80.14 vs fistula repaired 72.3), speech function (no-fistula 74.53 vs prior fistula repair 66.60), and eating and drinking (no-fistula 30.09 vs prior fistula repair 30.7) were all higher in patients without a history of a fistula repair; however, none of these differences were statistically significant. The COHIP score (no-fistula 6.06 vs prior fistula repair 8.07) demonstrated a significant difference, indicating patients with a history of fistula repair had poorer oral health (P=.015).

Conclusions: When compared to patients without a history of oronasal fistula, patients who have undergone repair of an oronasal fistula

exhibit worse overall oral health, as indicated by an increased COHIP score. Other measures of oral health indicated decreased score in patients with a history of fistula repair consistently, but statistical significance was not observed in this study. One would expect that successful repair of a fistula would result in improved function and patient satisfaction, but the consistent trend toward lower scores in our study group suggests the possibility that residual effect remains that may be subtle enough to avoid detection with our sample size despite an extremely high fistula rate within this cohort.

### 195. Perceptions and Preferences of Laypersons in the Management of Positional Plagiocephaly

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Background/Purpose: Positional plagiocephaly is a common, benign condition in infancy. Uncertain clinical evidence for the treatment of positional plagiocephaly, especially with helmet therapy, creates difficulties in counseling parents of patients. This study investigates layperson perceptions and treatment preferences for positional plagiocephaly to provide patient-oriented evidence for management.

Methods/Description: Adult laypersons were recruited through crowd-sourcing to view digitally modified images of normal, mildly, moderately, or severely plagiocephalic infant heads. Participants provided demographic information and rated the infant's head shape and potential social difficulties in the future, likelihood of consulting a physician for treatment options, and likelihood of seeking helmeting treatment for the infant on a Likert scale. Statistical analysis was performed using Kruskal-Wallis, Mann-Whitney U, and Pearson  $\chi^2$  tests.

Results: A total of 945 individuals participated in the study. Perception of head shape, prediction of future embarrassment and social difficulties, likelihood of seeking physician evaluation, likelihood of choosing helmet therapy, and willingness to pay for helmet therapy were pairwise different between 4 plagiocephaly severities (adjusted P <.001 for all), except between normocephaly (n = 194) and mild (n = 194)334) plagiocephaly or between moderate (n = 203) and severe (n = 203) 214) plagiocephaly. Younger respondents were more likely to consult a physician (unadjusted P = .016) and choose helmet therapy (unadjusted P = .004) for infants with normocephaly or mild plagiocephaly. Parents of children with physical disabilities were 6 times as likely as other participants to choose helmet therapy for mild plagiocephaly (adjusted P = .036). Perception of head shape normality, likelihood of consulting a physician to discuss treatment options, and likelihood of choosing helmet therapy were independent of household income and gender of the participants.

Conclusions: Laypersons considered mild plagiocephaly as normal but perceived a significant deformity in moderate and severe plagiocephaly. Their treatment preferences followed the perceived level of deformation. Parents of physically disabled children were significantly more likely than other participants to choose helmet therapy for mild deformations, even though parents in general had a higher threshold than nonparents for defining a head shape as abnormal and for seeing a physician to discuss treatment options. Our findings provide medical professionals with lay perspectives on positional plagiocephaly that may facilitate effective counseling of parents.

## 196. Neurodevelopment and Behavioral Functioning of Pre-Kindergarten Children With and Without Craniofacial Microsomia

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Background/Purpose: Craniofacial microsomia (CFM) is characterized by underdevelopment of the ear(s) and mandible. Although school-age children with CFM have been found to be at elevated risk for learning difficulties compared to unaffected peers, few studies have investigated their early neurodevelopment. This study examines development in preschoolers with and without CFM as part of the multisite CFM: Longitudinal Outcomes in Children Pre-Kindergarten (CLOCK) study.

Methods/Description: Preschoolers with CFM (n = 92) and without CFM (n = 76) were evaluated at an average age of 38 months (SD = 1.9). Assessments were completed in English (73%), Spanish (8%), or both languages (19%) and included the Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III) and the Clinical Evaluation of Language Fundamentals Preschool, Second Edition (CELF P-2). Parents completed the Child Behavior Checklist (CBCL). Medical information was gathered from caregivers and chart review. Scores were converted to z-scores based on the controls' score distribution. Group differences were estimated using linear regressions adjusted for age, sex, socioeconomic status (SES), study site, race, and test language. In categorical analyses, a delay was defined as ≥1 SD below the mean.

Results: Most participants were male (55%), white (68%), and Latino (46%), and a range of family SES was represented. The phenotype of cases was categorized as microtia with mandibular hypoplasia (51%), microtia only (29%), and other CFM features (20%). Nearly 70% of cases received intervention services and 81% had hearing loss. After adjustment for covariates, scores were similar for children with and without CFM on the Bayley-III subtests and on the CELF P-2 receptive language subtests of Sentence Structure and Basic Concepts. Children with CFM scored significantly lower than controls on the CELF P-2 expressive language subtest of Recalling Sentences ( $\beta$  = -.40, P = .04) and receptive language subtest of Concepts and Following Directions subtest ( $\beta = -.57$ , P = .01). Among cases, those with microtia only and with other CFM features had lower language scores. An area of delay was present in 37% of children with CFM compared to 15% of controls. There were no differences in the CBCL composite scales.

Conclusions: On average, development in preschoolers with CFM was comparable to controls in most areas, with the exception of group differences on selected language measures. In categorical analyses, children with CFM were more likely than controls to have an area of delay. Relative to previous cohorts, the CLOCK sample was comprised of fewer participants with more severe presentations of CFM and had a high rate of early intervention, which may attenuate differences. In addition, learning/developmental differences may become more pronounced over time. Providers should monitor for language, learning, and behavioral concerns as children with CFM enter school and assist in advocating for appropriate school and community services.

## 197. Multinational Study of Children With Craniofacial Microsomia: Parental Reports of Psychosocial and School Status

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Background/Purpose: Craniofacial microsomia (CFM) is associated with phenotypic variability that typically includes underdevelopment of the ear(s) and mandible. The annual US birth rate for CFM is approximately 1100. Beyond experiencing complex health-care needs, children with CFM can be at increased risk of social and educational difficulties, which may be affected by speech and hearing status. The goal of this presentation is to describe parent report of CFM social experiences and educational placement, services, and overall grades.

Methods/Description: As part of CAUSE, an NIH-funded prospective study at 4 sites in the United States and 5 sites in South America, caregivers of children with CFM under age 18 were interviewed about their child's demographics, health history, their child's awareness of their diagnosis, experiences of teasing, educational placement and services, and overall school performance.

Results: Children with CFM (N = 175) had a mean age of 6.9 years (SD = 5.9), 60% were male, and most were born in the United States (41%) or Colombia (45%). Responses were largely similar across US and South American participants. Unilateral microtia (86%) was the most common CFM feature and 88% had hearing loss, with 73% using hearing aids. Parents reported children's mean age of first awareness of their diagnosis was 4.2 years; however, a third of parents felt their children were still too young to notice their diagnosis. Teasing was reported for 28%, which started at an average age of 6.6 years and took place at school/ daycare (76%) by classmates (75%) and community peers (20%). Teasing was most often described as taking place some of the time (47%). Of those enrolled in school, 81% in the United States and 98% in South America were in general education. Parents described their children's grades as well above average (35%), above average (28%), or average (20%). Individualized education programs were in place for 45% in the United States compared to 4% in South America, most often for speech, language, and/or hearing impairments.

Conclusion: Results from this multinational study are consistent with findings from smaller US-based studies of psychosocial and educational outcomes in CFM. As parents noted diagnosis awareness around age 4, with around a quarter of children experiencing teasing starting before age, team members are encouraged to integrate coping strategies for parents and children starting in their first team visit. The majority of children were reported to be average or above average academically in general education classes, and nearly half of US children had an IEP. However, given high rates of hearing loss, it may be that additional support services would be indicated and team members should be prepared to assist in linking and advocating for appropriate services.

# 198. A Comparison of Alveolar Bone Graft (ABG) Outcomes After an Inter-Center Comparison With Peer-Benchmarking Led to a Change in a Center's Protocol to Earlier Grafting in the Late Primary Dentition Prior to Orthodontic Intervention

Catherine Lowry (1), Ross Long (2), Jennifer Giltner (3), Kathleen Russell (4), Jean-Charles Doucet (4), John Daskalogiannakis (5), Patricia Beals (6), Stephen Beals (6), Ana Mercado (7), Ronald Hathaway (8), Lexi Weaver (2)

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Background/Purpose: A variety of treatment protocols for ABG is used. Peer benchmarking by intercenter comparison has identified a range of ABG outcomes. Poor graft outcomes in one center using a traditional protocol to mixed dentition ABG compared to other centers using variations of that approach led to a change of protocol at that center, including earlier grafting before any orthodontic intervention. This is a similar intercenter comparison after that change, to compare outcomes between 2 groups with complete clefts treated at that center: one with the original grafting protocol (8-10 years and after pregrafting orthodontic intervention); the other with a new protocol to earlier secondary ABG (5-7 years and before orthodontic intervention). A third group from the original comparison was included to represent the outcomes possible with the change in protocol to earlier, preorthodontic treatment ABG.

Methods/Description: Sample was 99 consecutively grafted patients with complete clefts from 2 North American Centers forming 3 groups. Two groups (30 and 29 patients) from center 1 had undergone either "original" protocol of mixed dentition grafting (mean age: 10.2 years) after pregrafting orthodontic treatment or the "new" protocol of earlier, late primary dentition grafting (mean age: 6.5 years) prior to initiation of orthodontic treatment, respectively. The third group of 40 patients from center 2 was the benchmarked sample for earlier grafting (mean age: 6.7 years) from the initial comparison. Using occlusal radiographs, the Americleft SWAG scale from 0 (failed graft) to 6 (ideal) was used to rate graft outcome at a mean of 15.5 months post-ABG. Six trained and calibrated raters blindly scored each radiograph twice, with the average of the 12 ratings being used as the final score. Reliability was assessed using weighted kappa statistic. The significance of differences between groups was determined using the Kruskal-Wallis test and Dunn test for pairwise comparisons (P < .05).

Results: Inter-rater reliability was good (0.631). Intra-rater reliability was excellent (0.817). There was a tendency for improved total ABG outcome in center 1's early grafted group compared to the later grafted group, with the improvement being significantly different in only the coronal third of the early, preorthodontic ABG group (1.13-1.83; P=.009). However, although improved, the difference was not statistically significant for the total graft (3.63-4.17; P=.283). Outcomes from both center 1 groups however were significantly poorer than the early grafted group from center 2.

Conclusions: A change in grafting protocol as a result of an internal audit of bone graft outcomes through comparison with other centers and peer benchmarking (earlier age and prior to orthodontic treatment) led to an improvement in graft results as assessed with the SWAG method. The improvement was limited to the coronal third of the graft with no measurable difference in middle and apical thirds.

## 199. Effects of Nasoalveolar Molding in Children With Unilateral Cleft Lip and Palate: A Systematic Review and Meta-Analysis

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Background/Purpose: Nasoalveolar molding (NAM) is an increasingly common presurgical treatment option for children with unilateral cleft lip and palate (UCLP). Two systematic reviews on NAM published in 2011 and 2012 revealed a scarcity of comparative studies and inconsistent reporting of aesthetic outcomes, which prevented meta-analysis or definitive treatment recommendations. Since 2012, multiple large studies examining long-term treatment effects of NAM have been published, often with conflicting results. The aim of this systematic review and meta-analysis was to investigate the effects of NAM on nasal form and craniofacial development in children with UCLP.

Methods/Description: Manuscripts were included in this review if they met all of the following criteria: (1) involved children with UCLP who received NAM; (2) included a comparison group consisting of children with UCLP who either received non-NAM presurgical infant orthopedic appliances (PSIOs) or who did not receive any PSIO; (3) reported at least one objective or validated measure of nasolabial, craniofacial skeletal, or palatal form; and (4) had patient follow-up beyond 4 years of age. Outcomes were considered for meta-analysis if they were reported in at least 3 studies. Otherwise, outcomes were included in a qualitative data synthesis. A meta-analysis was performed, which assessed nasal form, nasolabial profile, and vermillion border according to Asher-McDade Index scores.

Results: A total of 11 studies met inclusion criteria for this review, involving 242 patients treated with NAM, 111 patients treated with non-NAM PSIOs, and 271 patients treated without PSIOs. When compared to children who did not receive PSIOs, those who underwent NAM therapy were more likely to have good to excellent Asher-McDade Index scores for nasal form (OR: 2.4, 95% CI: 1.24-3.68) and vermillion border (OR: 1.8, 95% CI: 1.19-2.71). No comparisons between NAM and non-NAM PSIOs achieved statistical significance. The preponderance of evidence from the qualitative synthesis suggests that NAM improves long-term nasal form but does not have clear long-term effects on craniofacial skeletal development.

Conclusions: This systematic review and meta-analysis consolidated evidence from multiple recent studies on the treatment effects of NAM in patients with UCLP. Our findings suggest that NAM produces benefits in nasal form when compared to no presurgical treatments. However, there is not yet sufficient evidence to conclude whether NAM produces similar benefits when compared to other PSIOs. This review also highlights limitations in reporting that must be overcome to guide management of patients with UCLP.

## 200. Pre-Surgical Orthopaedics in Cleft Lip and Palate: A Case Series from a Sub-Saharan African Country

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Background/Purpose: Presurgical orthopedics (PSO) is the gradual moulding of the nasolabial structures and alveolar segments of an infant born with cleft of the primary palate prior to lip repair. PSO is an uncommon practice in the developing world, and no publication was found describing the clinical practice of PSO in Africa. This report describes the clinical findings and the experience of a cleft center in Africa that offers PSO services as a component of comprehensive cleft care. To the best of our knowledge, this is the first of such report from sub-Saharan Africa.

Methods/Description: Parents of infants younger than 1 month who attended our cleft clinic between April and June 2019 were counseled on the merits and demerits of PSO. Infants with unilateral cleft of the primary palate alone or primary and secondary palate were recruited to have PSO done after obtaining informed consent from their parents. Pre and post PSO photographs were taken with a metric guide, while maxillary dental casts were obtained from impressions taken with rubber base material. Lip strapping was commenced while a nasoalveolar moulding (NAM) plate was fabricated in clear acrylic. The NAM plates were delivered with verbal instructions to the parents on how to use and how to clean them. Following familiarization with the use of the NAM plate, a nasal stent was added to complete the NAM device. The use of NAM device was continued to the time of lip repair. The infants were seen on a weekly basis and their NAM devices were adjusted with soft acrylic liner with the aim of passively moving the cleft alveolar segments toward each other. Mid-cleft soft tissue width, columella angle, and anterior alveolar cleft width were measurements obtained from the pre and post PSO photographs and the maxillary casts. The complications and challenges encountered were also documented.

Results: Eight infants with unilateral cleft of the primary palate with or without cleft of secondary palate were recruited. The mean age at the commencement of PSO was 17.6 (SD  $\pm$  5.5) days. One infant defaulted, 2 infants had strapping alone, and 5 infants had strapping with nasoalveolar moulding. There were 40.0% and 42.3% reductions in mid-cleft soft tissue width and anterior alveolar cleft width, respectively, while the columella angle increased by 26.4%. The complications encountered were transient abrasions (4/7) and hypopigmentation of the cheek (6/7). Distance of patient's place of residence to our center, frequent clinic visits and difficulty of the team in securing some materials such as the infant-size impression trays were the major challenges observed. However, the enthusiasm and motivation of the parents were very encouraging to the cleft care team.

Conclusion: Presurgical orthopedics in cleft lip and palate anomaly is a feasible practice with surmountable challenges in a sub-Saharan African country. Barriers to uptake of PSO services should be researched with a view to encouraging its practice our environment.

## 201. Stability and Outcome After Bimaxillary Surgery for Skeletal Class III with Lip or Occlusal Cant: Comparison Between Different Surgical Designs

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Background/Purpose: Lip or occlusal cant is not uncommon in patients with asymmetric class III deformity. The aim of this study was to compare the stability and outcome after bimaxillary surgery using 2 different surgical designs in patients with skeletal class III deformity and lip or occlusal cant. The predictors for lip cant correction were also evaluated. The hypotheses to be tested were (1) The surgical stability was different between the 2 different surgical designs. (2) The surgical outcome was different between the 2 different surgical designs.

Methods/Description: Fifty adults with class III asymmetry and lip or occlusal cant who consecutively underwent bimaxillary surgery were studied. Two groups were examined: 31 received asymmetric posterior maxillary impaction on both sides (II group), 19 received posterior maxillary impaction and extrusion on different sides (IE group). Conebeam computed tomography was used to assess surgical stability and outcome in terms of facial midline and lip cant at 3 time points: before treatment (T0), one week after surgery (T1), and after treatment (T2, finish of orthodontic treatment and at least 12 months after surgery). The predictors of lip cant correction were also investigated.

Results: After surgery the mandible moved and rotated upward in both groups, and the upward rotation was larger in the IE group than the II group. Significant improvement in facial asymmetry was found in both groups; however, majority of the II group patients remained undercorrection of the facial midline deviation. The correction of lip cant was related to the initial lip cant and the change in mandibular roll rotation.

Conclusions: Mandibular stability was different but symmetry outcome was similar in patients receiving 2 different surgical designs for correction of class III asymmetry and lip or occlusal cant. The correction of lip cant was related to its initial severity and the correction of mandibular roll asymmetry.

## 202. The Association Between Dental Anomalies and Non-Syndromic Cleft Lip and/or Palate: A Meta-Analysis

Tamer Marzouk (1), Igor Lemos Alves (2), Cynthia Wong (1), Lisa DeLucia (1), Christy McKinney (3), Brian Howe (4), Mary Marazita (5), Sean McLaren (1), Dorota Kopycka-Kedzierawski (1), Clinton Morrison (1), Hans Malmstrom (1), Hongyue Wang (1), Erin Shope (1)

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Background/Purpose: Conduct a systematic review and meta-analysis to assess whether individuals with nonsyndromic cleft lip and/or palate (CL/P) display a higher frequency of dental anomalies (DA) compared to individuals without CL/P.

Methods/Description: The authors conducted a literature search of indexed databases (PubMed, Cochrane, Web of Science, Embase, Science direct, and LILACS) without language restriction up to and including September 1, 2019, and used cross-referencing to further identify articles. The authors contacted several cleft teams across the United States and Europe to obtain the unpublished data. The eligibility criteria were as follows: (a) observational studies with original data that statistically compared (b) individuals with CL/P without syndromes to (c) those without CL/P on (d) any type of DA in primary and/or permanent dentition. The authors performed meta-analysis using Cochrane Community Review Manager 5 software and used odds ratio with 95% confidence intervals as a measure of the association between CL/P and DA. The authors used random effects meta-analysis with a Mantel-Haenszel estimator to obtain an overall summary estimate of the association between DA and CL/P. The authors assessed the methodological quality of the studies according to the modified Newcastle-Ottawa Scale.

Results: The literature search generated 974 records, and 51 full-text articles were reviewed. Twenty-one studies comprising 12 143 individuals, with ages between 1 and 74 years, from all the world regions fulfilled our selection criteria and were included in the meta-analysis. Thirteen studies reported the female percentage across the study and control groups which ranged from 30.0% to 50.6% and 36.5% to

63.6%, respectively. The overall risk of bias was "low" in a study, "moderate" in 15 studies, and "high" in 5 studies. The meta-analysis revealed significant associations between CL/P and tooth agenesis (odds ratio [OR] = 13.9; 95% confidence interval [CI] = 8.3-23.2), supernumerary teeth (OR = 4.4; 95% CI, 2.9-6.8), developmental enamel defects (OR = 4.2; 95% CI, 2.9-6.3), microdontia (OR = 13.4; 95% CI, 3.1-57.1), peg-shaped anterior teeth (OR = 12.1; 95% CI, 3.5-41.1), taurodontism (OR = 1.9; 95% CI, 1.2-3.0), thick-curved maxillary central incisors (OR = 42.2; 95% CI, 8.1-218.9), tooth malposition and transposition (OR = 6.3; 95% CI, 2.6-15.2), and tooth rotation (OR = 1.6; 95% CI, 1.2-2.2). Nonsignificant associations were observed between CL/P and fusion and gemination, dens invaginatus, root dilaceration, and tooth impaction.

Conclusions: Our findings suggest that individuals with CL/P are more likely to have a range of DAs than their unaffected peers.

#### 203. How Does the Bandeau Grow: Quantifying Post-Operative Changes in the Bandeau Over Time After Fronto-orbital Advancement

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Background/Purpose: Temporal hollowing is a late sequelae of fronto-orbital advancement (FOA) surgeries. Bony manipulation including devascularization and undercorrection may contribute to temporal hollowing. We sought to objectively assess how such bony morphology changes over time.

Methods/Description: A multicenter, retrospective study identified craniosynostosis (CS) patients treated with FOA between 2008 and 2018 with both early and late (>12 months) postoperative head CT scans. Scans were reconstructed, oriented, and manually segmented into surgical fragments that delineated the osteotomies of interest for a given patient. Thirty-two data points and 35 discreet metrics were collected from each patient and evaluated for changes over time.

Results: Twenty patients matched inclusion criteria (12 female, 8 male). CS subtypes included metopic (7), unilateral coronal (6 right, 3 left), and other (4). Mean age at surgery and time to follow-up scan was 1.4 and 2.8 years, respectively. Average biparietal growth was 3.3%. The bandeau AP length increased 18.2%, and height increased 29.5%. Although average bandeau bitemporal width increased 4.7%, anterior width decreased by 4.1%, leading to an anterior transverse deficiency. The average initial bandeau orbital width was 3.8 mm wider than the midface orbital width and decreased by 2 mm ( $\sim 50\%$  loss of overcorrection). Data were significant by paired t test.

Conclusions: The long-term shape and position of the bandeau determines surgical success of FOA. We found that the skull continues to widen bitemporally after surgery; however, widening at the anterior temporal region is negligible. This is the first comparative demonstration of the bony contribution toward temporal hollowing in early and late postoperative patients.

### 204. Long-term Growth Following Trigonocephaly Repairs: Are Overcorrections Necessary?

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Background/Purpose: Does bifrontal width change with growth following trigonocephaly corrections? Postoperative long-term growth was assessed, along with the impacts of phenotypic severity, surgical timing and operative technique, in order to determine how wide to surgically set bifrontal width.

Methods/Description: A retrospective review of all trigonocephaly repairs was performed. Exclusion criteria included syndromic conditions, incomplete records, and follow-up younger than 1 year. Anthropometric measurements taken through completion of growth were evaluated and compared to sex- and age-matched normal standards for Z-score conversion.

Results: Of 370 consecutive patients undergoing repairs, 95 had sufficient anthropometric data. The mean surgical age is 10.8 months (range, 2 months to 7 years) and mean follow-up is 54.3 months (range, 12 months to 17.8 years). Sequential measurements revealed progressive increases in bifrontal width. However, following conversion to Z scores, the initial overcorrection (mean overcorrection = 8.7 mm; mean Z-score = +2.3) steadily diminished to an undercorrection (mean measurement = -5.5 mm; mean Z-score = -1.1). Compared to treatment at an older age (10-12 months), repairs performed under 8 months showed worse growth (P = .004). Those more severely affected (lowest bifrontal Z scores) had similar growth to those more mildly impacted. Only 2 (2.1%) patients underwent secondary procedures for recurrences. No correlation was found between anthropological measurements and observers' severity assessments.

Conclusions: Subnormal bifrontal growth occurs following trigonocephaly corrections, especially with earlier corrections. Our repairs performed around 11 months of age had to be overcorrected by approximately 1.5 cm to produce a normal bifrontal width at maturity.

## 205. Anomalous Venous Drainage Preventing Safe Exposure for Cranial Vault Remodeling for Multi-Suture Syndromic Craniosynostosis

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Background/Purpose: Craniosynostosis is a condition in which one or more of the cranial sutures fuse prematurely. The incidence of craniosynostosis is 1 in 2000 to 2500 births. It often occurs in isolation; however, approximately 20% of cases are observed in the context of a predisposing genetic lesion that is associated with visceral and limb abnormalities. These cases almost always involve multiple cranial sutures and often are associated with anatomic abnormalities of the cranial base. The most common genetic alteration in syndromic craniosynostosis is abnormal tyrosine kinase fibroblast growth factor receptor pathway signaling. The most common syndromic craniosynostosis disorder is Crouzon syndrome, which is characterized by multisuture fusion, exorbitism, and facial deformity. Isolated nonsyndromic craniosynostosis is not associated with known genetic alterations. In cases of multisuture craniosynostosis, patients are susceptible to craniofacial abnormalities and neurologic sequelae including cognitive delay, increased intracranial pressure, hydrocephalus, and abnormal development of the cranial and intracerebral venous drainage system. Surgery is a definitive treatment option for patients with craniosynostosis. Operative intervention is often required to promote normal craniofacial development and to treat or prevent neurologic sequelae. Anomalous venous drainage is often observed in patients with craniosynostosis, and this has a significant impact on preoperative evaluation and surgical treatment planning. The detection of abnormally enlarged venous emissaries has served as a contraindication for surgery in some cases. Optimal surgical treatment strategies

must weigh the risks and benefits of intervention with patient- and disease-specific factors. When considering open cranial vault remodeling for craniosynostosis, preoperative imaging that evaluates skull, brain, and venous anatomy is an important modality for assessing surgical risk. This is particularly true for patients with syndromic craniosynostosis and multiple fused sutures as they are more susceptible to hemorrhagic complications during surgery and intracranial venous hypertension due to aberrant venous anatomy.

Methods/Description: We report a case of a patient with Carpenter syndrome and multisuture synostosis who was found to have significant superficial venous tributaries with deep venous drainage. The presence of this abnormal venous pattern served as a contraindication for surgery due to the grave risk of hemorrhage and/or venous infarction during exposure for cranial vault remodeling. The venous abnormalities were detected on CT venography. Preoperative venous vascular imaging is a necessary component of a thorough preoperative evaluation and is required to assess risk for surgery and avert potentially lethal injury to vital venous structures on exposure.

## 206. A Subtype of Sagittal Craniosynostosis Has Enhanced Anterior Dysmorphology: Pointers for Surgical Management

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Background/Purpose: Sagittal craniosynostosis has some variability in presentation. Some patients appear to be affected more anteriorly, while others are more affected posteriorly. Others are quite affected bidirectionally. The aims of this article are to describe a subtype of sagittal craniosynostosis which presents with a retrocoronal temporoparietal transverse band-like constriction, to demonstrate enhanced anterior cranial vault dysmorphology in these patients, and to elucidate upon reconstructive solutions for this problem.

Methods/Description: For the years 2015 to 2018, 51 patients with nonsyndromic single-suture sagittal craniosynostosis were operated upon by the authors. All patients were followed in a multidisciplinary craniofacial program including genetics. Patients routinely underwent a preoperative CT scan and pre- and postoperative photographs. A retrospective chart review captured the results described below.

Results: Four of these patients had severe retrocoronal transverse bilateral temporoparietal band-like constrictions. Four more patients were affected moderately. These patients all had a worsening of their anterior cranial vault deformity including enhanced bitemporal narrowing and increased frontal bossing. Of the 4 severe deformities, 3 underwent a subtotal cranial vault remodeling at a mean age of 9 months (range, 8-12 months). These 3 patients had overcorrection of the transverse expansion intraoperatively as well as subtotal resection of the greater wing of the sphenoid bone. The fourth patient underwent strip craniectomy at 3 months of age with widened lateral wedge ostectomies taken anteriorly, followed by helmeting. Mean followup was 23 months (range, 13-37 months). All 4 had excellent results with 2 residual mild bitemporal constrictions. The 4 moderate deformities all did well with 2 treated with cranial vault remodeling both at 8 months of age, and 2 treated with strip craniectomy and postoperative helmeting at a mean age of 2.5 months. One of these patients had mild frontal bossing and mild bitemporal constriction at 16 months postoperatively. There were no perioperative complications in these 8 patients.

Conclusions: Sagittal craniosynostosis patients with a significant retrocoronal transverse temporoparietal transverse band-like constriction have a worsened anterior dysmorphology. Principles of operative correction should include overexpansion transversely and takedown of the greater wing of the sphenoid bone bilaterally when done as an open procedure. Surgical follow-up of almost 2 years in the more severe patients demonstrates satisfactory outcomes. When a strip craniectomy with bilateral wedge ostectomies and postoperative helmeting is performed, the width of the anterior wedge ostectomy should be increased.

### 207. Incidental Diagnosis of Craniosynostosis at the ER: Case Series of 331 Patients

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Background/Purpose: Craniosynostosis (CS) is usually diagnosed in infancy based on characteristic changes in cranial shape. Latent forms of craniosynostosis, often presenting with little or no changes in the cranial form, have been described. The lack of clinical findings makes detection nearly impossible, and many cases are identified incidentally when imaging of the head is performed for another reason. We sought to quantify the prevalence of incidentally identified craniosynostosis in an otherwise healthy, normal general pediatric population seen in the emergency department for reasons other than head shape.

Methods/Description: Three hundred thirty-one computerized tomography (CT) scans from children between 1 and 5 years old (mean age  $2.4 \pm 1.3$  years) were retrospectively reviewed. These images were taken at the emergency department from June 2005 to October 2016. Reasons for the imaging included: trauma, seizures, and headaches, among others. Patients with a past medical history of shunt placement and CS associated syndromes were excluded. CS was diagnosed via review of CT scans by either 3 plastic and reconstructive surgeons or 1 neurosurgeon. Suture fusion, radiology report, the reason for CT scan and past medical history was recorded as covariates. Cranial shape analysis was performed on the patients with premature suture fusion.

Results: A total of 11 (3.3%) patients were found to have CS. Of these, 18.1% were reported by the radiologist. All the fused sutures were found to be sagittal. Indications for CT in patients who were found to have CS were trauma (63.6%), followed by seizures (18.1%), headaches (9.1%), and dog bite (9.1%). Most of the patients had no significant past medical history. Cranial shape analysis was performed in 5 of these patients. The cranial shape in all patients was grossly normal, but the shape analysis found that 4 had subtle phenotypic changes along the spectrum of scaphocephaly. The remaining case was deemed normal by shape analysis.

Conclusions: Craniosynostosis without the characteristic phenotypic changes was present in 3.3% of normal healthy children 5 years old and younger. This finding was noted by the radiologist in only 2 patients. The clinical significance of this finding is unclear but these patients may require careful clinical monitoring for elevated ICP and neurocognitive manifestations. We identified subtle phenotypic changes that can help identify these patients, but this requires sophisticated shape analysis algorithms that are not widely available. We have adapted our shape analysis algorithms for use with 3-dimensional photogrammetry, and this may provide a radiation-free method of detection in future.

## 208. Incidence and Perioperative Management of Von Willebrand Disease in Craniosynostosis Operative Cases

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Background/Purpose: Von Willebrand disease (VWD) is one of the most common inherited bleeding disorders. The disorder is autosomal dominant and occurs due to an absent or defective von Willebrand factor (VWF), an adhesive glycoprotein. VWF binds to platelets and sites of vascular injury to aid in platelet adhesion and also assists in fibrin formation through the reduction in factor VIII inactivation. VWD often presents as excessive bleeding after trauma or surgery. In order to minimize bleeding and reduce transfusion requirements during highly invasive surgical procedures, such as craniosynostosis repair, preoperative identification of these patients can be helpful.

Methods/Description: Prior to undergoing operative repair of craniosynostosis, patients were screened for von Willebrand disease as a part of the pediatric blood management protocol. The laboratory screening tests used were von Willebrand factor antigen, ristocetin cofactor, and von Willebrand factor multimers. Those who tested positive for von Willebrand disease were evaluated by a hematologist and the pediatric blood management team. These patients received Humate-P immediately preoperatively and tranexamic acid intraoperatively. Postoperatively, Amicar was given and ristocetin cofactor levels were checked on postoperative day 1 and 2. Humate-P was given for low ristocetin cofactor levels.

Results: Of the 100 patients screened from 2014 to the present, 14 patients tested positive for von Willebrand disease. At 14%, this is significantly higher than the up to 1% prevalence in the general population. All of these patients were found to be type 1, the mildest and most common form of the disorder.

Conclusions: Identification of patients with von Willebrand disease prior to cranial vault surgery allows for appropriate preoperative, intraoperative, and postoperative measures to be taken to minimize blood loss. By following our team protocol, these patients did not require an increased transfusion rate. Given the high prevalence of VWD found in this group of patients with craniosynostosis, further studies are needed to investigate a potential genetic link between von Willebrand disease and craniosynostosis.

## 209. Is Postoperative Intensive Care Unit Monitoring Necessary Following Spring-Mediated Cranioplasty for Sagittal Synostosis?

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Background/Purpose: In recent years, spring-mediated cranioplasty (SMC) has emerged as an effective and safe technique to treat patients with sagittal craniosynostosis. Despite this, SMC patients are routinely cared for in the pediatric intensive care unit (PICU) postoperatively due to beliefs that these patients are at high risk of neurologic and hemodynamic deterioration. This study aims to determine whether PICU care is truly indicated for SMC patients.

Methods/Description: A retrospective chart review of all patients treated with SMC or cranial vault reconstruction (CVR) for sagittal craniosynostosis from 2011 to 2018 was conducted. Postoperative outcomes were recorded, with a focus on complications that would necessitate PICU-level care, such as hemodynamic instability or altered neurological status, in addition to hospital stay information, such as length of stay and total time spent in the PICU. Standard descriptive statistics were used.

Results: A total of 65 patients underwent SMC, while 41 patients underwent CVR during the study period. In the postoperative period, 2 SMC patients (n = 65, 3.1%) and 2 CVR patients (n = 41, 4.9%) showed signs of hemodynamic instability. Zero patients in the SMC group showed signs of altered mental status, compared to 2 patients in the CVR group (n = 41, 4.9%). The most common complication was postoperative anemia, 6 patients (n = 65, 9.2%) in the SMC group requiring transfusions, compared to 7 CVR patients (n = 41, 17%).

Conclusions: Both SMC and CVR are safe procedures to treat sagittal craniosynostosis, and most patients do not require PICU care in the postoperative period. Postoperative anemia is the most common complication but is generally detected with routine blood work and can be well-managed outside of an intensive care setting.

## 210. A Unique Small Group Approach for Speech Development for Children Before and After Palate Repair

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Background/Purpose: Parents are introduced to good and bad speech habits at the time the cleft palate is identified. Information is provided in writing and a family-oriented book is recommended and sometimes provided. There is written communication with the family and early intervention (EI) provider via group e-mail when EI is in place. And yet at the time of preoperative appointment, some families are still confused about which sounds to focus on presurgery and why. With the support of myFace.org and a grant from the New York Community Trust, we were able to run a 2part program for families before and after their child's cleft palate repair. A 2-part speech workshop series in July 2019 and February 2020 is designed provide speech and sound stimulation techniques to 6 families and children younger than 13 months who were born with a cleft palate or cleft lip and palate. This provided support to those families possibly struggling with understand what was required of them and assistance to those families that have yet to qualify for EI services.

Methods/Description: The first workshop focused on preventative speech and sound stimulation for children and families presurgery; while the second workshop served as a follow-up with the same group postsurgery to evaluate the progress made. This workshop series provides a novel approach to teaching parents about speech and sound stimulation by bringing families together in a community setting and creating a sense of empowerment. Families going through nasoalveolar molding have a chance to connect with other families, however for parents' of children with cleft palate only, there are fewer opportunities to meet other families. This allows for an intimate small group and a supportive environment to learn in. This program is easy to duplicate requiring a quite comfortable room with AV equipment, yoga mats, light snacks, and if you choose written information or books to share with families.

## 211. Adaptive Early Intervention for Children with Cleft Palate: When to Focus on Speech Sound Production?

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Background/Purpose: Young children with repaired cleft palate are a diverse clinical population, who vary in their acquisition of speech and vocabulary skills during early development following palatoplasty. The purpose of this study was to examine an early intervention database for participant characteristics associated with changes in speech and vocabulary growth of children with repaired cleft palate who received an Enhanced Milieu Teaching Plus Phonological Emphasis (EMT + PE) (Scherer & Kaiser, 2010) intervention compared to children in a "business-as-usual" (BAU) control condition who were receiving routine interventions and follow-up in the community.

Methods/Description: Participants: Twenty-nine children with repaired cleft palate were randomly assigned (stratified by gender and age) at initial screening to a business-as-usual (BAU) condition or to the EMT + PE intervention condition. Participants were between the ages of 15 and 36 months, received their primary palatoplasty prior to 12 months of age, and had cognitive scale composite scores of 80 or greater on the Bayley Scales of Infant and Toddler Development-III. Measures and Assessments: The language and speech skills of the children were evaluated at 3 time points: (a) preintervention, (b) immediately postintervention, and (c) follow-up 3 months after postintervention. Assessments included: (1) A parent-completed form describing the child's medical, developmental, and family history. (2) The Profiles of Early Expressive Phonological Skills (PEEPS: Stoel-Gammon & Williams, 2013), consonant inventory, percent consonant accuracy (PCC) for total words and by manner. (3) Preschool Language Scale (PLS-4), (4) Total number of words (TNW) and number of different words (NDW) derived from the automated Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2008). (5) MacArthur Communicative Development Inventories (MCDI: Fenson et al, 2007) intervention procedure in the Enhanced Milieu Teaching (EMT; Kaiser et al., 1998) plus Phonological Emphasis (EMT + PE) condition a trained SLP used naturalistic intervention strategies to facilitate vocabulary and speech development (Scherer & Kaiser, 2010). Participants in the EMT + PE group received individualized therapy during 30-minute clinic sessions twice weekly for 6 months (48 sessions).

Results: Preintervention PLS-4 scores, PCC, and words per minute (WPM) showed large effect sizes (ie,  $r^2 > 0.68$ ) MCDI raw score and NDW had moderate effect sizes ( $r^2$  between 0.60 and 0.65) were correlated with better outcomes postintervention. Given the importance of rate of word use as demonstrated by WPM's high correlation with intervention outcome, we examined the descriptive statistics for WPM in the whole sample (M = 6.81, SD = 5.94, range = [0-17.10], median = 6.10). We split the treatment and controls samples at 10 WPM and completed a series of post hoc analyses comparing the changes from pre- to postintervention for place and manner.

### 212. The Development of Stop Consonants in Children with Cleft Palate from 9 to 39 Months of Age

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Background/Purpose: Children with cleft palate  $\pm$  lip (CP  $\pm$  L) exhibit difficulty with the production of oral stops prior to primary palate repair. The absence of stop consonants post palate repair is an important indicator of the need for early intervention and/or additional surgical management for velopharyngeal dysfunction. Although we expect that stop production may be delayed for young children with CP  $\pm$  L, little data are available about either the age or course of emergence of these sounds in children with cleft palate compared to noncleft children (NC) of the same age. The purpose of this study was to compare the acquisition of stop consonants in children with CP  $\pm$  L and NC children of the same age. Comparisons will be made between 2 groups of children with CP  $\pm$  L; those with suspected/confirmed velopharyngeal dysfunction (VPD) and those with adequate velopharyngeal function (AVPF).

Methods/Description: Participants included 72 children (49 children with CP  $\pm$  L and 23 noncleft (NC) children. No children with syndromes, sensorineural hearing loss, or cognitive impairments were included in the study. The children's spontaneous speech samples were collected and phonetically transcribed at 9, 13, 21, 27, 33, and 39 months of age. The samples were analyzed using the Logical International Phonetics Programs (LIPP). Descriptive statistics were used to characterize the emergence of sounds in the child's phonetic inventory (produced at least 2 times) and stable usage (70% correct production) of oral stops. Multilevel modeling for repeated measures (ie, multilevel growth modeling) will be used to compare the 3 groups of children across time.

Results: The results indicated that voiced stops /b, d, & g/ occurred in the consonant inventories of 50% of the NC participants by 9 months but were not present in the inventories of 50% of the children with CP  $\pm$  L until 17 months of age. The voiceless stops emerged later, with /t & k/ appearing at 13 months and /p/ at 17 months for the NC children. These sounds were not seen in the inventories of 50% of children with CP  $\pm$  L until 21 months of age. Stable consonant production (70% correct usage) also differed between the 2 groups of children. The children with CP  $\pm$  L showed stable usage 3 to 6 months later than the NC children, with /b/ showing stable production at 27 months, /d/ showing stable production at 33 months, /b & k/ and /p & g/ at 39 months of age. The NC children showed 70% correct usage for /b/ at 21 months, /p, d, & k/ at 27 months, and /g/ at 33 months. Finally, children with suspected/confirmed VPD showed later emergence and lower percent correct usage for all stops.

*Conclusions*: Production of stops was delayed for children with CP  $\pm$  L regardless of velopharyngeal status. The findings from the multilevel modeling analysis will enable us to better understand the group growth trajectories and catch-up of children with CP  $\pm$  L. This information has implications for clinical management and goal selection.

## 213. The Effect of Early Intervention Supplemented with Quantitative Language Feedback for Children with Cleft Palate

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Background/Purpose: The literature supports the effectiveness of parent-implemented early interventions on positive changes in the speech and language development of children with cleft palate (CP). The key to effective parent-implemented interventions is whether parents can use language stimulation skills consistently and appropriately when interacting with their children in their daily lives. It is essential to encourage and monitor parents to use language-stimulation skills for maximum gains of intervention. The Language Environment Analysis (LENA) enables clinicians to monitor parents' language

interactions with their children using a full-day recording. The LENA measures could serve as a type of biofeedback, enabling parents to focus on improving their linguistic interactions, and maintaining their provision of intervention. This study aims to explore the effects of training parents to administer focused stimulation interventions and providing quantitative language feedback using the LENA on the speech and language performance of children with CP.

Methods/Description: Ten children with CP and their parents participated in the study. Standardized and informal assessments were performed at the initiation of the study, while the following intervention occurred 3 months after the pretest and the follow-up test occurred 6 months after the intervention. Parent trainings were provided through 5 sessions over a 3-month period. Parents mainly learned focused stimulation skills to facilitate the use of words that contained stop consonants. After each training session, parents were asked to complete LENA recordings at home. Quantitative measures from the LENA automated analysis of each recording were used as language feedback reviewed with parents at the subsequent training session. This study also included another 10 children with CP from a broader longitudinal study. They did not receive the intervention and were included to compare the effects of the intervention against the course of natural development in children with CP over the same period. Children's language and speech measures from pre- and postintervention tests were compared between the groups.

Results: Seven children with CP have completed to participate in the entire sessions including intervention and the postintervention assessment at present. The preliminary results indicate that children with CP in the intervention group show a significantly greater extent of change in expressive vocabulary size and consonant inventory. We will track speech and language performance in children with CP who receive the intervention to determine the long-term effect of the intervention with quantitative language feedback using the LENA. That is, we will discern whether the impact of the intervention will move the children with CP to the normal developmental trajectory for speech and language performance.

Conclusions: This study addresses the benefits of the parentimplemented intervention supplemented with quantitative language feedback.

### 214. Outcome of ACPA Visiting Scholar Program in Mongolia

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Background/Purpose: In 2007, the School of Dentistry, Health Sciences University of Mongolia (present Mongolian National University of Medical Sciences) with the support of the Japanese Cleft Palate Foundation, established the first Speech Treatment Clinic for children with clefts, pioneering the development of speech pathology nationwide. Absence of speech therapist's training in Mongolia challenged and pushed us to search for a network and possibilities to cooperate at international level.

Methods/Description: In 2009, enrollment in the ACPA Visiting Scholar program became kickoff point for the future development of clinical speech therapy in Mongolia. Based on the experience within the ACPA Visiting Scholar, a multidisciplinary team was founded at the National Center for Maternal and Child Health

(NCMC). Within the process several key baseline activities was implemented, including establishment of a full-time position for a speech therapist for children with clefts at the NCMC. Also, due to existing need, since August 2009, our team were involved in cochlear implantation postsurgery rehabilitation and successfully started audio verbal therapy (AVT) for the very first congenitally deaf child with cochlear implantation in Mongolia by laying foundation for future progress of AVT nationwide. Cross-sectional retrospective study was done based on hospital data of a total of 301 patients to summon the outcome of the Visiting Scholar Program in Mongolia.

Results: Of total 301 patients, 203 (67.8%) gone through speech treatment for children with clefts and 98 (32.2%) received AVT after cochlear implantation. Speech treatment rendered to 65.8% of children, diagnosed with hypernasality. Of all participants, 77.0% had gone surgery on restoration of congenital cleft lip and/or palate. Of total 98 cochlear implanted hearing loss cases, 36 (36.7%) showed congenital etiology and 62 (63.3%) showed adventitious etiology. The causes of adventitious etiology hearing loss were ototoxic, 20 (32.3%) and meningitis, 18 (29.0%). From total 98 patients, 41 (42.0%) visited the audioverbal therapy sessions.

Conclusions: Our aim is to develop and strengthen an evidence-based speech pathology in Mongolia intended to carry out preliminary and population-based studies beside clinical practice in the future. Even though in the past 12 years tangible progress was made in the area of speech therapy for disabled Mongolian children, comprehensive studies and assessments are still limited. Future building of human capacity, strategic planning at the governmental level, joint research projects, publication, and distribution of study results are still in priority. Follow-up activities, continuing cooperation, training, and active practical networking for previously selected visiting scholars are needed. Future consideration on this matter is suggested for ACPA management team as a next level to strengthen already existing valuable platform—Visiting Scholar program.

## 215. Is playing a Woodwind or Brass Instrument Associated with Development of Velopharyngeal Insufficiency in Patients with Cleft Palate?

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Background/Purpose: Velopharyngeal insufficiency (VPI) occurs in 5% to 36% of patients following primary cleft palate repair and is defined as the inability to fully seal the velopharyngeal sphincter. Since most sounds in the English language require complete closure of the velopharyngeal sphincter, surgical treatment is often required for full rehabilitation. Stress velopharyngeal insufficiency (SVPI) occurs when high intraoral pressure prevents the soft palate from sealing and is reported in 7% to 34% of collegiate brass and woodwind musicians. Although normal speech seldom causes intraoral pressures to exceed 5 to 6 mm Hg, playing a woodwind or brass instrument can cause pressures to surpass 130 mm Hg. This study investigated the impact of woodwind and brass instrument use on VPI in patients with a history of cleft palate.

Methods/Description: Patients aged 8 to 24 years with a history of a cleft palate reconstruction and woodwind or brass instrument playing were recruited from a craniofacial center. Participating patients and families were administered a questionnaire pertaining to their musical instrument usage and speech competence. A subsequent retrospective

chart review utilizing the electronic medical records system was then performed.

Results: Participants ranged in age from 10 to 17 years with an average age of approximately 10 years. On average, the patients had played for about 2 years and practiced less than 2 hours per day and 5 hours per week. Approximately 75% of participants had a history of speech pathologies requiring speech therapy with the average time spent in therapy being slightly less than 4 years. None of the participants noticed new voice changes such as hypernasality or nasal air escape after playing their instrument.

Conclusions: Low time commitment (<2 h/d) woodwind or brass instrument playing does not appear to put patients with a history of a repaired cleft palate at greater risk of developing VPI. Therefore, school-aged participation in these instruments should not be restricted unless the patient becomes symptomatic.

### 216. Do Cleft Related Speech Outcomes Differ According to Payer Status?

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Background/Purpose: At our institution, patients with cleft palate are referred to speech-language therapy through a standardized preoperative and postoperative protocol with the goal of obtaining excellent speech outcomes for all children. Previous literature has demonstrated that race and socioeconomic factors, such as payer status, may influence outcomes in surgical care (such as trauma, pediatric surgery, gynecology, and others). This has been hypothesized to be due to implicit bias within the health-care system and/or lack of resources for these marginalized groups to navigate the system. Our hypothesis is that patients with cleft who have greater financial resources (using private health insurance as a proxy) may have improved speech outcomes. The purpose of this study is to examine whether speech outcomes vary with payer mix.

Methods/Description: This study consists of a retrospective review of all patients with cleft lip  $\pm$  cleft palate who were referred for NAM treatment prior to primary cleft lip repair at our multidisciplinary Craniofacial clinic from 2008 to 2018. To evaluate the role of socioeconomic status on speech outcomes, insurance payer data were collected as a proxy for socioeconomic status and classified as private versus public payer. Any patient who did not receive a 3- or 5-year-old speech evaluation at our institution was excluded from the study. All speech therapy visits were conducted by 1 of 3 speech-language pathologists on staff at our clinic. Specific speech outcomes were extracted and compared between the 2 cohorts. Statistical analysis was conducted in Microsoft Excel utilizing Student t test.

Results: A total of 215 patients were referred for NAM and were at least 3 years of age at time of data collection; 142 (66%) had speech data at 3- or 5-years of age; 60% of patients with a 3-year visit were public payers with 40% private. For those with a 5-year visit, 53% were public payers with 47% private. Private payers had greater intelligibility than public payers at ages 3 (68% vs 59%, P=.04) and 5 (87% vs 78%, P=.01). Public payers had greater need for speech therapy at age 3 (75% vs 57%, P=.04) and less nonpressure air sounds at age 5 (5.68 vs 5.95, P=.01) when compared with private payers. At the 5-year visit, both public payers who completed NAM (78% vs 87%, P=0.01) and did not complete NAM (80% vs 90%, P=0.04) had less intelligibility compared to private payers.

Conclusions: Our study found statistically significant differences in speech outcomes between payer status at 3- and 5-year speech evaluations in cleft lip/palate patients. Although public payers comprise the majority of our patient population, they demonstrate less percent intelligibility at ages 3 and 5 with fewer non-air pressure sounds at age 5 when compared with their private payer counterparts. These results implore us to further examine the cause of this difference (bias, access, education, coverage options) and improved awareness among medical staff that socioeconomic variables may influence speech outcomes.

#### 217. Early Outcomes of Speech Therapy by a Short-Term Trained Cleft Care Provider

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Background/Purpose: Cleft of the palate is a congenital anomaly that presents as a structural defect which may or may not involve the lip and the alveolus. An infant with this condition is first challenged by feeding and subsequently articulation of speech. If left unrepaired the infant may adapt to the feeding challenges over time but the speech challenges remain and often may not resolve following surgical intervention thus necessitating speech therapy. Speech and language pathologists with training in cleft speech pathology are scarce in Africa; therefore, the speech needs of individuals with cleft palate who have had palatal repairs have been left unmet until the recent initiative of Smile Train to provide short-term training in cleft speech pathology to cleft caregivers. This report is to describe the initial findings of a beneficiary of the Smile Train training as well as add to the sparse information on speech errors and outcome of speech therapy from the developing world.

Methods/Description: Patients who had palatal repairs between 2010 and 2019 were contacted and informed of the opportunity to have a free speech assessment. Consenting patients were assessed individually for speech errors based on conversational speech (speech intelligibility as described by Henningsson), their ability to count 1 to 10 and make the high pressure sounds P, B, T, D, K, G, F, and S clearly and consistently. A maximum score of 18 points was assigned when speech was without errors. The frequencies of articulation errors such as glottal stops, pharyngeal stops, midpalatal stops, pharyngeal fricative, and nasal emissions on each high-pressure sound were also assessed. The speech assessments were done individually by 2 cleft caregivers who participated in the short-term training by Smile Train.

Results: Seventy patients had palatal repairs done at our center within the 10-year period. Twenty-three (32.9%) consented to have a speech assessment and possible speech therapy. Seven (30.4%) patients were found to have normal speech and not needing speech therapy. Sixteen patients commenced speech therapy with a baseline mean score of 4.8 (SD  $\pm$  3.6). The frequency of the types of speech intelligibility was 12.5% for mild, 50.0% for moderate, and 37.5% for severe. The P sound was the least impaired in 25.0% of the cases, and nasal emission was the most frequent error occurring in 68.8%. Eight of the 16 patients attended the sessions regularly, and 2 patients of these have completed their therapy both with a score of 18 points each and normal speech intelligibility.

Conclusion: The early outcomes of the Smile Train initiative speech intervention in our environment are very encouraging with much gratitude from our patients who would have otherwise been underserved.

### 218. A Team in Flux: Navigating Changes in Cleft Team Members as Patients Keep Coming

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Background/Purpose: Craniofacial teams are dynamic in many different ways. In our Craniofacial team, the number of patients seen has increased over the years. The number of providers has not increased proportionately, and at times has decreased due to vacancies or leaves of absence. During a 2-year period, 41% of team providers took an extended leave or vacated their position, and 7 new providers were trained. Our team has been forced to make adjustments and compromises to meet patient demands without sacrificing the quality of care.

Methods/Description: The Craniofacial team coordinators (nurse practitioners) identified which patients needed to see which providers in order to meet patients' needs while avoiding overwhelming different disciplines and creating excessive wait times. Each discipline was given the opportunity to review their list of prospective patients and encouraged to edit their list as needed. Patients and families were offered individual appointments with providers not seen to bridge the gap until their next team visit. Certain identified problems were managed by phone when appropriate. Patients and families were notified in advance that providers would be absent and given the option to reschedule if preferable.

# 219. Helping Families Face an in Utero Diagnosis: A Panel Discussion on Prenatal Consultations for Families Expecting a Child with a Craniofacial Difference

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Background/Purpose: Prenatal ultrasound has made in utero diagnosis of congenital anomalies a routine occurrence; however, for the families receiving the news, it is often devastating and stressful. One of the key functions of a multidisciplinary cleft and craniofacial team is to provide prenatal educational counseling to the expectant parents, to identify psychosocial barriers and needs as early as possible, and to ensure a smooth and easy entry into clinical care after the baby arrives. There are no established guidelines or "best practices" for prenatal consultations. In this talk, multidisciplinary members of a midsize cleft and craniofacial center provide pearls and pitfalls related to the prenatal consultation.

Methods/Description: In this 20-minute panel discussion, we will provide the perspectives of the team coordinator, social worker, speech-language pathologist, and surgical team (surgeon, craniofacial orthodontist, and nurse practitioner). The panel will provide a brief overview and history of prenatal consults and discuss why our team came to prefer a specific order for the parents to see providers. We highlight common concerns and stress points experienced by parents: for example, how to discuss diagnosis with friends/family, what feelings parents may have in the delivery room, role of nasoalveolar molding, timing of operations, cost of care, and worries about their child's future. We conclude with tips on ensuring close follow-up and a smooth transition to the postnatal clinic evaluation with the team. A video will be presented to provide family perspectives on the utility of the prenatal consultation in preparing them to meet their babies.

### 220. Nursing Care Saves Lives: Providing Quality Nursing Care for Patients with Clefts

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Background/Purpose: International cleft lip and palate repair surgeries often occur in settings where nurses have low professional status and inadequate training. However, skilled nursing supports optimal postoperative outcomes following cleft surgery. Although postoperative sentinel events occur at very low rates for patients with clefts, they are most frequently respiratory in origin and most likely to occur when nurses are the primary care givers. The provision of adequate postoperative monitoring and nurse-initiated interventions should complications arise is imperative. Recognizing that in resource-limited settings, nurses may lack the abilities and confidence to provide optimal care following cleft surgery, the international cleft charity Smile Train developed a training program to teach nurses the necessary skills to anticipate, recognize, intervene in, and reduce postoperative complications. Knowing that empowerment and high morale are drivers for successful nurse-led intervention, another goal of this program is to improve the (self-) perception of nursing capability. This presentation will outline nursing-related problems faced in postoperative cleft care in resource limited settings and present a program aimed to address these challenges.

Methods/Description: In 2011, Smile Train launched a 3-day, classroom-based program: "Nursing Care Saves Lives." This presentation will highlight the program's curriculum, which involves lectures, skill-building exercises, simulation, and group problem-solving centered around 4 pillars: patient assessment, postoperative nursing care, postoperative complications, and postoperative nursing interventions. Particular focus is given to early recognition of postoperative complications and essential nurse-initiated actions. Pre- and postprogram tests are administered to identify knowledge gaps and assess improvement. Participating nurses are expected to develop individualized action plans to improve nursing care after the course. Additionally, nurses are instructed how to disseminate their new knowledge among their colleagues on return to their home hospitals. Since its inception, the Nursing Care Saves Lives course has been taught in 24 countries and directly trained over 1500 nurses. Nurses have consistently reported increases in knowledge and skills following the course. Through these improvements, this course has the potential to not only improve the postoperative care patients receive but also to increase nurses' confidence and normalize nurse-led intervention in countries with historic stigmatization of the nurse's role. Further measures of program success might include improvements in (1) the level of postoperative monitoring, (2) the number of nurse-initiated interventions, and (3) the rate of preventable respiratory complications following cleft surgery.

## 221. Qualitative Analysis of Patient and Parent Orthognathic Surgery Experiences: Implications for Team Providers

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(1) Children's Hospital of Los Angeles, CA

Background/Purpose: Orthognathic surgery can be a difficult procedure for patients with craniofacial diagnoses and their families, especially as patients are often at critical developmental transition to young adulthood. Challenges during the postoperative recovery period include weight loss, adjustment to diet restrictions, weight loss, absence from work/school, and restricted that can lead to mood and

behavioral symptoms. A more thorough understanding of patient and parent experiences can help better inform the health-care team in how to improve comprehensive pre- and postoperative education, strengthen coping skills, and support families. The purpose of this presentation is to describe a qualitative analysis of patient and family experiences and outline implications for care across specialties.

Methods/Description: Six structured focus groups (average 65 minutes) were held with 12 patients ages 17 to 23 (M = 19.5, SD = 1.8) and their mothers (n = 10) and fathers (n = 3), who were 41 to 60 years old (M = 50.3, SD = 5.6). Diagnoses included cleft lip and palate (50%), class II or III malocclusion (33%), Treacher Collins syndrome (8%), and muscular dystrophy (8%). Surgeries completed were LeFort I (42%), LeFort I and BSSO (42%), and BSSO (16%). Half of the participants were female and their ethnicities were 50\% Latino, 25% European American, 18% African American, and 8% Asian American. Household languages were 50% English, 42% Spanish, and 8% bilingual. Participant families represented the range of Hollingshead socioeconomic status (SES) categories: I (highest) 33%, II 17%, III 8%, IV 25%, and V (lowest) 17%. Focus groups were audio recorded, transcribed, and translated. Thematic analysis was completed to generate suggested intervention strategies. Themes included benefits of family anticipatory guidance, coping, social support, pain management, and adapted postoperative dental hygiene. They described concerns about nutrition, weight loss, sleep, breathing, logistical stressors, family impact, and missed school/employment. Based on participant and family experiences, strategies for team providers are summarized for presurgical preparation, management during admission, and postoperative monitoring and support. Qualitative analysis of patient experiences can serve to inform clinical care delivery and ensure alignment with family-centered care practices.

Learning Objectives: Each learner will identify 5 stressors associated with orthognathic surgery. Each learner will list 5 strategies to support patient and family positive postoperative adjustment.

#### 222. Utilizing a Nursing and Physical Therapy Interdisciplinary Clinic Model for the Evaluation Plagiocephaly and Torticollis

Bethany Sloane (1), Lisa Crupi (1)

(1) Oregon Health and Science University, Portland, OR

Background/Purpose: The Craniofacial disorders (CFD) clinic at Oregon Health & Science Universities' Child Development and Rehabilitation Center (CDRC) has been seeing children with deformational plagiocephaly (DP) and congenital muscular torticollis (CMT) for the past 25 years by our nurse practitioner team. With the successful implementation of the "Back to Sleep" campaign, the prevalence of DP has increased from 5% in the 1990s to 20% to 30% presently. Clinically, DP is mostly seen as a cosmetic condition; however, there has been research that shows that these children may be at risk for developmental delay and require further screening by clinicians. The incidence of CMT ranges from 0.3% to 2% of newborns but has been reported as high as 16%. The incidence of DP ranges from 18% to 19.7%; DP is strongly associated with CMT, as high as 70% to 95%. Over the past 5 years, a physical therapist was added to the team due the high incidence of CMT and DP occurring together. Early referral to physical therapy can lead to prevention of secondary sequelae, reduce treatment duration, and avoid more invasive interventions, which in turn proves to be most cost effective. If CMT is left untreated in rare cases, it can lead to botox injections and/or surgery for correction of movement limitations and consequent facial asymmetries in older children.

Methods/Description: During this session, we plan to discuss our interdisciplinary clinic model and specific evaluation components. Further we plan to explain the observed benefits of the interdisciplinary team approach such as decreasing health-care costs, reducing needing intervention, and providing a more comprehensive evaluation. Over the past year, our clinic began to collect data on the patients who attend this clinic by a creation of repository and utilizing our electronic medical record system. The repository has helped our clinic team gather data in order to show trends, analyze outcomes, improve quality of care, show overall impact of our clinic program, and better match children with plagiocephaly and/or torticollis to appropriate treatment protocols. These data will in turn improve continuity of care and documentation of children with torticollis and/or plagiocephaly within our practice setting at OHSU and can be carried forward to multiple settings. During this presentation, we plan to share clinic outcomes utilized, how we perform clinical research, and summarize our current findings. Further during this presentation, we plan to demonstrate the effectiveness, impact, and outline the steps we took to implement a statewide prevention plan to educate families and providers. The amount of intervention and the time it takes to resolve CMT and plagiocephaly is dependent on the severity and when the infant is first diagnosed and referred for treatment. Research shows that when infants are identified early (3-6 months) has required shorter treatment durations and less invasive interventions as compared to those who are referred after 3 to 6 months.

### 223. Practice Pearls in Implementation of Nurse Practitioner Lead Nasoalveolar Molding

Bethany Giacobbe (1), Carolyn Walborn (1), Katherine Fautsch (1), Christian Vercler (1), Steven Kasten (1), Steven Buchman (1)

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Background/Purpose: Nasoalveolar molding (NAM) can be a challenge for both patient and families as well as the practitioners providing NAM. Utilizing nurse practitioners is a great way to implement NAM at any institution. Our Nurse Practitioner lead NAM with the assistance of a dental assistant has made NAM an option for our patient population and assists our surgeons with cleft lip and palate repairs. Implementing new treatment options always has a learning curve for the providers. Several lessons were learned during the implantation of NAM. The lessons learned include setting up a NAM program and working with families to make NAM successful.

Methods/Description: This presentation with review lessons learned over the past 5 years as our institution implemented Nurse Practitioner lead NAM through past cases and experiences. Discussion topic include an overview of implementing Nurse Practitioner lead NAM team model, reviewing guidelines set up to make NAM successful and challenges along the way.

### 224. It's Party Time! Tips for Planning an Event for Your Patients and Families

Helen Huff (1)

(1) Children's Mercy Kansas City, MO

Background/Purpose: Parenting children diagnosed with a craniofacial condition can sometimes feel overwhelming and isolating. Children born with a craniofacial condition often feel like they are the only ones dealing with looking or sounding different from others. Parties

bring children, families, and staff together in a fun atmosphere to play, make friends, celebrate, and support each other. The goal is to bring patients, families, and staff together to celebrate with an event that is low cost, simple to coordinate, and so much fun that families want to return year after year.

Methods/Description: The power point presentation provides an overview detailing the planning process used to organize a team and family picnic for 250 people. The following items to consider when planning your event will be discussed: vision, budget, date, location, activities, food, giveaways, obtaining hospital or center permissions, invitations/RSVP, and family response.

## 225. Anatomic and Surgical Predictors of Velopharyngeal Insufficiency Following Primary Cleft Palate Repair

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Background/Purpose: A primary goal of cleft palate repair is normal speech, and velopharyngeal insufficiency (VPI) is a significant outcome following cleft palate repair that affects normal speech development. The purpose of this study was to assess the variables associated with VPI as an outcome following cleft palate repair.

Methods/Description: Data were prospectively collected for all non-syndromic patients with a cleft palate undergoing primary cleft palate repair by a single surgeon between 2002 and 2016. The following outcomes were recorded: fistula rates, incidence of VPI, and the rates of secondary surgery for VPI after failing speech therapy. Outcomes were correlated with the following patient and surgical parameters: preoperative cleft and palate width, preoperative palate length, changes in palatal length, palate classification, and the type of surgical repair used.

Results: Two hundred forty-six patients (116 male and 130 female) were identified. Mean age at primary palate surgery was 11.9 months. Mean cleft width was 9.68 mm (0-18 mm) and mean palate width was 29.5 mm (20-41 mm). Cleft lip and palate was associated with greater mean cleft and palate width (P < .01, P < .01).01) compared to cleft palate only. Of the patients with both preoperative and postoperative palatal length measurements, 64 and 55 had Furlow and von Langenbeck repair, respectively. Average percentage of soft palate lengthening following surgery for the Furlow group was 65.3% and for von Langenbeck was 65.5% (P = .46). Postoperative fistula was observed in 4 (1.6%) patients, with 1 (0.4%) being symptomatic and needing surgical intervention. Of 169 patients who were old enough to be tested for VPI, 20 of these patients required secondary surgery for VPI (11.8%). Patients with VPI had significantly larger preoperative cleft widths compared to patients without VPI (P = .02), but preoperative palate length (P = .08), overall postoperative lengthening (P = .08).31), and incidence of postoperative fistula (P = .13) were not significant. Statistical tests included  $\chi^2$  tests and 1-tailed t tests with a significant P value defined as <.05.

Conclusions: In this study looking at a single surgeon's cohort of 246 patients, cleft palate repair resulted in 11.8% of patients requiring surgery for VPI. In these patients, VPI was associated with preoperative cleft width, but not with changes in palatal lengthening following surgery or with postoperative fistula.

### 226. Predicting Velopharyngeal Insufficiency in Patients with Cleft Palate

Brady Anderson (1), Kasra Fallah (1), Joseph Moffitt (1), John Teichgraeber (1), Kim-Loan Luu (2), Courtney Stout (2), Phuong Nguyen (1), Matthew Greives (1)

(1) McGovern Medical School at The University of Texas Health Science Center at Houston (UTHealth), TX, (2) Children's Memorial Hermann Hospital, Houston, TX

Background/Purpose: Velopharyngeal insufficiency (VPI) after primary palatoplasty has been associated with various patient and surgical factors, including cleft size, genetic conditions, and fistula formation. Despite this information, a predictive risk stratification tool has not been developed for factors associated with VPI development. Although VPI affects the speech of up to 50% of patients undergoing primary palatoplasty, diagnosis requires long-term follow-up after speech development. We reviewed over 15 years of primary cleft repairs to examine predictive factors for VPI.

*Methods/Description*: A retrospective review of patients who underwent primary palatoplasty from 1999 to 2014 was performed. Inclusion required follow-up past age 5 and speech production. Patient demographics, Veau class, medical history, surgical details, and follow-up information were collected. The primary outcome was VPI, defined as revision palatoplasty or recommendation for surgery by a speech-language pathologist. Genetic diagnosis was defined as positive genetic testing for a craniofacial syndrome. Univariate analysis was performed, and variables with a P < .20 were included in a multivariate regression analysis.

Results: Of 274 patients included, 158 (57%) were male. Median age at primary repair was 1 year (0.9-1.1) with a median age of 8.1 at last follow-up. One hundred four (38%) patients developed VPI at a median age of 4.9 years (3.8-6.5); 11\% of African-American patients developed VPI, compared to 39% of Hispanic patients and 45% of white non-Hispanic patients (P < .05). VPI was 65% in patients who developed posterior fistulae (Pittsburgh 1-4) compared to 13% in those without (P < .01). VPI was lower following Furlow (7\%, n = 14) than straight-line repairs (40%, n = 260; P < .05). VPI in patients with Pierre-Robin was higher (55\%, n = 38) than those without (35\%, n = 236; P < .05). Following a bidirectional stepwise selection for a linear model, factors remaining associated with VPI were African-American race (OR: 0.18, 0.04-0.66), posterior fistula (OR: 12.2, 6.6-23.6), and genetic diagnoses (OR: 3.2, 1.2-9.3). There were no differences associated with demographic factors, birth complications, or cardiac issues.

Conclusions: VPI following palatoplasty is a known complication. Development of a posterior palatal fistula was associated with increased odds of revision surgery, likely due to persistent nasal regurgitation refractory to speech therapy. Although limited in number, lower rates of VPI among patients receiving Furlow palatoplasty are promising for improved outcomes, warranting further investigation into follow-up and implementation rates. Lower rates of VPI in African-American patients and higher rates in patients with a genetic diagnosis may suggest a genetic component.

### 227. Modern Practice Patterns for Surgical Management of Velopharyngeal Insufficiency (VPI)

Thomas Sitzman (1), Erica Weidler (1), M'hamed Temkit (1) (1) Phoenix Children's Hospital, AZ

Background/Purpose: Numerous surgical procedures have been described for the treatment of velopharyngeal insufficiency (VPI).

Although pharyngeal flap and sphincter pharyngoplasty are the most established procedures, palatoplasty (including straight-line re-repair and Furlow techniques) and palatal lengthening are more recently proposed alternatives. We sought to characterize the current utilization of each VPI procedure in the US.

Methods/Description: A retrospective cohort study was performed using the MarketScan Commercial and Multi-State Medicaid databases for 2015 to 2017, which contain a nationally representative sample of health-care delivery in the US. Patients undergoing VPI surgery were included if they had a diagnosis of hypernasality or VPI and were 3 to 24 years old. Procedure type was determined by physician procedure billing.

Results: Information was available on 573 VPI surgeries performed between 2015 and 2017, representing approximately 10% of all VPI surgeries in the US during that period. Pharyngeal flap was the most frequently performed procedure, accounting for 40.3\% of all VPI surgeries. Sphincter pharyngoplasty was performed in 25.0% of surgeries. Palatal procedures were less frequent with palatoplasty performed in 14.5\% of surgeries and palatal lengthening in 5.4\%. Combinations of these procedures were performed in 14.8% of surgeries. The average age at surgery was 8.8 years (SD 4.5), but there were significant differences in age between the procedure types (P <.0001): patients undergoing palatoplasty were younger (7.7 years [SD: 4.3]) while those undergoing sphincter pharyngoplasty were older (9.6 years [SD: 4.9]). Procedure type did not vary significantly by year of surgery (P = .14), gender (P = .96), or for patients with a diagnosis of 22q11 deletion syndrome (P = .28). Among surgeons performing more than one VPI surgery during the observation period, 85\% performed more than one type of procedure and 65% performed palatoplasty and/or palatal lengthening in at least some patients.

Conclusions: Pharyngeal flap and sphincter pharyngoplasty are the most frequently used procedures for treating VPI in the US. Although palatoplasty has been broadly adopted in the United Kingdom, and multiple surgeons have promoted use of palatoplasty for treatment of VPI in patients with small velopharyngeal closure patterns or sagittal levator muscle orientation, utilization of palatoplasty is much lower than pharyngeal flap and sphincter pharyngoplasty. This may be due, in part, to an absence of studies directly comparing the effectiveness of these VPI surgical procedures.

### 228. Square Root Palatoplasty: A New Modification of Double Opposing (Furlow) Palatoplasty

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Background/Purpose: Double opposing palatoplasty has gained wide popularity for primary palatoplasty in the US since its first description by Dr Furlow in 1976. The advantages of Furlow palatoplasty are better repositioning of levator muscle, palatal lengthening, tighter nasopharyngeal sphincter, and decreased risk of longitudinal scar contracture. However, the desired gain in length is created at the expense of shortening in the transverse axis, and therefore, it is difficult to achieve tension-free closure in wide clefts. Senior author of this study developed a modification of Furlow palatoplasty which allows repositioning of palatal flaps with less tension and better vascularity. Our aim in this study is to introduce this new modification of Furlow repair and compare its surgical outcomes to straight-line repair with intravelar veloplasty.

Methods/Description: In this technique, an incision similar to square root shape is planned in the soft palate. Anteriorly based oral mucosal

flap, which forms the transverse axis of the square root sign, is almost 90° to the central limb along the cleft edges. This design allows better mobilization and vascularity of this flap and prevents accidental tearing of the oral mucosal flap toward its base. Posteriorly based oral myomucosal flap, which forms the smaller "V" of the square root sign, can be easily transposed with this design. The levator veli palatini sling is reconstructed under the operating microscope and a spacer (Alloderm) is placed between oral and mucosal layers. An IRB-approved retrospective review was performed of a prospectively collected database of all cleft palate patients who underwent palatoplasty by a single surgeon between 2010 and 2016. Exclusion criteria included loss of follow-up, syndromic diagnosis, or Robin sequence. Variables reviewed included demographic data, Veau classification, surgical technique, fistula, speech analysis, and need for secondary speech surgery.

Results: A total of 136 patients were operated in that time frame. Of those, 58 were excluded resulting in 78 patients for review. Thirty-two (41%) patients underwent straight line repair with intravelar veloplasty and 46 (59%) underwent modified Furlow repair. Distribution of sex, cleft type, and presence of associated syndromes were similar between the groups. Veau 3 clefts were the most common between the 2 groups. Between straight line and modified Furlow, fistula occurred in 31.25% and 8.7% (P=.016), of which 7 and 1 patient needed repairs, respectively. Between straight and modified Furlow, 56% and 85% had normal VP function (P < .0001). Of these, secondary speech surgery was required in 12 (37.5%) patients and 2 (4.35%) patients, respectively (P=.0002).

Conclusion: Modified Furlow double opposing z-plasty along with levator veli palatini retropositioning under operating microscope demonstrated superior perceptual speech outcomes than straight-line repair and intravelar veloplasty, with similar fistula rates.

## 229. Complications and Revisions After Secondary Speech Surgery-A National and Longitudinal Claims Analysis

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Background/Purpose: Although most patients attain normal speech after cleft palate repair and speech therapy, up to 20% require secondary speech surgery. Despite the frequency of these procedures, perioperative complications and rates of revision secondary speech surgery after all procedure types have never been reviewed using national, longitudinal data.

Methods/Description: We examined insurance claims from Clinformatics Data Mart (OptumInsight) between 2001 and 2017. Using CPT codes, cases were categorized as palatal-based procedures (revision palatoplasty, secondary lengthening, palatal island flap) or pharyngeal-based procedures (pharyngeal flap, dynamic sphincter pharyngoplasty; n = 694). Continuous enrollment from 180 days prior to 30 days after surgery was required for inclusion. Patients were excluded if they underwent surgery at less than 3 years without an accompanying speech diagnosis. Primary outcomes included 30-day complications and rates of revision secondary speech surgery. Multivariable logistic regression was used to evaluate the relationship between procedure type and complications.

Results: In this cohort, 67.7% underwent pharyngeal-based procedures, and 32.3% underwent palatal-based procedures. Perioperative complications occurred in 10.4% of patients and included respiratory complications (3.3%), bleeding (1.3%), dehiscence (3.8%), and

critical care episodes (2.6%). There was no difference in complications between procedure types (OR = 0.72, 95% CI, 0.43-1.23; P = .22), but patients with increasing comorbidities had significantly higher odds of complications (OR = 1.39, 95% CI, 1.12-1.73, P = .003). Interestingly, 12.5% of patients required a revision speech surgery. In patients with 3 years of postoperative enrollment, 19.7% required revision speech surgery.

Conclusions: Although complication rates were comparable to recent literature, rates of revision procedures were quite high, especially in patients with long-term follow-up. Persistent velopharyngeal dysfunction in one-fifth of patients despite secondary speech surgery demands further research directed toward quality improvement and reduction in patient morbidity.

## 230. Outcome Assessment of Facial Orthopedics with Taping for Cleft Lip Deformities Using 3D Stereophotogrammetry

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Background: Prior to surgical correction of a cleft lip deformity, facial taping is a type of presurgical orthopedics (PSO) that is used to improve soft tissue alignment and facilitate surgical repair. Dynacleft and 3M taping are 2 commercially available taping systems that can be utilized with or without nonalveolar molding. Determining how these tapes differ in their ability to manipulate soft tissue is useful in optimizing the care of cleft lip and/or palate patients.

*Purpose*: The purpose of this study was to evaluate 2 cleft lip taping devices, 3M and Dynacleft, for their ability to impact nasolabial shape and symmetry during the treatment of the cleft lip deformity.

Methods/Description: A retrospective cohort study of 70 CLP patients was conducted including patients who have received either Dynacleft or 3MTM facial taping as a component of their PSO treatments. 3D stereophotogrammetry images were obtained at 3 time points: prior to treatment, after receiving taping, and after surgical repair. 3D images were annotated with 31 landmarks on the nasal and upper lip region using 3DMD Vultus software. The landmarks were then used in conventional morphometric analysis with previously validated facial measurements (linear distances, ratios, and angles) to describe and compare the 2 cohorts at each stage. Geometric morphometrics using Procrustes ANOVA analysis was also conducted to compare the nasolabial asymmetry between the 2 taping groups.

Results: Both taping devices demonstrated progressive improvement in multiple facial metrics after taping and after surgery. Although variability in the degree of improvement was noted in the evaluated facial metrics after PSO between the 2 taping groups, no differences were found between these metrics after surgery. ANOVA comparison of the nasolabial region after Procrustes analysis also found no significant difference in facial shape between the 2 taping cohorts after surgical cleft lip repair (P = .80). However, nasolabial shape after PSO (P = .02) and after surgery (P = .02) was found to be differ depending if patients received formal PNAM treatment with their taping or taping alone. Conclusions: PSO with facial taping reduces the severity of facial deformity prior to surgical correction across multiple facial measurements. Both 3M and Dynacleft taping devices result in similar facial alignment postoperatively suggesting either can be successfully utilized. Given the significant cost differences between the 2 systems, our data may provide support for greater uptake of 3M taping for PSO.

#### 231. The Nasoalveolar Molding Cleft Protocol: Long-Term Results From Birth to Facial Maturity

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Background/Purpose: We present the first long-term outcomes analysis of the nasoalveolar molding (NAM) treatment protocol on patients with a cleft followed from birth to facial maturity.

Methods/Description: Single-institution retrospective review of all patients with a cleft who underwent NAM between the years 1990 and 2000. All study patients completed cleft care treatment at the same institution and were followed by the same team members. Our institution's treatment protocol offers NAM to patients with a significant cleft nasal deformity and/or widely displaced alveolar segments. All patients underwent primary cleft lip and nasal repair prior to the age of 6 months. Gingivoperiosteoplasty (GPP) is performed, when possible, at the time of lip repair. Cleft palate repair is performed by 1 year of age. Collected data include surgical and orthodontic outcomes of cleft care such as cleft lip and palate repair, GPP, alveolar bone grafting (ABG), speech surgery for velopharyngeal insufficiency (VPI), palatal fistula repairs, orthognathic surgery, and revision surgery to the nose and/or lip.

Results: A total of 135 patients met the inclusion criteria. Mean length of follow-up was 18.8 years. Eighty-nine patients presented with a unilateral cleft (UNI) and 46 with a bilateral cleft (BI); 84% (113/135) of patients underwent GPP (UNI: 78% [69/89]; BI: 96% [44/46]), 43% (58/135) of patients underwent ABG (UNI: 40% [36/89]; BI: 48% [22/46]), 18% (24/135) of patients underwent speech surgery for VPI (UNI: 14% [12/89]; BI: 26% [12/46]), 3% (4/135) of patients underwent palatal fistula repair (UNI: 0% [0/89]; BI: 9% [4/46]), 31% (42/135) underwent orthognathic surgery (UNI: 22% [20/89]; BI: 48% [22/46]), and 11% (15/135) underwent revision surgery to lip, nose, or both prior to facial maturity (UNI: 9% [8/89]; BI: 15% [7/46]]. Of the patients who underwent GPP, 61% (69/113) did not require ABG (UNI: 65% [45/69]; BI: 55% [24/44]) and 42% (48/13) required neither ABG nor orthognathic surgery (UNI: 51% [35/69]; BI: 30% [13/44]).

Conclusions: Clinical outcomes of the NAM treatment protocol from birth to facial maturity demonstrate a low rate of revision surgery to the lip and nose, as well as a low fistula and VPI rate. The frequency of orthognathic surgery reported in this study is consistent with published data. In addition, 42% of patients who underwent NAM with GPP required neither ABG nor orthognathic surgery.

## 232. Etiology and Clinical Outcomes in Robin Sequence Based on Presence and Morphology of Cleft Palate

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Background/Purpose: Robin sequence (RS) is defined as the constellation of micrognathia, glossoptosis, and airway obstruction with or without cleft palate. Although cleft palate is not required for a diagnosis of RS, many have questioned whether those with RS without

cleft palate have differences in etiology or clinical outcomes. Inclusion criteria in prior studies on RS vary, making comparisons challenging. Most distinguish between isolated RS (iRS), syndromic RS (SyndRS), and Stickler syndrome (SS).

*Methods/Description*: The present study included retrospective chart review of 295 patients with RS who received care at 2 centers. Suspected or confirmed genetic etiology and syndromic group (iRS, SS, or SyndRS), medical and developmental comorbidities, and mortality were compared with the presence of cleft palate and cleft morphology (U-shape, V-shape, submucous, unilateral cleft lip and palate). Mortality analysis was limited to 1 institution (n = 201).

Results: In our cohort, 38% had iRS, 7% SS, and 55% had a syndrome other than Stickler (SyndRS). The genetic etiologies of SyndRS in our study were similar to prior studies. A cleft palate was seen in 97% of iRS, 95% of SS, and 70% SyndRS. An U-shaped cleft palate was seen in 86% of iRS, 82% of SS, but only 27% of SyndRS (P < .0001). Many patients with SyndRS had other medical contributors to respiratory failure, which was not observed in iRS or SS. Eleven patients died, at ages ranging from 10 days to 20 months. Of the 10 patients with documented cause of death, all died due to complications of their underlying syndrome, except 1 tracheostomy-related death. Each child who died had a distinct genetic syndrome, and no patients with iRS or SS died (P = .001). Only 27% of those who died had an U-shaped cleft palate, and all had multiple and significant medical comorbidities. There were 3 syndromes with >3 patients in our RS cohort: Stickler syndrome (n = 21), Treacher Collins syndrome (TCS, n = 5), and 22q11.2 DS (n = 5). An U-shaped cleft palate was seen in 82% of Stickler syndrome, 40% of Treacher Collins, and 0% of 22q11.2 DS (P = .003). All had non-cleft-related medical contributors to respiratory failure, and the two with no cleft had tracheostomy.

Conclusions: Patients with iRS and SS had predominantly U-shaped cleft palate and had a very low risk of death. Patients with SyndRS had a variety of cleft types or no cleft and had documentation of multiple contributors to respiratory failure (eg, aspiration, hypotonia, central apnea). Increased risk of mortality was confined to SyndRS. Molecular etiology is associated with cleft type. Our data support 2 etiologic groups: (1) Robin sequence (isolated, secondary to Stickler or to another syndrome), in which airway obstruction is caused by glossoptosis arising from failure of descent of the fetal tongue, and (2) Robin physiology, in which the diagnostic features of RS are met in parallel rather than in series, due to an underlying genetic syndrome.

#### 233. Mechanistic Dissection of Causal Gene Relationships in Syndromic Cleft Lip and Palate Formation in a Patient Derived Stem Cell Model

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Background/Purpose: Genome-wide association studies have greatly contributed to our understanding of the correlation of certain mutations with cleft lip and palate formation. Unfortunately, this contributes little to the mechanistic understanding of development. Due to the pertinence of such comprehension to preventative and therapeutic measures, we attempted to model a complex syndromic orofacial cleft of unclear pathogenesis in a dish. Deriving embryonic-like pluripotent stem cells from a patient born with a malformation allows for a retracing of the arc of development and grants the ability to dissect the regulatory networks involved in the formation of the cleft.

Methods/Description: Patient and parent samples were used to generate induced pluripotent stem cells (iPSCs). We infected peripheral blood mononuclear cells derived from a whole blood draw of the patient and her father with Sendai viruses leading to an overexpression of 4 factors highly expressed in early embryonic development: OCT-4, KLF-4, c-MYC, and SOX2. Following a 21-day period of cellular reprogramming, several developing colonies were chosen for the patient and the father. Following 5 passages, the 3 superior looking colonies from patient and father were expanded. We supplemented our downstream experimental design by performing all inquiries in a sexand ethnicity-matched batch of fully characterized iPSCs as a control. At passage 10, we tested the genetic stability, cellular pluripotency, and genomic purity of our cells. Furthermore, we analyzed the iPSCs ability to follow down the paths of differentiating cellular lineages relevant in craniofacial development.

Results: We derived induced pluripotent stem cells (iPSCs) from a single child born of unrelated parents in Honduras and her father. The girl was born with an oblique facial cleft with anophthalmia and a coloboma, developmental delays, and mild ventriculomegaly. The child's and her father's iPSCs displayed properties typical of embryonic stem cells, such as persistent self-renewal and the expression of markers of pluripotency. Furthermore, their iPSCs proved capable of 3 germ layer differentiation and differentiation into neural crest cells. Whole-genome sequencing and subsequent bioinformatic analysis identified missense mutations of TDGF1 and TLE2 as potential causes. The iPSCs of the patient and her father proved equally capable to differentiate into chondrocytes, adipocytes, Schwann cells, and osteoblasts.

Conclusions: We could demonstrate that the gene regulatory networks involved in brain and face development appear to be dispensable for iPSC reprogramming, as well as the formation of embryoid bodies. Utilizing CRISPRa/i technology will allow us to further dissect the networks involved in the interwoven processes of brain and face development. The identification of the regulatory networks involved will help us to better understand the developmental biology of syndromic cleft lip and palate and lay the ground work for future innovation.

## 234. Gene Expression Analysis and Genetic Disruption of Transcription Factor IRF6 and Dependent Genes in Palate Development

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Background/Purpose: Mutations of the IRF6 transcription factor are strongly associated with the development of cleft lip and/or palate (CL/P). Since transcription factors function by activating gene expression of downstream genes, we can study the mechanism of cleft palate development by identifying and understanding the function of IRF6-dependent genes. Modern high-sensitivity assays such as RNAscope in situ hybridization allow us to visualize spatiotemporal expression patterns in wild-type and mutant embryos. However, early embryonic lethality in irf6 mutant zebrafish models precludes assessment of palate phenotype and gene expression analysis of potential downstream genes in mutant fish. We hypothesize that embryonic lethality can be circumvented by controlling irf6 loss of function through optogenetic regulation.

Methods/Description: We applied RNAscope to analyze gene expression patterns of irf6 in craniofacial structures during zebrafish palate

development at 48 and 72 hours post fertilization. For optogenetic irf6 loss of function, zebrafish embryos were microinjected with mRNA encoding for VP16-EL222, a protein responsive to 465 nm light, and a plasmid encoding for a dominant-negative form of the irf6 protein under control of the C120 promoter (C120-irf6-ENR). The embryos were grown in the dark for 10 hours to allow for normal development through epiboly and subsequently exposed to 465 nm light or grown in the dark for 72 hours. Palate phenotype was then assessed by staining the craniofacial cartilages with alcian blue.

Results: VP16-EL222- and C120-irf6-ENR-injected embryos developed a hanging-jaw phenotype and cleft palate when compared to uninjected controls or injected embryos grown in the dark. Baseline RNAscope expression of irf6 was restricted to epithelial structures including the oral epithelium at 48 and 72 hours post fertilization.

Conclusions: An optogenetic system of irf6 loss of function was successfully implemented in zebrafish and resulted in the development of cleft palate. In addition, gene expression of irf6 was visualized in epithelial structures at 48 and 72 hours post fertilization in zebrafish. This system is critical in studying gene expression patterns of irf6 mutant fish, including downstream genes such as ghl3. Further, a high-sensitivity gene expression baseline was established that can be correlated with expression patterns of other downstream genes. Such analysis will be critical in understanding intricate interactions between epithelial and chondrogenic cell populations that give rise to the palate.

## 235. Factors Associated With Delay in Presentation for Cleft Surgery a Tertiary Children's Hospital in a Major U.S. Metropolitan City

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Background/Purpose: Current guidelines recommend cleft lip (CL) and cleft palate (CP) repair prior to 6 and 12 months of age, respectively. Given limited availability of cleft providers, international guidelines extend recommendations to 1 and 2 years of age, respectively. However, even when cleft surgeons are available, some patients continue to present late for cleft surgery. This study aims to identify factors associated with late presentation for cleft surgery in an urban environment.

Methods/Description: Retrospective review was performed of all patients undergoing primary CL repair (CLR) or CP repair (CPR) at Children's Hospital Los Angeles (CHLA) over 9 years. Patients with submucous CP were excluded. Variables included demographics, cleft type, insurance type, driving distance, and time to hospital. Patients presenting in timely and delayed fashion were compared. Both institutional protocol (CLR before 6 months, CPR repair before 1 year) and international guidelines (CLR before 1 year, CPR before 2 years) were used as thresholds for late presentation. Statistical analysis, including Student and Mann-Whitney U tests or  $\chi^2$  test, was performed in SPSS 17 with significance at P < .05.

Results: A total of 805 patients—503 (62.5%) presenting for CLR, 302 (37.5%) for CPR—were included. Of patients seeking CLR, 32.4% had isolated CL and 67.9% cleft lip and palate (CLP). Based on CHLA protocol, 14.3% of patients presented late. Delayed patients were more likely to be female (48.6% vs 33.6%, P < .05), Hispanic (63.9% vs 44.1%, P < .01), have government insurance (80.6% vs 55.5%, P < .005), a bilateral cleft (29.2% vs 24.8%, P < .005), CLP (70.8% vs

66.8%, P < .05), or be syndromic (31.9% vs 7.2%, P < .001). Using international guidelines, 5.4% of patients presented late. Female gender (55.6% vs 34.7%, P < .05), syndromic diagnosis (22.2% vs 10.1%, P < .05), and median cleft (7.4% vs 0.6%, P < .05) were associated with delay. Patients were less likely to have bilateral CL (18.5% vs 25.8%, P < .05). For CPR, 79.5% of patients had isolated CP and 20.5% had CLP. Using CHLA guidelines, 60.3% of patients presented late. Delayed presentation was associated with CLP (24.7% vs 14.7%, P = .024) or syndromic diagnosis (44.5% vs 25.8%, P = .002). Using international guidelines, 17.5% of patients were delayed and were more likely to be Spanish speaking (30.2% vs 18.5%, P = .032). No significant difference was observed in race, or distance and time to hospital between cohorts for either CLR or CPR.

Conclusions: In a major metropolitan city, delay in presentation for cleft surgery occurs most often for CP repair. Female gender and syndromic diagnosis are most associated with late CLR. However, Hispanic ethnicity, government insurance, bilateral CL, or CLP diagnosis are also significant factors. Syndromic and CLP diagnoses and being Spanish speaking are associated with late CP repair. Improved patient education and communication with referring physicians and insurance providers may be needed to improve timeliness of cleft surgery in urban environment.

# 236. Mandibular Measurements at the 20-Week Anatomy Ultrasound as a Prenatal Predictor of the Severity of Respiratory and Surgical Interventions Associated With Pierre-Robin Sequence

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Background/Purpose: Pierre-Robin sequence (PRS) is defined as micrognathia and glossoptosis leading to upper airway obstruction and possible respiratory distress. Prenatal diagnosis of PRS is key in preparing the delivery team for an airway emergency. Routine 20-week ultrasounds screen facial features, allowing for the evaluation of maxilla—mandibular relationships and mandible size. This study aimed to evaluate 20-week ultrasounds of infants diagnosed with PRS to determine whether specific facial measurements could predict PRS disease severity.

Methods/Description: A retrospective review of patients with PRS born between December 2014 and March 2019 was performed. NICU and surgical data were collected. Respiratory and surgical interventions were recorded and scored for severity. Midsagittal profile images of the anatomy ultrasound were reviewed for each patient, and 3 parameters were measured to assess for micrognathia: facial maxillary angle (FMA), facial nasomental angle (FNMA), and alveolar overjet.

Results: Anatomy results: 30 patients were reviewed. Thirteen (43%) had an FMA below 66°, suggesting micrognathia (range: 47°-82°, mean: 65.17°) and 23 (77%) had an FNMA below 136°, suggesting micrognathia (range: 104°-154°, mean: 130.9°). Mean alveolar overjet was 3.6 mm (range: 2-7 mm).

Respiratory outcomes: Of 30 patients, 12 required no respiratory support (40%), 11 needed supplemental oxygen by nasal cannula or CPAP (36.7%), and 7 were intubated (23.3%). As respiratory support severity increased, median FMA decreased and alveolar overjet increased, though statistical significance was not met. In the respiratory independent group, median FMA was 68.5° (range: 59.5°-74.75°), while the CPAP/NC group had a median FMA of 66° (range: 57°-66°) and the intubated group had a median FMA of 65° (range:

52°-66°). Alveolar overjet for patients with no respiratory support revealed a median 3.1 mm overjet, CPAP/NC patients had 3.2 mm, and intubated patients had 3.8 mm.

Surgical intervention: 11 patients had no surgical procedures (36.7%), 5 underwent supraglottoplasty (16.7%), 13 received mandibular distraction or tongue–lip adhesion (43.3%), and 1 required tracheostomy (3.3%). There was no statistically significant difference in mean ultrasound measurements between surgical and nonsurgical patients. However, surgical patients tended to have smaller FNMAs and greater overjet compared to nonsurgical patients; median FNMA was 131° versus 132°, and median overjet was 3.2 versus 3.1 mm, respectively. Five patients had no respiratory or surgical interventions.

Conclusions: While a majority of patients with PRS at this gestational age had normal FMAs, most patients had abnormally acute FNMAs. Alveolar overjet, previously not described in prenatal literature but routinely assessed on neonatal clinical evaluation, is measurable and may have utility in prenatal diagnosis. It is important to note that the mandible keeps growing until the end of gestation, so 20 weeks may be too early to detect micrognathia as the disarray of growth rate may not manifest until the third trimester.

### 237. The Epidemiology of Cleft Lip and/or Palate in Northern Ireland—A 24-Year Review

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Background/Purpose: To determine the epidemiology of cleft lip and palate in Northern Ireland over a 24-year period: between 1994 and 2017. This includes the overall incidence as well as the incidence of babies born with individual cleft subtypes, sex ratios, and trends occurring throughout this time period.

Methods/Description: A retrospective review was performed using both the Northern Ireland Cleft lip and palate database and data from the Northern Ireland Statistics and Research Agency's Birth Registry. The overall incidence and subgroup analyses were then performed based on these data.

Results: Over the 24-year period, there were 978 babies born with a subgroup of cleft lip and palate. In the same time period, there were 568 196 live births recorded in Northern Ireland. The average incidence over this time period was 1.72 per 1000 live births. The most common cleft subgroup throughout this time period was incomplete cleft palate (iCPO), with 214 babies born with iCPO and a further 180 babies born with complete cleft palate (CPO). A total of 84 babies were born with bilateral cleft lip and palate (BCLP) and 185 babies were born with a unilateral cleft lip and palate (UCLP). A further 49 babies were born with separate clefts of both the lip and palate. There was an overall male predominance with a sex ratio of 1.3:1. For cleft lip and palate (unilateral, bilateral, and separate), there was a male predominance of 2.3:1, but for cleft palate alone (complete and incomplete), there was a slight female predominance of 1.3:1. For cleft lip with or without alveolus, there was a male predominance of 1.4:1.

Conclusions: This epidemiological study of cleft subtypes across an entire country has provided results which can be compared to other countries reported in the literature with the aim of both supporting our current knowledge and gleaming new insights. The average rate quoted for the United States is approximately 1 per 1000 and our rate is surprisingly much higher than this despite the similar population. When compared to other countries with a similar, predominantly Caucasian population, for example, the Czech Republic, the incidence is similar at 1.64 per 1000 live births. The role of racial variation in the development of cleft lip and palate is also supported when our

incidence rate is compared to a population in Uganda, with a much lower incidence of 0.73 per 1000, and at the other end of the spectrum, higher rates are reported in Pakistan, with a rate of just under 2 per 1000 births. Country-specific reports and birth rates are an important method of monitoring the burden of care in relation to cleft lip and/or palate throughout the world as well as allowing the identification of trends and potentially preventable risk factors in countries with a higher incidence.

## 238. Epidemiology of Cleft and Craniofacial Pathology in Vietnam Using SMS and Electronic Data Entry: A Feasibility Study

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Background/Purpose: Cleft and craniofacial pathology in Vietnam has historically been targeted by humanitarian groups due to a perceived higher incidence. Current disease surveillance practices in Vietnam are insufficient for timely standardized data collection of craniofacial and cleft pathologies. Concurrently, the World Bank reports mobile cellular subscription rates (per 100 people) in Vietnam to be 128, placing it as one of the top mobile phone—using nations in the world. Herein, we discuss our experience developing and launching an SMS-based surveillance system based in the capital city of Hanoi, Vietnam.

Methods/Description: Vietnam National Children's Hospital (VNCH) is a pediatric tertiary care center comprised of 13 clinical departments and 1300 beds, serving approximately 30 provinces and 12 million children. The Department of Plastic Surgery is comprised of 4 surgeons, all who possessed SMS-enabled mobile phones prior to project introduction and were trained in data entry over the course of 1 week. The SMS database was designed using FrontlineSMS, a 2-way mobile gateway software application with automated messaging capabilities. Inbound messages can be downloaded as comma-delimited .xslx files; outbound messages prompting data entry are programmed based on participant input. VCNH has served as the pediatric pilot site for the introduction of electronic health records (EHRs) in Vietnam since 2007. Individual demographic data for patients with cleft and craniofacial pathologies seen by the department was obtained for 2010 through 2019.

Results: Seven patient records were logged using the SMS data input program over a 4-month period. Physicians were found to have an average of 43.88 seconds of down time between patients during clinic hours for data input; the time to record completion was on average 31.8 minutes. Actual cleft and craniofacial patient volume over the same 4-month period as obtained via the electronic record was 266, indicating a 2.63% capture rate. Cases input included 3 cleft lips, 1 cleft palate, and 3 cases of Apert syndrome. Our retrospective analysis yielded 2794 patients with a cleft or craniofacial diagnosis. Of those, 2529 had sufficient geographic, demographic, and diagnostic data available. Five provinces were statistically significantly (P < .01)overrepresented in our cohort in proportion to their respective populations: Ha Nam, Bac Ninh, Bac Giang, Nam Dinh, and Vinh Phuc, all bordering Hanoi. Underrepresented provinces included Thanh Hoa, Nghe An, Hai Phong, and Dong Nai. Prevalence of cleft lip and palate was found to be 0.136\% (or 1 in 735), a value corrected for previous findings that 43% of patients with cleft lip and palate rely on mission groups for surgical care. This is in line with previously reported findings.

Conclusions: To our knowledge, this is the largest epidemiology study of cleft and craniofacial pathology in Vietnam and the first to use mobile health. Regional and pathology-specific analyses will further strengthen understanding of risk factors.

## 239. Regrafting and Psychosocial Outcomes in Early Alveolar Bone Grafting for Cleft Lip and Palate Patients

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Background/Purpose: Alveolar bone grafting (ABG) is a treatment modality for patients with cleft lip and palate (CLP), often performed around age 9 during the mixed dentition stage but prior to canine eruption. However, we recently showed that surgical burden in CLP patients in age groups 8 to 10 was associated with poorer psychosocial outcomes. Therefore, we sought to evaluate whether alveolar grafting at an earlier age affects psychosocial functioning and complication rates in children with CLP.

Methods/Description: Patients with CLP who underwent ABG from the University of California, Los Angeles, craniofacial clinic, and the Orthopaedic Institute for Children (n = 77) were prospectively enrolled and administered the Patient-Reported Outcomes Measurement Information Systems (PROMIS) short-form measures. Patient demographic, operative, and follow-up data were retrospectively collected. Complications were determined as requiring alveolar bone regrafting. To evaluate the potential long-term psychosocial effects of early grafting, only patients who underwent initial ABG at ages 0 to 10 and had a minimum 1-year follow-up were included (n = 51), and patients were grouped into early (0-7 years) or standard (8-10 years) ABG age range. The  $\chi^2$  test with Fisher exact correction was used to analyze categorical variables and t test was used to analyze continuous variables. A significance level of .05 for 2-sided tests was used.

Results: Overall, 51 patients with CLP (22 males, 43.1%) underwent initial ABG between ages 0 and 10. The mean age of patients at the time of initial ABG was  $8.3 \pm 1.5$  years (range: 4-10). Unilateral CLP was the most common cleft diagnosis (n = 36, 70.6%), followed by bilateral CLP (n = 15, 29.4%). The average number of surgeries in this cohort was  $6.1 \pm 3.0$  (range: 2-15). PROMIS was administered at  $14.1 \pm 3.0$  years (range: 8-23 years). Of the patients who received alveolar bone regrafting (n = 13, 25.5%), regrafts were done at age  $10.8 \pm 2.0$  (range: 8-15). Compared to patients who had standard ABG (age 8-10), patients who had early ABG (age 0-7) demonstrated no significant differences in demographics, diagnoses, surgical timing, and regrafting rates; however, time between initial ABG and PROMIS assessment was longer in the early ABG group  $(7.0 \pm 3.1)$ vs 5.3  $\pm$  2.9, P=.02). In terms of psychosocial functioning, no difference was found in anger, depression, and peer relationships between the 2 groups, but patients who had early ABG had significantly lower levels of anxiety (41.0  $\pm$  7.9 vs 47.4  $\pm$  9.6, P = .03). Furthermore, while regrafting rates between the 2 groups were comparable, of those who required regrafting, patients who had early ABG had significantly lower anxiety (39.1  $\pm$  7.7 vs 50.6  $\pm$  8.5, P = .01) and depression (38.6  $\pm$  5.4 vs 48.2  $\pm$  8.7, P = .03) scores compared to patients who had standard ABG.

Conclusions: Earlier alveolar bone regraft surgery is associated with improved psychosocial outcomes without affecting regrafting rates in children with CLP.

## 240. Cost Analysis and Operative Time of Demineralized Bone Matrix Versus Autograft Bone Graft for Cleft Alveolar Reconstruction

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Background/Purpose: Iliac crest bone graft (ICBG) as an autologous source of bone remains the gold standard for alveolar bone graft (ABG) reconstruction in cleft lip/palate. Novel allograft sources of cancellous demineralized bone matrix (DBM) have demonstrated similar rates of graft healing without donor site complications associated with ICBG. Concerns have been raised regarding both clinical outcomes and additional cost of the allograft materials. This study examines operative time and total cost associated with DBM when compared to ICBG.

Methods/Description: A retrospective chart review from January 2015 to January 2019 was performed for patients undergoing ABG by a single surgeon. Cohorts were selected based on source of bone graft (ICBG vs DBM). Demographics, surgical outcomes, and costs were collected. Graft success was determined by a blinded orthodontist based on postoperative cone-beam imaging. Costs from both surgeon and hospital sources (direct and indirect costs) were collected from the hospital billing department. Univariate analyses were performed.

Results: In our analysis, 39 patients underwent ABG with ICBG versus 14 patients with DBM. Average age was similar between cohorts (9.6  $\pm$  2.0 years vs 9.5  $\pm$  4.2 years; P=1.00). In the DBM group, surgical time was reduced compared to the ICBG group (102.6  $\pm$  30.4 minutes vs 65.6  $\pm$  25.2 minutes; P=.002). DBM was utilized more frequently in bilateral cleft lip/palate when compared to ICBG (42.9% vs 31.0%; P=.41). Overall graft success rate was higher in the ICGB group (92.3% vs 86.6%; P=.52). There were no complications in either group for fever, infection, drainage, or return to the ER. Cost of DBM material was \$473.20 per patient (from hospital billing data). Average total cost was less using DBM when compared to ICBG (\$15.019  $\pm$  \$4604 vs \$12.960  $\pm$  \$4064; P=.16).

Conclusions: Patients tolerated both ICBG and DBM sources for ABG with minimal complication rates, with higher graft success rate in ICBG when compared to DBM. Operative time was reduced with DBM. Despite the added cost of DBM material, the average cost was less both in OR cost and total cost when compared to ICBG, though the difference is not statistically significant. From these data, DBM offers shorter operative time with cost benefit; however, graft success remains higher with ICBG.

### 241. Materials for Cleft Alveolar Bone Grafting: A Systematic Review and Network Meta-Analysis

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Background/Purpose: In recent years, numerous materials have been proposed for use in reconstructing patients with a cleft maxillary alveolus, challenging the gold standard of using iliac crest bone grafts. Given the mixed evidence for use of these materials, the objective of this systematic review is to compare the success rates of the various materials used for secondary cleft alveolar bone grafting during the mixed dentition phase.

Methods/Description: A comprehensive search of prospective studies evaluating 2 or more materials was applied to the following electronic

databases: MEDLINE, EMBASE, Cochrane Library, Scopus, and Google Scholar. To identify grey literature, abstracts from select conferences as well as references of included studies were searched. No limitations were placed on language. The search and data extraction were conducted in duplicate. Network meta-analysis was undertaken using the Bayesian random-effects framework to estimate the treatment effects of the included interventions. Additionally, surface under the cumulative ranking (SUCRA) values, used to determine the probability of each treatment being the best, were calculated.

Results: From an initial yield of 2280 studies, 18 studies of 19 interventions were included. Evaluating percentage bone fill of the defect following 3 to 12 months of follow-up, platelet-rich fibrin in combination with an iliac crest bone graft, hydroxyapatite in combination with collagen, or iliac crest graft alone were ranked as the most efficacious means of reconstruction (9 studies and 181 patients; 74.1% [95% CrI: 59.3-90.7], 64.6% [95% CrI: 49.1-80.7], 59.9% [95% CrI: 52.8-66.7], respectively). When evaluating bone volume gained at 6 to 12 months, iliac crest graft alone was suggested to be superior to other methods (10 studies and 192 patients; 0.431 mL [95% CrI: 0.402-0.458], compared to 0.342 mL [95% CrI: 0.303-0.383] and 0.186 mL [95% CrI: 0.132-0.240] for hydroxyapatite in combination with collagen and platelet-rich fibrin in combination with an iliac crest graft, respectively).

Conclusions: Results of this study suggest the superiority of iliac crest with or without platelet-rich fibrin or hydroxyapatite in combination with collagen for the reconstruction of cleft alveolar defects. Given the high risks of bias and low sample sizes of all of the included studies, further high-quality studies are necessary to make definitive conclusions regarding the efficacy of these grafting materials.

# 242. Efficacy of Demineralized Bone Matrix (DBX) for Alveolar Bone Grafting in Patients Previously Treated With Bone Morphogenic Protein 2 (BMP2)

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Background/Purpose: Autologous iliac crest bone grafting is the gold standard for alveolar reconstruction in patients born with cleft lipnose-palate, but donor site morbidity has prompted the search for bone graft substitutes. Two such alternatives include bone morphogenic protein 2 (BMP2) and demineralized bone matrix (DBX). Early studies showed BMP2 equivalent to iliac bone grafting, but concern for potential long-term complications prompted the FDA to caution against its use in pediatric patients. DBX alone has been shown noninferior to autologous bone in secondary alveolar bone grafting (ABG) in cases where primary reconstruction has failed. However, the use of DBX in patients with previous BMP-2 to their alveolar cleft site has not been investigated. This study examines whether patients previously treated with BMP-2 with persistent alveolar cleft have similar rates of successful repeat bone grafting (determined as no need for repeat ABG procedure) with DBX alone compared to use of bone autograft or a DBX-autograft mix.

*Methods/Description*: In our IRB-approved study, our center's cleft team medical records were searched from 2013 to 2019 for patients who received alveolar bone grafting (ABG). Charts were analyzed for type of primary bone grafting, prior BMP2 intervention, and graft revision rates. Data were analyzed using Fisher exact test.

Results: From 2013 to 2019, 33 children with cleft underwent either primary or secondary alveolar bone grafting, with average follow-up

time of 5.3 years. Of these children, 10 were previously treated with BMP2 for alveolar closure but had a persistent cleft defect requiring additional alveolar bone grafting. Within this BMP2 subset, 2 children were treated with autograft alone, 7 with DBX alone, and 1 child with a mix of autologous bone and DBX. Eight children had successful secondary ABGs, including both children who received only autologous graft. Two children, one with DBX alone and one with DBXautologous mix, failed and required repeat grafting. Fisher exact test comparing success rates of each of the primary grafting options given prior BMP2 intervention was not significant (P = .22). When children grafted with prior BMP2 use were compared to those undergoing primary ABG without prior BMP2 intervention (n = 15), they exhibited a similar success rate (73.3% vs 80% in children with previous BMP2, P = .18). This also held true when comparing the success rate of children with prior BMP2 intervention undergoing ABG with DBX alone versus those undergoing primary ABG with DBX alone (85.7% vs 88.9%, P = .99).

Conclusions: Use of DBX for alveolar reconstruction is associated with similar success rates to gold standard autograft for alveolar bone grafting in children with residual bony defect after BMP2 treatment. This is the first time these results have been presented for children with history of BMP2 grafting for alveolar cleft.

# 243. Our Treatment Strategy With Nasoalveolar Molding, Gingivoperiosteoplasty and Furlow Palatoplasty for Bilateral Complete Cleft Lip and Palate Patients

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Background/Purpose: We have been using our own treatment for bilateral cleft lip and palate (BCLP) patients. After presurgical orthopedics (PSO), the subsequent surgical procedures depend on the result of PSO. The procedures consist of PSO, gingivoperiosteoplasty (GPP), Furlow double opposing Z palatoplasty (FDOP), and maxillary protraction appliance (MPA). Our purpose is to evaluate the outcomes including speech, alveolar bone formation, and the maxillary growth after our treatment.

Methods/Description: Twenty-seven nonsyndromic BCLP patients without mental disorders and serious anomalies were treated by PSO, cheiloplasty, GPP, and FDOP at Kanagawa Children's Medical Center. After the treatments, their speech outcomes, alveolar bone formations, and maxillary growths were evaluated by CT imaging at 5 years of age. MPA was used as part of the protocol for 6 months to 1 year for postoperative retardation of maxillary growth cases.

## 244. Vascularized Composite Pericranial Bone Flap for Immediate Reconstruction of a Hemi-Maxillectomy Defect in a Pediatric Patient

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Background/Purpose: Surgical resection of maxillary tumors can result in defects that can be difficult to reconstruct using conventional methods due to the complex functional and anatomic nature of the midface area. Choice of maxillectomy reconstruction depends on the location and extent of the defect requiring reconstructive surgery. Various reconstructive methods have been used to repair partial or total maxillectomy defects including prosthetics and allogenic or autogenous nonvascularized bone grafts in combination with local flaps or osteocutaneous free flap reconstruction. Yet, no single technique has been able to achieve all of the reconstructive goals. The purpose of this study is to present a new technique for hemi-maxillectomy defect reconstruction in a pediatric patient using a vascularized composite flap consisting of temporalis muscle, pericranium, and parietal bone.

Methods/Description: The patient had a successful reconstruction of his right maxilla with the newly described technique. Stable skeletal fixation with adequate orbital support was maintained over a 3-year follow-up period. A new method of reconstruction using a vascularized composite perioranial bone flap consisting of parietal bone pedicled to perioranium and temporalis muscle is a reliable reconstructive option for large maxillectomy defects in pediatric patients. This technique can result in improved outcomes and reduced morbidity.

## 245. Advantages and Disadvantages of Surgical Techniques for the Treatment of Ankyloglossia: A Preliminary Study

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Background/Purpose: To compare postoperative and histological aspect of frenotomy made by 3 different techniques: scalpel as a "gold standard," CO<sub>2</sub> laser, and diode laser.

Methods/Description: Frenotomy was performed by scalpel, CO<sub>2</sub>, and diode laser. Specimens were classified into 3 groups according to tool used for surgical excision. The rats from 3 experiment groups with 20 rats in each group will be killed for one of the following time intervals: 3, 7, 21, 35 days. Biopsy specimens and full-thickness specimen containing the frenulum from both sides will dissected out of the 2 surgical sites on each rat. Histological evaluation was evaluated using light microscopy and assessed for epithelial and collagen arrangement and presence of inflammatory cells. All the rats were weight in the beginning of the experiment and monitored by weighing 3 times a week. The rats were checked for their physiological and behavioral state.

Results:  $CO_2$  and diode lasers showed immediate hemostasis compared to scalpel. Mean weight loss in the first week was highest in diode group followed by  $CO_2$  and scalpel. During the first 3 weeks, all the groups gain weight about 17% relative to the initial weight without any statistically significant difference within the 3 groups. Histologically, diode show highest collateral damage followed by  $CO_2$ . The scalpel group shows rearrangement, while the lasers group shows new arrangement of epithelial and collagen tissues.  $CO_2$  and diode both show increased amount of collagen fibers compared to the scalpel group.

Conclusions:  $CO_2$  and diode had better results in hemostasis parameter but have disadvantage such as lateral heat damage and tissue necrosis. Conventional scalpel treatment is better option in terms of faster reepithelization. The preliminary results enhance the hypothesis that laser causes new arrangement in the epithelial and collagen tissue that leads to less recurrent grow of the frenulum.

# 246. Skeletal, Soft Tissue and Globe Position Changes Following Le Fort I + III Surgery in Patients With Mid-Facial Hypoplasia and Proptosis

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Background/Purpose: Our study quantifies changes in skeletal, soft tissue profile, and globe position in patients with syndromic cranio-synostosis after Le Fort I + III (LF I + III) surgery.

Methods/Description: Patients with syndromic craniosynostosis who underwent LF I + III at the time of facial maturity were followed for at least 1 year. Each lateral cephalometric radiograph was traced using Dolphin Imaging software and superimposed at the sella. Changes in positions of the different landmarks at T0 (preoperatively), T1 (immediate postoperatively), and T2 (1 year postoperatively) were measured by the software. Sixty-seven soft tissue and skeletal landmarks were digitized and measured. LF III skeletal changes were measured by changes in lateral orbit and orbitale. LF I skeletal changes were measured at the A point and U1. Corresponding soft tissue profile and globe position were studied. All data were measured along the x-axis. Results: Twelve patients included in our study have the following syndromes: Crouzon (n = 6), Pfeiffer (n = 2), Apert (n = 1), Antley-Bixler (n = 1), cleidocranial dysplasia (n = 1) and frontonaso dysplasia (n = 1)1). Nine patients had previous history of LF III distraction. Standard descriptive statistics was used. Data were analyzed using paired T test. Lateral orbit advanced 5.49 mm (T0-T1) on average, with a P value of  $1.3^{-5}$ , and 5.94 mm (T0-T2) on average; 0.45 mm (T1-T2) change with a P value of .96 suggests the lateral orbit is stable. Similar advancement at orbitale is observed at 5.68 mm (T0-T1) and 6.42 mm (T0-T2). The globe moved anteriorly by 1.98 mm (T0-T1) with a P value of .025 and anteriorly by 0.944 mm (T0-T2). The change between T2 and T1 is 1.04 mm (P value: .26), which suggests the globe moved backward after postsurgical swelling subsided. The ratios of movement (globe to lateral orbit) between T0-T1 and T0-T2 are 31% and 16%, respectively. The decrease in ratio can be attributed to the reduction in soft tissue swelling at T2. Restoring position of the globe relative to the lateral orbit decreases the risk of exposure keratitis, keratoconjunctivitis sicca, and corneal ulceration. Anterior nasal spine and point A were advanced by 9.38 and 10.08 mm, respectively, between T0 and T1, and 9.01 mm and 8.51 mm, respectively, between T1 and T2. At the occlusal level, U1 advanced 10 mm and L1 moved back 1.45 mm between T0 and T1. Menton moved back 1.25 mm (T0-T1) but advanced by 2.48 mm (T0-T2). This change in direction is due to splint use at T1 as it rotates mandible clockwise.

 $\label{lem:conclusions: In our cohort, LFI + III surgery improved both midface deficiency and proptosis in those with syndromic craniosynostosis. Combined Le Fort I + III surgery allows surgeons to perform differential corrections of the midface at the orbital and the dentition level. This is ideal for proptosis correction and establishing optimal jaw relationship.$ 

# 247. Unilateral Box Osteotomy: An Under-Utilized Versatile Corrective Procedure of Fronto-Orbito-Zygomatic Hypoplasia and Orbital Dystopia in Hemifacial Microsomia

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Background/Purpose: Surgical correction of fronto-orbito-zygomatic (FOZ) hypoplasia and orbital dystopia in patients with hemifacial microsomia (HFM) has focused on camouflage procedures limited to soft tissue and bone augmentation by using autologous or synthetic options (fat grafting, bone grafts, and implants). These procedures do not address the primary underlying problem: hypoplasia of the fronto-orbito-zygomatic (FOZ) complex. We sought to describe the utility, technical aspects, and surgical/aesthetic outcomes of unilateral 4-wall box osteotomy in simultaneous correction of FOZ hypoplasia in patients with HFM.

Methods/Description: An IRB-approved retrospective review of patients who underwent unilateral, 4-wall box osteotomy to address FOZ hypoplasia was performed in patients with HFM between 2010 and 2019. Data collected include demographic data, surgical indications, extent of FOZ complex hypoplasia, intra- and postoperative complications, and short- and long-term surgical and aesthetic outcomes. We also address the technical modifications and concept of altering the height of frontal craniotomy, degree of asymmetric frontal bone advancement, and level of maxillary/zygomatic osteotomy to account for individual phenotypic variation of the extent of FOZ complex hypoplasia and orbital dystopia.

Results: During the study period, a total of 4 patients (100% female) underwent unilateral, 4-wall box osteotomy to address hypoplasia (left in 3 and right in 1) involving the frontal, supraorbital, lateral orbital rim, and zygomatic regions and unilateral vertical dystopia. Patients underwent the surgery at a mean age of  $16 \pm 4.4$  years, with a median follow-up duration of 21 months (range: 2 weeks to 9 years). Mean operative time was  $6.5 \pm 1.5$  hours. Mean estimated blood loss was  $375 \pm 84$  mL, with 2 patients (50%) requiring intraoperative blood transfusion. Mean hospital stay was  $4.25 \pm 0.4$  days. None of the patients had intraoperative or postoperative complications. Review of postoperative images and postoperative CT scans showed excellent correction of FOZ complex hypoplasia and orbital dystopia in all patients with a mean difference in anteroposterior projection of 1.8  $\pm$  1.8 mm in the frontal, 1.2  $\pm$  1.1 mm in supraorbital, 1.5  $\pm$  0.7 mm in lateral orbital rim, and  $4.0 \pm 3.8$  mm in zygomatic eminence regions when compared to the contralateral side. All patients maintained a stable skeletal correction at their last follow-up.

Conclusions: Unilateral, 4-wall box osteotomy is a safe, versatile, and powerful surgery that allows simultaneous correction of FOZ complex hypoplasia and orbital dystopia in patients with HFM and can be tailored to address variable degrees of hypoplasia affecting the frontal, supraorbital, lateral orbital rim, and zygomatic regions via adjustments of the height of frontal craniotomy, asymmetric frontal bone advancement, and the level of maxillary/zygomatic osteotomy.

### 248. Conformity of Virtual Surgical Plans to Actual Post-Operative Outcomes in Craniofacial Surgery

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Background/Purpose: Virtual surgical planning (VSP) is a powerful tool for planning reconstructive procedures in craniofacial surgery. The purpose of this study is to compare VSP predicted to the actual postoperative results for 5 distinct procedure categories performed by a single surgeon. We hypothesize that the virtual surgical plans for implant cranioplasty will be most conforming to the postoperative outcomes, and those for mandibular distraction will conform the least.

Methods/Description: Stereolithography formats were obtained and analyzed for the virtual surgical plan images and the postoperative computed tomography scans from patients who underwent one of the 5 following types of craniofacial surgery: implant cranioplasty (I), cranial vault remodeling (V), orthognathic surgery (O), mandibular reconstruction (M), and mandibular distraction (D). Digital renderings were imported and analyzed in the operated area of the craniofacial skeleton using volumetric overlays and linear data. Each category cohort was then compared and stratified. Statistics included ANOVA testing, with P < .05 used for statistical significance. Post hoc Bonferroni comparison was carried out for groups that displayed significant variance.

Results: The study included 125 patients (61 orthognathic; 15 mandibular reconstruction; 32 cranioplasty for craniosynostosis; 8 implant cranioplasty; 9 mandibular distraction) with VSPs and postoperative CT scans. Average volume discrepancies (%) in each group were the following: 28.4 11.7 (I); 30.5 11.2 (M); 45.0 7.3 (O); 54.2 22.2 (D); 77.8 11.9 (V). Implant cranioplasty was found to be significantly more conforming when compared to mandibular reconstruction, calvarial vault remodeling, orthognathic surgery, and mandibular distraction (*P* < .001 for all comparisons).

Conclusions: Conformity between VSP and actual postoperative results is greatest for implant cranioplasty and least for cranial vault remodeling. Despite imperfect conformity between VSPs and postoperative CT images in all categories, clinical end points were universally excellent. These results indicate that successful craniofacial reconstruction guided by VSP still relies on intraoperative assessment and artistic judgement developed from experience by the individual surgeon, particularly in cranial vault remodeling and mandibular distraction.

# 249. Cephalic Index Norms Differ Between Racial Groups: Evaluating a Diverse Cohort in the Wake of Back to Sleep Campaign

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Background/Purpose: In 1992, the Back to Sleep Campaign was instituted by the American Academy of Pediatrics. Although this public health campaign profoundly decreased infant mortality from sudden infant death syndrome, positional plagiocephaly incidence has subsequently increased at least 5-fold. Insurers authorize treatment for plagiocephaly with helmet therapy based on deviation from normative cephalic index (CI), the ratio of head width to length. The current norms for cephalic index are derived from the Farkas data set, which was established prior to the Back to Sleep Campaign and measured only Caucasian children with an overall small sample size. This study seeks to evaluate whether CI values of infants and children vary between different racial groups.

Methods/Description: Children between the ages of 0 and 3 months, 4 and 6 months, 9 and 12 months, 2 and 3 years, and 12 and 14 years were recruited at their well child appointment. Exclusion criteria were diagnosis of premature birth, craniosynostosis, positional plagiocephaly, craniofacial abnormality, hydrocephalus, gross developmental delay, or torticollis. Calipers were used to measure occipotofrontal and biparietal dimensions of the head. Cephalic index was calculated for each racial group and values were compared with Student *t* test and ANOVA analysis.

Results: A total of 657 patients met inclusion criteria. Overall, the population was racially diverse, 36.7% Caucasian (n = 243), 26.6% Hispanic (n = 175), 19.9% black or African American (n = 131), 8.8% Asian (n = 58), and 7.6% multiracial (n = 50). In the critical age group of 3 to 6 months, Asian children had a higher cephalic index (CI: 89.0) than Caucasian children (CI: 85.1), Hispanic children (CI: 84.3), and African American children (CI: 84.5). Cephalic index was statistically significantly higher in Asian children than in Caucasian children at 3 to 6 months (P = .012). There was no statistically significant difference in CI between racial groups at 0 to 3 months (P = .19). In all racial groups, the cephalic index peaked at 3 to 6 months of age and subsequently decreased with age.

Conclusions: The mean cephalic index of Asian children in this sample aged 0 to 3 months and 4 to 6 months was 84.7 and 89.0, respectively. These values are higher than those of other racial groups, and this difference is statistically significant at the critical 3- to 6-month time point when families and craniofacial surgeons are deciding about possible helmet therapy. In this series, the peak in cephalic index occurs in all racial groups in the 3- to 6-month age range, which is earlier than the 9- to 12-month CI peak in the Farkas data set. This is consistent with maximal occipital flattening in first few months of life, prior to babies' improved head control and the mobility typically obtained by 6 months of age. Prior normative cephalic index data included measurement of only Caucasian children, and this new data grow our understanding of cephalic index norms for all racial groups in the wake of the Back to Sleep Campaign.

### 250. Diagnosis and Management of Post-Op Subgaleal/Epidural Abscess in Pediatric Craniosynostosis Surgery

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Background/Purpose: Infection after pediatric craniofacial surgery is a rare but potentially serious complication. Subgaleal/epidural abscesses may lead to sepsis, meningitis, bone loss, and the need for additional surgery. A literature review failed to identify management guidelines for epidural abscesses in synostosis surgery. The purpose of this study is to describe one center's experience with epidural abscesses including preoperative risk factors, clinical course, and the management protocol of these patients over 20 years.

Methods/Description: An IRB-approved, single-surgeon, retrospective audit of pediatric transcranial craniosynostosis procedures from 1997 to 2018 identified 384 consecutive patients. Six patients (1.6%) developed postoperative epidural abscess. Records were abstracted for diagnosis, procedure, clinical course, daily Tmax, WBC, CRP, culture results, and management. Outcomes were assessed for bone resorption and need for additional surgery. Continuous variables not normally distributed as a median were summarized with interquartile range (Q1, Q3). Median differences were performed using the Mann-Whitney U test or Wilcoxon signed-rank test. Continuous variables were summarized as means with standard errors (SE) and compared using the Student t test. Categorical data were compared using  $\chi^2$  or Fisher exact test as appropriate. We performed analyses in SAS for windows version 9.4.

Results: A total of 384 consecutive pediatric patients underwent 413 surgical procedures. There were 315 cases of single suture and 69 cases of multiple suture synostosis. Six patients required treatment for epidural abscesses. Infection presented during the initial hospital stay in 4 patients, while 2 patients were readmitted after discharge at

postoperative day 10. A prior history of posterior vault distraction was noted in 2, with skin compromise (severe eczema) in another. Presentation included fever >102F with a daily Tmax that remained above the mean of a control group and was associated with increasing edema, elevated WBC, CRP, and sedimentation rate. CT confirmed the abscess. All patients with epidural abscesses had undergone bilateral fronto-orbital advancement, with no infections seen in the calvarial vault remodeling group (P=0.03). Infections were managed with surgical drainage, washout and drain placement, plus 6 weeks of culture directed antibiotic therapy. Bone salvage was attempted in all 6 patients. One patient required return to the OR for secondary reconstruction due to bone resorption.

Conclusions: Epidural abscesses occurred only in patients undergoing bilateral fronto-orbital advancement and presented during the initial hospital stay in 4 of 6 cases. Prior surgery and skin compromise were risk factors. Presentation included an elevated Tmax and increased edema. Infections were managed with washout, deep drains, and 6 weeks of antibiotic therapy. Bone salvage was attempted in all 6 cases. One patient has undergone secondary reconstruction for defect in the frontal bone.

# 251. A Comparison Between Autologous Bone Graft and Carbonated Calcium Phosphate Cement for Secondary Cranioplasty; A Long-term Institutional Experience

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Background/Purpose: While the mainstay of cranioplasty remains autologous bone graft (ABG), several alloplastic substitutes have emerged in order to provide a renewable source, mitigating the inherent drawbacks of autologous tissues. Cranios, a carbonated calcium phosphate (CCP) bone cement, has been suggested as a suitable alternative due to its structural similarities to mineralized bone and ability to be resorbed and replaced by natural bone. We compare our long-term institutional experience using Cranios and autologous bone grafting for secondary cranioplasty refinements in pediatric and adult patients.

Methods/Description: A retrospective cohort study was conducted of subjects undergoing secondary cranioplasty for forehead reshaping utilizing autologous bone or Cranios between June 2007 and August 2018. Exclusion criteria included less than 1-month follow-up. The primary outcome variable was aesthetic correction of head shape, while secondary outcome variables includes complication and reoperative rates.

Results: In all, 159 subjects met inclusion criteria, with 112 and 47 subjects underwent secondary cranioplasty using CCP and ABG, respectively. Median age at the time of secondary cranioplasty differed between cohorts, with patients undergoing ABG at a younger age (14.0 vs 15.5 years, P < .006). Common indications for cranioplasty included cranial vault reshaping in infancy (78.6%) and trauma (10.1%), which did not differ between cohorts (P < .09). The majority of bony defects were limited to the frontal region (89.9%), although midface/nasal defects were more likely to be reconstructed using ABG (P < .01). Wound class (P < .02) and ASA grade (P < .02) significantly differed between cohorts with sicker subjects undergoing ABG-based reconstruction. Length of procedure was 1.5 hours greater on average in the ABG cohort (P < .0001). Subjects in the ABG cohort presented more often with full-thickness cranial defects near the frontal sinus (P < .0001).

< .0001). Subjects in the CCP cohort were more likely to undergo reconstruction of partial-thickness cranial defects in the onlay position (P < .0001). Larger defects were reconstructed utilizing CCP (96.0 vs 31.0 cm², P < .0002). Complications rates in the ABG and CCP cohorts were 19.2% and 11.8%, respectively (P < .31). The majority of complications in the CCP cohort related to infection versus volume loss in the ABG cohort. Factors associated with complications in the CCP cohort included clean-contaminated wound class (P < .001) and proximity to the frontal sinus (P < .03).

Conclusions: Cranios remains an efficacious alloplastic bone substitute with an acceptable complication profile in patients undergoing secondary cranioplasty when compared to gold standard autologous bone. There are inherent advantages and disadvantages associated with use of autologous bone versus alloplast that must be considered on a case basis. Complications may arise months to years following implantation, warranting long-term postoperative follow-up.

## 252. Influence of Radiographic Soft Tissue Findings on Clinical Entrapment in Patients With Orbital Fractures

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Background/Purpose: Radiographic herniation of extraocular muscles through orbital fractures has become a means of diagnosis for orbital entrapment. This study aims to evaluate the value of radiographic soft tissue findings, specifically extraocular muscle herniation, in predicting "true" clinical orbital entrapment after orbital fracture.

Methods/Description: A single-institution, retrospective review at a regional level 1 trauma center was conducted. Patients with a diagnosis of "fracture of orbital floor" were identified and CT reports were queried for the terms "herniation," "herniated," "entrapped," and "entrapment." The first 400 records were analyzed.

Results: Forty-three percent of radiology reports mentioned "entrapment," while the incidence of clinical orbital entrapment was 2.8%. Radiographic extraocular muscle herniation had a positive predictive value of 7.9% for clinical orbital entrapment. Fat herniation alone and muscle contour irregularity had predictive values of 4.2% and 4.8%, respectively. Irrespective of clinical exam, 50% of patients with extraocular muscle herniation and 43% of patients without extraocular muscle herniation ultimately required surgery.

Conclusions: Radiographic soft tissue findings, specifically extraocular muscle herniation, do not predict true clinical orbital entrapment. Diagnosis of orbital entrapment should be made based on physical exam alone. The assumption that radiographic findings correlate with orbital emergencies may result in unnecessary interfacility transfers, subspecialist consultations, and ultimately, operative procedures.

### 253. Identifying Predictors of Levator Veli Palatini Muscle Contraction During Speech Using Dynamic MRI

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Background/Purpose: The levator veli palatini (LVP) muscle is the primary muscle responsible for velar elevation and achieving velopharyngeal (VP) closure during speech. Computational modeling has identified the importance of closure force, muscle activation and velocity in muscle fatigue, and complete VP closure (Inouye et al., 2015). Few dynamic MRI studies have successfully quantified changes in LVP muscle length during real-time speech tasks. Only one study has reported LVP contraction velocity during speech production, with a sample of 6 subjects with normal anatomy (Pelland et al., 2019). Findings by Pelland et al. (2019) and Inouye et al. (2015) identified VP port depth as the most influential predictor of closure force among 6 adult subjects. Further exploration is needed to confirm these findings using a larger sample size, identify other significant predictors, and control for sex effects. Furthermore, LVP contraction velocity in individuals with repaired cleft palate has never been reported. This study aims to identify significant predictors of LVP contraction using a larger sample size of adults with normal anatomy, while controlling for sex effects. A secondary aim is to report contraction velocities of individuals with repaired cleft palate and make preliminary comparisons between contraction velocities of cleft and noncleft anatomy.

Methods/Description: This study included 27 Caucasian English-speaking adults. Participants in the noncleft group included 11 males and 11 females between the ages of 19 and 33 years (M = 22.8, SD = 4.1). Five participants (2 males and 3 females) between the ages of 19 and 36 (M = 26.6, SD = 7.1) with a history of repaired cleft palate were recruited. All participants had velopharyngeal competence and normal speech characteristics. Dynamic MRI images were obtained in the oblique coronal planes during production of "ansa" using the FLASH multishot technique and sliding window reconstruction to achieve the frame rate of 30 fps (Sutton et al., 2010). Amira 6.0.1 Visualization and Volume Modeling Software and MATLAB were utilized for image and data analysis.

Results: Significant predictors (P < .05) of LVP shortening during velopharyngeal closure include mean extravelar length, origin-to-origin distance, hard palate length, and velar knee to posterior pharyngeal wall distance. Intravelar segment was the only significant predictor of maximum contraction velocity during velopharyngeal closure. No significant sex differences were found (P > .05). No significant difference (P > .05) in the LVP muscle shortening and maximum contraction velocity was found between the cleft population with velopharyngeal competence and noncleft population.

Conclusions: This is the first study to explore sex differences in LVP contraction velocity, as well as differences in velocity between the cleft and noncleft population. This study identified 5 predictors of LVP contraction and can be useful for predicting LVP function when dynamic MRI is not readily available.

## 254. Influence of Voice Focus Adjustments on Hypernasal Speakers' Oral-Nasal Balance in Speech and Singing

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Background/Purpose: It is difficult to teach speakers with hypernasality related to cleft palate how to improve the closure of their velopharyngeal sphincter because conscious proprioception is limited. The present research investigated in how far global voice focus adjustments can help speakers lower their nasalance scores. Forward voice focus is characterized by a raised larynx, a protruded tongue, and a narrowed pharynx. Backward voice focus is characterized by a lowered larynx, a retracted tongue, and a widened pharynx. Three previous studies on normal speakers (de Boer & Bressmann, 2016; de Boer et al., 2016; Santoni et al., in press) have indicated that forward voice focus increases nasalance scores and backward voice focus decreases nasalance scores in both speech and song. The present study investigated the effect of voice focus adjustments on the oral–nasal balance of hypernasal speakers.

*Methods/Description*: Using both forward and backward voice focus adjustments, seven hypernasal speakers (3 males/4 females) aged 5-12 years (SD 2.91) repeated phonetically balanced, nasal and oral sentences, and sang a short musical passage. Nasalance scores were collected with the Nasometer 6450.

Results: From the average baseline of 28.95% (SD: 10.47) for the oral sentence, nasalance scores increased to 37.14% (SD: 17.07) in the forward and decreased to 27.45% (SD: 8.53) in the backward focus condition (biggest individual reductions of -11% for forward and -10% for backward focus). From the average baseline of 64.81%(SD: 3.77) for the nasal sentence, nasalance scores increased to 64.86% (SD: 12.08) in the forward and decreased to 54.05% (SD: 11.97) in the backward focus condition (biggest individual reductions of -22% for forward and -32% for backward focus). From the average baseline of 50.67% (SD: 5.99) for the balanced sentence, nasalance scores increased to 55.05% (SD: 10.18) in the forward and decreased to 45.29% (SD: 7.69) in the backward focus condition (biggest individual reductions of -8% for forward and -11% for backward focus). From the average baseline of 41.50% (SD: 7.64) for the song, nasalance scores increased to 59.17% (SD: 2.79) in the forward and decreased to 39.17% (SD: 18.13) in the backward focus condition (biggest individual reduction of -11% for backward focus).

Conclusions: For the group, forward focus resulted in higher and backward focus resulted in lower nasalance scores. There were exceptions: 2 participants produced lower scores in the forward and 1 participant produced higher scores in the backward focus condition. The pattern of results for the singing task was similar to that of the balanced speaking task. The study demonstrated that voice focus adjustments may have potential as a therapy technique for hypernasal speakers. Future research should include nasopharyngoscopic or videofluoroscopic imaging to visualize the effect of voice focus adjustments on the velopharynx. The longer term effectiveness of the intervention as a therapy technique should also be investigated.

## 255. Is Velopharyngeal Ratio Really an Accurate Clinical Determinant of Velopharyngeal Function?

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Background/Purpose: Velopharyngeal (VP) ratios are continually used to evaluate normal VP anatomy and normal VP function. VP ratios are calculated by dividing the entire velar length by the pharyngeal depth. Individuals with normal anatomy present with VP ratios ranging from 1.2 to 1.43 (Subtelny, 1957; Tian et al., 2010). Individuals presenting with a VP ratio below the average range have previously been thought to have a disadvantageous VP mechanism which could lead to VP incompetence (VPI). However, Perry et al. (2018) reported individuals with a repaired cleft palate and normal resonance with a VP ratio of 0.7 to 1.21. This suggests that VP ratios alone cannot explain acceptable VP function for speech. Tian and Redett (2009) introduced the idea that an effective VP ratio may be a more appropriate indicator of normal parameters for speech. This ratio is calculated with the effective velar length, the distance from the posterior nasal spine to the velar eminence and includes the region of the

velum that is activated during speech. The aims of this study are to examine if the VP ratio is preserved across the age span or if it varies with changes in the VP portal and to analyze if the effective VP ratio is more stable across the age span.

Methods/Description: Magnetic resonance imaging (MRI) was used to analyze velopharyngeal anatomy of 271 participants. Based on cranial growth rate, the participants were divided into the following groups for statistical analysis: infants, children, adolescents, and adults. The VP ratio and effective VP ratio were calculated using previously described methods. ANOVAs and a Games-Howell post hoc test were used to compare variables between groups.

Results: There was a statistically significant difference (P < .05) in all measurements between the 4 age groups. Pairwise comparisons reported statistically significant adjacent group differences (P < .05) for velar length, VP ratio, effective velar length, adenoid depth, and pharyngeal depth. No statistically significant differences between adjacent age groups was reported for effective VP ratio. Visually, velar length was noted to increase significantly from infancy to adulthood, while effective velar length grows at a slower rate, with minimal changes from adolescents to adults.

Conclusions: This study provides insights into differences in effective VP ratio and VP ratio across the age span. Effective VP ratio was not statistically significant between adjacent age groups, while VP ratio was statistically significant between adjacent age groups from the infant to child group and adolescent to adult. Therefore, the VP ratio changes significantly more across the age span in comparison to effective VP ratio. This may be due to velar length increasing in length throughout life, while the effective velar length does not experience a large increase in growth following adolescents. This study suggests that effective VP ratio is more correlated with VP function than VP ratios.

#### 256. Phoneme Specific Nasal Emission and Phoneme-Specific Hypernasality in Patients With and Without Structural Disorders of the Velopharyngeal Mechanism

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Background/Purpose: Compensatory speech sound errors are frequently associated with velopharyngeal dysfunction and palatal anomalies. However, compensatory speech errors can also be observed in patients who do not have a history of cleft palate—patients with velopharyngeal mislearning. A unique error that frequently arises in these cases involves phoneme-specific nasal air emission (PSNE) and phoneme-specific hypernasality (PSH). The purpose of this study is to describe the occurrence of PSNE and PSH in patients referred to a craniofacial center and to examine the relationship of this speech behavior to physical findings and to coexisting phonologic processes and the differences in patterns of occurrence of PSNE and PSH between patients with and without structural disorders of the velopharyngeal mechanism.

*Methods/Description*: In this retrospective case analysis, 1252 patients were grouped into 3 groups: group A—patients with overt palatal anomalies (n=350) having cleft lip and palate (n=126) or cleft palate only (n=126), group B patients with occult anomalies of velopharyngeal mechanism (n=736) having occult submucous cleft palate (n=345), submucous cleft palatal (n=167), and patients with velopharyngeal insufficiency but no sign of palatal cleft anomaly (n=226), and group C—patients with PSNE and PSH in the absence of

structural anomaly of the velopharyngeal sphincter (n = 166). Perceptual VPD tests including Hebrew articulation and phonological assessment following endoscopy were conducted. Differences were analyzed by  $\chi^2$  test.

Results: Thirty-six percent of the studied patients had phonological disorders. Thirty-five percent of patients in groups A and B had compensatory articulation disorders. Patients in group A with compensatory disorders had significantly (P < .006) lower prevalence of phonological errors (49%) in comparison to the prevalence of phonological errors (63%) in patients with compensatory articulation disorders in group B. In group A, 6% had PSNE, of which 53% had phonological errors; 2% had PSH, of which 25% had phonological errors. In group B, 4% had PSNE, of which 25% had phonological errors, and 3% had PSH, of which 64% had phonological errors. In group C, 56% had PSNE, of which 42% had phonological errors, and 44% had PSH, of which 42% had phonological errors. Patients in group C had significantly (P < .000) higher prevalence of atypical backing phonological error (22%) in comparison to 18% in group B, and no backing error in group A.

Conclusions: The current study provides additional data about the etiology and the use of phoneme-specific nasal resonance and/or nasal air emission. Our results about relationship between phonology errors and compensation disorders in relation to PSNE and PSH in patients with and without structural disorders of the velopharyngeal mechanism can help clarify the etiology of this specific speech disorder as an articulatory structural constraint or an underlying phonological/organizational disorder. Understanding the etiology of PSNE and PSH can affect treatment options for patients.

# 257. Evaluation of the Levator Veli Palatini Muscle Asymmetry in Noncleft Population Using MRI and Correlation to Nasopharyngoscopy Findings

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Background/Purpose: The orientation and position of the levator veli palatini (levator) muscle bundles are significantly altered in children born with cleft palate (Perry et al., 2014a). Having insight about the levator muscle morphology is critical for improving knowledge about typical and atypical function of the velopharyngeal (VP) mechanism (Perry et al., 2013). Studies have quantified the levator length and thickness in the right and left muscle bundles and demonstrated asymmetry in cleft anatomy. However, studies were limited by sample size (less than 10 subjects) and the effect of sex has not been considered. Also, the typical degree of asymmetry for the length and thickness of the levator muscle in noncleft anatomy is undetermined. The first and second aims of the study were to measure the right and left length and thickness of the levator muscle bundles in males and females and determine the typical degree of asymmetry. Findings about the levator muscle features are important to understand how variations in this muscle impact VP closure. Asymmetrical VP closure has been reported to be observed in typical anatomy (Moreira et al., 2008) without VP dysfunction. No study has examined the correlation between the levator muscles findings through MRI to the function of the VP closure through nasopharyngoscopy. The third aim was to explore the relationship between the anatomic findings and VP closure function.

*Methods/Description*: The participants included 89 adults (46 females, 43 males) with typical VP anatomy between 18 and 36 years of age. Participants were imaged through MRI at rest and analyzed using Amira 5.5 software. Flexible nasopharyngoscopy was used to determine the function of the VP closure. Matched paired T tests (P < .05)

were conducted to compare differences in the right and left levator length and thickness in males and females separately. The spearman correlation analysis was administered to determine the correlation between MRI findings and nasopharyngoscopy observations. To investigate intra-rater reliability, all subjects for nasopharyngoscopy and 40 subjects for MRI were randomly selected to repeat measurements

Results: Matched paired t test revealed nonsignificant differences (P = .36, P = .09) between the mean length and thickness of the right and left levator muscle bundles for male and female groups. A nonsignificant negative correlation ( $\rho = -0.106$ , P = .52) was found between the function of VP closure and MRI findings.

Conclusions: Although the differences of the levator muscle bundles features were minimal, the range highlights that typical anatomy can be associated with a sizeable (up to 10 mm) difference in the levator muscle length. Also, findings suggest observations through nasopharyngoscopy do not correlate with the longer length of the levator muscle from MRI.

### 258. The Speech Outcome Evidence Base and the Speech Pathologist Reviewer

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Background/Purpose: Research in velopharyngeal function dating from 1986 has shown that compensatory articulations including glottal stops, pharyngeal fricatives, and nasal fricatives are produced with incomplete velopharyngeal closure, even when the velopharynx is capable of complete closure with appropriate articulations. Further studies showed that patent oronasal fistulas can influence velopharyngeal movement and closure. Therefore, valid assessment of velopharyngeal function, perceptual or instrumental, should control for both fistulas and compensatory articulations. Failure to do so could cause invalid assessment leading to unnecessary surgery or inadequate speech therapy. Studies that do not stipulate exclusion of compensatory articulations or fistulas may be of questionable validity. At ACPA 2018, we reported the result of a literature sample of 158 published articles from 42 journals to evaluate how well the literature on velopharyngeal assessment or management outcome stated control for fistulas and compensatory articulations. Only 6% of the articles stated control for both fistulas and compensatory articulations; 12% controlled for one or the other, but not both. Thirty-seven percent described speech sample validity by describing the speech pathologist (ie, "experienced"); 24% made no statement concerning the speech sample used. Were these articles peer reviewed by speech-language pathologists (SLPs) who may be more sensitive to speech sample validity? The purpose of this study was to determine how many of the journals from the prior study used SLPs as peer reviewers.

Methods/Description: The editorial staffs of the 42 identified journals were contacted with a standard e-mail message to inquire if a submitted manuscript concerning cleft palate speech or surgical speech outcome would be peer reviewed by an SLP. Responses were recorded in both Word logs and Excel database. Nonresponding journals were sent follow-up queries, with 3 standard queries sent over a period of 5 months.

Results: Sixteen journals (38%) did not respond, or declined to give information, including 5 journals that have either been discontinued or have merged with other publications. Nine journals (21%) confirmed SLP reviewers; 9 journals (21%) stated possible use of SLPs. Four journals (9.5%) responded unlikely use of SLP reviewers, and another 4 (9.5%) reported that they do not use SLPs. The articles that

described full speech sample validity came from just 4 journals: 2 which confirmed SLP peer reviewers, 1 stated likely use of SLPs, and 1 nonresponsive journal.

Conclusions: Fewer than half of the 42 journals that have published cleft palate/velopharyngeal speech assessment or outcome articles use, or possibly use, SLPs as peer reviewers. These results suggest that discernment and caution may be valuable tools for the reader of the evidence base concerning speech and surgical outcomes in children with cleft palate and velopharyngeal dysfunction.

### 259. System Identification of Velopharyngeal Closure and Tongue Elevation in Fricative Sound Articulation

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Background/Purpose: Speech-language-hearing therapists usually communicate with the patient to collect important information such as sound, voice meaning, expression, and physical condition. They examine the patient's overall articulation attitude (eg, listening to the voice while observing the mouth). They observe both voice and mouth clarity to determine the states of articulation disorders. In addition, if they have knowledge of the aeroacoustic mechanism of acoustic articulation, they may be able to make better decisions based on the physics. Fricative sound /s/ in a VCV sequence is produced by 2 main musculoskeletal activities, which is occasionally a syllable to be clinically treated. (1) Velopharyngeal closure is a musculoskeletal function that changes the topology of the vocal tract from a branched channel to a single tube. (2) A movement of the tongue and lower jaw changes the shape of the oral cavity. Both of these activities change the shape of the vocal tract. This causes 2 major physical changes (resonance and airflow effects). To elucidate the effects of tongue and soft palate elevation speed and glottic flow control on fricative sound /s/ production in a VCV sequence, this study conducted experimental measurements using a simplified vocal tract model.

Methods/Description: A simplified model was constructed based on 5 cross-sectional shapes of the vocal tract of /s/. The tongue ascent rate and volumetric flow rate at the model's glottal entrance were controlled using a stepping motor and an electromagnetic proportional control valve, respectively. The flow velocity in the gap between the incisors and the sound propagating from the model was measured simultaneously with a hot-wire anemometer and a microphone.

Results: By controlling the tongue ascent rate and flow rate in the simplified model, the spectrogram of /s/ was reproduced. As a result of the flow measurement, it was shown that velocity fluctuation and sound propagation started almost simultaneously with the reproduction of the spectrogram. In contrast, the soft palate had to be raised prior to tongue elevation to achieve velopharyngeal closure. By reducing the rate of tongue elevation under conditions where velopharyngeal closure was achieved, velocity fluctuations preceded the appearance of sound production. This indicates that the tongue rising speed has an influence on the control of sound source generation of /s/ in the word pronunciations.

Conclusions: When producing syllables containing fricative sounds, velopharyngeal closure needs to be achieved prior to the tongue rising. If velopharyngeal closure is achieved, raising the tip of the tongue to the front of the palate results in /s/. Meanwhile, the elevation speed

affects the spectral characteristics of /s/ and the continuity with the pronunciation of subsequent vowels.

## 260. Administrative Steps in the Execution of Surgical Outreach Programmes for Facial Cleft Repair: Experience of an Indigenous Foundation

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Background/Purpose: Facial cleft is a congenital malformation of the face, an anomaly that is recorded everywhere in the world. The etiology is complex and likely to have both genetic and environmental factors. It is recommended that surgical repairs of the lip and palate be carried out in the early age of life. This is a mirage in the developing world due to poverty and dearth of professionals; therefore, medical outreach programs mainly by foreigners has been the major source of assistance to poor victims of the disease in developing countries. Most of the medical outreach interventions focus mainly on the clinical aspect, with little or infinitesimal attention being given to administrative aspect involved. This paper hopes to highlight the administrative steps involved in the administration of medical outreaches in Nigeria, as experienced by an indigenous organization, Cleft and Facial Deformity Foundation (CFDF). This could serve as a guide for interested individuals who might want to venture into the business of surgical outreach programs.

Methods/Description: The administrative steps are broadly divided into 3: premedical outreach, medical outreach, and postmedical outreach activities. (1) Premedical outreach activities—planning: project timelines activities, budget; organizing stakeholders meetings; information management; procurement management; logistical arrangement. (2) Medical outreach activities proper—logistical arrangement; data collection and processing; screening of patients; arrangement of laboratory investigations; assigning responsibilities to participants. (3) Postmedical outreach activities—review of the outreach activities; data analysis; project report; establish communications with operated patients.

# 261. Administrative Challenges of Organising Surgical Missions for the Care of Cleft Lip and Palate in Nigeria: Eight Year Experience of Cleft & Facial Deformity Foundation, Nigeria

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Background/Purpose: Facial clefts are congenital malformation of the face, whose etiology is unknown but could be genetic or environmental. Due to poverty and dearth of surgical specialists, the care of the affected patients in Nigeria and other developing countries has essentially been through surgical missions. Majority of the surgical missions in Nigeria have been by foreigners, but in recent time, indigenous surgical foundations are springing up, including Abujabased Cleft and Facial Deformity Foundation. Over the years, administration has played important roles in the progress and success of an organization; these roles includes the ability to develop administrative procedures, administrative budget, planning, and control. Administration is one of the key elements associated with high-level productivity and efficiency. Sometimes, it is thought that the role of administration is not important in an organization and thereby neglected. The involvement of administrative activities in surgical mission is as important

as the surgical activities, to the success of the program. Most studies and presentation have always been on clinical aspect of surgical missions rather than administrative involvement; this seeks to outline administrative challenges of surgical missions and how to mitigate them, with a view to educate practitioners on the field of surgical missions.

Methods/Description: Administrative challenges in organizing surgical missions in Nigeria: poor cooperation from host communities, government bureaucracy, delay of supply of medical equipment and consumables, staff/volunteer welfare, traditional/religious beliefs, security of people and materials, poor terrain, accident, and inadequate funding. The foundation has been able to mitigate against these challenges with successful surgical missions for the past 8 years, having a record of 23 medical missions conducted where 1634 patients have benefited. These challenges were mitigated by ensuring proper authority before every activity is carried out, timely notifications of institutions involved, seeking relevant security information about outreach location, timely procurement process, provision for alternative/multiple suppliers, emphasize the importance of the project in the various communities, ensure adequate provision in the budget, and provide security briefing for staff before moving to communities. Others include provision of safety facilities, insurance policy on properties, seeking adequate information and appropriate travel plan, adhere strictly to all safety regulations, engagement of religious and traditional leaders, and ensure proper enlightenment about the scourge across religious and ethnic backgrounds. Constant review of administrative police guideline by the foundation over the years has been able to effectively achieve result.

### 262. #Cleftlip/Palate: What is the World Talking About?

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Background/Purpose: Social media has infiltrated all aspects of society, including the medical community. Studies have begun analyzing how the world converses on social media platforms about different medical and surgical topics. No study to date has examined how cleft lip and palate, 2 of the most common birth defects in the world, are discussed on Twitter.

*Methods/Description*: Tweets were identified by searching Twitter's official website, using any of the following keywords or hashtags: cleft, cleft lip, cleft palate, #cleft, #cleftlip, #cleftpalate. Eight months between 2017 and 2018 were analyzed.

Results: A total of 1222 tweets were included. #Cleft was the most common hashtag (71%), and it was significantly associated with more retweets (P = .03). Twenty-seven countries tweeted, with the United States (34%) and India (27%) producing the most. Charities (36%), hospitals (14%), and physicians (13%) were the most common authors. Oral maxillofacial surgeons produced the most tweets out of all physicians (7%). Over three quarters of tweets were selfpromotional. The top content included charity information (22%), patients' cleft stories (14%), and surgical service trips (13%). The accuracy of educational tweets (n = 309) was 61% high quality, 38% low accuracy, and 1% inaccurate. One hundred forty-nine tweets (12%) discussed a published research article, but 41 of these tweets did not share a link to read the article. Links to blog posts (29%), links to websites (21%), charity images (21%), unrepaired cleft lip/palate images (14%), and repaired cleft lip/palate images (13%) were the most common multimedia. A minority of tweets ( $\leq 4\%$ ) was also shared on other social media platforms.

Conclusions: Charities dominate the cleft lip/palate "Twitterverse." Physicians make up a minority of contributors. Most tweets were self-promotional, and over a third of the tweets that were educational were low accuracy. The ethical debate of sharing patient images on social media remains highly relevant to the cleft lip/palate world, as pre- and postoperative images were in the top 5 multimedia shared. There is an opportunity for more health professionals to engage on Twitter and to contribute more accurate education on this important topic, including sharing published research articles (along with the link to read the article). As the cleft social media community continues to grow, we recommend the use of the hashtag #cleft as well as sharing tweets across multiple social media platforms in order to reach a wider audience.

## 263. Clinical Dashboards for Monitoring Feeding, Nutrition, and Growth in Infants With Clefts of the Lip and Palate: An International Collaboration

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Background/Purpose: Infants with clefts of the lip and palate are at increased risk of feeding difficulties and malnutrition and require close monitoring of growth and feeding in the first 12 months. Identification of those infants who demonstrate weight faltering allows for early implementation of interventions to optimize nutrition. Additionally, families are dealing with the impact of social, economic, and environmental factors influencing a child's health status. These family characteristics must also be identified to increase the success of any intervention to treat weight faltering. Our objective was to develop a sustainable method to monitor growth and nutrition in infants with orofacial clefting and allow for identification of the characteristics of infants with orofacial clefts who require intervention to optimize nutrition.

Methods/Description: A craniofacial nurse, dietitian, feeding therapist, pediatrician, social worker, and speech-language pathologist reviewed the data typically collected in the medical documentation for infants with clefting. We held a series of focus groups to identify the data elements that are most useful for population monitoring as well as those used to identify children at risk for malnutrition. We cross referenced metrics used in published studies of growth and nutrition in infants. We then created a dashboard using data from a research study population of 45 infants enrolled at our institution in the infants with clefts: Caregiver Observation of Outcomes study. Data are displayed in a simple, reproducible Excel format using pivot tables and graphs, as well as an Access database in CRUX system at an international site.

Results: We review the data elements selected for the dashboard and data collection procedures. We present visual displays for descriptive statistics on individual and population metrics, including trends for weight, Z score for weight, weight for length, number of days to return to birth weight, and diagnosis of malnutrition using Aspen research-based criteria. The dashboard includes metrics for health-care interventions, such as feeding method, percentage of children seen by a dietitian, feeding therapist and/or lactation consultant, need for increased caloric density, supplemental feeding tube, and/or medical hospitalization. We will share data elements and methods for data collection in simple formats, including Excel and RedCap.

Conclusion: We created a method for tracking clinically relevant characteristics in a population of infants with orofacial clefting to facilitate identification of children at risk for malnutrition. Ultimately, we hope that this tool will facilitate improved recognition of infants who demonstrate growth faltering. We also hope to use this tool to facilitate future hypothesis-driven, practice-based research and quality improvement efforts by members of the multidisciplinary team.

## 264. Breastfeeding and Breast Milk Feeding Experiences of Mothers of Children With a Cleft Lip and/or Cleft Palate

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Background/Purpose: Breastfeeding has numerous far-reaching benefits in early life. Children with oral clefts are often unable to feed exclusively at the breast, yet little is known about the mother's early experiences with feeding-related stress and breast milk feeding that does not occur at the breast. We set out to learn about the extent, motivation, and stress related to breastfeeding and breast milk feeding of mothers of infants with a cleft lip only (CLO) or with a cleft palate only or cleft lip with palate (CP/CLP).

Methods/Description: We conducted a case series study among mothers of infants with CLO or CP/CLP. We administered a web-based survey on feeding experiences in the infant's first 4 months of life and abstracted the chart for clinical details. Mothers of infants with CLO or CP/CLP up to 36 months of age were eligible to participate in the study. We generated descriptive statistics and compared experiences between infants with CLO and CP/CLP using t tests, Fisher exact, and  $\chi^2$  tests. We stratified findings based on CLO and CP/CLP because we anticipated cleft palate involvement would have a substantial effect on feeding. Breast milk feeding was defined as feeding a baby milk from the mother's breast using a method other than at-breast.

Results: Our sample included 75 mothers and had a response rate of 30%. Over 40% of mothers were >35 years old, had a 4-year degree or greater, and no prior live births. Among infants, 61% were male, 81% were white, 41% had a CLO, and 59% had a CP/CLP. Most mothers (84%) intended to exclusively breastfeed prior to learning of their child's diagnosis and 77\% were disappointed when they learned they may not be able to do so. Based on a 0 to 100 stress scale, feedingrelated stress was higher among mothers of an infant with CP/CLP than CLO at week 1 (78.8 vs 41.5, P < .001) and at week 4 (41.0 vs 32.4, P < .001) after the infant's birth. When the infant was 1 week old, mothers of infants with a CLO versus CP/CLP had higher prevalence of any breastfeeding (71% vs 12%) and exclusive breastfeeding (52% vs 2%). At 16 weeks, mothers of infants with CLO versus CP/CLP also had higher prevalence of any breastfeeding (61% vs 7%) and exclusive breastfeeding (35% vs 2%). The number of mothers predominantly breast milk feeding their infant decreased between 1 and 16 weeks for infants with CLO (42%-30%) and infants with CP/CLP (77%-64%). Fewer mothers of infants with CLO versus CP/CLP ever used a breast pump (74% vs 96%, P = .01). More mothers with CP/CLP versus CLO used a hospital grade pump (50% vs 26%, P = .06).

Conclusions: There are opportunities to improve access to mother's own breastmilk for infants with CLO and CP/CLP. Mothers of infants with CLO may benefit from strategies to improve duration and exclusivity of breastfeeding. Mothers of infants with CP/CLP may benefit from strategies that increase their ability to provide breast milk, including obtaining a hospital grade pump and providing additional support throughout the first months after birth.

### 265. Introduction of Solids for Infants With Unrepaired Clefts

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Background/Purpose: There is ample evidence-based information regarding bottle feeding an infant with an orofacial cleft, but comparatively little research is available for introduction of solids in the unrepaired cleft population. Therefore, families are left without standard guidelines regarding introduction of solids as well as cup drinking, leading some families to delay trials of a variety of tastes and textures until after the palate is repaired. These reduced exposures of solids during appropriate developmental stages may result in oral aversions and possible delays in oral motor skills necessary for eating and drinking. Practitioners often have similar questions about the progression of feeding and would benefit from some uniform guidelines for feeding older infants and toddlers with unrepaired clefts so that they can better educate families (SLPs, OTs, pediatricians, nurses, inter-team vs intra-team consistency, etc).

Methods/Description: A review of literature was completed to compile information for introduction of solids in the typical population for the age range in question. These results were then incorporated with evidenced-based knowledge of the cleft population to create recommendations. A family-friendly handout was developed to share this information with patients in a consistent manner.

### 266. Determination of Ethnic Variation in Infant Nasolabial Anthropometry Using 3D Photographs

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Background/Purpose: The goal of a synchronous bilateral cleft lip/nasal correction is to achieve normal dimensions and growth for the nasolabial subunits. The majority of work done to design this operation has been derived from normative anthropometric data published by Farkas et al. These data represent normative anthropometric measurements of Caucasian children. We aimed to asses if infant's nasolabial anthropometry exhibited significant ethnic variabilities, which would motivate variations in surgical correction. Specifically, we sought to investigate a common assertion that a long columella is a Caucasian feature, and therefore, accepting a short columella and/or delayed columella lengthening is a suitable strategy, particularly in ethnic patients.

Methods/Description: Thirty-three pediatric infants without craniofacial pathology (10 African American [AA], 7 Hispanic [H], and 16 Caucasians [C]), ages 3 to 8 months, presenting to the Johns Hopkins All Children's General Pediatric Clinic were recruited. Four separate 3D photographs (2 submental and 2 frontal) were taken using the Vectra-H1 handheld camera. Significant differences were noted between nasolabial anthropometry for African American patient. Eighteen linear facial distances were measured using Mirror 3D analysis. ANOVA coefficients with the Bonferroni post hoc comparisons, at  $\alpha=.05$ , were used to measure significant differences between ethnic groups. ANCOVA adjusted for sex was also used. Pearson correlation was used to determine intra- and inter-rater reliability. All statistical analyses were carried out using SPSS version 21.0, with statistical significance set at P < .05.

Results: Significant differences were seen between ethnic groups in nasal width (sbal-sbal [C-AA, P = .02]; ac-ac [C-AA, P = .00; H-AA, P = .04), and al-al [C-AA, P = .00; H-AA, P = .001]), as well as

labial length (sn-ls [C-AA, P=.041); sn-sto [C-AA, P=.005]; Cphs-Cphi L [C-AA, P=.015] were noted between ethnic groups. African American infants exhibiting wider noses and longer lips and Hispanic infants representing intermediate values between Caucasian infants and African American infants. Nasal projection (sn-prn) (P=.974) and columella length (sn-c) (P=.99) did not differ significantly between groups. Correlation coefficients for both intra- and inter-rater reliability were good to excellent and were significantly correlated for all 18 measures.

Conclusions: Significant differences were noted between nasolabial anthropometry for African American patient and Caucasian infants. Nasal width and lip length were greater in African American patients. Our study demonstrates that slightly wider intra-alar differences may be tolerated in ethnic patients and as much lip length should be preserved for all patients. No difference was noted in nasal projection and columella length, indicating that these structures should be corrected during the primary cleft lip and nasal repair for all patients and should not be differed to secondary correction.

## 267. 10 Year Outcome Analysis of Minimally Invasive Secondary Cleft Lip Reconstruction Using Autologous Fat Grafting

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Background/Purpose: Secondary cleft lip deformities are frequent following primary cleft lip repair. We have been performing minimally invasive, secondary cleft lip reconstruction for the last 10 years. The objective of this study is to present our long-term results and to identify types of cleft lip secondary deformity which achieve a long-lasting benefit from this reconstructive technique.

Methods/Description: We performed a retrospective analysis from January 1, 2008, to January 1, 2018, of all cases of secondary cleft lip revision using fat grafting techniques. We identified 549 cases with at least 12-month follow-up. Patients were examined for type of cleft, age sex, location of secondary deformity, fat graft type and location, amount of fat injected, percentage of fat graft survival, speech or feeding difficulties, and long-term need for additional secondary lip repair. Overall morbidity and mortality were also examined for the lip and fat donor site. Fat grafting was successfully performed in 100% of cases. There was no mortality or wound infections noted. Mild bruising in the donor was seen in 95% of patients. This resolved in less than 7 days. Lip edema was noted in 100% of patients but did not result in any problems with speech or oral intake. Two percent (12/549) of patients had a greater than 50% loss in the graft after 12 months which required additional grafting. The best fat graft improvement was noted in the vermilion tubercle, followed by the lip vermilion lateral to the cleft repair. One percent of patients (5/549) had excess fat growth in the lip following weight gain, which required ultrasonic liposuction to remove fat from the lip several years following the initial grafting. Secondary open lip revision was required in 4% of patients who requested additional improvement in lip contour. No increase in scaring was noted, and in fact 92% of patients reported a sense that their lip scar was improved overall by the operation. Ninety-eight percent of patients were pleased with the overall result. This long-term study shows that fat grafting can be successfully performed in the cleft lip for the reconstruction of the cleft lip secondary deformity. The grafted fat lasts for long term and in some cases can exhibit overgrowth in weight gain. The risk of this procedure is low with a high long-term rate of patient satisfaction. Fat grafting should be considered when long-term correction of the secondary lip deformity via a minimally invasive approach is desired.

## 268. The Unilateral Cleft Lip Nasal Deformity (Uclnd) Revisited: Uncovering Fundamental Misconceptions Using 3D Image Analysis

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Background/Purpose: Current models of the uCLND are based upon subjective observations and study of a limited numbers of cadavers. The various descriptions are confusing, contradictory, and often lack objective data to validate them. Given that optimal treatment of the uCLND remains a challenge, a better understanding of the deformity is critical to devising ways to improve care. The purpose of this study was to develop a model of the uCLND that is based upon objective 3D data and that spans the spectrum of presentation.

Methods/Description: We assessed landmark displacements, anthropometric dimensions, and shape-base measures on 3D images infants with unrepaired unilateral cleft lip at 6 months of age (N=100). Lateral displacement of subnasale (sn), the surface landmark for the anterior nasal spine and caudal septum, had the greatest magnitude aberration of any measure and was therefore used as the independent variable in a linear regression analysis model. In order to accommodate for age-related alterations, we corrected for the age at image capture. Significance was assessed by ANOVA with P < .05 considered significant.

Results: Significant changes were expressed for each 1 mm deviation of sn from midline (P < .05). Nose: The columella tilts  $4.2^{\circ}$ , the cleft nostril widens (1.1 mm), and, to a lesser degree, the nose broadens (0.7 mm). The noncleft alar base is driven lateral, away from midline (0.8) mm), while the cleft alar base gets left behind and migrates posteriorly (0.6 mm). The nasal dorsum follows sn deviation, swinging 1.8° from midline, toward the noncleft side. The noncleft alar base does not deviate as much as subnasale resulting in constriction of the noncleft nasal sill (-0.2 mm). Collapse of the dome occurs progressively with these changes with measureable alterations in the nose tip volume ration (+0.15) and alar-cheek definition (+2.09). Lip: The entire philtrum deviates away from midline, toward the noncleft side. The lip heights adjacent to the cleft diminish (-0.2 mm) and the lateral lip lengths shorten (-0.3 mm). Meanwhile, the noncleft medial lip height grows (+0.2 mm). Face: Facial height and width do not change, however, interendocanthal distance widens (+0.4 mm).

Conclusions: Deviation of sn can be used as a measure of cleft severity to produce a progressive model of the uCLND and associated changes. Contrary to previous descriptions, there is no normal side, the cleft alar base is not displaced lateral, and the nasal dorsum does not deviate toward the cleft. Rather, displacement of the noncleft alar base, deviation of the dorsum that follows sn, and retrusion of the cleft alar base are associated with progressive collapse of the nasal form. Based on these data, treatment should aim to centralize the columella and balance the alar base relationships by moving the noncleft alar base medial and the cleft alar base anterior. Correction of the "twist" in the nasal foundation may be critical to long-term stability of primary repair.

## 269. Analysis of National Outcomes for Outpatient Versus Inpatient Cleft Lip Surgery

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Background/Purpose: Several peer-reviewed studies over the past decade have demonstrated safety of outpatient cleft lip repair in

patients without cardiopulmonary risk factors. The purpose of this study is to analyze national trends of outpatient cleft lip repair and its corresponding safety profile.

*Methods/Description*: A retrospective cohort study was conducted of primary cleft lip repair performed in the United States between 2012 and 2017 using the ACS NSQIP-P data set. Pearson  $\chi^2$  test and binary multivariate logistic regression were used to analyze categorical variables. Mann-Whitney U test and the Hodges-Lehman method were used to analyze nonparametric linear variables.

Results: Patients undergoing cleft lip repair as outpatients were significantly older (P < .001, 95% CI: 66-86), weighed more (P < .001, median difference: 3.44 lbs, 95% CI: 3.04-3.92), and were significantly taller (P < .001, median difference: 2.21 in, 95% CI: 2.00-2.54). Surgical procedure time was significantly longer for inpatients (P < .001, median difference: 15 minutes, 95% CI: 13-18). The proportion of procedures performed outpatient over the study period was stable (P = .954), with 55.7% (n = 505 of 906) in 2012 to 56.2% (n = 505 of 906) in 2012 to 56.2% (n = 505897 of 1597) in 2017 performed outpatient. Risk factors including asthma (P = .015), pulmonary abnormality (P = .046), digestive system abnormality (P < .001), cardiac risk factors (P < .001), seizure disorder (P = .024), central nervous system abnormality (P < .001), and supplemental nutritional requirement (P = .031) were significantly overrepresented in cleft lip repairs with inpatient admissions. Patients without any risk factors were significantly (P = .001) more likely to be treated outpatient at 57.10% (n = 3089 of 5411) than be admitted overnight postoperatively. Complications were significantly (P = .002) higher for inpatients (1.50%, n = 60 of 3892) than for outpatients (0.80\%, n = 41 of 4894). Readmission was significantly (P < .001) lower in the outpatient cohort (2.60%, n = 128 of 4894) compared to the inpatient cohort (4.70\%, n = 183 of 3892), such that inpatients were 1.83 times (95\% CI: 1.50-2.31) more likely to be readmitted in 30 days for any reason. Over the years, the readmission rate overall has significantly (P < .001) decreased from 4.90% (n = 44of 906) in 2011 to 2.30% (n = 37 of 1597) in 2017. Likewise, the readmission rate in outpatients has significantly (P = .001) decreased from 4.40% (n = 22 of 505) in 2012 to 1.3% (n = 12 of 897) in 2017. Conclusions: Despite the published safety of outpatient surgery for

Conclusions: Despite the published safety of outpatient surgery for cleft lip repair, the percentage of patients treated as outpatients has remained stable from 2012 to 2017. Complication rates and readmission rates appear to be declining slightly, perhaps indicating that surgeons are appropriately risk-stratifying patients for inpatient surgery. Nevertheless, there may be additional cleft lip patients that could be candidates for outpatient surgery.

## 270. Does Revision Palatoplasty Have an Increased Risk of Complication Compared to Primary Palatal Repair?

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Background/Purpose: For patients with cleft palate, primary palatal repair is typically performed by age 1. Patients with challenging cleft morphologies are at increased risk of developing fistulas and persistent velopharyngeal insufficiency later in life. Presently, it is unclear if these patients continue to experience poorer outcomes even after secondary repair. Therefore, the purpose of this study was to determine if revision palatoplasty was associated with increased rates of postoperative complication and fistula compared to primary palatal repair.

Methods/Description: This was a retrospective cohort study of patients with cleft palate undergoing palatoplasty. The records were obtained from the Kids' Inpatient Database between 2000 and 2014. The primary predictor was surgery type and was classified as either primary or revision using ICD-9-PCS code terminology. Secondary predictors included demographics, congenital comorbidities (cardiovascular, neurological, chromosomal), and cleft characteristics (lip involvement, laterality, completeness). The study outcomes were the post-operative complication and fistula rates during the admission. Univariate logistical regression was performed to identify relationships between predictors and study outcomes. To calculated adjusted associations, multiple logistical regression models for complications and fistulas were created.

Results: A total of 13 931 admissions were identified and included in the final sample; 10 778 (77.4%) admissions were for primary palatal repairs and 3153 (22.6%) admissions were for revision palatoplasties. The mean age of primary repair was 0.7 years compared to 6.2 years for revisions. The revision cohort had a greater prevalence of cleft lips (63.5% vs 43.9%, P < .01). Compared to primary repairs, revision palatoplasties had a relative complication rate of 2.2 (rate of complications: 7.6% vs 3.4%, P < .01) and a relative fistula rate of 19.7 (rate of fistulas: 5.9% vs 0.3%, P < .01). In the multivariate model, after controlling for the effects of other associated variables, revision palatoplasty was still accompanied by greater relative odds of short-term complication (OR: 2.6, P < .01) and fistula (OR: 21.4, P < .01).

Conclusions: Revision palatoplasty carries higher rates of inpatient complication and fistula formation than primary repair. Failure of a primary repair may portend an increased risk of future failure with subsequent surgeries.

### 271. Degree of Asymmetry Between Complete and Incomplete Clefts

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Background/Purpose: Anthropometric study of the primary unilateral cleft deformity has been limited due to technical limitations resulting from the patient's age. Recent advancement in digital three-dimensional (3D) photogrammetry now allows us to photograph and study unrepaired infants with cleft lip and palate. Surgical outcomes for more severe complete clefts are not as ideal as those achieved for milder phenotypes. The technical challenges of repairing a wide complete cleft account for much of this variability. We hypothesized that in addition to the greater width of the cleft, patients with complete cleft lip and palate exhibit a greater degree of hypoplasia and asymmetry in other surgically relevant nasolabial dimensions. To investigate this, we used 3-dimensional digital photogrammetry to compare patients with unilateral complete cleft lip and palate and incomplete cleft lip  $\pm$  cleft palate to determine the degree of asymmetry present in surgically relevant nasolabial features.

Methods/Description: We conducted a cohort study of 28 pediatric patients with unilateral complete (14 patients) and incomplete (14 patients) cleft lips that were treated at Johns Hopkins All Children's Hospital. Fourteen linear facial distances were obtained using the Vectra H1 handheld camera (Canfield Scientific) and Mirror 3D analysis software (Canfield Imaging Systems). An unpaired t test was used to compare linear measurements of asymmetry between patients with complete and incomplete clefts. Standard deviations (SD) were determined for each measurement and laterality. The  $\chi^2$  tests confirmed all SD <1 mm. Pearson correlation was used to determine intra-rater reliability on duplicate 3D images. All statistical analyses were

completed using Statistical Analysis System (SAS). Statistical significance was set at P < 0.05.

Results: The degree of asymmetry of the nasal base, measured with subnasale to alare (Sn-al) and subnasale to subalare (Sn-sbal), was significantly greater for patients with complete clefts (P < .001, P < .001). Other anthropometric measures: Cheilion to crista philtra inferioris (Ch-cphi), nasion to endocanthion (N-en), subnasale to crista philtra inferioris (Sn-cphi), and labial fullness of the lateral and medial lip elements were not statistically significantly more asymmetric in complete or incomplete clefts. Intra-rater reliability was high for all 14 facial measures, ranging from 0.70 (N-En on the cleft side) to 0.99 (Sn-Al on the cleft side).

Conclusions: As expected, patients with the diagnosis of complete cleft lip and palate exhibit a greater width of the clefts than patients with incomplete cleft lip  $\pm$  palate. However, there is no difference in the degree of hypoplasia and asymmetry in other surgically relevant dimensions between these 2 groups.

# 272. The Functional Palate Suspension: A Technique to Expand the Versatility of the Double Opposing Buccal Flap Procedure for Palatal Lengthening

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Background/Purpose: The Double Opposing Buccal Flap Procedure for Palatal Lengthening (DOBFP) has become a proven alternative to the nonphysiologic pharyngeal flap (PF) and sphincteroplasty (SP). Traditional PF and SP procedures tether the velum specifically, the levator veli palatini, rendering it essentially immobile. Alternatively, the DOBFP maintains a natural levator panlanti and velar unit to act as the closing mechanism for the nasal pharyngeal space. While the DOBFP lengthens the anterior portion of the velum in patients with wider velar gaps, the lengthened palate invariably falls inferiorly with each millimeter gained posteriorly. As this progression continues, the point of velar contact on the posterior pharyngeal wall also falls progressively downward and the closure may be rendered incompetent. This is one reason why some patients with wide nasal pharyngeal gaps fail to achieve effective closure with the DOBFP alone. The functional palate suspension (FPS) expands the gap range of the DOBFP in order to treat patients with wider gaps and shorter palates. The FPS is a new way in practice to use posterior pharyngeal tissue that does not tether or immobilize the velar muscles. Clefts are often missing most or all of the musculus uvulae, which pulls the palate upward to reach the proper point of contact. Because we cannot replace the musculus uvulae, we must instead replace its upward pulling function with a small tissue suspension. The FPS elevates the soft palate and allows the velar muscles to remain functional and positioned to achieve the natural point for closure. The purpose was to demonstrate how the effectiveness of the DOBFP procedure can be expanded by adding the FPS for patients who fail to achieve normal speech with the DOBP procedure

*Methods/Description*: The speech results and complications of 17 patients were retrospectively reviewed. The occurrence of obstructive sleep apnea (OSA) symptoms was also reviewed.

Results: Of the 17 patients who underwent the FPS, the complication rate was 0% for syndromic (n = 6) and nonsyndromic (n = 11) patient populations. The development of normal speech status post FPS was 66.7% for syndromic patients and 90.9% for nonsyndromic patients. There were no reports of OSA among either patient population.

Conclusions: The FPS is an effective alternative reconstructive option to treat VPD. The FPS achieves velar closure by utilizing the natural

levator sling at the appropriate point of closure without narrowing the nasopharyngeal space. This approach falls within the Anatomic Palate Restoration Philosophy (APRP).

## 273. Three-Dimensional Analysis of Bilateral Cleft Lip and Palate Nasal Deformity: Presentation to Completion of Nasal Growth

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Background/Purpose: Understanding the changes in nasal morphology posttreatment and growth is useful in assessing outcomes and in designing future interventions for the BCLP patient population. Three-dimensional photogrammetry (3DP) is a rapid and reproducible image capture method that can be used to longitudinally evaluate nasal morphology at time points from before cleft lip repair to the completion of nasal growth. The purpose of this study is to use 3DP to assess nasal morphology of patients with BCLP at presentation and at multiple time points during growth with comparison to age-matched controls.

Methods/Description: A retrospective review of 3D photographs of 112 nonsyndromic bilateral cleft lip and palate patients were analyzed at 7 time points over the course of cleft care. An equal number of agematched control patients were analyzed for comparison. The time points used included patient presentation at 2 weeks of age, post-nasoalveolar molding (NAM) therapy, post-primary cleft rhinoplasty, and at 1, 5, 10, and 15 years of age. The nasolabial angle, nasal length, nasal height, columella height, columella width, nasal tip width, alar width, and alar base width were measured at each time point. The mean, standard deviation, and *t* tests were calculated for each measurement at all time points.

Results: The nasal height and columella height averages progressively increased over time in the BCLP (6.21-21.53 mm; 1.14-10.62 mm) and control (9.72-19.86 mm; 4.12-9.59 mm) groups. The control group nasal height and columella height were significantly greater than the BCLP group from pre-NAM through the 5 years of age time point (P < .01). The columella width in the BCLP group was significantly greater than the control group from pre-NAM through the 5 years of age time point (P < .01). The nasal tip width progressively increased over time in the BCLP (14.03-25.55 mm) and control (11.55-20.41 mm) groups. The BCLP group nasal tip width was significantly greater than the control group from 2 weeks through the 15 years of age time point (P < .05). The alar width and alar base width averages progressively increased over time in the BCLP (27.14-38.14 mm; 31.42-36.86 mm) and control (20.11-35.43 mm; 22.35-34.47 mm) groups. The BCLP group alar width and alar base width were significantly greater than the control group from pre-NAM through the 10 years of age time point (P < .01). The mean nasal lengths progressively increased over time in the BCLP (15.04-40.02 mm) and control (15.45-42.50 mm) groups but were not significantly different at any time point (P > .05).

Conclusions: BCLP patients had significantly shorter nasal height and columella height through 5 years of age. The columella width was significantly greater through 5 years of age. The alar width and alar base width were significantly greater through 10 years of age. The nasolabial angle and nasal tip width were significantly greater in the BCLP group at all time points. Nasal length was not significantly different at any time point.

## 274. Harnessing the Osteo-Inductive Property of JAGGED1 as a Maxillary Bone Regenerative Intervention

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Background/Purpose: Maxillary bone deficiency (MBD) occurs due to aberrant maxillary bone development. Bone morphogenetic protein (BMP2) is commonly used to ameliorate MBD, in adults, but in pediatric cases, it leads to pain, erythema, and inflammation. Therefore, we investigate BMP2-independent treatments such as JAGGED1's (JAG1) ability and mechanism to induce bone formation. JAG1, a membrane-bound NOTCH ligand, is required for normal craniofacial development, and Jagged1 mutations in humans cause maxillary bone hypoplasia and Alagille syndrome. We previously recapitulated this phenotype in Wnt1-cre; Jagged1f/f (Jag1CKO) mice and demonstrated deficient maxillary osteo- and vascular-genesis by conditional deletion of Jagged1 in maxillary mesenchymal cells. We also discovered a noncanonical JAG1-NOTCH1 signaling target, JAK2, which is essential to induce differentiation of neural crest cells (osteoblast [Obls] precursors) during intramembranous ossification for maxillary development. Thus, we hypothesized that JAG1 delivery to craniofacial bones will induce bone formation.

*Methods/Description*: We first investigated the targets downstream of JAG1-JAK2 in O9-1 cells, a cranial neural crest (CNC) cell line. O9-1 cells stimulated with recombinant JAG1-Fc bound to beads (5 μM), Fc-beads alone (5 μM), or BMP2 (3.84 nM) for 30 minutes and 12 hours in the absence or presence of the NOTCH canonical pathway inhibitor; DAPT was subjected to RNAseq. To test the feasibility of JAG1 delivery for induction of bone formation, we delivered JAG1-Fc-Beads complex (5, 10, or 20 μM), beads alone, and BMP2 (2.5 μM) incorporated in 4% PEG-MAL Hydrogels  $\pm$  CNCs by maxillary onlay injection in mice as 3 separate doses (initial dose, week 4, week 8). After 12 weeks, we quantified differences in ectopic bone formation between experimental groups by μCT analysis. Data presented as mean  $\pm$  SD (n > 3) were subjected to ANOVA and Tukey posttest (P < .05).

Results: RNAseq revealed enhancement of multiple genes within the NOTCH pathway (Hes1) and those involved in bone homeostasis (CXCL1, Snail1, Id1) at 30 minutes as well as 12 hours after JAG1 treatment alone. JAG1, in the presence of DAPT, induces genes essential for Obl differentiation (Id4) evidencing JAG1's noncanonical role in neural crest cells. We also observed increased bone deposition at 12 weeks postdelivery, demonstrating the efficacy of PEG-MAL hydrogel delivery of JAG1.

Conclusions: Collectively, these data suggest that JAG1 stimulates bone formation in vivo and the identification of JAG1-induced Oblspecific, NOTCH canonical and noncanonical targets, including genes that regulate bone homeostasis and essential craniofacial transcription factors in CNC cells that contribute to Obl-differentiation and bone mineralization. Our findings demonstrate the feasibility of using JAG1 to induce bone formation, independently. The coupling of NOTCH noncanonical targets and the delivery of JAG1 could change conventional interventions for maxillary hypoplasia in diseases like Alagille syndrome.

### 275. Three-Dimensional Facial Asymmetry in Non-Syndromic and Muenke Syndrome Associated Unicoronal Synostosis

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Background/Purpose: The purpose of this study was to quantify and compare differences in facial asymmetry (FA) in children with non-syndromic unicoronal synostosis (NS-UCS) and Muenke syndrome associated unicoronal synostosis (MS-UCS). It was hypothesized that children with MS-UCS present with significantly more severe FA compared to children with NS-UCS.

Methods/Description: The sample consisted of 20 children with UCS (7 MS-UCS and 13 NS-UCS) acquired from the primary care centers in Copenhagen and Aarhus, Denmark (N = 14) and Helsinki, Finland (N = 6). Data consisted of CT scans taken preoperatively (mean age: 0.6 years; range: 0.1-1.4 years) from which soft tissue surfaces were constructed. Guided by 21 manually placed soft tissue landmarks, a symmetric facial template was deformed to match each patient's surface, obtaining spatially detailed left-right point correspondence in all patient surfaces. FA was defined as a vector A between a point on one side of the midsagittal plane (MSP) and the corresponding point on the other side after mirroring across the MSP. The length of A provided the total amount of asymmetry (in millimeters), while its Cartesian components provided asymmetry in the transverse, vertical, and sagittal directions, respectively. Mean FA values were calculated for the full-face region and for 6 subregions: forehead, eye, nose, cheek, mouth, and chin. Comparisons of means were carried out using Mann-Whitney U test (level of significance: 5%).

Results: For the full-face region, a significant difference between means of 2.4 mm (P=.001) was calculated between the 2 groups and the largest difference was documented in the transverse direction (1.3 mm; P<.001). Region-wise, the largest significant difference between means was seen in the chin (4 mm; P<.05) and forehead (3.3 mm; P<.01) regions, followed by the eye (2.4 mm; P<.01), cheek (2.4 mm; P<.01), mouth (1.9 mm; P<.01), and nose (1.3 mm; P<.01) regions. The transverse direction presented with the largest significant difference in the following regions: chin (4.9 mm; P<.01), forehead (1.8 mm; P<.01), mouth (1.9 mm; P<.01), nose (1.3 mm; P<.01), and cheek (1 mm; P<.01). In the eye region, the largest significant difference was found in the sagittal direction (1.3 mm; P<.01).

Conclusions: The hypothesis that children with MS-UCS presented with significantly larger FA in all studied regions compared to children with NS-UCS could not be refuted. The largest significant difference between means was found in the forehead and chin regions, where the transverse direction presented with the largest significant difference.

### 276. Deep Craniofacial Phenotyping of Patients With Complete Congenital Arhinia

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Background/Purpose: Complete congenital arhinia is a rare embryologic defect characterized by absence of the soft tissue and skeletal structures of the nose. It is often associated with ocular and reproductive defects (Bosma arhinia microphthalmia syndrome) and has been genetically associated with a rare SMCHD1 gene mutation. The exact pathogenesis mechanism remains unclear. Arhinia is compatible with life and individuals with arhinia usually adapt to oral breathing. Surgical reconstruction is postponed until pre-school-age or later. In total, 88 cases have been reported in the literature so far, most of which describe neonates or include limited information regarding the craniofacial phenotype. The purpose of this study is to present for the first time a detailed craniofacial, oral, and dental phenotypic characterization of individuals with arhinia.

Methods/Description: Eight arhinia probands (4 males and 4 females), with a mean age of 28.4 (18-53) years, were enrolled in an IRB-approved study (protocol: 16-D-0040) and were examined at the NIH Craniofacial and Dental Clinic. A clinical examination, 2D, 3D photos, and head CBCTs were used for their craniofacial analysis. 2D cephalometric and geometric (3D) morphometric analyses were performed. Age-, gender-, and ethnicity-matched orthognathic control group was used for the needs of the 3D analysis.

Results: Clinically, a complete absence of natural soft and skeletal tissue nasal structures, ossification of the nasal aperture, absence of nasopharyngeal airway, midface hypoplasia, and anosmia were identified in all probands. Other significant clinical findings included longer facial third, mandibular prognathism mainly due to chin prominence, increased inner canthal distance, soft tissue asymmetry, whistle lip deformity, and a high arched and narrow palate. The dental phenotype included hypodontia, class III malocclusion, and anterior and posterior crossbite. CBCT examination revealed complete absence or severe hypoplasia of the paranasal sinuses, nasolacrimal duct stenosis or atresia, and ossification of the cribriform plate in all cases. Cephalometric analysis results supported the clinical findings and revealed a short anterior cranial base length and obtuse cranial base angle in most cases. Morphometric analysis indicated an overall difference in skull size, shape, and degree of asymmetry between arhinia and control subjects.

Conclusions: The complete absence of the nose has a significant impact on the development of the midface structures and the cranial base. However, facial width, total facial height, and vertical dental relationships were not as severely affected. Our future goal is to further understand the role of SMCHD1 gene in craniofacial development.

#### 277. Delayed Maturation and Reduced Crown Width of the Permanent First Mandibular Molar in all Subgroups of Cleft Lip and Palate

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Background/Purpose: Numerous studies have reported deviations in the development of the permanent dentition in cleft children. Most studies have, however, been carried out after surgical treatment of the cleft condition, and the treatment could possibly have influenced dental maturation negatively. A few studies have investigated the development of the permanent first mandibular molar in cleft children prior to surgery and only in the most severe types of cleft lip and palate and have found both delayed maturation of this tooth and reduced crown width. The aim of the present study was to investigate if these findings are representative for the total cleft lip and palate (CLP) population, including both complete and incomplete forms.

Methods/Description: A consecutive longitudinal sample consisting of 240 children examined at both 2 and 22 months of age: 196 children with CLP (66 with complete CLP [47 UCCLP; 19 BCCLP] and 130 with incomplete CLP [81 UICLP; 49 BICLP[) as well as 44 children with unilateral incomplete cleft lip (controls, UICL). In lateral cephalometric X-rays, maturation (MA) of the permanent first mandibular molar was assessed according to Haavikko (1970) at both 2 and 22 months of age. The follicle width (FW) was measured at 2 months of age, and the width of the crown (CW) was measured at 22 months of age. Intra-rater reliability was determined by duplicate measurements in 30 subjects. Relationship between dental developmental stages (MA) in the groups were tested using Mantel-Haenszel test. Differences between group means of FW as well as of CW were tested using Wilcoxon rank sum test. Level of significance: 5%. Lip closure was performed after examination and palatal closure after examination.

Results: Intra-rater errors: MA kappa = 0.87 (excellent agreement); FW and CW coefficient of variation = 1.8%; Dahlberg's s(i) = 0.2 mm. In general, it was found that gender did not influence follicle/tooth maturation significantly at this early age and females and males were accordingly pooled in all groups. MA was significantly delayed in all CLP groups (UICLP, UCCLP, BICLP, BCCLP) compared to UICL at both 2 and 22 months of age (P < .01). Mean FW was significantly delayed in all CLP groups (UICLP, UCCLP, BICLP, BCCLP) compared to UICL at 2 and 22 months of age (P < .01). CW measured at 22 months of age showed significantly decreased values for all CLP groups (UICLP, UCCLP, BICLP, BCCLP) compared to UICL, and BCCLP showed significantly decreased CW values compared to the other CLP groups (UICLP, UCCLP, and BICLP).

Conclusions: Delayed maturation, decreased follicle width, and crown width of the permanent first mandibular molar were found in all subgroups of CLP at this early age before any surgical treatment had been performed. These traits may, therefore, be considered characteristic of the CLP population, and it is likely the case in other permanent teeth as well. Delay in maturation of permanent teeth is of clinical importance in pediatric dentistry and should thus be given close attention.

## 278. Craniometric and Volumetric Analyses of Cranial Base and Cranial Vault Differences in Patients With Sagittal Craniosynostosis

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Background/Purpose: The emergence of powerful analytic technologies has allowed for more accurate 2- and 3-dimensional segmentation and measurements of stacked computer tomography (CT) data. With this technology, craniometric analyses of patients with metopic and unicoronal craniosynostosis demonstrated statistically significant differences in craniometric angles, distances from midline, and cranial vault volume measurements. We hypothesized that these

measurements differ significantly between patients with nonsyndromic sagittal synostosis (NSSS) and unaffected controls.

Methods/Description: This case—control study included infants with NSSS and unaffected, normocephalic controls. CT scans of the head were analyzed using Mimics (Materialise) to calculate cranial indices, distances to landmarks from midline, intracranial angles, and segmented cranial vault volumes. For the craniometric analysis, mean measurements by laterality were compared for each group; mean differences in measurements by laterality were compared between the NSSS and control groups. Statistical analysis was performed using Kruskal-Wallis, Mann-Whitney *U*, and paired *t* tests. For the volumetric analysis, anterior, middle, and posterior vault volumes were normalized to total cranial vault volume and compared between groups using a beta regression model controlling for age.

Results: Twenty-two patients with NSSS and 19 controls were identified. Petrous ridge angles (PRA) were significantly larger in NSSS compared with control (P left side = .002, P right side = .019). Interoccipital angles (IOA) and right and left euryon to zygomaticofrontal suture angles (EzFA) were significantly more acute in NSSS than in control (P < .001, P = .002, and P = .004, respectively). Cranial Index was significantly smaller in NSSS than in control (P < .001). NSSS showed no significant transverse lateralization of skull base structures relative to midline. Normalized anterior vault volume was 63% greater in NSSS than in control (P < .001). Normalized posterior vault volume was significantly (17%) smaller in NSSS than in control when not controlling for age (P = .012); however, the difference failed to reach statistical significance when controlling for age (P = .068). Normalized middle vault volumes were not significantly different between the 2 groups.

Conclusions: Our study provides a true, objective craniometric analysis of the cranial vault and endocranial base in patients with NSSS. While showing no significant transverse lateralization of structures of the anterior skull base relative to midline, patients with NSSS had significant, symmetric differences in craniometric angles, including IOA, EzFA, and PRA, as well as a larger anterior vault relative to total cranial volume. Further investigation is needed to determine the implications of cranial vault remodeling on cranial vault and cranial base dimensions.

## 279. Evaluation of Facial Morphology in Patients With Parry Romberg Syndrome and Linear Scleroderma

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Background/Purpose: Parry-Romberg syndrome (PRS) is a rare craniofacial disorder defined by progressive unilateral facial atrophy. PRS is often described as an autoimmune disease that rests on the same spectrum as linear scleroderma (LSD). The current literature categorizes PRS as mild, moderate, or severe based on the presence of atrophy in one or more areas of trigeminal nerve (CN V) sensory branch distribution. This study aims to use 3D soft tissue analysis to illustrate the most common areas of disease involvement and demonstrate the amount of facial asymmetry that can occur in specific anatomical regions.

Methods/Description: Twenty-eight patients with Parry Romberg syndrome/linear scleroderma (PRS/LSD) were enrolled in the study. We stratified our patients into mild (7), moderate (4), and severe categories (17) based on whether PRS involved 1, 2, or all three regions

of CN V sensory branches. We obtained 3D geometric measurements of anatomical landmarks on both sides of the 3D facial photo. Mathematical models were also developed to assess the asymmetry in the 3D photographs. Asymmetry was measured at the frontotemporal region, cheek (further divided into preauricular, malar, and lower regions), nose, and mouth. Finally, we used 2-sample *T* tests assuming unequal variance to determine statistical significance between grouped data.

Results: On average, the frontotemporal region demonstrated the maximum amount of asymmetry (4.7  $\pm$  1.8 mm) and followed by the cheek (3.7  $\pm$  1.9 mm), nose (2.8  $\pm$  1.1 mm), and mouth (2.8  $\pm$  1.4 mm) regions. Subanalysis showed that the preauricular and lower regions of the cheek contributed most to the cheek's total asymmetry. Whereas the malar section demonstrated the least amount of asymmetry. Mild patients demonstrated significantly less asymmetry in the cheek and mouth areas when compared to severe patients. Moderate patients showed significantly less asymmetry in the forehead when compared to severe patients. However, there were no significant differences in asymmetry between mild and moderate patients.

Conclusions: Our results suggest that the frontotemporal region is most significantly affected in patients with PRS/LSD. The total cheek followed closely behind the frontotemporal region in contributing to total facial asymmetry. Understanding the most common areas of anatomical involvement will aid the process of correcting facial dysmorphology in patients with PRS/LSD.

## 280. Improving Care Adherence in Patients With 22q11.2 Deletion Syndrome: A Quality Improvement Initiative

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Background/Purpose: 22q11.2 deletion syndrome (22q11DS), a condition associated with multiple health and neurodevelopmental sequelae, is best managed through comprehensive centers that offer coordinated interdisciplinary team care. Potential benefits of team care include ensuring completion of evaluations and screenings recommended by the International 22q Care Guidelines (Bassett et al., 2011). A quality improvement (QI) project was initiated at our institution to increase 22q guideline adherence among patients with 22q11DS. Using electronic medical record (EMR) and clinic-based interventions, we aimed to increase the proportion of patients with 22q11DS who were adherent to key 22q guidelines.

Methods/Description: Patients with confirmed 22q11DS seen by the 22q team between October 2017 and January 2018 were included in the project. The QI team assessed adherence to 8 22q care guidelines including those satisfied by one-time assessments and those requiring repeat surveillance (eg, labwork). Adherence was measured as a "bundle" (adherence to all 8 guidelines). A cause-effect analysis was completed and key drivers identified. The project aimed to increase the proportion of patients with 22q11DS with adherence to 8 key care guidelines from 71% (baseline) to 95% (goal). Several educational and EMR-based interventions were implemented. These included (1) an expanded after-visit summary explaining the value of 22q surveillance testing to families and (2) EPIC-based Best Practice Advisory (BPA) alerts for providers. These included a Health Maintenance BPA reminding providers to order surveillance testing when indicated, as well as a Referral BPA to encourage referrals to the 22q Center. Monthly BPA reports were generated to track utilization of EMR tools as a process measure. Pre-post assessments of one-time and repeat surveillance assessments were performed. The proportion of 22q11.2DS patients adherent to all 8 guidelines increased from 71% to 84% at 10 months postintervention. Stratification based on the frequency of assessments showed adherence for repeat surveillance measures (eg, labwork) increased from 76% at baseline to 89% postintervention. In just 5 months, the Referral BPA fired 301 times, leading to 68 referrals to the 22q Team. The Health Maintenance BPA was acted upon 74 times (11.7% provider interaction). Educational and EMR-based interventions for both patients and providers have improved 22q care guideline adherence in a short period of time at our 22q Center. Variations in guideline adherence for one-time screenings versus repeat labwork will be discussed, as well as new interventions which are currently underway to improve adherence to specific care recommendations. EMR-based interventions offer 22q centers new options to employ automated technology to facilitate increased referrals to team care and improve provider adherence to 22q surveillance testing at appropriate intervals.

### 281. Detecting Early Differences in Speech-Language Development in Infants and Toddlers With 22q1lds

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Background/Purpose: While communication disorders are a hallmark feature of 22q11.2 deletion syndrome (22qDS), few have studied speech-language development of 22qDS in the first few years of life (Scherer et al., 1999; Scherer et al., 2001). Children with 22qDS may display limited vocabulary, restricted sound repertoire, and general language delays; yet little is known about their early vocal characteristics, social interactions, and parent vocal input. The purpose of this study is to examine these early communicative characteristics in young children with 22qDS.

Methods/Description: Thirty children with 22qDS (17 males) and 21 typically developing peers (TD group, 8 males), ages 10-35 months (mean 22 months), were enrolled in this prospective study. All participants completed a day-long audio recording (mean 13 hours) in their home using a Language Environment Analysis (LENA) digital language processor. LENA software algorithms and statistical modeling were used to analyze conversations and estimate AVA scores (an index of speech output complexity that reflects the degree of organization of a child's vocalizations as speech-like sounds) and quantify child vocalizations, conversational turns, and caregiver vocal input. Data regarding the child's home environment, vocabulary size, therapies, and hearing history were also collected. Mann-Whitney U tests were used to examine group differences.

Results: Children with 22qDS produced significantly less frequent vocalizations (115/hour vs 160/hour, P < .022) and conversational exchanges as compared to TD peers (35/hour vs 52/hour, P < .007). Age was not predictive of quantity of child vocal output (P = .25). AVA speech complexity scores were significantly lower in children with 22qDS (83.7 vs 100.6, P < .001). In the 22qDS group, there was a significant negative correlation between age and AVA scores (r = -.453, P < .05). Quantity of child vocal output was not associated with AVA scores (r = .092). Children with 22qDS were exposed to less caregiver input than TD peers, but this did not reach significance (1087/hour vs 1356/hour, P = .063). While maternal education level was similar across groups (P = .269), higher maternal education level

was correlated with higher caregiver vocal input, conversational interactions, and child vocal output (r = 0.36, 0.43, 0.32, all P < .01).

Conclusions: Young children with 22qDS are significantly less vocal and interactive than TD peers starting at very early ages. Speech output complexity is also significantly reduced in young children with 22qDS. An unexpected finding of this study was the negative relationship between age and AVA scores in children with 22qDS, suggesting that older children in this study had less complex speech output compared to the younger children. The AVA score may serve as a very early index of emerging speech disorders in infants and toddlers in 22qDS. Implications for early diagnosis and intervention for speech-language disorders in 22qDS will be discussed.

### 282. Functional Variations of the Velopharyngeal Mechanism in Children With 22g11.2DS

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Background/Purpose: Dysmorphology in velopharyngeal structures has been shown to have significant negative implications on speech among individuals with 22q11.2 deletion syndrome (22q). Velopharyngeal dysfunction associated with 22q has a complex etiology. MRI investigations so far in the 22q population have reported variations in velopharyngeal structures among individuals with 22q, including a short, thin, asymmetric levator muscle with an increased angle of origin and a significantly shorter origin-to-origin distance. However, the majority of these studies utilized sedation, limiting the information to static data. As such, no studies to date have assessed the internal velar musculature during speech activity in children with 22q. The purpose of this study was to evaluate variations of the velopharyngeal mechanism during rest and sustained phoneme production in children with 22q, using an innovative, unsedated MRI protocol.

Methods/Description: Ten children with 22q and 10 children with normal velopharyngeal anatomy (ages 4-12 years) participated in the study. All participants were native English speakers and were imaged in the supine position using a Siemens 3 tesla MRI scanner. Images were collected during rest and sustained phoneme production. There was no use of sedation for the MRI exams. Paired t tests were used to assess variations between rest and sustained phoneme production conditions. Independent samples t tests were used to examine differences between experimental (22q) and control (children with normal anatomy) groups.

Results: Magnetic resonance imaging data were successfully obtained using the child-friendly scanning protocol. Quantitative and qualitative differences of the levator muscle and associated velopharyngeal structures were noted across both tasks and between the 2 groups. The levator muscle contracted differently across different phonetic contexts. Levator muscle length and angle of origin were reduced on the sustained phonation (P < .05). Across all subjects, the length of the levator muscle reduced going from rest to /i/ production. However, preliminary results suggest that the change was less evident in the 22q group compared to the control group, which may be due to hypoplasia of the levator muscle. For levator angle of origin, the angle was reduced going from rest to /i/ production across both groups; however, the change was more evident in the 22q group compared to the control group.

Conclusions: Results from this study suggest that individuals with 22q present with unique velopharyngeal muscle variations that may contribute to the high rate of velopharyngeal dysfunction associated with

this syndrome. This is the first study to demonstrate the successful utilization of MRI in obtaining functional 3D imaging data on velopharyngeal structures in children as young as 4 years with 22q, without the use of sedation.

## 283. 22q11.2 Deletion Syndrome and Postoperative Hypocalcemia After Pharyngeal Flap: Report of Three Cases

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Background/Purpose: Hypocalcemia and hypoparathyroidism are common features of 22q11.2 deletion syndrome (22q11DS) and are classically considered a transient feature in the neonatal period. Latent hypocalcemia and hypoparathyroidism have been reported, possibly triggered by acute biologic stress. The published guidelines for managing children with 22q11DS reference the risk of developing hypocalcemia during times of biologic stress, highlighting surgery as an example. Despite this, little has been published on the effect of surgery on children with 22q11DS and their risk of postoperative hypocalcemia. To date, there are no known reports of postoperative hypocalcemia in noncardiac surgeries for children with 22q11DS. We present 3 cases of children with 22q11DS who developed postoperative hypocalcemia after undergoing posterior pharyngeal flap.

Methods/Description: This is a case series of 3 patients. Written consent was obtained from the patients' families. As this is a case series of only 3 patients, IRB approval was not required. Chart review was completed for each patient including demographics, surgical data, and preoperative and postoperative details such as lab values and medications.

Results: Three pediatric patients with history of 22q11DS, hypernasality, velopharyngeal insufficiency (VPI), and a neurogenic soft palate underwent posterior pharyngeal flap for treatment of VPI. All three patients were prepubertal. One patient had a history of congenital hypoparathyroidism and transient hypocalcemia as an infant. At the time of the procedure, none of the patients were taking any calcium supplementation or calcitriol. Preoperative serum calcium was obtained on all patients and found to be in the normal range. The surgical procedures themselves were uneventful and operative time ranged from 146 to 170 minutes. All 3 patients had a significant postoperative decline in serum and/or ionized calcium. At the time of hypocalcemia, intact parathyroid hormone (iPTH) was obtained and found to be inappropriately normal in each patient. Mild neuromuscular symptoms were observed in 2 patients, yet no severe signs or symptoms of hypocalcemia were encountered. Pediatric endocrinology was consulted for each patient and medical therapy was initiated in 2 of the 3 patients. All patients had normalized their calcium by postoperative day 4 and were discharged home without any complications.

Conclusions: This case series is the first to highlight the presence of postoperative hypocalcemia in patient with 22q11DS after undergoing posterior pharyngeal flap. The hypocalcemia was transient and not associated with significant signs or symptoms. Surgeons caring for children with 22q11DS should be aware of the risk for postoperative hypocalcemia and consider checking calcium both pre- and postoperatively. Future studies are needed to better understand the physiologic cause, who is at risk, and the clinical significance of postoperative hypocalcemia in patients with 22q11DS.

#### 284. 22q11.2 Duplication Syndrome: Expanding What We Know About the Clinical Presentation

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Background/Purpose: 22q11.2 duplication syndrome has a frequency of  $\sim 1/700$  in the intellectual disability (ID) population. Despite this frequency, there is limited information on the variable clinical presentation. Findings can include a range of developmental delays (DD), ID, growth retardation, and hypotonia. Although the incidence of congenital anomalies is well described for 22q11.2 deletion syndrome, it is not well understood for 22q11.2 duplication. Additionally, with 22q11.2 deletion syndrome,  $\sim 90\%$  of cases occur de novo, where the majority of the 22q11.2 duplications are inherited from a parent with a normal or near-normal clinical presentation. This study reviews a large clinical series of patients with 22q11.2 duplication to define the incidence of associated features to help guide medical management.

Methods/Description: The cytogenetics database and 22q Multidisciplinary Clinical Repository were queried for patients with 22q11.2 duplications that included the TBX1 critical region, diagnosed by microarray analysis and qPCR from 2009 to 2019. A retrospective chart review was completed, documenting relevant clinical history.

Results: Forty-five patients were identified, 29 males and 16 females. One had triplication of 22q11.2 and was included; another with a larger unbalanced chromosomal complement was excluded from analysis. Average age at diagnosis was  $\sim 3.5$  years, with one fetus identified prenatally. The duplication was inherited in 69% of cases. Nine (20.5%) were identified with palatal anomalies, including high palate, overt cleft palate, and submucous cleft palate. Gastrointestinal complications were noted in 23 (52.2%) patients. Eighteen patients (41%) had vision abnormalities; endocrine dysfunction was noted in similar frequency. Less frequent congenital anomalies included cardiovascular (36.4%), musculoskeletal (31.8%), and neurologic anomalies (22.7%). Psychiatric illnesses were seen in 20 patients (45.4%). The majority (66%) had DD/ID.

Conclusions: This study represents a large single institution series of 22q11.2 duplication syndrome. While acknowledging the ascertainment bias, the data suggest the incidence of congenital anomalies may be higher than previously reported. Affected individuals have an increased risk of gastrointestinal complications, endocrine dysfunction, ophthalmologic abnormalities, palatal anomalies, congenital heart disease, musculoskeletal differences, and neurologic abnormalities. The high incidence of DD/ID and psychiatric illness is similar to previous reports. Where parental testing was completed, most individuals had inherited the duplication from a parent. Given the risks of the associated findings, we recommend that management following diagnosis should minimally include evaluations by plastic surgeon, otolaryngologist, gastroenterologist, endocrinologist, ophthalmologist, cardiologist, and genetic counselor. Parental testing is also strongly recommended. Individuals with 22q11.2 duplication syndrome would benefit from care coordination by an interdisciplinary care team.

# 285. Pathogenic Variants in CDC45 on the Remaining Allele in Patients With a Chromosome 22q11.2 Deletion Result in a Novel Autosomal Recessive Craniosynostosis Syndrome

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Background/Purpose: The 22q11.2 deletion syndrome is the most common microdeletion in humans, with highly variable phenotypic expression. The deletion results in haploin sufficiency of  $\sim 50$  genes in those patients (84%) with a standard LCR22A-LCR22D deletion. The remaining patients have a "nested" deletion involving fewer genes within this region, such as those extending from LCR22A-LCR22B, LCR22A-LCR22C, LCR22B-LCR22D, and LCR22C-LCR22D. Although palatal anomalies, immunodeficiency, congenital heart disease, feeding and swallowing difficulty, hypocalcemia, vertebral anomalies, cognitive differences, and neuropsychiatric illness are observed in over 50% of individuals with 22q11.2 deletion syndrome, a subset of patients present with additional atypical findings which are generally rare, such as craniosynostosis and anorectal malformations. Moreover, we previously reported the association of deletions or mutations in the intact chromosome 22q11.2 allele unmasking an autosomal recessive condition in some patients with atypical features. Recently, pathogenic variants in the CDC45 (cell division cycle protein 45) gene, located within the LCR22A-LCR22B region of chromosome 22q11.2, were noted to be involved in the pathogenesis of craniosynostosis and suggested to be part of the Meier-Gorlin syndrome.

Methods/Description: Thus, we performed next-generation sequencing on the DNA from 15 patients with a previously confirmed chromosome 22q11.2 deletion, using fluorescence in situ hybridization, multiplex dependent ligation probe amplification, array comparative genomic hybridization or SNP microarray, and atypical phenotypic features such as craniosynostosis, short stature, atypical skeletal differences, and anorectal malformations.

Results: These studies identified 4 novel rare nonsynonymous variants in CDC45 in 5 of 15 patients with 22q11.2 deletion syndrome and craniosynostosis and/or other atypical findings which resulted in a concomitant autosomal recessive condition.

Conclusions: This study supports CDC45 as a causative gene in craniosynostosis, as well as a number of other congenital anomalies. We suggest that this association results in a condition independent of Meier-Gorlin syndrome, perhaps representing a novel condition and/ or a cause of features associated with Baller-Gerold syndrome. In addition, this work confirms that the phenotypic variability observed in a subset of patients with 22q11.2DS may be due to pathogenic variants on the nondeleted chromosome, and therefore, all patients with atypical features should be screened for mutations on the remaining intact allele not only to provide appropriate medical management but also for accurate genetic counseling.

#### 286. Atypical Deletions Involving the 22q11.2 Region: One Laboratory Microarray Review 2009-2019

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Background/Purpose: Health-care providers involved in multidisciplinary 22q deletion syndrome clinics are often referred patients or contacted by parents of children with chromosomal microarray abnormalities involving the 22q11.2 region that are not the typical recurrent deletions associated with 22q11.2 deletion syndrome. These providers should be familiar with the basic principles of microarray interpretation, especially in the context of copy number variants (CNV) in the 22q11.2 region. Nationwide Children's Hospital (NCH) performs ~1500 chromosomal microarrays per year. Review of NCH laboratory and institutional experience will help to characterize atypical deletions and identify any recurrent challenges or trends.

Methods/Description: Two laboratory genetic counselors reviewed all postnatal chromosomal microarrays (n = 25 667) performed in the NCH cytogenetics laboratory dating from 2009 (when oligonucleotide platform was introduced) to the present. Any microarray with a deletion involving the 22q11.2 band met the inclusion criteria. A microarray result was determined to be an atypical deletion if it involved the 22q11.2 band but was not either the common 3 or 1.5 Mb recurrent deletion that include TBX1 and HIRA. For atypical deletion patients, genotype and phenotype characteristics were recorded.

Results: A total of 136 microarray results meeting inclusion criteria were identified; 100 were typical 22q deletions representing 22q11.2 deletion syndrome; 36 were atypical deletions. Genotype and phenotype information was available for 32 of these 36 patients. Of the microarray findings, 8 were likely pathogenic (LP), 18 variants of unknown significance (VUS), and 8 likely benign copy number variation (LBCNV). Two patients had 2 separate CNVs both involving the 22q11.2 region. Where inheritance could be determined, 1 of 1 LP was de novo, 5 of 6 VUS were inherited, and 3 of 3 LBCNV were inherited. Mean size of CNVs were as follows: LP 1310 ± 857 kb, VUS 55.9  $\pm$  20.4 kb, and LBCNV 30.8  $\pm$  4.6 kb. Five out of 8 LP results were the same  $\sim$  700 kb deletion spanning from breakpoints LCR22-B to LCR22-D. A 29.73 kb deletion that includes the 3' portion of the putative gene c22orf25 was identified in 12 patients and categorized as a VUS 7 times and LBCNV 5 times. A 37.58 deletion that includes the 3' end of TANGO2 and the 5' end of DGCR8 was identified in 4 patients and categorized as VUS twice and LBCNV twice.

Conclusions: Deletions involving the 22q11.2 region are found frequently on clinical chromosomal microarray analysis. In our single institutional experience, a typical recurrent deletion was found approximately 3 times more commonly than an atypical deletion. Although less common than typical deletions, a few recurrent findings in this study provide insight into the ongoing challenge of the clinical interpretation of atypical 22q deletions.

### 287. How Far Is Far Enough? Assessing Long-Term Efficacy of Mandibular Distraction Osteogenesis in Treating OSA in Infants With Micrognathia

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Background/Purpose: The use of mandibular distraction osteogenesis (MDO) to surgically correct obstructive sleep apnea (OSA) in infants is well-documented. The anterior mandibular movement and subsequent repositioning of the tongue and pharyngeal muscles produces more stable air flow and relieves obstruction at the level of the tongue base. While jaw surgery in infants centers on improving function, we have also observed that initial overdistraction may result in more balanced facial proportions at long-term follow-up. Whether micrognathia is associated with a syndrome or with isolated Pierre Robin sequence, there is a greater degree of growth retardation of the mandible. To that end, we "overcorrect" the mandible to a skeletal class III relationship so when maxillary growth nears completion, a skeletal class I relationship is more achievable. The purpose of this study is to confirm the long-term efficacy of MDO in reducing AHI and oxygen requirement as well as to evaluate the effect of mandibular overdistraction on long-term facial balance.

Methods/Description: A retrospective chart review of patients from January 1, 1995, to May 22, 2019, with OSA associated with micrognathia either from an underlying syndromic cause or isolated Pierre Robin sequence was completed. Patients must have undergone MDO

before 12 months of age. Patient information included demographic characteristics, diagnoses, previous treatments, existing comorbidities, and details of the preoperative, intraoperative, and postoperative distraction protocols and polysomnograms (PSG). Lateral photographs were reviewed for skeletal classification by 2 senior physicians at routine follow-up visits.

Results: A total of 82 patients met inclusion criteria. The mean age at distraction was  $63.32 \pm 71.40$  days. Patients were distracted at a mean rate of 1.73  $\pm$  0.24 mm/d to a mean distance of 27.51  $\pm$  4.37 mm. Preoperative PSG demonstrated a mean AHI of 34.06  $\pm$  33.48 (n = 57) with lowest oxygen desaturation and mean oxygen  $(O_2)$  requirement of  $80\% \pm 8.13\%$  and  $0.63 \pm 0.81$  L, respectively. Compared to preoperative findings, the first postoperative PSG (mean 69.68  $\pm$ 54.931 days, n = 66) demonstrated a decreased AHI to 5.76  $\pm$  5.63 (P < .0001), an increased lowest desaturation of 85.81%  $\pm$  7.90% (P = .064), and a mean  $O_2$  requirement of 0.26 L  $\pm$  0.53 (P = .027). A PSG at the last follow-up after surgery (1058.68  $\pm$  1009.241 days, n = 38) when compared to the first postoperative PSG showed a decrease in AHI to 2.40  $\pm$  3.09 (P = .016), an increase in lowest desaturation to  $86.06\% \pm 6.81\%$  (P = .67) and a decrease in  $O_2$ requirement to 0.05  $\pm$  0.18 L (P = .045). At last follow-up, 84% of patients who had been overcorrected to class III were no longer prognathic.

Conclusions: MDO is safe and predictable for the treatment of OSA in infants. MDO decreases AHI, reduces the need for supplemental O<sub>2</sub>, and decreases oxygen desaturation during critical sleeping stages for infants. Overdistraction to skeletal class III can help patients achieve and maintain a more balanced skeletal profile as growth progresses

### 288. Does Mandibular Distraction Vector Influence the Rate of TMJ Ankylosis?

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Background/Purpose: Since its advent by McCarthy et al [1] in 1992, mandibular distraction has become the primary choice for treatment of patients with moderate to severe Robin Sequence [2]. Based on the Ilizarov principle of bone lengthening [3], mandibular distraction relieves airway obstruction by lengthening the mandible. Despite its effectiveness, a potential yet problematic complication of mandibular distraction is TMJ ankylosis. Previous studies report TMJ ankylosis rates of up to 10\% [4], while other studies have shown virtually no incidences of TMJ sequelae [5]. A theory on this difference relates to distraction vector; a vertical vector is more likely to lead to TMJ ankylosis because of the cranially directed pressure withstood by the TMJ during activation, as compared to horizontally or obliquely directed vector. Historically, our center has used a vertical distraction vector with a more recent conversion to an obliquely oriented vector. The purpose of this presentation is to discern if there is difference in rates of TMJ ankylosis between vertical and oblique distraction groups.

Methods/Description: After IRB approval, a retrospective chart review was performed of all patients who underwent mandibular distraction at Children's Mercy Hospital from 1997 to 2015. All operations were performed by 3 surgeons. Ankylosis rates were compared between the 2 groups.

Results: A total of 94 patients were reviewed. The average age of presentation was 103 days. Seventy underwent vertical distraction, while 24 underwent oblique distraction. TMJ ankylosis was recorded in 12 cases, all in the vertical vector group, a 17% rate of ankylosis.

There were no cases of ankylosis in the oblique vector group. The average age at diagnosis of TMJ ankylosis was 6.5 years. When excluding all syndromic patients in both groups, 48 patients remained. Thirty-four underwent vertical distraction versus 12 for the oblique group. There still was a 12% rate of ankylosis, all in the vertical group. *Conclusions*: Vertical mandibular distraction carries a significantly increased risk of TMJ ankylosis and should be avoided.

#### 289. Early Mandibular Distraction Reduces Relapse in Orthognathic Surgery for Craniofacial Microsomia

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Background/Purpose: The optimal surgical treatment for the Pruzansky II mandible in craniofacial microsomia (CFM) is controversial. While some surgeons advocate to forego distraction and to perform definitive orthognathic surgery at skeletal maturity, others suggest that mandibular distraction osteogenesis during childhood is useful for increasing bone stock and providing improvements in symmetry during a critical period of psychosocial development. In this work, we evaluated the long-term orthognathic surgery outcomes of Pruzansky II mandibles in CFM.

*Methods/Description*: CFM patients with Pruzansky type II mandibular hypoplasia (age  $22.0 \pm 6.7$  years) were retrospectively reviewed for demographics, operative characteristics, surgical outcomes, relapse, and other complications. Patients who received distraction during childhood (<9 years of age) were compared to those who were not distracted in terms of orthognathic surgical outcomes using  $\chi^2$ , Fisher exact, Kruskal-Wallis, and Mann-Whitney U tests.

Results: Thirty-one patients were identified who met the study's inclusion criteria. Twenty-four patients underwent mandibular operations, and 21 underwent distraction osteogenesis at a mean age of  $8.6\pm3.6$  years. Repeat distraction was required in 2 (9.5%) patients. Orthognathic surgery was performed in 10 (32.3%) patients. Relapse following orthognathic correction was observed in 3 (30.0%) of patients. Patients who underwent early mandibular distraction received significantly fewer orthognathic operations overall (P=.02) and were significantly less likely to experience relapse following orthognathic surgery (P<.01).

Conclusions: In CFM patients with Pruzansky type II mandibular hypoplasia, early distraction both decreases the frequency of orthognathic surgery at skeletal maturity and relapse following orthognathic surgery.

## 290. Salutary Soft Tissue Changes After Maxillary Distraction With a Rigid External Distraction Device in Patients With Bilateral Cleft Lip and Palate

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Background/Purpose: Patients with bilateral cleft lip and palate often develop maxillary hypoplasia and class III malocclusion. In severe cases, the maxillomandibular relationship can have a negative overjet greater than 10 mm. Other characteristic facial findings include malar deficiency, redundant malar soft tissue envelope, plunging nasal tip, acute nasolabial angle, and a thin, unsupported upper lip. Limited upper incisal display at repose and an overclosed, brachycephalic appearance are also common. The current treatment to correct the

maxillomandibular relationship is orthodontic decompensation and Le Fort I osteotomy. A high winged modification of the traditional Le Fort I osteotomy addresses the malar deficiency component of the dentofacial deformity. Distraction osteogenesis is recommended for maxillary advancements greater than 10 mm to reduce the risk of relapse by opposing forces from the overlying soft tissue functional matrix. In these cases, we recommend use of a rigid external distractor (RED) device as it allows for greater degrees of freedom while correcting the maxillary position during distraction. A high winged Le Fort I with RED distraction allows for correction of the midface deficiency while restoring optimal aesthetic convexity and maximally filling the soft tissue envelope of the face. The purpose of this study is to demonstrate the salutary soft tissue changes in patients with bilateral cleft lip and palate who underwent maxillary advancement with a high winged Le Fort I osteotomy and distraction osteogenesis with an RED device.

Methods/Description: Patients with bilateral cleft lip and palate with severe maxillary hypoplasia who underwent maxillary distraction with a high winged Le Fort osteotomy and rigid external distraction device between 2017 and 2019 were reviewed in a retrospective fashion. Patient demographics, age at the time of surgery, and length of distraction stages (latency, activation, and consolidation) were examined. Patients with less than 3 months follow-up after RED device removal were excluded. Preoperative and postoperative lateral cephalograms were taken, and bony and soft tissue cephalometric landmarks were measured. Preoperative and postoperative photographs were evaluated by independent reviewers and rated for their changes in soft tissue position and esthetic appearance. Study findings will be presented.

## 291. Comparative Study of Skeletal Stability Following Mild, Moderate, and Severe Lefort I Advancement in Patients With Cleft Lip and Palate

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Background/Purpose: Le Fort I advancement surgery is challenging in patients with clefts because of the palatal scar tissues. In this study, we investigated the outcome of Le Fort I advancement surgery (mild, moderate, and severe groups) and 1-year skeletal stability in patients with cleft lip and palate.

Methods/Description: A retrospective chart review was performed to identify patients with nonsyndromic unilateral or bilateral cleft lip and palate who underwent maxillary Le Fort I advancement at skeletal maturity from 2013 to 2019. To satisfy the inclusion criteria, all patients had to have diagnostic quality cone beam computed tomography (CBCT) prior to surgery (T0), immediately postoperative (T1), and at 1-year follow-up (T2). A total of 59 patients (unilateral n = 34, bilateral n = 25) who underwent Le Fort I advancement was identified. Nineteen of these 59 patients were excluded due to insufficient radiographic records; thus, 40 patients with complete records were included in the study. The sample was comprised of 9 females and 31 males, with an average age of 19.1  $\pm$  3.21 years at the time of the surgery. Lateral cephalograms were extracted, traced, and superimposed using Dolphin Imaging software (V 11.95). Horizontal surgical movement (T0-T1) and postoperative relapse (T1-T2) at skeletal and dental level were quantified as linear changes at point A and upper incisor edge (U1-tip), respectively. Patients were divided into 3 groups according to the severity of surgical movement: mild (<5 mm, n = 9), moderate (5-10 mm, n = 20), and severe (>10 mm, n = 11). The statistical analysis was performed using 2-way repeated-measures

ANOVA to test the difference of surgical movements and postoperative relapse between groups.

Results: The mean advancement (T0-T1) of all patients at point A was  $8.1\pm2.8$  mm and at U1-tip was  $7.7\pm2.6$  mm. In the mild, moderate, and severe groups, the mean advancement at point A were  $4.6\pm1.3$  mm,  $7.7\pm1.1$  mm, and  $11.6\pm1.2$  mm, and at U1-tip were  $5.7\pm2.9$  mm,  $7.4\pm1.3$  mm, and  $10\pm2.6$  mm, respectively. There were significant skeletal and dental advancements in all the 3 groups following Le Fort I surgery (P < .0001). At 1-year follow-up, the mean relapse (T1-T2) at point A was  $1.2\pm1.1$  mm and at U1-tip was  $0.07\pm1.9$  mm. When analyzed within the mild, moderate, and severe groups, the mean relapse at point A were  $0.8\pm0.7$  mm,  $1.2\pm0.9$  mm, and  $1.9\pm1.5$  mm and at U1-tip were  $-0.4\pm1.6$  mm,  $0.4\pm2.1$  mm, and  $-0.2\pm1.5$  mm, respectively. There was no significant difference in the relapse amount between the mild, moderate, and severe groups at skeletal and dental components (P > .05).

Conclusions: Le Fort I advancement surgery successfully corrected maxillary hypoplasia in patients with cleft lip and palate in all the 3 groups. This study also demonstrated that larger advancement in the severe group can result in equivalent skeletal stability when compared to the mild and moderate advancement. Though mild skeletal relapse was observed in all the 3 groups, none of the patients had to be reoperated.

## 292. Effects of Single-Dose Pre-Operative Pregabalin on Post-Operative Pain and Opioid Consumption in Cleft Orthognathic Surgery

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Background/Purpose: The current opioid epidemic has placed postoperative pain management under scrutiny. Limiting postoperative pain can decrease overall opioid usage in the recovery period, especially after orthognathic surgery. Several studies have illustrated the efficacy of pregabalin in decreasing postoperative pain and opioid usage in adults undergoing orthognathic surgery. We aim to study the effects of a single dose of preoperative pregabalin on postoperative pain and total opioid consumption after orthognathic surgery in individuals with cleft lip and palate.

Methods/Description: This was a retrospective review of consecutive patients with diagnosis of cleft lip and palate seen between June 2012 and July 2019. We took advantage of our institution's implementation, beginning in 2016, of a one-time dose of preoperative pregabalin for Le Fort I midface advancement. All patients underwent Le Fort I midface advancement performed by one of 2 surgeons at a single institution. The treatment group received a one-time preoperative dose of pregabalin. The control group did not receive pregabalin. Total morphine milligram equivalents (MME) consumption was calculated by adding intraoperative opioid administration and postoperative opioid consumption during admission. Postoperative opioid analgesia consisted of intravenous (IV) hydromorphone or morphine and oral oxycodone. Duration of hospitalization and pain scores based on the pain intensity numeric rating scale (0-10) were also recorded. The mean postoperative pain assessment scores during admission were calculated for each patient. The median of these individual mean pain assessment scores for each group was subsequently computed.

Results: Twenty-six patients (15 males, 11 females) were included in this study; half of patients received pregabalin (median dose: 150 mg,

range: 100-200 mg). Mean age (years) at operation of the pregabalin (18.4  $\pm$  1.9) and control groups (17.8  $\pm$  1.9) were also equivalent (P=.442). Mean hospital stay for both groups was 1.23 days. The pregabalin group had significantly lower consumption of total opioids during admission (total MME median: 26.00; Q1: 10.75-Q3: 83.35) compared to the control group (median: 67.40 MME; Q1: 45.5-Q3: 117.84) (Mann-Whitney U=44; P=.039). Although pain scores in the treatment group (median: 2.63; Q1: 0.97-Q3: 4.46) were lower than in the control group (median: 3.44; Q1: 1.81-Q3: 5.97), the difference was not statistically significant (Mann-Whitney U=67.00; P=.390).

Conclusions: Based on the results, a one-time preoperative oral dose of pregabalin before orthognathic surgery in patients with cleft lip and palate reduced total opioid consumption during admission. However, there was no difference in average length of stay or pain scores within the 2 groups. A single preemptive oral dose of pregabalin should be considered an effective adjunct to pain management protocols in patients undergoing orthognathic surgery.

### 293. Surgical Treatment of Unilateral Condylar Hyperplasia During the Active Phase Leads to Improved Facial Symmetry

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Background/Purpose: There is no consensus regarding the best treatment for facial asymmetry due to unilateral condylar hyperplasia (UCH). Different management strategies are available based on disease activity. Active UCH may be addressed with high condylectomy with or without concurrent or staged orthognathic surgery. Alternatively, a delayed approach may be used, in which orthognathic surgery alone is performed after the disease process has "burned out" or arrested. Support for these options has largely been based on case reports rather than definitive evidence. Therefore, this study sought to quantify and compare facial asymmetry in UCH patients before and after corrective surgery for active or arrested disease in an effort to inform future treatment guidelines.

Methods/Description: An approved retrospective review was performed on patients who underwent surgery for UCH with at least 6 months of follow-up. Pre- and postoperative 3-dimensional (3D) images obtained using a 3D camera system were available for 11 patients with active disease and 9 patients with arrested UCH. The images of UCH patients were compared to images of 20 age- and gender-matched controls without concerns of facial asymmetry. Facial asymmetry was analyzed and quantified by calculating the root mean square deviation (RMSD) between points on the native faces and those constructed by mirror image for all patients. Paired Student t tests were performed to compare the RMSDs of pre- and postoperative images between the UCH groups and the control group (P < .05 was considered statistically significant).

Results: Prior to surgery, patients with arrested UCH were more asymmetric than those with active disease (P=.011). Surgical treatment resulted in improved facial asymmetry for both groups (P=.0069 and P<.001 for active and arrested UCH, respectively), based on images obtained at longest follow-up (mean  $1.0\pm1.1$  years). However, patients with arrested UCH remained notably more asymmetric compared to symmetric controls despite surgery (P<.001). There was no significant difference between symmetry measurements in those treated during active UCH and the same controls (P=.089).

Conclusions: These findings indicate that facial asymmetry due to UCH is more likely to be corrected if treatment is pursued while the condylar hyperplasia is still active. High condylectomy not only arrests condylar activity but also corrects vertical deformity of the mandible to restore facial symmetry. A delayed approach may produce significant improvements, but facial asymmetry may persist. Management strategies should therefore focus on intervening while UCH is active rather than waiting until the disease process has arrested.

#### 294. Assessing Risk of Speech-Language Disorders in Children With Cleft Lip/Alveolus

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Background/Purpose: While it is well known that children with cleft palate (with or without cleft lip) are at higher risk for speech and language difficulties than the general population (Riski & Delong, 1984; Scherer & D'Antonio, 1995), little is known about the speech-language profiles of children with cleft lip with or without cleft alveolus (CL/A) (without a cleft palate). The risk of communication disorders in the general population of children without clefts is approximately 5% for articulation disorders and 3% for language disorders (Black et al., 2015). Vallino et al. (2008) reported 13% of children with nonsyndromic CL/A demonstrated articulation skills below age expectations and 18% demonstrated language delays. Deelder et al. (2011) reported similar findings; thus, these reports suggest that children with nonsyndromic CL/A have an elevated risk of speech-language disorders. The purpose of this study was to determine the relative rates of occurrence of speech-language disorders in a consecutively evaluated group of children with CL/A, as well as examine the types of speech and language difficulties and impact of co-occurring syndromes and other diagnoses (eg, otitis media, submucous cleft palate) on risk for communication disorders.

Methods/Description: A retrospective chart review was completed with children from a single pediatric academic medical center, ages 12 to 72 months, with a cleft lip or cleft lip and alveolus, without an overt cleft palate. Participants included 86 children (58 males, 28 females). Forty children had cleft lip and alveolus; 46 had isolated cleft lip only. Five children also had a submucous cleft palate. Eight children had multiple congenital anomalies including 1 with CHARGE association and 1 with craniofacial microsomia.

Results: Fifty-three percent of children with CL/A had received at least 1 speech-language evaluation and 51% had received at least 1 team visit (including at least a surgeon, SLP, and orthodontist). The average age at first speech evaluation was 26.7 months. Overall, 37% (n = 17) of children assessed presented with some type of communication disorder; 26% (n = 12) were diagnosed with a language disorder and 21% (n = 9) were diagnosed with an articulation disorder. The presence of ear tubes or submucous cleft palate in children with CL/A was not significantly associated with the diagnosis of an articulation disorder (P = .22 and P = .19, respectively).

Conclusions: The results of this study are congruent with past reports suggesting children with CL/A have a higher risk of communication disorders relative to the general pediatric population. The risk rates in this cohort were higher than expected based on past reports, which may be related to methodologic differences across studies; however, all continue to suggest an increased risk of speech-language disorders in this patient population. Team care for children with CL/A should

include speech-language assessment starting at an early age, with ongoing monitoring to ensure management needs are identified and addressed in a timely manner.

### 295. Assessing Speech Intelligibility Using the Intelligibility in Context Scale in Children With Cleft Lip/Palate

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Background/Purpose: Speech articulation disorder is more prevalent in children with cleft lip/palate (CL/P) compared to their peers (Chapman et al., 2018), making it critical to assess functional speech outcomes across development. Additionally, children with CL/P may demonstrate concerns with velopharyngeal dysfunction, which may also impact speech intelligibility. The Intelligibility in Context Scale (ICS) was used to examine speech intelligibility across a range of communication contexts in a sample of children with CL/P between the ages of 3;0 and 8;11 and to explore the relationship with speech articulation skills and resonance. 1. What is the mean score for children with CL/P on the ICS? 2. What is the relationship between total score on the ICS and standard score on the GFTA-3? 3. What is the relationship between total score on the ICS and perceptual ratings of hypernasality? 4. Which of the communication contexts on the ICS significantly predict standard score on the GFTA-3 in children with CL/P?

Methods/Description: Children with nonsyndromic cleft lip/palate who had received a speech evaluation at a metropolitan cleft team were included in this study. The Intelligibility in Context Scale (ICS) is a 7-item scale that asks caregivers to rate their child's speech intelligibility over the last month across communication partners (McLeod, Harrison, & McCormack, 2012). For the current study, the ICS was completed by the caregiver(s) during the speech evaluation clinical encounter. The Goldman-Fristoe Test of Articulation, Third Edition (GFTA-3, Goldman & Fristoe, 2015) Sounds-in-Words subtest was administered by a certified speech-language pathologist during the clinical encounter.

Results: Speech intelligibility ratings were examined for children with CL/P between the ages of 3;0 and 8;11. The mean ICS score across communicative contexts was 3.84 (SD = 0.48), indicating children with CL/P were sometimes-to-usually understood by their communicative partners. Children with CL/P presented with a lower mean ICS score compared to the normative data for the ICS (McLeod, Crowe, & Shahaeian, 2015). A significant moderate correlation was observed for the association between speech intelligibility and performance on a standardized articulation assessment in children with CL/P (r = 0.407, P = .003). For children with CL/P, the ICS ratings significantly predicted GFTA-3 standard score,  $F_{7,\,41} = 3.149$ , P < .05, accounting for 35% of the variation in GFTA-3 standard scores with adjusted  $R^2 = 0.239$ . In this regression model, the intelligibility rating for strangers was a significant predictor of GFTA-3 standard score.

Conclusions: Children with CL/P demonstrated reduced speech intelligibility measured by caregiver ratings of intelligibility across communication contexts. The ICS is an appropriate measure to include in a comprehensive assessment of speech skills, especially due to its role in describing how a child's speech influences functioning in their natural environment.

# 296. Exploratory Analysis of the Relationship of Clinician-, Family-, and Patient-Reported Outcome Measures for Speech in the Care of Children With Cleft Lip/Palate

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Background/Purpose: Cleft palate affects speech function, overall intelligibility, and verbal communication competence. Given the significant functional impact of speech/intelligibility across all areas of an individual's life and high risk of speech impairments in those born with cleft palate, comprehensive long-term evaluation and management is essential to facilitate normal speech and communication abilities. In recent years, patient-reported speech assessment has been proposed as a supplement to traditional clinician-reported speech appraisal. Despite the advent of validated patient-reported outcome measures (such as the CLEFT-Q), it remains unknown whether and how these measures relate, practically, to traditional clinical assessment. This project was a pilot study to explore possible relationships of interest.

Methods/Description: This was a retrospective analysis of prospectively collected clinical data from children with cleft palate (with/without cleft lip) evaluated by a mid-volume multidisciplinary cleft and craniofacial center between November 2016 and May 2018. We examined the relationships among outcomes reported by the clinician (ie, articulation and velopharyngeal function), family (ie, overall speech intelligibility in context), and patient (ie, CLEFT-Q Speech Function and Psychosocial Distress Related to Speaking subscales). Box plots and scatter plots allowed visual inspection of the relationships of the observed distributions.

Results: The final data set consisted of 140 patients. Clinician-reported scores for articulation and velopharyngeal function appear to be related. Family-reported overall speech intelligibility in context (ICS) appeared to be related to clinician-reported appraisal of articulation (PCC scale) and velopharyngeal function (VPC scale). Family-reported ICS also appeared related to patient-reported outcomes (CLEFT-Q scales). While patient-reported outcomes (CLEFT-Q) appeared related to clinician-reported assessment of articulation (PCC), CLEFT-Q scores did not appear to be related to clinician-reported assessment of velopharyngeal function (VPC). Finally, the 2 patient-reported CLEFT-Q subscales (ie, speech function and psychosocial distress related to speaking) were observed to be positively related to each other.

Conclusions: This study provides an essential early exploration of the relationship between the various perspectives of speech outcome assessment: that is, from clinicians, family, and patients. Issues captured by family- and patient-reported instruments have been shown to be valuable adjuncts to (but not replacements for) traditional clinician-reported ratings of articulation and velopharyngeal function. This project also generated several hypotheses regarding the causal relationship between speech constructs and relevant implications of these that warrant ongoing research in this area through future prospective studies.

## 297. Issues in Percent Consonant Correct as an Outcome Measure for Cleft Speech in an Intervention Study

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Background/Purpose: In cleft speech outcome studies, narrow phonetic transcription and the reporting of rater reliability/agreement are acknowledged as the gold standard. However, cleft speech brings special challenges to this task. Percent consonant correct (PCC) is typically used as an outcome measure in non-cleft speech disorder intervention studies and reporting outcomes in cleft speech studies. The aim of this study was to evaluate the reliability and use of PCC in a cleft speech therapy intervention study.

Methods/Description: Two independent experienced listeners analyzed 119 recordings from 46 participants, aged 2.9 to 7.5 years, in a randomized controlled trial. Recordings were randomly selected from 5 data points before, during, and following intervention. The listeners attended a 1-day training course following which they analyzed 69 speech samples. A modified PCC score was calculated for words and phrases/sentences. Analysis indicated that further training was indicated. Following this, the listeners analyzed other 70 speech samples, 20 of which were duplications.

Results: The initial results showed poor reliability for the number of targets elicited for words (ICC = 0.07) and sentences/phrases (ICC =0.42). Differences in classification of errors as glottal stops and consonant deletion accounted for this. Other differences regarding transcription of active nasal fricatives and nasal emission/turbulence accompanying sounds were also noted. Despite this, reliability of the modified PCC was good (ICC word = 0.80; ICC phrases/sentences = 0.79). Results of the second reliability study showed improvement in the number of targets elicited in words and phrases/sentences, with very good inter-rater reliability for the modified PCC score on the word data set (ICC = 0.9; 95% CI = 0.84-0.94) and the phrases/ sentence data set (ICC = 0.88; 95% CI = 0.8-0.93). Very good intra-rater reliability (ICC = 1.0; CI = 0.98-1.00) for the modified PCC score in both words and phrases/sentences for each listener was found. One listener consistently gave higher modified percent consonant correct scores.

Conclusions: The modified PCC was reliable. However, reliability of the number of targets elicited should be reported in cleft speech intervention studies. Listeners need to distinguish between glottal articulation and consonant deletion, in order that the PCC score is meaningful. Attention should be paid to where listeners are reliable, but where one listener rates consistently higher than another, as this can have serious consequences in an intervention study. More research is needed on approaches to measure the resolution of articulation difficulties in cleft intervention studies.

### 298. Reliability of Untrained, Lay Listener Ratings of Global Measures of Speech Performance

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Background/Purpose: In formal clinical research, audit, benchmarking, and outcome studies, the perceptual rating of performance for specific speech constructs including hypernasality, nasal emission,

and nasal turbulence, as well as the phonetic transcription of speech disorder is undoubtedly the role of a highly trained, experienced speech-language pathologist (SLP). While SLPs can also make judgments about global measures of speech performance for constructs such as speech understandability and speech acceptability, it is argued that lay or naive listeners represent a more ecologically valid group of raters for judging these concepts. A comparison of lay listener and SLP ratings of global speech measures from the same speech samples could also potentially determine how valid SLPs are in making global judgments about speech performance. The aim of this study was to investigate the inter-rater and intra-rater reliability of untrained lay listener judgments of global measures of speech performance from spontaneous (connected) speech samples from children with differing degrees of velopharyngeal function and to compare the reliability of lay listener ratings to a group of SLPs' ratings of the same speech samples.

Methods/Description: Audio recordings of spontaneous speech stimuli were elicited from 40 children aged between 4 and 14 during routine attendance at a cleft palate clinic. The speech samples were selected to reflect the typical clinical presentation of velopharyngeal function (and velopharyngeal insufficiency). Ordinal and visual analog scale (VAS) ratings of both speech understandability and speech acceptability using scales from the Rhinocleft Speech Assessment were made by a group of 4 SLPs and 12 adult lay listeners. Reliability of ordinal scale measures were estimated using kappa and Fleiss' kappa statistics and VAS measures were estimated using the intraclass correlation coefficient (ICC). Spearman's rho was used to estimate the strength of correlation between lay listener and SLP mean VAS ratings.

Results: Intra-rater reliability was fair to good for ordinal scale ratings (kappa = .25 to .72) and good to excellent for VAS ratings (ICC: .69 to .95). Inter-rater reliability was fair to moderate for ordinal scale ratings (Fleiss' kappa = .32 to .51) and excellent (ICC: .97 to .99) for VAS rating ratings. There was a high to very high positive correlation (Spearman's rho: .89 to .93) between the mean VAS ratings for the lay listener and SLP ratings.

Conclusions: This study has established that untrained, lay listeners demonstrated good to excellent degrees of intra-rater reliability and excellent inter-rater reliability for VAS ratings of global measures of speech performance which were superior to the results achieved on ordinal scale measures. Furthermore, these reliability estimates were consistent with the estimates derived from the experienced SLPs' ratings of the same speech samples using the same rating scales.

## 299. Speech Skills of Monolingual Spanish Speaking Children With and Without Cleft Palate: Preliminary Findings from Peru

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Background/Purpose: There are 41 million people in the United States who speak Spanish. Speech-language pathologists (SLPs) need to have knowledge of how speech differences could potentially be a dialect difference instead of a speech disorder. Many studies have documented speech errors in children who speak English with and without cleft palate (CLP); however, little is known about speech errors in children with CLP who speak Spanish. There is only one study to date which reported speech errors from 5 children with CLP who were monolingual Spanish speakers from Mexico (Guillen,

2005). While this study contributed new knowledge of speech errors in children with CLP who spoke Spanish, it did not include a group of typical peers without CLP (TD). A comparison group is essential to identify which errors may be associated with CLP versus which represent dialect differences in Spanish. The current study included a larger sample of children with CLP, a smaller age range, and a TD group.

Methods/Description: Fourteen monolingual Spanish-speaking children from Peru ages 4 to 7 (7 with CLP and 7 TD matched for age and gender) participated in this study. The following speech tasks were recorded: conversation, counting, phrase repetition, and single words on the Goldman Fristoe Test of Articulation-3 Spanish Edition (GFTA-3 Spanish). Recordings from the GFTA-3 were phonetically transcribed. Percentage consonants correct (PCC) from the GFTA-3 Spanish was compared across the 2 groups with an independent samples *t*-test. Speech samples were also analyzed qualitatively to identify error patterns for each sound and sound classes in Spanish.

Results: Participants in the TD group had higher PCC values on the GFTA-3 Spanish (M = 89.29, SD = 9.70) than the group with CLP (M = 63.06, SD = 24.66), t(12) = 2.618, P = .022, d = 1.40. Children with CLP produced more speech errors including glottal stops, glottal fricatives, pharyngeal stops, pharyngeal fricatives, nasal substitutions, mid-dorsum palatal stops, nasal fricatives, and backing errors. The TD group did not demonstrate the compensatory errors aforementioned; however, backing and nasal substitutions were present in both groups. Both groups also demonstrated stopping, interdental placement of /s/, cluster reduction, /l/ for the trilled /r/, and final consonant deletion.

Conclusions: This is the first study to prospectively assess the articulation skills of children with CLP and TD who are monolingual Spanish speakers. Preliminary findings suggest that compensatory errors in Spanish-speaking children with CLP are similar to those that have been documented in English. The Spanish phonology system is similar but different from English and differences exist regarding the developmental acquisition of phonemes in Spanish. This study contributes new knowledge about speech errors in children who speak Spanish with and without CLP. This information is critical for SLPs when evaluating children who speak Spanish as some speech differences could be dialectal and some errors require speech therapy.

#### 300. Development and Implementation of a Cleft Speech Scoring System That Complements Existing Speech Assessment Protocols

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Background/Purpose: The SickKids Cleft Speech Scoring System is the result of collaboration between cleft surgeons and speech-language pathologists. The purpose of the system is to produce a concise and comprehensive score that provides an overall impression of speech status for individuals with cleft and velopharyngeal dysfunction-related speech disorders. There has been discussion in the cleft palate literature regarding the need for a scoring mechanism that provides both a total outcome measure and separate scores for distinct speech parameters. The SickKids Cleft Speech Scoring System, which generates a SickKids Cleft Speech Score, offers a solution. It is both a summary score and a code that can be rapidly "decoded" to reveal the specific features contributing to an individual's velopharyngeal dysfunction-related speech status. Speech-language pathologists can apply the SickKids Cleft Speech Score to speech samples in any

language, for children or adults, using existing speech assessment protocols.

Methods/Description: In the first phase of development of the Sick-Kids Cleft Speech Scoring System, a single-center reliability study was conducted. Five speech-language pathologists with clinical expertise in cleft/velopharyngeal dysfunction speech assessment evaluated 18 two-minute controlled speech samples on 2 occasions, 1 month apart. The results showed good or excellent reliability for all the parameters, and all raters described the tool as a practical and useful complement to perceptual speech assessment. Having developed a reliable and valid tool for cleft speech reporting, the second phase of development involved preparing the system for distribution to other cleft centers. As such, a study to assess the sensibility of the scoring system was initiated to obtain feedback from speech-language pathologists in multiple cleft centers across North America. The study aimed to answer the following questions: What are the current methods used in North American cleft centers to record and communicate a child's speech outcome/status? Does the SickKids Cleft Speech Scoring System have good sensibility, content validit, and clinical utility? Do speech-language pathologists believe that the scoring system is successful in detecting differences between severity of cleft speech characteristics? Would speech-language pathologists use the new scoring system for reporting perceptual speech outcomes in daily practice? The objectives of this presentation are to (1) describe the SickKids Cleft Speech Scoring System, (2) discuss the process involved in the development of a new tool, and (3) provide an update on the status of the multicenter sensibility study.

## 301. What Matters Most?: Assessing Patient Family Identified Priorities for a Multidisciplinary Cleft Lip and Palate Team Visit

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Background/Purpose: This presentation will focus on an effective and quick procedure to determine family-identified priorities for multidisciplinary cleft care as well as data regarding specific familyidentified priorities gathered through a quality improvement initiative at a Midwestern multidisciplinary cleft lip and palate team. This team utilizes half-day multidisciplinary visits during which patients see any combination of the following providers: plastic surgery, ENT provider, social work speech language therapy, audiology, genetic counseling, pediatric psychology, orthodontics, prosthodontics, dentistry, nursing, and pulmonology. The specific providers who will see the patients for their team visit are identified ahead of time through chart review and standard practice; however, scheduling is flexible and team members may be added or removed throughout the day based on patient/family preferences, scheduling, and team member recommendations. This model of care is in accordance with ACPA parameters of care calling for an interdisciplinary team of specialists to provide management of patients with craniofacial differences; however, it does not take into account family-identified priorities in any standardized manner. In an attempt to ensure incorporation of familyidentified priorities into the multidisciplinary visit, this team began asking families "What matters most today?" at the start of their team visit. The goal of this presentation is to review the procedure this team implemented as well as to present data regarding family-identified priority on the day of their child's multidisciplinary team visit gathered through asking this question.

Methods/Description: All patients and families presenting for a multidisciplinary cleft lip and palate team appointment were asked "What matters most today?" at the start of their team visit with the intent of identifying their main concern(s) for that visit. The current sample includes responses from 237 patient families with data collection ongoing (expect to double sample size by the time of the ACPA conference). Families were asked this question by the provider who saw them first on the multidisciplinary clinic day. Attempts were then made to address these identified concerns as soon in the course of the visit as possible, which at times included adding an additional team provider to their clinic visit that day depending on the response. At the end of each visit, the last team provider seeing the patient would then confirm that the family's main concern for the day had been addressed and/or the family was aware of a plan in place to address this concern in the future. This presentation will review the above-described procedure, rational for this procedure, and data regarding familyidentified priorities gathered through asking this question, as well as discuss future directions for research related to family-identified priorities for a multidisciplinary cleft lip and palate clinic visit.

### 302. Reducing No-Show Rate and Increasing Team Capacity in a Multidisciplinary Cleft and Craniofacial Center: A Quality-Improvement Project

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Background/Purpose: When patients do not present to team clinic ("no-show"), it wastes clinical and financial resources and creates the extra burden of following up with these families to reschedule these appointments. Our team performed an informal audit and discovered we had an unacceptably high 32% no-show rate in 2017, despite the routine reminder calls provided by our institution. In response, we piloted a systematic communications program in an attempt to reduce the no-show rate. A secondary aim was to improve team efficiency and capacity through the use of a "waiting list" that would allow us to easily substitute patients in the event of cancelations/reschedulings.

Methods/Description: A rapid improvement event (RIE) project was modeled according to the Lean framework and Institute for Healthcare Improvement (IHI) model for improvement. In designing the plan-do-study-act (PDSA) cycle, we convened an extended-team focus group to identify the key drivers related to no-shows and to explore related areas of potential intervention for achieving our goal of reducing no-shows to <10% within 2 months. The principal intervention was early, direct verbal communication from the team coordinator with the family 2 weeks prior to their scheduled multidisciplinary team appointment. The secondary intervention was the creation of a waiting list, such that when conflicts were identified (via the reminder calls), alternate patients could be contacted to potentially fill empty slots. The intervention occurred in April, 2019. A one-month "washout" period was allowed after intervention before data collection. Results were tracked using a control chart.

Results: Historically, the no-show rate from 2017 was 32%. In the 40 clinics immediately prior our intervention, the no-show rate had already improved to  $17.8\% \pm 12.1\%$  (range: 0%-50%), indicative to some degree of the Halstead effect, given that we had increased attention on the matter of reducing no-shows. After intervention, the no-show rate across 14 clinics decreased to  $5.7\% \pm 6.0\%$  (range: 0%-12.5%; P < .0001). Secondarily, the intervention allowed for rescheduling 13 patients who would have otherwise been no-shows, and the waiting list allowed for the substitution of 5 patients into these slots (38.4% fillage). Team total capacity (maximum number of patients that the team could manage to see in one clinic) did not significantly increase.

Conclusions: This simple intervention allowed us to meet our principal aim of reducing no-show rate to <10% within 2 months, as well as our secondary aim of filling slots that would have otherwise gone unused. Although the additional phone calls required significant time on the part of the team coordinator, it effectively amounted to time shifting from reactive to proactive communication: that is, these were phone calls that would have been made anyway in order to track down and reschedule patients had they no-showed. The reduction in no-shows allowed for improved efficiency, reduced waste, and added value.

## 303. Is Team Care Standard Care for Children with Cleft Lip/Alveolus? A Survey of Current Team Practices and Perceptions

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Background/Purpose: Management of children with cleft lip and/or palate is best provided by an interdisciplinary team (ACPA Parameters of Care, 2018). Although the team care needs of children with cleft palate, with or without cleft lip (CP/L), are well known, there remains ambiguity whether children with cleft lip/alveolus without cleft palate (CL/A) require the same care pathway. The aims of this study are to (1) describe current cleft team care practices for children with CL/A, and (2) assess team coordinator perception of the care needs of children with CL/A.

Methods/Description: A REDCap-based survey was distributed by e-mail to all ACPA-approved team coordinators in the US. Survey questions focused on whether children with CL/A routinely received team visits, which team members routinely saw children with CL/A, differences in team care delivery for children with CL/A versus CP/L, type and frequency of assessments offered to children with CL/A, and so on. Coordinators were asked about their perception of the risk of speech, hearing, dental, and appearance concerns in children with CL/A. Responses were collected anonymously. Data were exported into SPSS for analysis.

Results: Fifty-seven surveys were completed (35% response rate), with 71% of surveyed coordinators indicating that they were a nurse; 19.6\% of team coordinators reported they were not familiar with the ACPA Parameters of Care; 78.9% indicated they routinely see children with CL/A for team visits, with 40% seeing them annually. Children with CL/A were most likely to see the plastic surgeon (94%), feeding specialist (90%), and orthodontist (78%). Half of teams employ a "different" approach to team care for children with CL/A compared to children with CP/L; 50% routinely referred children with CL/A for hearing testing and 35% reported that they receive a speech evaluation every 1 to 2 years; 47% of teams only evaluated speech "as needed," yet 48% of coordinators thought children with CL/A had a higher risk for speech disorders than the general population. The majority felt that children with CL/A had a high risk of appearance- and dental-related concerns. Qualitative analysis of open-ended responses revealed that most coordinators felt children with CL/A should receive help with psychosocial needs (28%), dental/jaw needs (59%), and speech-language or hearing (38%). Overall, 70% of respondents felt that all children with CL/A should receive routine team visits, regardless of need.

Conclusions: There is considerable variability in current team care practices for children with cleft lip/alveolus, without cleft palate,

among ACPA-approved teams. Teams may benefit from further education regarding the care needs of children with CL/A, and current parameters of care. Screening tools that can assist with needs assessment may also be helpful to guide team care decision-making.

### 304. Evaluation of Provider-Specific Outcomes Through a Multidisciplinary Team Clinic for Patients with Isolated Cleft Palate

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Background/Purpose: Children with isolated nonsyndromic cleft palate (CP) often receive long-term follow-up through multidisciplinary cleft team clinics, in accordance with consensus recommendations of the American Cleft Palate-Craniofacial Association (ACPA). However, follow-up protocols for these patients remain heterogeneous, in part because their longitudinal health needs have been underdescribed. This study aims to inform team assessment schedules for patients with CP with the goal of improving resource allocation and reducing costs. Methods/Description: This was a retrospective cohort study of children with CP evaluated by an ACPA-approved team at a tertiary academic children's hospital between January 2000 and July 2019. Medical records were reviewed for demographic information, age at primary cleft repair, team composition at each visit, and follow-up duration. Study outcomes included incidence of secondary palatoplasty, tympanostomy tube insertion, speech therapy, hearing loss in at least one ear, dental/orthodontic treatment, and psychology interventions. Provider-specific outcomes were calculated for patients at ages 0 to 3, 3 to 5, and >5 years.

Results: One hundred forty-two children met inclusion criteria. Median age at the final team visit was 7.0 years (IQR: 3.3-11.8 years). At their last team assessment, 42% of patients still had conductive hearing loss (12% slight, 22% mild, and 8% moderate). The rate of isolated tympanostomy tube insertions, that is, those not done alongside a palatoplasty, was highest for ages 3 to 5 (210/1000 person-years) and dropped significantly after the introduction of new AAO-HNS guidelines in 2013 (P = .015). A total of 76 children (54%) received speechlanguage therapy during follow-up. Speech therapy initiation rates decreased after age 5 (P < .001). Palatoplasty, psychology intervention, and dental/orthodontic treatment were all less common than speech therapy or tube insertion (P < .01). Secondary palatoplasty was performed in 31 (22%) patients. Most of these procedures were for velopharyngeal insufficiency (53%) or fistulae (43%). Rates of psychology intervention were 46/1000 person-years. Rates of dental/ orthodontic treatments were 55/1000 person-years. Median followup was longer for patients who received speech therapy, dental/orthodontic treatment, or psychology intervention than for those who did not (9.8 vs 2.1 years, P < .001).

Conclusions: Patients had lower rates of secondary palatoplasty, psychology intervention, and dental/orthodontic treatment compared to rates of tube insertion and speech therapy. Half of the patients terminated team follow-up by age 7, suggesting that burden of care outweighs perceived benefits of continued follow-up for many families. However, patients with a speech, psychology, or dental/orthodontic intervention continued to follow-up for longer. These results can be used by cleft teams to establish protocols for children with CP and provide value-based care.

# 305. Capturing (and Getting Paid) for Those Non-Face-to-Face Hours of Care: Thinking About CPT 99358 & CPT 99359 for Craniofacial Psychology Practice

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Background/Purpose: Craniofacial psychologists provide many clinically important services outside of the traditional face-to-face patient encounter. Increasingly, psychologists are being held to physician models of fiscal accountability and productivity. Services to enhance patient care, such as coordinated treatment planning with craniofacial team members, extended review of outside records, collaboration with schools or other treatment providers, IEP/504 consultation, or preparation of highly individualized treatment plans are often not captured in a psychologist's billing and productivity measures. Prior to 2017, these clinically important, but non-face-to-face services, were not reimbursed by Medicare or private insurances. In 2017, Medicare quietly "opened" 2 important CPT codes for reimbursement by psychologists: CPT code 99358 (prolonged services without face-to-face contact before and/or after direct patient care; first hour) and add-on CPT code 99359 (prolonged services without face-to-face contact before and/or after direct patient care; additional 30 minutes beyond first hour). This change allows psychologists to bill for some of the clinically important non-face-to-face services we provide as craniofacial team members. This brief talk will review the potential use of CPT codes 99358/+99359 from the perspective of an outpatient craniofacial team psychologist.

Methods/Description: In 2017, Medicare began reimbursing psychologists for CPT code 99358 (Prolonged services without face-to-face contact before and/or after direct patient care; first hour) and add-on code 99359 (prolonged services without face-to-face contact before and/or after direct patient care; additional 30 minutes beyond first hour) under the Medicare Physician Fee Schedule. Many private insurers have followed suit. With a national average reimbursement rate of \$113 per unit of CPT 99358 and a national average reimbursement rate of \$55 per unit of CPT + 99359, these clinically important services should be captured as part of daily productivity and billing in a psychologist's practice. This power point presentation will (1) review the definition of CPT codes 99358/+99359, (2) describe their potential uses in craniofacial psychology practice, (3) identify the time limits and restrictions on using these codes, (4) illustrate their use with a relevant case example, (5) offer recommendations for documenting the use of these codes, and (6) provide resource links to further explore the viability of utilizing these codes in your own practice.

Conclusions: With the opening of CPT codes 99358/+99359, Medicare now allows payment for some of the clinically important but indirect services that psychologists have always provided in the medical setting. With proper utilization and documentation, these 2 codes can positively impact craniofacial psychologists' fiscal accountability and viability as members of a specialty team.

#### 306. A Retrospective Review of Medical Insurance Coverage from a Single Private Orthodontic Office for Patients with Cleft and Craniofacial Conditions

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*Background/Purpose*: Multiple phases of orthodontic treatment are often necessary as part of the rehabilitation of a patient born with cleft or craniofacial condition. One of the burdens of orthodontic care is the

cost. Insurance coverage for orthodontics is often not well understood by providers or patients, varies between states and companies, and confusion exists between orthodontics being covered by medical and/or dental insurance. In addition, not many orthodontic offices are set up to file medical insurance claims. The purpose of this retrospective review is to evaluate the outcome of billing private medical insurance for orthodontic care in patients with cleft/craniofacial conditions in a private practice orthodontic office.

Methods/Description: A financial chart audit of patients with the diagnosis of a cleft/craniofacial condition who started treatment between 2009 and 2019 in a single private orthodontic practice in Portland, Oregon, was performed by the same financial coordinator that managed each account. An initial insurance claim (and a first appeal when necessary) using CPT codes 21110 and 99214 was submitted along with a letter of medical necessity. The overall claim approval rate, the percentage of claims requiring appeal, and average amount of the benefit paid were calculated. These variables were also evaluated relative to the passage of HB 4128 in 2012 which required medical insurance companies in Oregon to cover medically necessary dental procedures.

Results: A total of 178 patients with a cleft/craniofacial diagnosis were identified. There was an 88% overall approval rate for getting medical insurance to provide coverage for orthodontic treatment. First-level appeals were required in 51% of the cases, but this dropped to 26% after the passage of HB 4128. The first-level appeal was successful in 75% of the cases. The insurance companies paid an average of 25% of the treatment fee.

Conclusions: In Oregon, billing medical insurance from a single private orthodontic office for patients with cleft/craniofacial conditions can be successful and helps to reduce the financial burden of the care. Although the initial claim may be denied, the first-level appeals were usually successful. The need for an appeal was reduced by half after the passing of HB 4128 in Oregon. An average of 27% of the treatment fee was paid out.

Discussion: High deductibles were most directly responsible for the relatively low percentage of coverage of the overall orthodontic treatment fee. Second level and higher appeals were not included in this study. They were handled by each patient family and often not followed through with. Further work could be done to find out if other CPT codes could be more appropriate or effective in this type of billing, and if it would be possible to get CPT codes created specifically for orthodontic care. State-specific rules and managing the billing learning curve should be considered when evaluating how this type of billing may be applicable in another office.

#### 307. The Team Takes Care of Everyone. Who Takes Care of the Team?

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Background/Purpose: Health-care providers experience high levels of burnout and compassion fatigue. Additionally, working in a health-care setting, providers are 4 times more likely to be victims of work-place violence than those working in private industry. Providers may also find themselves in situations that are ethically questionable or morally distressing. These experiences result in increased levels of stress. Craniofacial teams also have their own unique stressors, with providers caring for complex patients with multiple diagnoses who may be difficult to treat, possibly in the setting of social or cultural barriers to care. Families who seek out information on the Internet or through social media may have unrealistic expectations or demands,

leading to families feeling disappointed in outcomes or frustrated by perceived shortcomings in care. The purpose of this presentation is to acknowledge the challenges of providing care to patients with Cranio-facial differences in the 21st century and to identify the ways in which providers can protect against stress and burnout.

Methods/Description: A review of current data regarding stress among health-care providers was conducted. Resources to support health-care providers in various types of adversity, both locally and nationally, were reviewed and compiled. The main objective of the presentation is to reach providers who are experiencing professional burnout and help them identify their need for self-care, provide suggestions for supporting their Team members and to be an advocate for institutional wide "caring for the caregiver" programs.

### 308. What is Normal Newborn Sleep? A Characterization of Sleep Patterns in Neonates with and Without Airway Obstruction

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Background/Purpose: Polysomnography is vital in evaluating neonatal airway obstruction. Although many institutions use sleep data to select patients for mandibular distraction osteogenesis (MDO), no "normal" published references exist for this age. We present normative polysomnography data for newborns age 0 to 1 month. We also compare this normative reference to pre- and postoperative data of infants this age undergoing MDO.

*Methods/Description*: Following IRB approval, normative subjects were recruited from our NICU to undergo nap polysomnography. Included were infants without airway obstruction, gestational age 37 to 42 weeks, and age less than 28 days. Data included apnea—hypopnea indices, pulse oximetry, CO<sub>2</sub>, EEG, and ECG. One blinded sleep physician read all studies. Sleep data for newborns undergoing MDO were collected prospectively (2016-18). All data were collected and analyzed using REDCap and SPSS software.

Results: Twenty-one neonates without airway obstruction provided normative sleep data; median age at polysomnography was 4.5 days and median sleep time was 181 minutes. Median total apnea-hypopnea (AHI), obstructive apnea-hypopnea (OAHI), and central apnea indices (CAI) were 7.6, 5.0, and 0.8 events/hour. The median O<sub>2</sub> nadir was 91%. Polysomnography was done on 15 neonates with airway obstruction before and after MDO. Median age at preoperative study was 7 days and median sleep time was 328 minutes. Median AHI was 38.3, OAHI was 37.0, and CAI was 2.1. Median O2 nadir was 84%. Prior to undergoing MDO, neonates with airway obstruction had significantly worse AHI, OAHI, and O2 nadir than normative counterparts (P < .001). There was no significant difference in CAI. Postsurgical sleep data were collected after activation phase of MDO; median age was 59 days and median sleep time was 363 minutes. In this group, median AHI was 6.1, OAHI was 4.8, and CAI was 1.4. Median  $O_2$  nadir was 91%. Paired t tests demonstrated significant improvements in OAHI, AHI, and oxygen saturation nadir after MDO (P < .001). When comparing the normative group to neonates who underwent MDO, there was no significant difference in oxygenation or any apnea-hypopnea index.

Conclusions: In children, OAHI > 1 is considered abnormal; this norm has been extrapolated to neonates. Our findings demonstrate "normal" neonates have more obstructive and central apneic events than previously appreciated, with a median of 5.0 obstructive and 7.6 total events per hour. Furthermore, newborns without airway obstruction

still exhibit a wide range of "normal" OAHI values (1.6-35.7). Newborns with airway obstruction had significantly worse OAHI/AHI and  $O_2$  saturation nadir than their nonobstructed counterparts and exhibited improvement in normative levels following MDO. Each center with a multidisciplinary MDO team should consider collecting normative neonatal sleep data to reflect their regional population, enabling calibration of existing patient selection algorithms, and informing important discussions with anxious parents.

## 309. PSG-Based Phenotypical Characterization of OSA in Robin Sequence Including Arousal and Sleep-Stage Specific Indices

Julia Aziz (1), Gustavo Nino (1), Esperanza Mantilla Rivas (1), Monica Manrique (1), Daniel Lewin (1), Jenny Lew (1), Gary Rogers (2), Albert Oh (1)

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Background/Purpose: Robin sequence (RS) is a top cause of obstructive sleep apnea (OSA) in early life. Most studies have focused on understanding anatomic factors leading to OSA and changes in apnea—hypopnea index (AHI) on polysomnography (PSG), but little is known about other PSG features of OSA in RS. The goal of this study was to compare OSA features in newborns with RS versus infants with OSA using PSG-based determination of arousal responses and sleep-stage-specific respiratory indexes.

Methods/Description: We included 10 newborns with RS (mean age: 1 month) and 20 nonsyndromic infants aged 0 to 2 years (10 non-RS with severe OSA and 10 controls without OSA). We obtained standard OSA severity parameters (OAHI, AHI, and RDI) and used raw data to calculate sleep-stage-specific indexes (REM vs NREM) as well as position-specific indexes (supine vs nonsupine). For arousal responses, we used respiratory arousal indexes correlated with individual's AHI. Impact of OSA in gas exchange was assessed by SpO<sub>2</sub> values, CO<sub>2</sub> values, and correlation with AHI.

Results: All standard OSA parameters (AHI, OAHI, and RDI), gas-exchange values, sleep-stage-specific indexes, and position-specific indexes showed similar distribution in newborns with RS and in non-RS infants with OSA. AHI correlated strongly with intermittent hypoxemia (O2 desaturation index) in the non-RS group with OSA (R-sq = 93%, P < .01) but only moderately in RS newborns (R-sq = 62%, P = .01). In both OSA groups, AHI correlated with sustained hypoxemia (time < 90%; non-RS Rsq = 86%, P < .01; RS R-sq = 73%, P < .01) and with nadir SpO<sub>2</sub> (non-RS R-sq = 84%, P < .01; RS R-sq = 81%, P < .01). Non-RS infants with OSA exhibited significant correlation between AHI and respiratory arousal indexes (R-sq = 72%, P < .01); however, RS newborns showed no correlation (R-sq = 28%, P = .11), particularly during REM sleep (R-sq = 6%, P = .49)

Conclusions: Newborns with RS and OSA seem to have reduced arousal mechanisms to terminate apneic events, particularly during REM sleep. This may be due to young age and the trend of RS babies to have less intermittent hypoxemia, an important arousal trigger in OSA. We believe PSG-based characterization of arousal and sleep-stage-specific indexes in RS may contribute to determine the best candidates for surgical correction by identifying individuals with higher risk for severe OSA due to a combination of anatomical factors and reduced arousal mechanisms to terminate apneic events.

#### 310. Sphincter Pharyngoplasty Surgery Simulation Using Finite Element Computational Modeling

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Background/Purpose: Sphincter pharyngoplasty is one of the most common surgical treatments for hypernasality in patients with velopharyngeal (VP) dysfunction (Ysunza et al., 2004). The goal of the surgery is to augment the posterior pharyngeal wall (PPW) to facilitate improved velopharyngeal closure (Sie et al., 2009). Despite its wide use, the outcomes of these surgeries are variable and there remain several questions as why some surgeries fail. For example, does variation in the pharyngoplasty location influence the outcome? Does variation in the VP anatomy itself influence outcome? Although these questions are difficult to answer clinically, computational modeling has the ability to simulate the VP movement based on anatomical input scenarios, allowing us to answer these important questions. The goals of the study are to (1) develop a computational model of the sphincter pharyngoplasty and associated VP mechanism, and (2) use the model to simulate multiple scenarios in order to examine potential causes of a failed surgical outcome.

Methods/Description: A 3-dimensional model, including the velum, levator veli palatini (LVP) muscle, pharyngoplasty, and PPW, was developed based on MRI data from Mason et al. (2019). The subject's VP anatomy was imaged following sphincter pharyngoplasty, and the postsurgical inflammation and migration of the sphincter tissue was evaluated. Mechanical properties of muscle and soft tissue were incorporated into the finite element model based on previously studies (Blemker et al., 2005 and Inouye et al., 2015). The LVP muscle was activated to simulate VP closure at the multiple locations including pre and post sphincter migration locations and 5 hypothetical sphincter placements superior to the original placement. The model output of LVP muscle's minimum activation required for closure was used to examine the effectiveness VP closure mechanism.

Results: The model simulations resulted in high required muscle activation to achieve VP closure at pre and post sphincter migration location (24.7% and 25.5%, respectively) compared to healthy population (approximately 16% in Pelland, 2019). The surgical outcome could be explained by the lack of contact between the velum and sphincter tissue at both locations. Furthermore, the asymmetry in LVP overlap contributed to the increase in required activation for VP closure. Increasing the location of the sphincter by 1 to 5 mm above original placement decreased the LVP muscle's required activation (5.12%-1.71%) and negated the impact of any asymmetry in levator muscle sling to VP closure.

Conclusions: Simulations predicted that superior/inferior location of the sphincter had a significant impact on activation required for closure. The computational model was able to capture biomechanical properties of VP closure post-pharyngoplasty across multiple anatomic variations in pharyngoplasty location. Applications of computational modeling provide insight into factors that may improve surgical outcomes following pharyngoplasty.

## 311. The Regeneration Effects of Extracorporeal Shock Wave Therapy on Distraction Osteogenesis in Rats' Mandible

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Background/Purpose: To investigate whether extracorporeal shock wave therapy can accelerate bony consolidation and regeneration in distraction osteogenesis of the rat mandible. Second, at which stage of distraction osteogenesis EWST is most effective to accelerate bone consolidation and regeneration.

*Methods/Description*: Twenty-four male Sprague Dawley rats were subjected to DO of the right mandible (latency period, 3 days; distraction period, 10 days at a rate of 0.5 mm/d). Rats were divided to 3 groups: group I (control) without ESWT, group II received ESWT (0.18 mJ/mm<sup>2</sup>) at latency period, and group III received ESWT (0.18 mJ/mm<sup>2</sup>) at consolidation period.

Results: After 4 weeks of consolidation period animals were killed, and explants were removed for radiographic, histological, collagen orientation, micro-CT, and immunohistochemical (IHC) evaluation. Radiographic X-ray showed more radiopacity in both ESWT treatment groups compared with control. Histological evaluation detected intense capillary formation, osteocytes within the mature bone, and bone remodeling compared to other groups. The collagen orientation index showed more lamellar bone in group III, as opposed to control group which exhibited more woven bone. µCT of the distracted mandible showed significantly increased bone mineral density, bone volume fraction, and trabecular thickness in group III compared to control group (P < .05). Immunohistochemistry demonstrated significantly increased expression of bone morphogenetic protein-2, vascular endothelial growth factor, proliferating cell nuclear antigen, osteocalcin, and collagen type-1 proteins in group III compared to control group.

Conclusions: The present study demonstrated that ESWT application at consolidation period during DO in rat mandible enhances bone formation, extracellular bone matrix protein, osteogenic and angiogenic growth factors; improves bone mechanical properties; and accelerates bone mineralization.

## 312. Implementation of a Clinical Care Pathway for Cleft Palate Surgery Reduces Opiate Consumption and Length of Stay

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Background/Purpose: Cleft palate repair is a painful procedure and achieving adequate postoperative analgesia is challenging. The optimal perioperative management approach for patients undergoing primary cleft palate repair is unknown. Comprehensive clinical care pathways can improve postoperative pain control and decrease length of hospital stay. We instituted a clinical care pathway for patients undergoing cleft palate repair and evaluated its impact on specific clinical outcomes.

Methods/Description: A novel pathway was created through an iterative process, combining literature review with expert opinion and discussions with institutional stakeholders. The pathway focused on multimodal analgesia throughout the perioperative course, including intraoperative suprazygomatic bilateral maxillary nerve blocks and scheduled acetaminophen and ibuprofen postoperatively. Medical records of 40 consecutive patients undergoing primary cleft palate repair prior to pathway implementation (January 2016 to October 2018) were retrospectively reviewed as controls; 15 patients treated with the pathway were prospectively reviewed. Primary outcomes were (1) length of hospital stay, (2) cumulative opiate consumption, and (3) time to initiation of oral intake. This work received ethics

approval from the Conjoint Health Research Ethics Board (CHREB) at the University of Calgary.

Results: There were no differences in baseline demographic data between the treatment and control groups. The patients following the pathway had shorter mean length of stay (32 vs 57 hours, P < .001), decreased cumulative morphine consumption (73 vs 727 µg/kg, P < .001), and shorter time to initiate oral intake (10 vs 22 hours, P = .01). There were no differences in length of total anesthesia time, total surgical time, or complication rates between the control and treatment groups. Statistical analysis was conducted using SPSS (version 24.0).

Conclusions: Implementation of a perioperative clinical care pathway for cleft palate repair is safe, feasible and associated with reduced length of stay, reduced opioid consumption, and faster time to oral intake. Consequently, the pathway is now the standard of care at our institution and will form the basis for the development of further protocols for other surgical procedures. These findings could also provide a foundation for other pediatric centers to institute similar clinical care pathways.

## 313. Comparison of Airway and Feeding Outcomes of Mandibular Distraction, Tongue-Lip Adhesion and Conservative Management in Pierre-Robin Sequence

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Background/Purpose: Pierre-Robin sequence (PRS) is the triad of micrognathia, glossoptosis, and upper airway obstruction. Many patients also have feeding difficulties. In mild cases, conservative management, consisting of prone/side positioning, is sufficient. In more severe case, surgical intervention, consisting of mandibular distraction osteogenesis (MDO) or tongue—lip adhesion (TLA), may be necessary. Our goal was to evaluate airway and feeding outcomes of conservative management, MDO, and TLA in the treatment of PRS.

Methods/Description: A retrospective review of newborns treated for PRS between 2005 and 2018 was performed. Patients who had polysomnograms (PSG) obtained both before and after treatment were included. Patients treated conservatively were included if they had a baseline PSG and a follow-up PSG at least 3 months later. Preoperative and postoperative data, including syndromic status, time to extubation, time to discharge, complications, need for gastrostomy tube, failure to thrive (any weight percentile <5%), and PSG data, were collected. Outcomes of the 3 treatment modalities were compared using the Kruskal-Wallis test.

Results: The study included 67 patients with PRS, 26 (38.9%) of whom had a syndrome; 29 patients underwent MDO, 19 underwent TLA, and 19 were treated conservatively. Baseline AHI was similar between MDO (25.4) and TLA (20.1, P=.2), but was lower in patients treated conservatively (9.1, P<.05 compared to MDO and TLA). Postoperatively, there was no difference in AHI between patients who underwent MDO (1.3), TLA (4.2, P=.2) or conservative management (4.5, P=.3). With regard to the reduction in AHI, it tended to be larger with MDO than TLA, although this was not significant (94.8% vs 72.6%, P=.1). There was no difference in the proportion of patients treated with MDO and TLA who achieved a postoperative AHI less than 5 (96.6% and 89.5%, respectively, P=.3). There was a tendency toward a higher complication rate with MDO than TLA (31% vs 10.5%, P=.09). MDO complications

consisted of 8 infections, all of which resolved with oral antibiotics, and 1 readmission for opioid withdrawal. TLA complications consisted of one dehiscence requiring repeat TLA, and one persistent upper airway obstruction requiring tracheostomy. There was a tendency toward more gastrostomy feeding tubes in patients who underwent TLA compared to those who underwent MDO (31.6% vs 10.3%, P=.06). The number of days from surgery to extubation was shorter in patients undergoing TLA than those undergoing MDO (2.2 vs 9.7, P<.001). However, the number of days from surgery to discharge was similar (35.9 vs 26.6, P=.3).

Conclusions: Although MDO tended to lower AHI more than TLA, both procedures achieved AHI reduction to less than 5 in the majority of patients. MDO tended to have more complications, although most were mild. Patients undergoing TLA tended to require more gastrostomy tubes. Patients undergoing TLA had shorter time to extubation, but similar time to discharge, than those undergoing MDO.

### 314. Is the Message Clear? Evaluation of Readability Levels for Cleft Lip, Cleft Palate, and Craniosynostosis Websites

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Background/Purpose: Web-based health information has become the leading source of medical knowledge for patients and families. The American Medical Association (AMA) and US Department of Health and Human Services recommend patient reading material be at or below a sixth-grade reading level. Readability equations can be used to quantify reading ease and grade level for both printed and online materials. Caution must be taken with web-based information as content may vary in reliability, understandability, and bias. The value of these websites to provide parental understanding of cleft lip and palate, and craniosynostosis has never been studied. The goal of this study was to evaluate the reliability and readability of the most popularly searched websites providing parents information related to cleft lip, cleft palate, and craniosynostosis.

Methods/Description: A Google search of "cleft lip" and "cleft palate" (top 40 websites), and "craniosynostosis" (top 30 websites) was performed in an incognito window with the location set to the United States. Duplicate websites and those without text were excluded. Validated reading understandability tools Flesch Reading Ease Score (FRES) and Flesch-Kincaid Reading Grade Level (FKGL) were used to determine the ease of reading and reading grade level of the website content. For a sixth-grade reading level (easy to read, conversational English for consumers), the FRES is 80 to 90 and the FKGL is 6.0 to 6.9. The higher the FRES and the lower the FKGL, the easier the website is to read. Scores were stratified based on nonacademic (NA) versus academic (A) website cohorts.

Results: For the top 40 websites relating to "cleft lip/cleft palate," the average FRES was 58.5  $\pm$  12.1 (fairly difficult to read) and FKGL was 9.4  $\pm$  2.2 (plain English, easily understood). There was no significant difference between NA and A websites for FRES (56.1  $\pm$  14.8 vs 61.0  $\pm$  8, P = .21) and FKGL (9.7  $\pm$  2.6 vs 9.0  $\pm$  1.8, P = .32). There were 6 (15%) at or below FKGL of 6. For the top 30 websites relating to "craniosynostosis," the average FRES was 40.7  $\pm$  14.2 (difficult to read) and FKGL 11.8  $\pm$  2.5 (fairly difficult to read). There was no significant difference between NA and A websites for FRES (38.9 vs 41.7, P = .67) and FKGL (11.9 vs 11.6, P = .79). There was only one website at or below FKGL of 6. Overall, 7 websites (10% [4 A and 3 NA]) were found to be at or below the sixth-

grade level set by the AMA for web-based information related to cleft lip, cleft palate, and craniosynostosis.

Conclusions: Web-based medical information related to cleft lip, cleft palate, and craniosynostosis is on average at the FKGL 10th-grade reading level. This is higher than the AMA recommended sixth-grade readability for patient education materials with greater difficulty of reading (FRES > 65 recommended). These validated reading instruments may be beneficial for the development of improved online formation for patients with cleft lip, cleft palate, and craniosynostosis.

### 315. Barriers and Facilitators to Implementation of Standardized Outcome Measurements for Children with Cleft Lip and Palate

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Background/Purpose: Standardized outcome measurement is an essential component of evaluating and improving health-care services. Although cleft centers in the United Kingdom have implemented standardized evaluations of aesthetic, speech, and dental outcomes for children with cleft lip and palate, to date there has not been widespread adoption of standardized outcome measurements at cleft centers in the United States (US). To overcome this gap, we sought to identify facilitators and barriers to adoption of standardized outcome measurements in cleft care.

Methods/Description: Individual semistructured qualitative interviews were conducted with 24 providers and staff representing all treatment disciplines at a single US-based cleft center and 3 three key stakeholders from the United Kingdom. Interviews explored the barriers and facilitators to implementing standardized outcome measurements, focusing on collection of standardized facial photographs, dental models, and speech evaluations at age 5. Interviews were audio recorded, transcribed, and analyzed for content using the Consolidated Framework for Implementation Research (CFIR).

Results: Qualitative analysis identified multiple barriers and facilitators to standardized outcome measurements. Barriers to implementation fit into 2 general constructs—adaptability and complexity. Providers were concerned the inability to adapt the measurements to individual patients may prevent nuanced evaluations and, for speech measures, prevent evaluation of non-English speakers. Providers also felt that implementing standardized outcome measurement would be complex, requiring changes to systems for patient recall, standardization of record taking across providers, and addressing patients' fear and anxiety about the evaluation. Two facilitators to implementation were consistently described: providers viewed standardized outcome measurements as the gold standard with strong evidence supporting its adoption; they also prefer consistent, systematic approaches to evaluating children. In addition, providers believed standardized outcome measurements would facilitate evaluation of their treatment practices.

Conclusions: This is the first study to systematically evaluate barriers and facilitators to implementing standardized outcomes measurements in cleft care. Our results suggest that an implementation strategy, which facilitates patient attendance to clinic and reduces patient's fear and anxiety related to their evaluation, may increase adoption of standardized outcome measurements. Further, providers' engagement in the adoption process may be strengthened by building on their preferences for systematic evaluations in clinic and their desire for feedback regarding their treatment outcomes. Pilot testing of an implementation strategy incorporating these findings is an important next step in pursing the objective of continuous evaluation and

improvement of treatment outcomes for children with cleft lip and palate.

#### 316. Disparities in Access to Craniofacial Centers for Newly Diagnosed Craniosynostosis: The Influences of Urban Versus Rural Residence and Private Versus Government Insurance

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Background/Purpose: One of the goals of recent US health-care reform is to increase access for all citizens. The aim of this study is to assess disparity in access for craniosynostosis surgery for a single Midwestern United States craniofacial center in 2018 with an emphasis upon patient insurance status and urban versus local residence.

*Methods/Description*: The charts of all patients who underwent primary craniosynostosis repair in 2018 at our institution were reviewed for demographic factors, age at time of consultation, and surgical technique (open cranial vault remodeling vs endoscopic strip synostectomy).

Results: Fifty-four patients, ages 2 to 22 months, underwent primary craniosynostosis surgery at our institution in 2018; 34 underwent an open procedure, while 20 underwent a strip craniectomy, followed by helmeting; 47 of these patients were nonsyndromic. Twenty-eight (52%) patients had private insurance; 24 (44%) patients had statefunded Medicaid insurance, and 2 (4%) were without insurance. Twenty-nine (54%) patients lived in an urban environment; 25 (46%) resided rurally. Further results include (1) While 52% of patients had private insurance; 75% of patients who underwent strip craniectomy were privately insured; 25% were Medicaid or not insured; (2) 44% of patients who underwent open vault reconstructions were privately insured; 56% were Medicaid or uninsured. (3) While 54% of patients were urban-based, 74% of patients who underwent strip craniectomy were urban-based; 26% were rural; (4) 45% of patients who underwent open vault procedures were urban-based; 55% were rural. (5) For strip craniectomy patients, those with private insurance were first seen in consultation at a mean of 42 days of life. With Medicaid or uninsured, it was 58 days of life. (6) For insured urbanbased strip craniectomy patients, mean day of first consultation was 37 days. If rural-based, the mean was 57 days.

Conclusions: Disparities in 2018 health-care access for US craniosynostosis patients exist with delayed presentation in clinic for less-insured and/or rural patients as well as fewer strip craniectomy/helmeting procedures in these patients. We will also provide 2019 data at our presentation which we will compile at year's end.

#### 317. Cost Analysis of Cleft Care in the First Year of Life

Ingrid Ganske (1), Olivia Langa (1), Karl Sanchez (1), Jason Yu (1), John Meara (1)

(1) Boston Children's Hospital, MA

Background/Purpose: With increased attention to value-based health-care reform and potential bundled care payments on the horizon, it is important to understand the individual components of cost that go into multidisciplinary care models such as cleft care. Understanding the drivers of cleft care costs is also important for individual cleft centers focused on optimizing the cost/outcomes value equation.

Methods/Description: A retrospective review was performed from 2012 to 2019 identifying all infants who completed care for cleft lip

and/or palate (CL  $\pm$  P) in the first full year of life at our institution. Patients comprised 3 phenotypes: cleft lip alone (CL), cleft lip and palate (CLP), and cleft palate alone (CP). Nonsyndromic and syndromic cohorts were separated. Costs were calculated from charges obtained from hospital and department billing records. Costs attributable to the following components of cleft care were tabulated and averaged: inpatient hospitalizations, intensive care utilization, operating room (OR) services, physician billing, and ancillary services such as laboratory and radiology services. Total cost of care, including noncleft-related charges were also collected and averaged for both nonsyndromic and syndromic cohorts.

Results: Costs varied by phenotype and presence of syndromic diagnosis. All patients with nonsyndromic clefts completed all indicated procedures within the first 12 months of life; whereas 26% (n = 15) with syndromic clefts did not. Average total cost of cleft care for a child with nonsyndromic CL (n = 66) was \$22 550 (main components: 33% OR, 33% plastic surgeon, 11% inpatient, and 13% anesthesiologist); for CLP (n = 70) was \$61 200 (36% OR, 29% plastic surgeon, 16% inpatient, and 13% anesthesiologist); and for CP (n = 63) was \$19 450 (23% OR, 27% inpatient, 23% plastic surgeon, and 10% anesthesiologist). For patients with syndromic diagnoses, there was wide variation in cost of care, reflecting increased utilization of diagnostic services, treatments, prolonged admissions, and intensive care. The main cost driver for infants with syndromic orofacial clefting was intensive care, comprising 33% of costs for CL (n = 7), 21% for CLP (n = 18), and 32% for CP (n = 34). Separating cleft and noncleftrelated components of cost for infants with syndromic diagnoses was complicated by the concurrent and multidisciplinary care provided during lengthy hospitalizations. Plastic surgery physician billing was a small component of the overall cost of care for patients with syndromic clefting: 2.5% CL, 11% CLP, and 2.5% CP.

Conclusions: The average cost of care for infants with nonsyndromic CL  $\pm$  P is divided approximately evenly between hospital and physician billing and can be estimated for a defined 1-year period. The costs for syndromic CL  $\pm$  P are more difficult to project, given the variable completion of care during a standard time frame, higher utilization of intensive care, and comingling of costs for cleft care and other medical conditions.

#### 318. An Analysis of USA and European Craniofacial Center Websites: US Craniosynostosis Families are Being Given Fake News. European Families are Being Given Much Less but Accurate Information

Jeffrey Goldstein (1), Amanda Johnston (1), Michael Lypka (1) (1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: Craniofacial center websites are an early and significant source of education for parents. The aims of this presentation are also to analyze the information given by American and European craniofacial websites to selected questions about craniosynostosis and to assess the variability and accuracy of these answers.

Methods/Description: The Internet search phrases "craniosynostosis," "craniofacial center," and "craniosynostosis center" were employed, leading to 53 US center websites and 25 European center websites. The websites were reviewed. Answers to select questions were recorded and assessed.

Results: Answers to questions were highly variable between American websites with disparate statements noted, including: (1) 13% of websites state that surgery is required for all craniosynostoses. (2) 14% of websites state that if untreated, craniosynostosis mostly or always

leads to developmental delay. (3) 22% of websites only mention open surgical correction. (4) 46% of centers claim maximum age for endoscopic surgery to be under 4 months, 52% claim under 4 to 6 months, and 1 center claims under 8 months. (5) Multiple inaccurate marketing claims were made by American websites including transfusion-less surgery and concerning center reputation and stature. European craniofacial center websites were much more fundamental and rudimentary, often focusing on basics including registration and how to contact them. Less information was provided. A few were more detailed, but still not close to comprehensive. No significantly inaccurate claims were found.

Conclusion: The most search engine—optimized US craniofacial center websites often are not comprehensive and provide inaccurate and variable information to families. European websites are more fundamental and accurate but are more incomplete. The need for and influence of craniofacial center marketing in the US will be discussed.

#### 319. Access to Cleft Care Amongst Publicly Insured Patients with Cleft Lip and Palate

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Background/Purpose: Cleft lip and palate are the most common congenital craniofacial anomalies. A stepwise, multidisciplinary, and multiyear approach is necessary to provide optimal functional and aesthetic outcomes for this patient population. Repair of cleft lip and palate most commonly occurs within the first year of life, often without significant delays in care. One factor largely contributing to this early repair is near uniform insurance coverage of these sentinel procedures. For patients with cleft of the maxillary alveolus, timely bone grafting is necessary for optimal dental development. However, orthodontic treatment often precedes bone grafting, yet insurer coverage of orthodontic procedures is not uniform. The purpose of this study is to determine whether publicly insured patients are at risk for delayed alveolar cleft bone grafting and which factors contribute to this delay in treatment.

Methods/Description: A retrospective review of patients with complete cleft lip and palate treated at our regional cleft and craniofacial center was conducted. Patient demographics, insurance payer, median income, age of cleft lip, palate, and maxillary alveolar cleft repair were recorded. A total of 64 patients were included. Variations in age of alveolar cleft bone grafting and risk factors for delayed bone grafting were investigated between publicly and privately insured patients.

Results: Sixty-four patients were included in our analysis, 36% female and 64% male; 81% of patients had unilateral cleft lip and palate and 19% were bilateral; 83% of patients were privately insured while 17% were uninsured or publicly insured. Average age of bone grafting was 10.8 in privately insured patients and 11.4 in publicly insured patients. Delayed bone grafting and/or loss to follow-up prior to bone grafting was seen in 30% of privately insured patients compared to 63% of publicly insured patients.

Conclusions: Identification of patients at risk for delayed alveolar cleft bone grafting can allow for preventative interventions to optimize cleft care outcomes. Our preliminary results suggest uninsured and publicly insured patients with maxillary alveolar cleft are at risk for delayed bone grafting. This is likely multifactorial in nature, owing in part to lower socioeconomic status and difficulty accessing presurgical orthodontic care. Further investigation with larger study

populations is necessary to further identify specific risk factors for delayed bone grafting among publicly insured patients with cleft lip and palate.

### 320. Provision and Utilization of Team- and Community-Based Operative Care for Cleft Lip/Palate

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Background/Purpose: Cleft lip and/or palate (CL/P) is among the most common of birth defects, affecting one baby born in the United States every 2 hours. Children with CL/P may be functionally disabled (with regard to eating, speaking, breathing, and hearing) and may carry the visible stigma of being "different" unless provided appropriate care. Treatment is necessarily complex, delivered in stages across 2 decades of the child's life, and best coordinated by a multidisciplinary team. The American Cleft Palate-Craniofacial Association (ACPA) has an official process for approval of multidisciplinary teams in North America based on the team's structure and operational characteristics. Although there exist 175 ACPA-approved cleft teams in the United States, there are many geographic regions (and even entire states) where ACPA-approved team-based care is not available. To date, no one has evaluated how many children are receiving community-based care rather than ACPA-approved team-based care. In this study, we characterize the provision and utilization of cleft care in North Carolina.

Methods/Description: Cleft-related clinical encounters were identified in the AHRQ Healthcare Cost and Utilization Project's (HCUP) State Inpatient Database (SID) and State Ambulatory Surgery & Services Database (SASD) for North Carolina from 2012 to 2015 using ICD-9-CM diagnostic codes and CPT and ICD-9 procedural codes. Clinical encounters were then classified as "team-based" versus "community-based" care, according to county codes and hospital provider ID. Operative care patterns for each location were classified into 11 procedural categories that reflect the wide range of cleft operations. In a secondary aim, we reviewed concordance of team and community practices with an ACPA guideline related to coordination of care.

Results: Three ACPA-approved teams and 39 community providers performed 3010 cleft-related procedures across 2070 operative encounters. Teams performed 69.7% of total volume and the majority of "complex" craniofacial procedures. Community locations principally offered simpler procedures, such as myringotomy, but did account for the majority of cleft rhinoplasty, a complex procedure. Team care was associated with higher guideline concordance.

Conclusions: CL/P is a complex condition that requires longitudinal, coordinated, multidisciplinary treatment. The majority of CL/P encounters in North Carolina occurred in an ACPA-approved team setting. Interestingly, case mix and procedure mix were noted to be different in team versus community settings. This potentially reflects differences in clinical expertise, group structure, and interests of those providers. Guideline concordance related to coordination of care was noted to be higher in teams, suggestive of tighter integration between specialties. Future work will compare clinician-, family-, and patient-reported outcomes in ACPA-approved teams and other community providers.

#### 321. Costing Analysis of Pre-Surgical Infant Orthopedics (PSIO): A Critical Component of Establishing Value for Latham and NAM

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Background/Purpose: "Value-based health-care reform" is defined as aggregated outcomes divided by total cost. In cleft care, much attention is paid to the numerator of the value equation. For example, the debate around presurgical infant orthopedics (PSIO) has focused on safety, effectiveness, and relative advantages and disadvantages of different modalities of treatment; the denominator of the PSIO value equation has to date remained unexamined. In this project, we employ robust costing methods to quantify the cost of PSIO using the Georgiade-Latham appliance and nasoalveolar molding (NAM).

Methods/Description: This is a prospective study of patients with nonsyndromic cleft lip/palate (CLP) who underwent PSIO between 2017 and 2019. Two academic hospitals were studied—one using Latham, the other using NAM. The following costs were included: personnel, facility, equipment, and patient travel costs. Personnel costs were calculated using time-driven activity based costing methodology (TDABC). At each visit, researchers followed patients from moment to moment and recorded time spent with each personnel type (receptionist, nurse, dentist, surgeon), throughout their course of care, until they completed PSIO and underwent labial repair. The cost attributable to each personnel type was defined as his/her total time caring for patient multiplied by the national average hourly rate for his/her personnel type. Facility costs were estimated by multiplying time spent in the operating room (OR) by previously published average OR cost per minute. National averages for outpatient clinical space cost per minute were unavailable, so this was defined based on overhead costs specific to our hospital. Equipment costs were comprised of tallied third-party vendor charges. Travel expenses were used to estimate direct costs borne by the patient and family; indirect costs (eg, time off from work) were not considered.

Results: Twenty-three patients with CLP were treated with Latham, and 14 were treated with NAM. Total average cost for Latham was \$3968/patient, including \$1025 in personnel cost, \$634 for equipment, \$1305 in facility cost, and \$1004 borne by families for travel (average 6.5 visits). The total average cost for NAM of \$2356, comprised \$333 in personnel cost, \$151 for equipment, \$146 in facility cost, and \$1726 for travel (average 13 visits).

Conclusions: Latham and NAM are 2 methods of PSIO designed to accomplish the general principle of aligning alveolar segments prior to labial repair. NAM includes a secondary aim of elongating the columella and rounding the nasal ala. This study shows that both Latham and NAM can be provided at reasonably low cost: \$2300 to \$4000 per course of treatment. Latham and NAM distribute costs differently: whereas Latham heavily utilizes hospital resources, the majority of the cost of NAM is borne by families due to the number of visits required. Future investigation should focus on whether outcomes achieved by PSIO justify the \$2300 to \$4000 cost of these adjunctive procedures.

## 322. Interdisciplinary Management of Speech and Mandibular Defects in Infants with Pierre Robin Sequence

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Background/Purpose: Pierre Robin sequence (PRS) is a triad of congenital facial abnormalities that includes micrognathia, glossoptosis, and upper airway obstruction. Current evidence suggests that the incidence of PRS is 1 in 8500 live births with a mortality rate of between 10% and 30%. PRS potentially leads to life-threatening obstructive apnea and feeding difficulties during the neonatal period. Understanding potential airway lesions in concordance with potential feeding problems is important for determining appropriate treatment: observation, tongue—lip adhesion, mandibular distraction osteogenesis (MDO), or tracheostomy. MDO has shown superior outcomes in many studies, but there is currently not enough studies evaluating the effects of MDO on the treatment of feeding and airway obstructions in PRS patients.

Methods/Description: A retrospective review of patients presented with PRS at a tertiary referral pediatric hospital between January 2015 and December 2018 was performed. The patients were stratified into 2 groups based on the genetic information either isolated (iPRS) or syndromic (sPRS) PRS. Patients with associated clinical features or genetic anomalies were classified as sPRS. The primary outcome measure was improvement in airway and feeding outcomes as well as the presence of complication following treatment. The airway outcome measure included avoidance of tracheostomy (primary airway), decannulation, relief of OSA, improvement in oxygen saturation, and the repeat of procedure. The primary feeding measure included achievement of full oral feeds at latest follow-up (primary feeding), growth/weight gain, and improvement in gastroesophageal reflux. Complications included external scarring, infection, hardware exposure, device dislodgement, facial nerve problems, and tooth bud damage.

Results: PRS was isolated in 52% of patients and syndromic in 48%. Associated syndromes in sPRS patients were noted. The timing of the operative intervention, the avoidance of tracheostomy, the size of the mandible pre and post distraction, and the achievement of full oral feeds were reported. Approximately 83% of subjects treated with MDO achieved a positive feeding outcome and 91% achieved a positive airway outcome. We will also discuss in detail the type of MDO performed, the mandible measurement variability in PRS patients, and the use of latency and activation phase during distraction. Multivariate analyses were then conducted through logistic regression models involving respiratory disorders for one model, feeding disorders in a second model as binary dependent variables using IBM SPSS Statistics. Over the study period, we identified 26 infants with PRS who were treated with MDO and managed by a single surgeon.

Conclusions: The avoidance of an additional surgical intervention and decreasing time needed to care for a neonate are important potential benefits for patient quality of life and caretaker burden in PRS patients. Here, we present the short-term outcomes of the management of speech and mandibular defects in PRS.

### 323. Parent Reported Feeding Experiences in Infants with Craniofacial Conditions and the Relation to Health Outcomes

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Background/Purpose: Infants with craniofacial conditions (CFC) are at increased risk of feeding difficulties due to impaired sucking, swallowing, and breathing mechanics. Interventions are available to improve feeding quality and safety. Validated tools to assess feeding behaviors and experiences have potential for promoting successful feeding but are not standard and assessments vary across disciplines and institutions. Identification of parent-reported feeding experiences and behaviors that correlate with health and quality of life have the potential for guiding feeding interventions for infants with CFC. We hypothesize a simple, parent-reported characterization of feeding measured by the Baby Eating Behavior Questionnaire (BEBQ) is associated with relevant health outcomes.

Methods/Description: In this prospective cross-sectional study, we recruited infants with CFC (n = 16) and healthy controls (n = 12). Cases had varying risks for feeding difficulties: cleft palate only, craniofacial microsomia, Robin sequence, and Beckwith-Wiedemann syndrome. In an overnight visit, we collected demographic and anthropometric data, sleep data (parent-reported, polysomnogram), health and developmental characteristics, and parent-reported measures of feeding, parent confidence, parent sleep, and fatigue. The BEBQ is a validated 18-item questionnaire assessing multiple domains with 5 subscales: food responsiveness, enjoyment of food, satiety responsiveness, slowness in eating, and general appetite. Clinical characteristics and BEBQ responses were summarized for the cohort using descriptive statistics. Differences between cases and controls were assessed using Fisher exact test and Wilcoxon rank-sum test due to non-normal distribution. Regression analyses were used to estimate the association between BEBQ subscale scores and infant and parent factors.

Results: The cohort consisted of 28 infant–caregiver dyads. Median age was 4.9 months (IQR: 3.8-5.9), 46% were female, 70% of mothers had a college degree, oral feeding was the primary nutrition source for 90%, gastroesophageal reflux (GER) was present in 46%, developmental delay (DD) was diagnosed in 14%, and a range of obstructive sleep apnea (OSA) was detected by sleep study (median obstructive AHI 6 per hour). Parents of infants in the case group reported slower feeding than parents of controls (P=.03). Infants with less food enjoyment were more likely to have DD (P=.02). Infants with lower weight/length% were less food responsive (P=.04). Slowness in eating was associated with GER (P=.02), DD (P=.03), worse OSA (P=.04), and more fragmented sleep (P=.01). Parent-reported fatigue and confidence scores were not significantly associated with BEBQ scores.

Conclusions: These data reveal a simple set of BEBQ-derived questions associated with growth, OSA, and GER that may have potential craniofacial research and clinical applications. Standard assessment of parent-reported feeding behaviors has the potential to identify targets to improve holistic care and health outcomes for infants with CFC.

### 324. Experiences in Prenatal Cleft Lip and Palate (CL/P) Consultations: Our Center by the Numbers

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Background/Purpose: Previous studies have shown that prenatal diagnosis of a congenital anomaly is an independent predictor of parental psychological distress. Surgical prenatal counseling can decrease parental anxiety by helping families understand the surgical needs and potential outcomes of their infant. The purpose of this study is to present our experiences with prenatal CL/P consultations and clarify rates of prenatally diagnosed clefts and cleft-related terminations.

Methods/Description: A retrospective review was performed of mothers seen at our center for prenatal CL/P consultation from January 2015 through August 2019. Primary outcomes were number of consultations and CL/P repairs, and number of terminations and deaths. Secondary outcomes included demographic information.

Results: One hundred twenty-three patients had surgical consults for prenatally diagnosed CL/P during our study period (2015 n = 13, 2016n = 29, 2017 n = 31, 2018 n = 26, 2019 n = 24). The average maternal age at time of visit was 31.4 years (range: 16.1-42.9 years). An additional 23 patients were in contact prenatally with our nurse navigator but did not have a surgical consultation. Of those patients, who did not come in for consultation, 2 opted for termination (9%) and 12 followed up postnatally (52%). A subset of these prenatal consultations, those seen in 2015 and 2016, were further analyzed (n = 42). This subpopulation was selected because it represents the third of the population with the longest followup (mean 2.38 years from prenatal consultation, range: 0.94-3.81 years). The average gestational age at plastic surgery consultation was 30 weeks 2 days (range: 22 weeks 4 days-37 weeks 6 days); 43% of these patients (n = 18) were referred from our institution's fetal imaging center; 57% returned to our center for surgical repair postnatally (n = 24). All but one of the patients who returned for surgery had congruent pre- and postnatal diagnoses (n = 23). One patient, in addition to bilateral CL/ P, had atypical orofacial clefts involving the cheek that were not noted prenatally; 24% of patients (n = 10) were additionally seen prenatally by our institution's fetal heart center; 81% of patients had isolated CL/P (n = 34) while the remaining 8 (19%) patients had additional anomalies (CHARGE syndrome [n = 1], Hay-Wells syndrome [n = 1], complex Tessier clefts [n = 2], and cardiac and neurological anomalies [n = 3]). Two fetuses with multiple anomalies were terminated and 2 infants with CL/P and additional anomalies passed away before 3 years of age. In the period from 2016 to 2019, 1 additional fetus with multiple anomalies was terminated and 1 infant passed away soon after birth.

Conclusions: Prenatal consultation for CL/P is an important method of information transfer for expectant mothers. We have not identified any fetuses who were terminated for isolated CL/P after prenatal surgical consultation, perhaps suggesting that CL/P may be better accepted by parents than previously thought. Further analysis is needed to identify what aspects of prenatal consultation are most helpful to families in the prenatal period.

### 325. Socioeconomic and Racial Gaps in the Utilization of Secondary Cleft Surgeries

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*Background/Purpose*: Secondary surgeries are important for the long-term cosmetic and functional outcomes following cleft palate repair. The current study sought to analyze the impact of social and economic factors on the utilization of these surgeries in the United States.

Methods/Description: Primary cleft palate, revision cleft palate, and secondary rhinoplasty surgeries were identified in the Kids' Inpatient Database (KID) data released every 3 years from 2000 to 2009. Data were combed for patient demographics and socioeconomic indicators. Bivariable and multivariable analyses were performed to compare patient subgroups.

Results: There were significant differences in the racial and socioeconomic profiles of patients with primary cleft and those receiving secondary procedural care. Secondary rhinoplasty and revision repair patients were more likely to be white (P < .001), of higher income (P = .042) and pay using private insurance (P < .001) than patients receiving primary cleft palate repair. Nonwhite patients also received fewer concurrent procedures at the time of their revision repair (P < .003) and experienced greater total numbers of complications (P < .0211). Revision procedures were more likely to be considered elective (P = .045) and were more likely to be performed at hospitals of smaller bed size (P = .012) than primary repairs.

Conclusions: These findings suggest potential gaps in access to secondary rhinoplasty and revision cleft repair among nonwhite patients and patients of lower socioeconomic status. Among other interventions, improved long-term follow-up, more accommodative reimbursement policies, and increased diversity among treatment teams may help mitigate disparities in care.

#### 326. What is the Burden of Care of Nasoalveolar Molding?

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Background/Purpose: Nasoalveolar molding (NAM) reduces the severity of the cleft lip/nasal deformity and aligns alveolar segments prior to surgical repair. It requires a dental impression followed by fabrication of a molding plate and nasal stent that is adjusted weekly or biweekly. Numerous studies have demonstrated the benefit of NAM; however, due to the commitment required of families, barriers to care have been referred to as the "burden of care." Although frequently referenced, this concept, defined as the physical, psychosocial or financial problems that can be experienced by caregivers, has not been entirely studied in the context of NAM. This systematic review aims to objectively evaluate the burden of care of NAM using this accepted definition.

Methods/Description: Following Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines, 5 databases were searched from inception through April 22, 2019, for keywords and subject headings pertaining to cleft lip and/or palate and nasoalveolar molding. Two independent reviewers selected clinical studies on NAM with reference to physical (access to care, number of visits, distance traveled), psychosocial (caregiver perceptions, family interactions, breast milk feeding) and financial (direct and indirect costs) burden. Data were collected according to these variables.

Results: The search identified 997 articles. After title, abstract, and full-text screening, 107 articles remained for qualitative synthesis. Burden of care was discussed but not measured in 44% of articles and only 25% assessed burden of care through a primary outcome. Of these, 19 articles reported on physical, 7 articles on psychosocial, and 12 articles on financial burden. Greater than 30% of cleft teams in North America perform NAM. The mean reported travel distance was between 65 and 70 miles and average treatment duration up to 136 days and 15 clinic visits. Physical burden variables alone have been used as proxies for burden of care. Psychosocial burden was mitigated by opportunities for counseling, breast milk feeding, and development of coping strategies. Financial considerations were context-dependent and could be partially or completely covered by insurance. Several studies reported reasons caregivers decide to pursue NAM and reasons for completion or noncompletion of treatment. Alongside studies that publish noncompletion rates, others report no issues with treatment compliance.

Conclusions: NAM has been indiscriminately associated with burden of care in the literature. Although NAM may not be the ideal treatment option for all patients and families, the physical considerations are limited and short-lived when accounting for the observed psychosocial advantages. Financial burden appears to be offset by the benefit of limiting future surgery and improved quality of life but further research is required. Teams should more directly assess the impact of this early intervention on the well-being of caregivers and advance strategies that improve access to care.

### 327. Validation of Optical Coherence Tomography with Optimized Thresholds for Detecting Elevated Intracranial Pressure

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Background/Purpose: Craniosynostosis predisposes children to increased intracranial pressure (ICP). Subclinical elevation of ICP can have detrimental consequences, including neurocognitive impairment. Noninvasive measurement modalities of ICP remain inadequate, but a spectral domain optical coherence tomography (OCT) protocol showed promise to detect subclinical elevated ICP. The purpose of this study is to assess the validity of noninvasive OCT assessment of ICP with a novel cohort of patients with craniosynostosis.

Methods/Description: Pediatric patients undergoing cranial vault procedures were prospectively enrolled through an IRB-approved protocol. Spectral domain OCT was performed preoperatively after induction of anesthesia and ICP measured with a subdural catheter intraoperatively. The primary outcome was association of retinal OCT parameters with directly measured ICP. Comparison of newly acquired data with historical controls was used to provide validation.

Results: One hundred thirty-four patients met inclusion criteria (median age 12.5 months, range: 1.0-205). Combination of retinal nerve fiber layer maximum thickness (RNFL), maximal anterior projection of retina (MAPR), and retina maximum thickness (RMT) cutoffs established in previous studies yielded a sensitivity of 78% (95% CI: 45%-94%) and specificity of 53% (95% CI: 30%-75%). These are comparable to previously published values of 89% (95% CI: 69%-97%) sensitivity and 62% (95% CI: 41%-79%) specificity. Frequency of elevated ICP in craniosynostosis patients was more common in cases of sagittal or lambdoid synostosis than metopic or unicoronal synostosis (P = .047), a finding that previously trended toward

significance. Multiple cranial suture involvement was associated with higher rates of elevated ICP (P = .002).

Conclusions: Spectral domain OCT remains a promising noninvasive modality for assessment of subclinical elevated ICP among patients with craniosynostosis. These findings suggest the validity of previously defined OCT parameters and suggest the possibility of wider deployment of this detection modality.

#### 328. Challenging the Norm: Is Routine Use of Head CT in Evaluation of Craniosynostosis Necessary?

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Background/Purpose: Traditional teaching is that the gold standard for diagnosing craniosynostosis is a head CT. In most craniofacial centers, the preoperative workup for suspected craniosynostosis involves a head CT scan either by the referring pediatrician or the craniofacial team. The purpose of this study was to investigate the benefits of preoperative CT scans in the diagnosis and treatment of nonsyndromic single-suture craniosynostosis.

Methods/Description: An IRB-approved retrospective review was performed for all children who underwent surgery for nonsyndromic single-suture craniosynostosis at Southern California Kaiser Permanente from 2007 to 2018. Our preoperative assessment of single-suture nonsyndromic craniosynostosis is based on clinical examination, with imaging studies reserved only for cases where a diagnosis cannot be made clinically (group 1). However, some patients already have imaging studies performed prior to presentation (group 2). The objective data we collected included: age at presentation to craniofacial team, age at surgery, preoperative clinical diagnosis, any preoperative imaging and if before or after initial craniofacial visit, coexisting radiographic findings, and the correlation of the intraoperative diagnosis.

Results: A total of 138 patients were included in this study with a median age of 3.1 months at initial craniofacial visit and 7 months at time of surgery. Of these, 27 (19.6%) patients had imaging obtained before the first craniofacial visit (group 2): head CT in 6 patients and X-ray in 21 patients. The remaining 111 (80.4%) patients had no imaging studies obtained before their first craniofacial visit—group 1. Based on initial clinical assessment in group 1, single-suture synostosis was diagnosed in 102 (92%) patients, whereas in 9 (8%), the diagnosis was not clear. Of the patients with unclear diagnosis, one was successfully diagnosed clinically at a planned follow-up examination, and the remaining 8 (7%) patients were diagnosed using imaging studies-3 CT scans and 5 X-rays. In all patients, the preoperative diagnosis was confirmed to be accurate during intraoperative assessment. No radiographic abnormalities nor findings that changed operative plans were found in either group. There were no differences in the age of initial craniofacial visit (ANOVA; F = 0.604, P = .4) and surgery (ANOVA, F = 0.391, P = .5) between the groups.

Conclusions: The results of this study suggest that CT scanning is not necessary for diagnosis by experienced craniofacial surgeons in 93% of single-suture, nonsyndromic craniosynostosis. Imaging obtained before referral to our craniofacial clinic provided no additional benefits in identifying coexisting conditions, age at referral to craniofacial clinic, or age at surgery. Thus, we challenge the utility of routine CT scan in the evaluation of craniosynostosis and recommend use only by an experienced craniofacial team and only when clinical diagnosis is uncertain.

#### 329. The Sharing Circle – An Enrichment Activity of the Camp Experience

Jamie Idelberg (1), Jennifer Maybee (1)

(1) Children's Hospital Colorado, Aurora, CO

Background/Purpose: A camp experience is inspiring and rewarding for any child, but it can truly change the outlook for a child with a cleft or craniofacial condition by bringing them into contact with other children who have similar conditions and life experiences. For the past 20 years, our hospital's Cleft Camp Program has brought kids together for Winter and Summer Cleft Camp. Many campers report on post camp evaluations that the "Sharing Circle" is their favorite activity; they look forward to participating in it. The Sharing Circle is a safe place for campers to talk about what life is like living with a cleft condition—both the challenges and positive aspects. Campers can participate at their comfort level by writing an anonymous question ahead of time, sitting and listening to the conversation, or joining the conversation directly. The discussion is often facilitated by our older, teenage campers who serve as role models. For camp staff attending the Sharing Circle, the insights shared by the campers provide a rare and valuable insider view about living with a cleft condition.

Methods/Description: This visually stimulating presentation will highlight the benefits of a Sharing Circle as part of the camp experience. The Sharing Circle can also be included in the activities of a social/educational program for adolescents and teenagers. The purpose of this presentation are to (1) demonstrate the impact of bringing kids together for a cleft camp experience; (2) teach participants the power of adolescents and teenagers talking openly about their questions, fears, dreams, and what the future can be for them; and (3) explore what health-care professionals can learn from a Sharing Circle as we work with children who are navigating their life (or personal) journeys.

#### 330. The Evolution of Cleft Lip (& Nose!) Repair

Raj Vyas (1)

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Background/Purpose: The goal of this presentation is to review the evolution of principles and techniques in the repair of cleft lip and nasal deformity. In addition to historical perspective, this presentation seeks to highlight the nuances between the most common types of nasolabial repair in practice today—including the various presurgical dentofacial appliances. For each technique, there will be focus on relevant anthropometry and discussion of technique-specific advantages and trade-offs. Long-term, fourth-dimensional outcomes are compared and reviewed. Finally, this presentation will examine controversial aspects of presurgical orthodontics and nasolabial repair (extent of nasal repair, role of primary premaxillary setback, growth disturbance, perioperative care, etc).

*Methods/Description*: This presentation will meet the study session objectives with use of multiple illustrations, figures, photographs, and videos. Both sides of controversial subjects will be presented using the best available evidence for each.

### 331. Future Directions in Transition of Care for Young Adults with Cleft Lip and Palate: Clinical Education

Brenda Louw (1), Linda Vallino (2)

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Background/Purpose: A recent development in Cleft Palate Team Care is the transition of care for young adults with cleft lip and palate (CLP) and their families moving from child- to adult-centered care. CLP Teams are increasingly becoming aware of the needs of these young adults with CLP as they are discharged from the pediatric team. To date, few teams in the USA have acted on this need and provide a transition of care plan to their patients aging out of their pediatric team care (Bisceglia et al., 2017). Although the ACPA Parameters of Care (ACPA, 2018) allude to services for adults with CLP, transition of care has not yet been included in this document. In opening this door, Vallino and Louw (2017) proposed an evidence-based conceptualized model for the transition of care for young adults with CLP. Yet, there is more to do. In order for CLP team care to advance to meet the needs of patients across the age spectrum, there is an urgent need for future professionals to be trained in transition of care for young adults with CLP. Interprofessional education (IPE) is the ideal approach to follow in jointly preparing future professionals in transition of care since it is a team endeavor. According to the WHO (2010), IPE ensures that the future health workforce is collaborative-practice ready. It is timely for teams to embrace the concept of transition of care and to train future professionals to provide services that recognize the importance of personal and environmental factors in facilitating holistic transition planning and service delivery and are developmentally appropriate to young adults with CLP (Farre & McDonagh, 2017). Training future professionals can be approached through problem-based learning (PBL). Anchored to PBL is experiential learning (EL). Central to both is to encourage critical and independent thinking in the student. Teams who provide transition of a care to their patients and commit to training future professionals will contribute to sharing the national health burden of these young adults; strengthen the skills of future professionals, and contribute to an optimal health system with improved health outcomes. The aim of this presentation is to describe a model for preparing future professionals in the transition of team care for young adults with cleft palate within an ICF framework following student-centered approach.

Methods/Description: An overview of adolescents/ young adults with CLP and a service delivery model for their transition of care will be described (Vallino & Louw, 2017). The key components of the proposed IPE training model of transition of care to be discussed are (1) the concept of IPE; (2) the International Classification of Functioning, Disability and Health; (3) strategies for PBL and EL; and (4) suggestions to guide future professionals through the 4 integrated processes of EL. Practical strategies for teams to involve future professionals in developing and providing transition of care to young adults with CLP will be presented.

### 332. The Value of Nasometry in Supporting the Perceptual Evaluation of Velopharyngeal Function

Cara Werner (1)

(1) Cincinnati Children's Hospital Medical Center, OH

Background/Purpose: Although a perceptual evaluation of speech and resonance is essential in the assessment of velopharyngeal function, these measures are inherently subjective. By adding nasometry to the diagnostic protocol, the examiner can obtain objective data to support recommendations and outcomes measuring. Nasometery is advantageous to other procedures in that it is quick, noninvasive, and easy to administer, even with young patients. Nasometry is an indirect, instrumental procedure for evaluating velopharyngeal function. It measures the acoustic correlates of resonance, audible nasal emission, and velopharyngeal function through a computer-based instrument. Nasometry can be used to assist with differential diagnosis related to

velopharyngeal function and resonance disorders. Additionally, it can be used to compare an individual's nasalance score to normative data and their preoperative status and to compare speech and surgical outcomes through objective measures. The purpose of this presentation is to describe the diagnostic value of nasometry in supporting perceptual evaluation results. The presenter will discuss nasometric methods and how to use nasometry for the purpose of differential diagnosis related to phoneme-specific nasal emission or a symptomatic fistula versus VPI. Identifying evidence of upper airway obstruction through nasometry will be described. The presenter will also discuss the value of nasometry in determining speech outcomes following intervention and comparing outcomes among surgeons and surgical procedures.

Methods/Description: In this session, the presenter will provide a basic overview of nasometry as an instrumental procedure for evaluation of velopharyngeal function. This session will include techniques for achieving a successful examination, even in very young children, and how to interpret results. The presenter will explain how nasometry can provide objective measures to support a differential diagnosis of cause. Case studies will incorporated and discussed with participants. A practical laboratory for hands on practice will be provided. Finally, the presenter will describe how nasometry can be used to compare speech and surgical outcomes using objective data.

## 333. Endoscopic vs. Open Surgery for Craniosynostosis—Indications and Outcomes from the Kids' Inpatient Database

Danielle Rochlin (1), Clifford Sheckter (1), H. Peter Lorenz (1), Rohit Khosla (1)

(1) Stanford University, Palo Alto, CA

Background/Purpose: The optimal surgical treatment for craniosynostosis remains controversial. Although open techniques have been the historical norm, recent investigations have demonstrated the benefits of endoscopic surgery. Little is known regarding regional variation between these 2 techniques and whether disparities exist in access to endoscopic surgery. The purpose of this study is to evaluate national differences in inpatient outcomes and predictors of treatment type for endoscopic versus open surgery for craniosynostosis.

Methods/Description: A retrospective analysis of patients up to 3 years of age in the 2016 Kids' Inpatient Database who underwent endoscopic or open craniectomy for craniosynostosis was performed. Patients were identified using International Classification of Diseases, Tenth Revision (ICD-10) codes for both diagnosis and procedure. Logistic regression modeled endoscopic versus open treatment based on patient-level (gender, race, income, comorbidities, payer) and facility-level (bed size, United States region, teaching status) variables. Outcomes assessed by multivariable regression using a parsimonious approach included complications, blood transfusion, length of stay, and cost of hospitalization. Sampling weights were applied per Healthcare Cost and Utilization Project recommendations.

Results: The weighted sample included 514 patients, of which 81.5% were younger than 1 year; 83.0% of patients underwent open repair and 17.0% underwent endoscopic repair; 13.8% of all patients had associated diagnoses suggestive of syndromic craniosynostosis. Patients were more likely to be treated open if they were older (odds ratio [OR]: 3.2, P = .007) or syndromic (OR: 9.7, P = .026). There were no significant racial, socioeconomic, or geographic (Northeast, Midwest, South, West) disparities in predictors of treatment type. Patients who underwent open repair were more likely to receive red blood cell transfusions or blood component transfusions (23.4% vs

9.5%, P=.030) and have longer lengths of stay (mean: 3.0 vs 1.7 days, P<.001) and more costly hospitalizations (mean: \$25 674.8 vs \$14 734.0, P=.019). Complications did not significantly vary between procedure type, though patients with syndromes were more likely to have both systemic (OR: 4.2, P=.003) and local (OR: 3.7, P=.016) complications.

Conclusions: United States national data demonstrate that age and presence of syndromic comorbidities predict whether a patient with craniosynostosis received endoscopic versus open repair. Racial, socioeconomic, and geographic factors were not significantly associated with treatment type. Compared to open surgery, endoscopic surgery showed benefits including a lower risk of transfusion, shorter hospital stay, and lower hospital costs, without a significant change in the rate of postoperative complications.

### 334. The Impact of Suprazygomatic Maxillary Nerve Blocks on Postoperative Narcotic Use in Patients Undergoing Orthognathic Surgery

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(1) Emory University Hospital, Atlanta, GA, (2) Long Island Plastic Surgical Group, Garden City, NY, (3) Children's Healthcare of Atlanta, GA

Background/Purpose: The mainstay of analgesia in orthognathic interventions for maxillary hypoplasia is perioperative opioids; however, the side effect profile is broad with the potential for well-described deleterious effects. The suprazygomatic maxillary nerve block has been previously shown to be effective in decreasing pain associated with palatal surgery. To date, there have been no studies detailing the use of maxillary nerve blocks as an adjunctive pain control measure during correction of maxillary hypoplasia. Consequently, we sought to evaluate the efficacy of bilateral suprazygomatic maxillary nerve blockade in decreasing perioperative narcotic consumption in patients undergoing orthognathic surgery.

Methods/Description: Between January and August 2019, patients undergoing suprazygomatic maxillary nerve blockade for orthognathic correction of maxillary hypoplasia were prospectively collected and compared to controls. Patient demographics, narcotic use (represented as morphine milligram equivalents per kg; MME/kg), reported pain scales, and complication rates were compared.

Results: Over 8 months, 27 patients met inclusion criteria (n = 15 block; n = 12 control). Mean ages were  $16.5 \pm 2.5$  years and  $15 \pm 2.2$  years, respectively. The block group demonstrated a significant reduction in postoperative narcotic requirements for the first 24 hours when compared with the control group  $(0.02 \pm 0.03 \text{ MME/kg vs. } 0.10 \pm 0.11 \text{ MME/kg respectively; } P < .01)$ . Beyond 24 hours, a similar trend toward significance was noted. Corroboratively, self-reported pain scale assessments in the first 24 hours were significantly decreased in the block compared to control groups  $(0.8 \pm 0.85 \text{ vs. } 2.5 \pm 2.3, \text{ respectively; } P < .01)$ . Neither group-evidenced perioperative complication nor return to service within 30 days.

Conclusions: Administration of a suprazygomatic maxillary nerve block for patients undergoing orthognathic surgery for cleft-related maxillary deficiency demonstrated a significant reduction in both postoperative narcotic requirements and reported pain scales in the first 24 hours with a trend toward significance thereafter supporting its utility as a safe and effective analgesic adjunct.

## 335. Impact of Fronto-Orbital and Lateral Orbital Wall Advancement on Orbital Volume and Shape in Non-Syndromic Metopic Craniosynostosis

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Background/Purpose: Metopic craniosynostosis has been estimated to occur in approximately 25% of nonsyndromic craniosynostosis cases. Periorbital dysmorphology associated with metopic synostosis includes hypotelorism, lateral orbital wall recession, and trapezoidal shape to the orbital perimeter. The objectives of this study were to quantify orbital volumetric and shape changes perioperatively after performing fronto-orbital advancement (FOA) in combination with lateral orbital wall advancement and assess differences between the orbital morphology of children with nonsyndromic metopic craniosynostosis compared to healthy age-matched controls.

Methods/Description: This retrospective case series included nonsyndromic metopic craniosynostosis patients who underwent FOA with lateral orbital wall advancement over a 10-year period by a single surgeon. Pre- and postoperative orbital volume and morphology (ie, interzygomaticofrontal suture distance, length of lateral wall, angle between the lateral wall, and Frankfort horizontal plane) were measured via CT scans using a 3-dimensional (3D) segmentation software by a single rater. Next, preoperative orbital measures were compared to a cohort of healthy controls (ages 8-12 months). Descriptive statistics were performed.

Results: A total of 39 children were included, who had surgical intervention at an average age of  $1.02~(\pm 0.49)$  years. One (2.6%) patient had preoperative increased intracranial pressure. Statistically significant increase in bilateral orbital volume was obtained post-operatively (right:  $14~111~\pm~2206$  to  $16~857~\pm~2126~\text{mm}^3$ , P < .001 and left:  $13~927~\pm~2167$  to  $16~603~\pm~2184~\text{mm}^3$ , P < .001). Similarly, statistically significant increases were obtained in pre- to post-operative interzygomaticofrontal suture distances (P < .001), lateral wall length (P < .001), and angle between the lateral wall to the Frankfort horizontal plane (P < .001). When comparing preoperative metopic craniosynostosis (n = 34) orbital measurements with a cohort of healthy controls (n = 31), orbital volumes were not statistically different, but the lateral wall length (P < .001) and interzygomaticofrontal suture distances (P < .001) were found to be significantly smaller in the craniosynostosis cohort.

Conclusions: Beyond cranial vault reshaping and expansion with FOA in nonsyndromic metopic synostosis, attention focused on the surgical correction of the orbital dysmorphology was found to show improvements in morphometric outcomes as assessed by orbital volume and perimeter shape and normalization compared with age-matched controls. Further volumetric and morphological analyses are needed to assess long-term outcomes.

### 336. Endoscopic Internal Orbital Frontal Advancement: Experience with 100 Patients

David Jimenez (1), David Yates (1), Edward Santee (1) (1) El Paso Children's Hospital, TX

Background/Purpose: Standard open frontal-orbital advancement (FOA) includes bifrontal craniotomies and orbital bandeau advancement of several centimeters to correct the plagiocephaly associated

with coronal craniosynostosis. It is associated with significant dissection, facial swelling, blood transfusion, and postoperative pain. Long-term outcomes are inconsistent. Presented is a series of patients who achieved correction of this deformity without the need for a FOA procedure using a minimally invasive technique.

Methods/Description: One hundred infants underwent an endoscopic-assisted craniectomy to release the frontal bone from the parietal bone on the affected side of the craniosynostosis. The craniectomy extended from the anterior fontanelle to the squamosal suture. There were 72 females and 28 males. The mean age was 4.1 months and ranged between 1.5 and 7.2 months. All patients were place in custom molding cranial orthoses postoperatively. All patients had standard AP and lateral radiographs taken on the day of surgery, 8 weeks, 16 weeks, and 24 weeks post-op. Measurement of the craniectomy (in millimeters) was done by the radiologist reading the lateral films.

Results: In all cases, release of the frontal bone allowed the brain to move the orbitofrontal complex forward and downward. The mean osteotomy width at surgery was 5 mm, at 8 weeks was 15 mm (range: 13-17 mm); at 16 weeks was 26 mm (range: 22-28 mm); and at 24 weeks was 31 mm (range: 30-35 mm). In all patients, the plagiocephaly improved or corrected. There was marked and significant correction of the vertical dystopia. There were 2 skin infections treated with antibiotics only and one dural tear. There were no mortalities.

Conclusion: Endoscopic internal orbital frontal advancement leads to successful correction of frontal plagiocephaly and vertical dystopia with improved aesthetic outcomes. It is associated with significantly less trauma, swelling, and pain than traditional open techniques.

#### 337. Withdrawn

#### 338. Self-Perceptions of Social Adjustment and Stigma in Children with Cleft Lip and/or Palate

Claudia Crilly Bellucci (1), Canice Crerand (2), Farah Sheikh (3), Suzanne Woodard (4), Meredith Albert (5), Amy Conrad (6), Celia Heppner (7), Kathy Kapp-Simon (5)

(1) Shriners Hospitals for Children—Chicago and University of Illinois at Chicago, IL, (2) Nationwide Children's Hospital, Columbus, OH, (3) The Hospital for Sick Children, Toronto, Ontario, (4) Lancaster Cleft Palate Clinic, Lancaster, PA, (5) Shriners Hospitals for Children, Chicago, IL, (6) University of Iowa Children's Hospital, IA, (7) Children's Medical Center, Dallas, TX

Background/Purpose: Children born with cleft lip and/or palate (CL/P) are vulnerable to social difficulties including bullying and social inhibition, likely due to speech and/or facial appearance differences. Negative social experiences can impact overall psychosocial functioning and may increase risks for anxiety, depression, and poor quality of life. However, few studies have examined children's self-perceptions of social adjustment and stigma, while accounting for differences by sex or cleft type. To address these gaps, this study examined social adjustment and stigma in a large cohort of children with CL/P recruited from multiple centers in North America.

Methods/Description: As part of a larger study examining cleft care outcomes, 363 children ages 8 to 10 years old (50% male) with a confirmed diagnosis of CL/P (18% CP, 67% CLP, 15% CL) were consecutively recruited across 6 sites. Children completed the Pediatric Quality of Life Inventory (PedsQL) Social, Emotional, and School Subscales, PROMIS Stigma Scale, and the CLEFT-Q Social, Psychological and School Subscales in conjunction with a clinic visit. Analyses examining differences in social adjustment and stigma by diagnosis and sex were completed using multivariate analysis of

variance (MANOVA). Percentages of children having at-risk scores on the PedsQL and PROMIS stigma scale were determined using published cutoff points in the general population; comparisons to published norms of the CLEFT-Q were calculated.

Results: MANOVA results indicated that children with CP reported less stigma than children with CL and CLP with the difference reaching significance for CLP (P=.006). On the PedsQL Emotional Subscale, children with CL reported better functioning than children with CP and CLP with the difference reaching significance for children with CLP (P=.006). No significant differences by diagnosis or sex were found on the other PedsQL or CLEFT-Q subscales. On the PedsQL, 40% of the sample scored below the cutoff on the Emotional Subscale; 23% on the School Subscale, and 9% on the Social Subscale. On the PROMIS Stigma Scale, 9% fell in the at-risk range. For the CLEFT-Q subscales, 9% of the study children fell at least 1 SD below the mean on the Psychological Scale, 15% were below on the School Scale, and 14% on the Social Scale.

Conclusions: This study observed few significant differences among children based on cleft diagnostic group or sex, although children with CLP appeared to be most vulnerable to stigmatization and emotional issues. Across the sample, risks for emotional, school, and social functioning were noted, with some children reporting lower scores on the CLEFT-Q compared to cleft population norms. These findings underscore the need for routine screening in these domains across diagnoses and specifically among youth with CLP and illustrate the benefits of using both generic and cleft-specific instruments for assessments.

### 339. Child and Parent-Proxy Reported Quality of Life in Children with Cleft Lip and/or Palate

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Background/Purpose: Interrater agreement for health-related quality of life (HRQoL) has been found to be variable, particularly for child and parent-proxy reports. This construct has been explored in pediatric populations with various health conditions (eg, cancer, postoperative pain), but examination of interrater variance has been limited for youth with cleft lip and/or palate (CL/P).

Methods/Description: Participants (363 children, ages 8-10, and their parents) from 6 centers, recruited between August 2016 and May 2019, completed a measure of HRQoL, the Pediatric Quality of Life Inventory Generic Core Scales (PedsQL) as part of a larger study. Analyses included 315 children (147 male; 46 iCL, 55 iCP, and 214 iCLP), along with parent-proxy-reported scores from 258 female caregivers and 83 male caregivers (parent-proxy report scores were collected from 2 caregivers for 30 patients). Separate analyses were conducted for male and female parent-proxy report, such that each child report score was used only once in each set of analyses.

Results: Two-way mixed effect intraclass correlations (ICC) revealed poor to fair agreement (r < .04) between child and parent-proxy raters for most PedsQL subscales and composite scores. Of note, moderate agreement was detected for child and female caregiver ratings of

school functioning (r=0.43) and for both male and female caregiver proxy scores for the overall psychosocial composite (r=0.42) and 0.41, respectively). Overall means were also compared for child report and parent-proxy report by male and female caregivers; there was no difference between groups across all PedsQL subscales and composite scores. Child and parent-proxy reported scores were also compared to published normative data for the PedsQL Generic Core Scales for both chronically ill and healthy samples. Child report and parent-proxy report scores were significantly higher than scores from the chronically ill standardization sample across all domains (P < .01) for all); however, when compared with healthy controls, child and parent-proxy scores were significantly lower (P < .01), with the exception of the Physical Functioning Scale, which was not significantly different from healthy controls.

Conclusions: Although parent-proxy report may provide valuable information about children's observed functioning, the lack of agreement between parents and children with CL/P about the child's own level of functioning supports the need for both parent-proxy and child self-reported outcomes to be included when evaluating HRQoL for youth with CL/P. In addition to providing important information about children's own perceived HRQoL, differences between self-report and parent-proxy report may be clinically meaningful. Children with CL/P and their parents may endorse better HRQoL when compared to youth with other conditions defined as chronic illness; however, results also suggest that psychosocial functioning is negatively impacted for this population when compared with healthy youth.

#### 340. Implementation of a Suicide Screening Program for Cleft Patients: Early Results

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(1) Children's Mercy Hospital, Kansas City, MO

Background/Purpose: Suicide is the second leading cause of death among teenagers. Because many teenagers who die by suicide have seen a health-care provider in the months prior to their death, the Joint Commission (JC) has issued a recommendation for suicide screening in adolescent patients within a hospital environment. Patients diagnosed with a clefting condition are thought to have a higher risk for suicide. There is currently no data available investigating routine use of suicide screening in a pediatric cleft team. This study describes the implementation of a suicide screening program within a large-volume Cleft Clinic and the first 7 months data.

Methods/Description: The Cleft program began completing suicide screening for all patients aged 12 years and older in January 2019. This was preceded by employee training. The Ask Suicide-Screening Questionnaire (ASQ) is used to identify patients who are currently at risk of attempting suicide or who have been at risk historically. The ASQ is a validated tool developed for use in emergency departments, inpatient, and outpatient settings within pediatric institutions. Screening is performed by health-care providers with the consent of caregivers. Staff ask caregiver to exit the room while screening is completed privately with the adolescent.

Results: In the first month after implementation, there was a 50% screening rate of eligible patients. Further training and support was provided to frontline staff, and after the first month, there was improvement to 83% in the number of eligible patients identified and successfully screened. Fifty-eight patients were screened for suicide risk during this time. Of these, 3 (5.6%) patients were identified as at risk for suicide, 2 of whom were acutely at risk and 1 who was

historically at risk. Social Work addressed positive screens through completion of Mental Health Assessment and clinical intervention.

Conclusions: As suicide is the second leading cause of death in teens, the implementation of suicide screening at outpatient visits can be critical in identifying at-risk patients. This is further supported by the identification of 3 patients at a Cleft Clinic visit within the first 4 months of the implementation of the screening process.

#### 341. Outreach Programs: Providing Patients Programs to Meet the Needs of Your Center

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Background/Purpose: Outreach programs provide a means for people with similar medical conditions to create a community in which they can feel connected and find support. It is important to create programs that allow people who truly understand what it is like to live with a craniofacial difference to have the opportunity to support each other. Cleft and craniofacial centers have the unique opportunity to lead the outreach programs because they work with the families closely and have the ability to reach out and connect the families. Centers hold the contact information and are able to connect families. It can also benefit the center by connecting the families even more to the center. By connecting families outside of the hospital environment, in a relaxed atmosphere, families are open to sharing life situations and bonding through that connection. In this way, they have a shared interest in their treatment and the institution delivering that care. Outreach programs can be simple or expansive based on the man power and funding. A simple outreach program can host a holiday party, annual picnic, or walk annually. An extensive program can include monthly workshops for patients, summer speech programs, and educational workshops. Funding programs and manpower are the 2 typical barriers. The goal of the presentation is to educate the audience on 2 different center's experiences in outreach programming based on their region's needs. Defining what outreach programs look like in a calendar year, how those are funded, who takes on the responsibility. Each center has a different experience but both with outcomes that are meaningful and positive for the patient population. The audience can then use the information to begin or grow their own outreach community.

Methods/Description: Introduction of 2 different Programs-Outreach Program Definition: (1) What are the needs in your cleft and craniofacial community? (2) Defining the target participants and their needs? (3) What can be done in your cleft and craniofacial community given the manpower and finances? Starting an Outreach program: (4) Is there a leader/spearhead (staff or parent) that wants to direct the program? (a) Center A: Family wanted a picnic specifically for the cleft community but needed support (5) Getting started. How extensive is the program based on manpower and funding? (a) First year example: Center A/B: Holiday Party, Center A: cleft picnic Center B: Community program Examples of typical Outreach programs: (1) Calendar-A year in the Life of Outreach for the Center, (2) Holiday party (a) Same place/different place each time (zoo, resort, children's museum), (3) Monthly programs (a) Donor funded outings (escape room, Michael's baking class, pottery painting, canvas painting, Giving back day at the hospital waiting rooms) b. Community connections (professional baseball games, college baseball games, professional hockey, professional football).

#### 342. Surgical Workforce and the Global Burden of Orofacial Clefts

Benjamin Massenburg (1), Christopher Crowe (1), Shane Morrison (1), Nivaldo Alonso (2), Mert Calis (3), Peter Donkor (4), Prasetyanugraheni Kreshanti (5), Jie Yuan (6), Richard Hopper (1)

(1) Seattle Children's Hospital, WA, (2) University of São Paulo, Brazil, (3) Hacettepe University, Ankara, Turkey, (4) Knust-Kath, Kumasi, Ashanti Region,(5) Cleft and Craniofacial Center Cipto Mangunkusumo Hospital-Faculty of Medicine Universitas Indonesia, Jakarta Pusat, DKI Jakarta, Indonesia, (6) Shanghai 9th People's Hospital, China

Background/Purpose: Orofacial clefts are one of the most common congenital anomalies and require a multidisciplinary care team. However, this disease burden and the care providers are both inequitably distributed worldwide. Our hypothesis is that most of the burden of orofacial clefting falls on the countries with the smallest surgical workforce or lowest sociodemographic indices, rather than the countries with the highest prevalence of disease

Methods/Description: The Global Burden of Disease methodology was used to estimate prevalence and morbidity of orofacial clefting in 195 countries from 1990 to 2017. Disability-adjusted life years (DALYs) and prevalence were compared over time, geographically, and against the Socio-Demographic Index (SDI) and size of the national surgical workforce. Linear and logarithmic regressions were performed. Countries and regions with particularly high disease burden and low surgical workforces were identified as regions of need.

Results: From 1990 to 2017, the number of clefts worldwide decreased by 4.9% to 10.8 million, and the burden of this disease significantly decreased by 70.2% to 652 084 DALYs. In 2017, low- and middle-income countries experienced 83.5% of the DALY burden. The largest decreases in DALY were seen in East Asia and the Pacific (83.6% decrease) and Sub-Saharan Africa (73.1% decrease), while North America (14.2% decrease) and high-income countries (20.5% decrease) remained neutral. Prevalence was weakly positively associated with increasing SDI ( $r=0.43, r^2=0.18$ ), while DALYs were negatively associated with SDI ( $r=-0.79, r^2=0.48$ ). There was a logarithmic association between the estimated surgical workforce and the disease burden, with significantly fewer DALYs in countries that had a surgical workforce of more than 6 providers per 100 000 population.

Conclusions: The burden of orofacial clefts has decreased significantly despite steady prevalence over the past 28 years. Prevalence of disease was not significantly associated with sociodemographic index. Most of the burden of orofacial clefting is carried by low- and middle-income countries. Countries with inadequate surgical workforces could benefit from national policy changes to stimulate the education and training of surgeons equipped care for patients with orofacial clefts.

### 343. Factors for Reducing the Incidence of Palatal Fistula After Palatoplasty in Developing Countries

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Background/Purpose: We have been conducting medical collaboration programs for cleft lip and palate in Madagascar from 2011 to 2017. Controlling the increased fistula incidence is clinically very important in developing countries, and considering the factors related to the increases essential for achieving this outcome. The purpose of this study was to investigate the incidence of fistula after primary palatoplasty.

Methods/Description: Subjects were 43 consecutive patients (27 males and 16 females) who underwent primary palatoplasty for cleft palate in Madagascar from 2011 to 2017. The age at the time of surgery was 1 year and 2 months to 29 years (average, 7 years and 10 months), and the cleft types were 0 class I, 13 class II, 18 class III, and 12 class IV, by the Veau classification. With respect to cleft palate width, measurements were made on 4 patients in 2016 and 6 patients in 2017 from which maxillary plaster models could be taken. Measurement was conducted using digital calipers that can measure up to 1/100 mm. All surgeries for cleft palate were performed according to the modified 2-flap palatoplasty reported by Tosa et al. In the nasal mucosa, Z-plasty was performed, and the muscular layer was sutured in 3 layers according to the intravelar veloplasty technique. The follow-up period after palatoplasty averaged 3 years and 9 months, from 3 months to 6 years and 6 months. The postoperative palatal condition was always checked before individual patients were discharged. Thereafter, the palatal condition was confirmed by our team in the observation period and by a Japanese nun qualified as a midwife working in Madagascar. None of the 43 patients had complications resulting in fistula formation, and the incidence of postoperative fistula complications was 0%. The cleft width in deciduous dentition was 7.2 to 15.4 (average, 11.9 mm). The width in permanent dentition was 4.9 to 11.4 mm, averaging 9.1 mm. We investigated the incidence of palatal fistula, which is an early complication after primary palatoplasty, in 43 cases conducted in Madagascar over 7 years in which 100% follow-up during the acute phase was possible. Numerous papers have been published regarding the incidence of palatal fistula after primary palatoplasty, which is quite large at 1.6% to 77.8%. The 2-flap technique is a method reported by Bardach in 1967. Salyer et al. report that the 2-flap technique has shown good results over the course of 20 years. The postoperative fistula incidence rate is lower than other surgical techniques. Many patients undergoing volunteer surgery in developing countries have poor nutritional status and oral hygiene due to poverty. For this reason, there are many cases in such surgery where wound healing is generally poor, as the blood circulation condition of the wound site is bad and postoperative granulation does not proceed well. Modified 2-flap technique enables multiple-layer, tension-free, watertight closure which important for minimizing fistula rate.

## 344. Comparative Analysis of Weight-For Age in Infants with Cleft Lip and Palate in Sub-Saharan Africa with WHO Standards: A Cross-Sectional Study

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Background/Purpose: A weight of 10 pounds (4.6 kg) is generally used as the minimum weight requirement for surgical intervention in infants who are up to 10 weeks of age and have adequate hematological indices. Infants with cleft lip and palate usually have feeding challenges, which may translate to their attaining the required weight for surgery at an older age. This is especially so in sub-Saharan Africa. This study aims to determine the degree to which the weight for age of infants who had cleft repair compared with WHO standards. It also

aims to identify any association between the nature of the cleft and weight for age scores.

Methods/Description: A retrospective review of all infants who had primary repair of cleft lip and palate from 2007 to 2019 was obtained from a computerized surgical database. Data obtained were age in days at time of repair, weight, gender, nature of the cleft (unilateral cleft lip, bilateral cleft lip, cleft palate), and severity of the cleft (incomplete or complete). The 95% confidence interval for WHO standard means of weights for each age represented in the data set was calculated to obtain the desired range of normal. Z scores were obtained from the WHO standard, and the distribution of observed weights represented graphically. Descriptive analysis and  $\chi^2$  tests were used to compare weights of the infants with WHO standards and associations with the nature of the cleft, respectively.

Results: One hundred forty-five infants met the inclusion criteria. There was a male preponderance, 82 (56.6%). The mean age of the infants was 177.48 days ( $\pm$ 78.6), while the mean weight was 6.01 ( $\pm$ 3.29) kg. Only 56% of the infants were within WHO mean standard weight. For infants with unilateral cleft lip deformities, the mean age was 159.02 ( $\pm$ 66.74) days, mean weight was 6.24 ( $\pm$ 3.71), and 60.6% (N = 63) were within normal. The mean age of infants with bilateral cleft lip deformities was 182.88 ( $\pm$ 77.03) days, the mean weight was 5.12 ( $\pm$ 1.37) kg, and only 42.3% (N = 11) were within normal range. Infants who had palatal repair had a mean age of 296.07 ( $\pm$ 42.68) days, a mean weight of 6.67 ( $\pm$ 2.05) kg and just 46.7% (N = 7) were within normal range. These differences in weights with nature of the cleft did not reach statistical significance. There was no association between the weights and either the nature of the cleft or its severity.

Conclusion: Nearly 50% infants with cleft lip and palate at the time of repair did not fall within 95% confidence interval of WHO mean standard weights. Therefore, weight for age in children with cleft lip and palate from sub-Saharan Africa may be more accurate than absolute weights in designing protocols for surgical intervention.

#### 345. Incorporating Speech Pathology on an International Cleft Care Team

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Background/Purpose: Every year up to 250 000 new babies with cleft lip and/or palate are born in the world, many of whom are born in areas where resources are limited, scarce, or nonexistent (Mars, Sell, & Habel, 2008). A variety of international cleft care programs have been created in an effort to provide care for individuals who may not otherwise receive surgical intervention. Best practice guidelines assert that interdisciplinary team approaches should be utilized in providing care in international settings, and speech-language pathologists play a crucial role in supporting long-term outcomes for individuals experiencing craniofacial differences. Because of the underdeveloped nature of the medical infrastructure in many developing nations, rehabilitation services including speech-language pathology services may be scarce or unavailable. The inclusion of a speech-language pathologist on an international team allows for speech assessment of previously treated patients, improved triage of surgical candidates, family and community education on speech and language, and instruction of therapy techniques for patients of the program. Involvement of speech pathology may also improve outcomes, as some cases of velopharyngeal dysfunction are related to mislearned errors that can be remediated with speech therapy techniques alone.

Methods/Description: This presentation will discuss crucial elements of speech-language pathology programming abroad and the role of the speech pathologist on international cleft care trips, including screening surgical patients, evaluating nasal resonance balance and velopharyngeal function to inform surgical decision-making, and training families and caregivers in language and articulation. Case study examples will be utilized to exemplify these elements and the importance of collaboration between the surgical team and the speech pathologist in identifying appropriate surgical candidates. The American Cleft Palate-Craniofacial Association has also created guidelines to inform clinicians working abroad, noting standards for training requirements and an emphasis on sustainability with the goal of creating viable and ethical programs. Discussion will also include efforts for program sustainability, to include training family members and other local personnel in delivering speech-language pathology treatment programs. Unique ethical and training considerations when practicing internationally will also be discussed.

## 346. Anaesthetic Overview of the Management of Patients With Cleft Lip and Palate: The Experience of an Indigenous Medical Foundation in Nigeria

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Background/Purpose: Anesthetic consideration is a major challenge in surgical management of patients generally and especially babies with cleft lip and palate. This study aims to highlight the methods and challenges of anesthesia in cleft management as experienced by an indigenous surgical mission.

*Methods/Description*: This is a retrospective study, part of a larger study of patients seen during outreach programs across Nigeria from March 2011 to Dec 2017 by an indigenous surgical mission, Cleft & Facial Deformity Foundation. All the cases of operated orofacial cleft patients were included. Active training of anesthetist was part of the program.

Results: A total of 479 patients, 244 (51.0%) male and 235 (49%) female were operated during the study period, 350 (73.1%) under general anesthesia and 129 (26.9%) under local anesthesia. The age range was 0 to 65 with a mean 9.1 (SD) + 11.2 years. The mean age (+SD) for the LA and GA was 23.7 + 11.8 and 5.4 + 6.7 years, respectively. Complications were recorded in 31 (8.9%) patients. The complications recorded under GA were hypothermia 5(1.4%), hyperthermia 2 (0.6%), bronchospasm 6 (1.7%), tachycardia 10 (2.9%), bradycardia 4 (1.1%), and difficult laryngoscopy due to wide cleft lip 4 (1.1%). There was one case of mortality which was due to Mendelson syndrome. No significant complication was recorded with local anesthesia. Five anesthetist were trained during the study period.

Conclusion: Anesthesia is a major challenge, however, a lot of patients with such disabilities can be treated from such outreach program with better training of personnel, provision of equipment and proper planning.

### 347. Surgical Treatment of Velopharyngeal Insufficiency and Airway Management: A Delicate Balance

Jill Arganbright (1), Steven Goudy (2), Travis Tollefson (3), Siva Chinnadurai (4), Oksana Jackson (5), Dave Ingram (1)

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Background/Purpose: The most common types of surgery for velopharyngeal insufficiency (VPI) are pharyngeal flap, sphincter pharyngoplasty, Furlow palatoplasty, and more recently buccal myomucosal flap. These surgeries either add length to the soft palate, add bulk to the posterior pharyngeal wall, or physically block space in the back of the nose to prevent air escape with speech. Common to all these surgeries is the risk of airway obstruction and subsequent obstructive sleep apnea (OSA). Currently, there is no consensus on necessary airway workup and management either before or after VPI surgery, leading to a myriad of different practice patterns. While OSA is a common side effect of these procedures (pharyngoplasty more so than palatal lengthening), there are limited data on the treatment options and outcomes of these treatments for children with OSA following VPI surgery. The aim of this panel discussion will be to engage providers on different approaches to airway management before and after these surgeries, advantages and disadvantages of these approaches, and explore options for management of OSA when present postoperatively.

Methods/Description: Case based discussions will be used to guide the discussion. Complex cases will be presented and discussed that explore various aspects of airway management for children undergoing surgery for VPI as well as highlight the current variability that exists between providers on this topic. Continued dialog among the panelists and the audience using a case-based discussion will address the following topics: (1) How do you screen patients for obstructive airway symptoms preoperatively? (2) What are specific considerations for airway management in patients with 22q11.2 deletion syndrome or Robin sequence? (3) Does information from nasopharyngoscopy/ video fluoroscopy help to assist you in airway management for patients considering VPI surgery? (4) Do you obtain preoperative or postoperative sleep studies for patients undergoing VPI surgery? (5) Which patients benefit from staged adenotonsillectomy prior to VPI surgery? (6) What precautions during VPI surgery do you take to hopefully prevent the development of OSA? Lastly, a case-based discussion will allow panelists to discuss and debate management options for patients who have developed OSA following VPI surgery. Complex/advanced OSA management will be discussed including druginduced sleep endoscopy and its utility in surgical decision making. We will encourage audience participation, taking questions and hearing opinions of those in the audience.

#### 348. To Admit or Not to Admit, Part One: A Survey of North American Practices

Hillary Jenny (1), Xiya Ma (2), Jordan Steinberg (3), Richard Redett (4), Robin Yang (4)

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Background/Purpose: Hospital and intensive care unit (ICU) admissions following craniofacial procedures affect resource utilization and have a financial impact on the health-care system. We present a survey of current admission patterns following craniofacial surgery. By characterizing practice variability, we can compare current practices to the literature to encourage evidence-based admissions decisions.

Methods/Description: A 36-item survey was sent to craniofacial surgeons at 185 ACPA-approved Cleft Palate and Craniofacial Teams in the United States and Canada to elucidate their current admissions practice to the hospital and/or ICU following craniofacial procedures. The survey is de-identified and exempt from full Institutional Review Board review.

Results: A total of 180 centers had working e-mails and were successfully contacted. Teams were e-mailed the survey twice at a 12-day interval; the survey closed after 1 month. Fifty-three surgeons from 26 states and Canadian provinces completed the survey. Most surgeons report routinely performing cleft lip and cleft palate repairs (96%), cranial vault remodeling (76%), and orthognathic surgery (72%). The most common postoperative pathway following cranial vault remodeling involves ICU admission (72%) for a mean of 2 days (range 1-5). However, 18% report routinely admitting patients in a non-intensive care setting; mean overall hospital stay across all providers was 3.1 days (range 1-5). The majority of surgeons routinely admit both cleft lip (86%) and cleft palate (94%) repairs in a non-ICU setting, with a mean hospital stay of 1.0 (range 1-2) and 1.3 days (range 1-3), respectively. Eight percent report routinely admitting cleft palate repairs to the ICU, with a mean ICU stay of 1.3 days (range 1-2). Following orthognathic surgery, 65\% of respondents routinely admit patients in a non-ICU setting, and 14% admit to the ICU with a mean ICU and overall hospital stay of 1.3 (range 1-2) and 1.6 days (range 1-3). When asked how they feel their admitting practices compared to other surgeons, almost all respondents reported they are similar (93.9%), with 6.1% believing they are less likely to admit patients overnight or to the ICU, and no respondents believing they are more conservative with their admitting practices.

Conclusions: There is significant heterogeneity in hospital and ICU admission practices following craniofacial surgery. Strikingly, cleft palate is handled so disparately that some report a routine ICU admission that is as long as the average total hospital stay. Lastly, there appears to be a disconnect between surgeons' perception of standard management and the true variability in practice.

#### 349. To Admit or Not to Admit, Part Two: A Systematic Review of the Literature

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Background/Purpose: Given the significant variability in current hospital and ICU admissions practices across the United States and Canada, there is no current standard of postoperative care for many craniofacial procedures. We present a systematic review of the literature to better educate providers on factors that may indicate ICU admission.

Methods/Description: A systematic review was performed using the PubMed/MEDLINE, Embase, Cochrane, and Web of Science databases for papers describing admissions practices after craniofacial surgery published from January 1, 2009, to August 9, 2019. Articles were included if they involved craniofacial procedures performed by a plastic surgeon. Case reports, commentaries, systematic reviews, and database studies were excluded.

Results: A total of 1809 articles were identified; 60 were included after abstract screening, and 44 were included after manuscript review. After cranial vault remodeling, 19 of 26 papers discuss routine ICU admission, whereas 7 admit most or all patients to the floor. Average ICU and overall hospital stay for those admitted to the ICU were 1.8 and 5.1 days. Patients not admitted to the ICU had an average hospital stay of 2.3 days. Factors reported to be associated with a need for ICU-level care included open repair, ASA physical status 3 or 4, body weight less than 10 kg, intraoperative blood

transfusion > 60 mL/kg, and not giving intraoperative tranexamic acid. After orthognathic surgery, 8 of 11 papers report routine ICU admission. Length of ICU/hospital stay after mandibular distraction osteogenesis and other orthognathic surgery was 8.3/23.5 days and 8.2/10.1 days, respectively, compared to 3.4 days for non-ICU admissions. Factors associated with a longer length of stay or ICU needs included operative duration and surgery complexity (double jaw surgery or combining orthognathic surgery with other procedures such as septoplasty). After cleft palate repair, 2 of 6 studies mention routine ICU admission as their protocol dictates ICU extubation. Average ICU admission after cleft palate repair in the remaining studies averaged 4.3% and was driven primarily by airway issues.

Conclusions: Multiple studies demonstrate success with targeted, rather than routine, ICU admission following craniofacial procedures. As ICU stays lead to longer hospital stays, patient and procedural factors should be considered when creating postoperative plans to avoid unnecessary ICU admissions. These findings can be compared to the survey of actual practices in part one to identify areas of uncommon practices, for example, 8% of providers admit to the ICU after cleft palate repairs, which may represent the ICU extubation protocols identified in this review. Further research into these populations may help identify whether or not these ICU admissions for delayed extubation are beneficial.

#### 350. A New Technique for Perioral Muscle Reconstruction and Lip Lengthening in Complete Unilateral Cleft Lip

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*Background/Purpose*: Adequate skin lengthening and symmetry may not be consistently obtained in unilateral cleft lip repair, especially in patients with complete cleft. The purpose of this study was to present the model of muscle dissection and approximation to facilitate the lip lengthening and symmetry.

Methods/Description: The design followed the rotation-advancement method without skin measurement. A curvilinear skin incision was made from subnasale to the Cupid's bow peak. Muscle dissection was continued to the contralateral nostril floor beneath the columella base to facilitate downward rotation in the medial lip. Wide muscle dissection was performed in the lateral lip segment from the nasal mucosa passing the alar base. The lateral lip muscle was advanced and sutured to the medial lip muscle in a Z-plasty fashion. A small skin backcut was made above the Cupid's bow peak. Primary nasal correction was performed. A series of 138 patients with complete unilateral cleft lip and palate were included in this study. Standard photographs were collected for measurement in the nasolabial region.

Results: Adequate lengthening and symmetry of the lip was obtained. The ratio of vertical philtral height was  $0.99 \pm 0.05$  between the cleft and noncleft sides. The C flap was used for supplementary skin lengthening in 58% of cases. Postoperative lip retraction requiring massage occurred in 13%. Overall nasolabial appearance was satisfactory.

Conclusions: The new technique of perioral muscle reconstruction facilitated to obtain lip lengthening and symmetry in the repair of complete unilateral cleft lip.

### 351. Posterior Cranial Vault Distraction Osteogenesis: Surgical Technique to Maximize Safety and Optimize Outcomes

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Background/Purpose: Posterior cranial vault distraction osteogenesis is an established alternative to traditional posterior vault osteotomy for cranial vault expansion multi-suture craniosynostosis. The benefits of posterior vault distraction over traditional osteotomy include a more gradual controlled expansion, leading to reduced tension of the overlying scalp while allowing for maximal bone remodeling and decreased rates of relapse. Most described posterior vault distraction techniques place the inferior osteotomy above the torcula. In this study, we present a low occipital craniotomy extending to the foramen magnum for improved cosmesis and increased volume expansion of the narrow posterior cranial base that frequently accompanies multisuture craniosynostosis, as well as demonstrate its benefits and safety.

Methods/Description: We performed a retrospective chart review of all pediatric patients with multi-suture craniosynostosis undergoing posterior distraction osteogenesis surgery at the University of Michigan between 2012 and 2019. Patients were identified using the Electronic Medical Record Search Engine (EMERSE). All included patients underwent preoperative evaluation by neurosurgery, plastic surgery, ophthalmology, and neuropsychiatry. The CT and MRI brain and cervical spine with CSF flow imaging were obtained preoperatively to evaluate bony morphometry, venous sinus position, and potential Chiari malformation. Chart review was preformed to extract biographical data, perioperative and intraoperative surgical information, and postoperative complications. The technique, advantages, and complications are described.

Results: We identified 14 patients undergoing posterior vault distraction at the University of Michigan between 2012 and 2019. Thirteen patients had multi-suture synostosis. Clinical syndromes included Saethre-Chotzen, Crouzon, and Apert. The average age at time of posterior distraction osteogenesis surgery was 14.2 months (range 5 to 93 months). Blood loss was an average of 86 cc (range 20 cc to 200 cc), with 7 patients requiring transfusion. No patients had hyponatremia requiring treatment. The average hospital length of stay after surgery was 6.4 days (range 2 to 29 days) and all patients completed distraction of 30 mm. Three patients had Chiari malformation prior to posterior distraction; 2 improved and 1 remained stable postoperatively. Complications included distractor device failure requiring reoperation (1 patient), shunt exposure requiring operation (2 patients), and mild scalp wound infection requiring only local wound care (1 patient). Twelve patients underwent secondary fronto-orbital advancement at 8 to 14 months after completion of posterior distraction osteogenesis.

Conclusions: The utilization of a low suboccipital craniotomy and stepwise distraction is safe and effective. It produces a good cosmetic outcome, especially on turribrachycephalic patients and allows for potential decompression of the foramen magnum.

#### 352. A Long-term Evaluation on the Cupid's Bow Reconstruction in Bilateral Cleft Lip Repair

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Background/Purpose: Bilateral cleft lip, the most serious type of congenital cleft lip deformity, leading tissue defect, heterotopia and hypoplasia widely, is one of the challenges of comprehensive treatment of cleft lip and palate. Therefore, finding an ideal approach to repair is very necessary. The aim of this study was to evaluate the appearances of the lip and nose in patients with bilateral cleft lip and palate, repaired with the Cupid's bow reconstruction method in a long-term evaluation.

Methods/Description: Eighty patients with bilateral complete cleft lip and palate (BCCLP) and 80 patients with cleft palate only (control) were retrospectively enrolled. Facial casts of all subjects taken before and after cheiloplasty, before palatoplasty, and 3 to 4 years later since palatoplasty were used. Ten landmarks were measured by electronic caliper. Independent-sample *t* test was used in analyzing difference between the BCCLP and control, paired-sample *t* test among BCCLP groups, and Pearson correlation coefficient in analyzing the relationship between the landmarks and the surgical effects. The evaluation on scars includes the melanin, the vascularity, the height, and the pliability of them. Independent-sample *t* test was also used in analyzing difference between the left-side scar indexes and the right-side ones. The threshold of significance was set at .05.

Results: The nasolabial appearance is satisfactory. The columellar length (P < .001), peak-to-peak distance (P < .001), and prolabium length (P < .001) increased significantly with catching-up growth, while the nasal width (P < .001) decreased. The VSS scores of the left-side and right-side upper lip are 2.931 and 3.094 in 15 points, respectively, at 5 years after the bilateral cleft lip repair.

Conclusions: Nasolabial displacements were corrected partially by primary surgery, and catching-up growth happened since then. The columellar length is the key factor in bilateral cleft lip repair. Some patients still need scar revisions.

### 353. Early Feeding Outcomes in Infants With Robin Sequence

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Background/Purpose: Infants with Robin Sequence (RS) typically have impaired oral feeding. The efficacy of operations to improve airway obstruction has been well-described, and mandibular distraction osteogenesis (MDO) has been shown to yield superior airway outcomes compared to tongue lip adhesion (TLA). However, the effect of these operations on oral feeding has not been well-characterized. The purposes of this study were to: (1) evaluate feeding outcomes in patients with RS who had no operation, TLA, or MDO over the first 2 years of life and (2) determine whether infants who had early operations (<3 months-of-age) had better feeding outcomes than those that were treated later.

*Methods/Description*: This was a retrospective cohort study of patients with RS whose airway obstruction was managed by nonoperative treatment only, TLA, or MDO at Boston Children's Hospital from 1999 to 2019. A control group of patients with isolated cleft palate but without RS was also included. The primary predictor variables were type of operation (no operation, TLA, MDO) and age at the time of operation (> or < 3 months). The primary outcome variable was weight gain, measured by weight z-score (number of standard deviations from the population mean). One-way ANOVA with post hoc analysis and multiple independent t tests were calculated and statistical significance was set at P < .05.

Results: A total of 222 (56% female) subjects were included: no operation, n = 61 (27.5%); TLA, n = 78 (35%); MDO, n = 22 (10%); and control, n = 61 (27.5%). Patients that had MDO tended to have their operation significantly later than those that had TLA (mean ages 37-99 days for TLA and 247-312 days for MDO, P < .05). Most (96%) patients who had TLA and 59% of those that had MDO underwent the operation prior to 3 months of age. At 6 months of age, the MDO group had the lowest mean weight ( $Z = -2.34 \pm 1.88, P < .05$ ), and both TLA and MDO groups were significantly underweight compared to the control group (P < .05). By 9 months of age, there was no significant weight difference between the TLA and MDO groups, but both were still underweight compared to the control group (P < .05). By 24 months of age, there were no significant weight differences between any RS group and the control group. Subjects that had an operation prior to 3 months of age had significantly faster weight gain compared to those that had later operations (P < .05).

Conclusions: Patients with RS who had an airway operation in the first year of life had significantly poorer early weight gain than infants without RS but caught up to non-RS patients by age 2. Those in the MDO group had the lowest weight gain at 6 months, possibly as a result of this operation occurring much later than TLA; this difference between operative groups resolved by 9 months. Patients that had operations before 3 months of age had faster weight gain than those that had later operations.

### 354. Surgical Outcomes and Effects of Two-Flap Palatoplasty on Japanese Speech: A Retrospective Two-Center Study

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Background/Purpose: The two-flap palatoplasty (TFP) technique provides tension-free and multilayer repairs and has been used for many patients with cleft palate worldwide. This study aimed to assess surgical outcomes of TFP for cleft palate, effects on Japanese speech, and compare the findings with reports from other countries.

Methods/Description: This retrospective cohort study assessed records and images of patients with cleft palate treated using TFP between April 2007 and April 2017 at the hospitals. A single surgeon performed all operations using TFP. The main outcome was the oronasal fistula rate, and secondary outcomes concerned speech (hypernasality, nasal emission, articulation, intelligibility, velopharyngeal function [VPF], and the need for additional follow-up palate surgery rate). A perceptual speech evaluation was performed by 3 speech pathologists who were unaware of the conducted surgery. We received IRB approval.

Results: Of the 176 included patients, 27 non-syndromic patients (male:female = 17:10; bilateral cleft lip and plate:unilateral cleft lip and plate:cleft palate = 5:14:8) underwent TFP and their speech outcomes were assessed. The average patient age at surgery was  $14.3 \pm 2.0$  months and that at the time of speech evaluation was  $70.3 \pm 19$  months. The rate of occurrence of oronasal fistula was 11%. Speech was normal to mildly hypernasal in 89% of patients, and 70% had no or inaudible nasal emission. No compensatory articulation errors were noted in 63%. Secondary or minor articulation errors (palatalization or lateralization of consonant sounds) were observed in 22% of patients, and 15% had classic compensatory articulation errors (glottal stops, pharyngeal fricatives, or posterior displacement of consonants). In 89% of patients, intelligibility was normal or good. VPF was normal

in 78% of patients. Three patients required secondary palate surgery (11%).

Conclusions: Occurrence rate of the fistula was similar to that reported in studies from other countries. Hypernasality, intelligibility, VPF, and the need for secondary palate surgery were at the same levels as those reported in other studies. However, the nasal emission rate was slightly lower in this study than in other studies, and the normal articulation rate was clearly lower in our study than in other studies. The age at evaluation was possibly associated with these results because patient age was younger in this study than that in previous studies. Falseness of articulation may improve with aging. Racial differences in craniofacial morphology and sound differences while speaking between Japanese and other languages, such as English, are other causes for differences in outcome. Thus far, TFP alone has not been considered sufficient for Japanese-speaking patients with cleft palate, and additional procedures are required.

### 355. An Assessment of the Utility of Metopic Index in the Evaluation of Patients With Metopic Ridge and Metopic Craniosynostosis

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Background/Purpose: Premature closure of the metopic suture can result in a broad clinical and morphologic presentation ranging from metopic craniosynostosis with severe trigonocephaly to isolated metopic ridge. Isolated metopic ridge is detectable by inspection and palpation, but unlike metopic craniosynostosis, does not have hypotelorism or pterional constriction and does not usually require surgical correction. Pindrik et al proposed the use of various anthropometric cranial measurements, including metopic index, in the evaluation of children with craniosynostosis and reported mean values and reference ranges in normal children. However, mean metopic indices and reference ranges for metopic index have yet to be reported in patients with metopic ridge or metopic craniosynostosis. As a result, we aimed to measure metopic index in these patients at our institution, compare them to reference ranges in normal children, and assess the utility of this metric in the evaluation of patients with metopic ridge and metopic craniosynostosis.

Methods/Description: A retrospective review of patients with metopic ridge and metopic craniosynostosis was performed. The mid frontozygomatic diameter (MFZD) and maximal cranial width for each metopic ridge and metopic craniosynostosis patient were measured from head CT scans. Metopic index for each patient was calculated using the ratio of MFZD to maximal cranial width. Mean values and ranges for metopic index were calculated for these patients at 0 to 3, 4 to 6, 7 to 12, 13 to 18, and 19 to 24 months and compared to normal. Multivariate linear regression was performed to determine the impact of metopic ridge and metopic craniosynostosis on metopic index controlling for age and gender.

Results: Data were available for 18 metopic ridge patients and 75 metopic craniosynostosis patients. Mean metopic index was below the normal reference range at 0 to 3, 4 to 6, and 7 to 12 months of age in patients with metopic craniosynostosis (0.51 vs 0.55-0.63, 0.45 vs 0.54-0.62, 0.48 vs 0.50-0.62) and within normal limits at all ages except 4 to 6 months (0.64 vs 0.54-0.62) in patients with metopic ridge. Mean metopic index was significantly greater in patients with metopic ridge than in patients with metopic craniosynostosis at 4 to 6 months (0.64 vs 0.45; P = .0390) and at 7 to 12 months (0.56 vs 0.48; P = .0011) but not significantly different at other age ranges.

Conclusions: Metopic ridge and metopic craniosynostosis both result from premature fusion of the metopic suture; however, patients with metopic craniosynostosis have reduced metopic indices compared to normal while patients with metopic ridge have metopic indices within normal limits. Our study provides the first reference ranges for metopic index in patients with metopic ridge and metopic craniosynostosis and validates the metric proposed by Pindrik et al for use in the evaluation of patients with craniosynostosis. We hope that this metric will aid physicians in their diagnosis and management of patients with metopic craniosynostosis and isolated metopic ridge.

### 356. Surgical Management of Oronasal Fistulae in Patients With Cleft Palate: A Validated Management Schema

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Background/Purpose: Oronasal fistulae are a well-described complication of cleft palate repair with an overall incidence of 5% to 6%. There are many different techniques to repair oronasal fistulae; however, no one technique has been shown to be superior. The ideal technique likely depends on a variety of factors including fistula location and size and patient age and surgical history. Unfortunately, because fistulae are relatively uncommon, it is unlikely for any one surgeon to develop a comprehensive experience with all types of fistulae and techniques for repair. However, this can be overcome by pooling the experience of multiple surgeons with distinct experiences and approaches. The study has 3 objectives: (1) to describe the multiple surgical techniques available for fistulae repair, (2) to develop a validated schema for management of fistulae, and (3) to provide a review of the benefits and potential complications associated with each technique.

Methods/Description: Institutional review board approval was obtained to transcribe an audio recording from the 2018 Americleft meeting of 6 craniofacial surgeons discussing management of difficult fistulae. Based on this discussion, and using the Pittsburgh classification as the organizing principle, a schema was developed to guide management of various fistulae. Illustrations of the Pittsburgh classification were designed in addition to graphic illustrations to represent the fistulae of the patients discussed at the meeting. These illustrations were included in a survey sent to craniofacial surgeons from 10 different cleft centers to assess fistulae management techniques and the validity of the proposed schema.

Results: Six complicated fistulae were discussed at the Americleft meeting and used as a framework for developing the schema for treatment of fistulae. The management schema was organized by the 7 types of fistulae defined by the Pittsburgh classification and further delineated by the presence or absence of velopharyngeal insufficiency, bilateral or unilateral cleft lip and palate, and approaches to both nasal and oral lining reconstruction. Consensus was reached on important patient factors that should be considered when formulating a treatment plan, including: smoking status, age, dental status, prior surgeries, and future anticipated procedures. Sixteen surgeons completed a follow-up survey eliciting their management approach to 10 hypothetical cases with distinct fistulae; results from this survey validated the

proposed management schema and produced an exhaustive list of surgical approaches for each fistula type.

Conclusions: Using fistula location as the organizing principle, the present study provides a detailed schema for management of oronasal fistulae. By gathering the experiences and expertise from numerous craniofacial surgeons, this study offers a comprehensive tool guiding management of a challenging problem craniofacial surgeons face that is not otherwise provided in the literature.

#### 357. Novel 3D Shape Analysis Tool for Quality Assessment of Cleft Lip/Palate Repair

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Background/Purpose: In low- and middle-income countries (LMICs), the quality of cleft lip/palate (CLP) repair surgeries is primarily assessed by reviewing 2-dimensional (2D) images. This type of review is limited by its time-intensive and subjective nature. There has been little investigation into the use of 3-dimensional (3D) imaging modalities in cleft quality assessment in LMIC settings. 3D technologies may offer a cost-effective and superior alternative to their existing 2D counterparts. The aim of this presentation is to describe the potential application of a novel 3D method of facial shape analysis in the setting of cleft repair quality assessment.

Methods/Description: To begin this pioneering work, the international children's charity Smile Train partnered with engineers and statisticians to develop an inexpensive, user-friendly 3D camera specifically for this project. Researchers then built a software program capable of constructing 3D statistical models of human facial shapes based on the images captured by this camera. The software produces specific point clouds relevant to pre- and postoperative patients with clefts, then attaches color to each point cloud, and renders a 3D image via triangulation. This novel approach uses over 1000 3D points compared to the 23 points used in historical 2D analyses. A "closest control" method is then used to compare surgical patients to controls, and a resultant "patient scorecard" is generated that can be used to provide surgeons with immediate and informative feedback on their surgical outcomes. A full 3D surface representation, or "fitted facial model," can also be used to visually compare patient and control images. A pilot project at a Smile Train partner hospital in Uttar Pradesh, India used these novel 3D cameras and software to take both pre- and postoperative images of 164 patients with CLPs, as well as pictures of 254 individuals unaffected by CLPs (control population). Through the methods outlined above, this novel system has the potential to both improve the current model of international CLP repair assessment and also to create analogous software tools that can be utilized in separate applications and extended to other institutions across partner networks.

### 358. National Trends in Hospitalization Charges and Utilization of Services for Cleft Lip and Palate Rhinoplasty

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Background/Purpose: Many patients with cleft lip and palate will have a cleft nasal deformity requiring rhinoplasty. Due to the complexity of these procedures, significant health-care resources are necessary to

manage these patients. However, the national health-care burden and utilization trends of these procedures are currently poorly understood and may result in significant variation in health-care costs and barriers to access. The purpose of this study was to examine national trends in patient utilization and hospitalization charges associated with cleft rhinoplasty over multiple years.

Methods/Description: The Healthcare Cost and Utilization Project

National Inpatient Sample database was analyzed from January 2007 to December 2014. All patients who were diagnosed with cleft lip or palate who underwent rhinoplasty were included. Variables of interest included demographic data, hospital characteristics, hospitalization data, and total hospital charges. All charges were inflated to May 2019 value with the Consumer Price Index. Univariate and generalized linear models were used to examine associations between various factors of interest and the final adjusted hospitalization charge as well as the change in these factors over the multiple years included. Results: A 62 471 patients were diagnosed cleft lip or palate during hospitalization between January 2007 and December 2014. Of these patients, 905 (1.4%) received rhinoplasty during that hospitalization and were included in the study. Regression analysis showed that national procedure volume increased significantly during the study period (P = .015). Higher procedure volume (>10 cases/year) was associated with nonprofit private hospitals (P < .001) and urban teaching hospitals (P < .001). The database did not have records of any cases performed at a rural hospital. The mean total hospital charge for these patients was \$33 248.21 (interquartile range: \$19 369.16 to \$40 585.97). Hospitalization charges also increased significantly in this

Conclusions: Utilization of services for cleft rhinoplasty appears to center around urban teaching and nonprofit private hospitals. Although hospital length of stay appears be stable year to year, demand for the procedure and hospitalization charges associated with the procedure rose significantly during the period studied. Further studies are necessary to examine other factors that could be driving the increased hospitalization charges associated with this procedure in order to improve the health-care burden and patient access to this procedure, especially since the overall health-care burden for this procedure is expected to rise if demonstrated trends continue.

time period (P = .014), in conjunction with an average hospital length

of stay of 1.42 days that did not change significantly during the study

period (P = .957).

# 359. The Synostosis Research Group (SynRG) Outcomes Study: Preliminary Results from a MultiCenter, Prospective Consortium for the Study of Craniosynostosis Diagnosis and Treatment

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*Background/Purpose*: Craniosynostosis (CS) treatment is complex and varies widely. Large-scale outcome studies are difficult given the practice variation, low incidence of disease, and long time between intervention and final outcome. Established in 2016, the Synostosis

Research Group (SynNRG) is the largest multicenter consortium focused on prospectively evaluating the diagnosis and management of patients with CS. Here, we present a preliminary analysis of these data.

*Methods/Description*: Institutional review board at each SynRG institution approved this study prior to data collection. Patients diagnosed with CS who presented to any of 5 institutions from 2017 to present were enrolled in this study. Clinical data in 276 categories including history, diagnosis, radiographic imaging, intraoperative details, hospital course, and follow-up were recorded prospectively and stored in a REDCap database.

Results: Of 298 patients registered, 62.7% were male. Average age at registration was 10.4 months. Single suture CS accounted for 80% of patients and multisutural 20%; 3% of patients were syndromic. Mean age at surgery was 11.3 months; 46% underwent open vault reconstruction, 43\% underwent strip craniectomy, and 11\% underwent other types of reconstructions. Of those who underwent open reconstructions, 50.1% were fronto-orbital advancements. Of those who underwent strip craniectomy, 66.2\% were sagittal, 16.9\% metopic, and 13.6% coronal. Drains were used in 40% of patients. Antibiotics were given before incision in 98% of patients and continued post-op in 25% for a mean of 25 hours post-op. Tranexamic acid was used in 46% of patients and steroids in 60.5%. Intraoperative transfusion occurred in 42% of patients (80% in vault reconstructions and 11% in strip craniectomies). Postoperative hematocrit was on average 27.0, and 4.6% of patients required post-op transfusion. In-hospital complications were hematoma in 2.3%, early wound breakdown in 0.5%, and seizure in 0.5\%. No CSF leaks, infections, or deaths were reported. Early reoperations were necessary in 1.9% of patients. Mean length of stay was 2.7 days. Narcotics were prescribed at discharge for 73\% of

Conclusions: Large, prospective, multicenter studies of CS treatment have the potential to identify opportunities to optimize care and improve outcomes. This preliminary analysis of the SynRG data reveals clear trends in treatment of CS and will be useful in improving outcomes moving forward as the consortium continues.

### 360. The Burden of Nasoalveolar Molding: Presenting Early Cleft Lip Repair as a More Cost Effective Alternative

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Background/Purpose: Over the last 3 years, a shift at our institution has taken place in which patients originally designated for nasoalveolar molding (NAM) as an adjunct to cleft lip repair (repair after 3 months) have instead undergone early cleft lip repair (ECLR; 2-5 weeks of life) without NAM. Through our previous published studies, when indexed for severity, the 2 groups have demonstrated equivalent surgical outcomes. After implementing the ECLR program at CHLA, only a small subset of patients still undergo NAM with the standard surgical timing of repair. The financial and social impact of this potential paradigm shift has not been studied. We sought to examine the financial and cost-effectiveness of the ECLR protocol.

Methods/Description: We reviewed records for all patients who underwent NAM as an adjunct to cleft lip repair from November 2011 to June 2018. From November 2011 to February 2014, NAM with standard timing of lip repair was the only intervention offered to patients with wide cleft lip defects. From February 2014 to June 2018, ECLR without adjunctive NAM was offered as an alternative. Retrospective chart review of the 2 groups was conducted with emphasis on the following variables: NAM and ECLR cleft classifications, NAM dental visits, ECLR length of hospital stay, ECLR patients' cleft width ratio (CWR), and operative dates. ECLR patients who had a CWR of >0.5 and unilateral complete cleft lip (UCL) were identified as patients who would have originally been offered NAM as an adjunct to their cleft lip repair.

Results: NAM patients required an average of 11 dental visits, accounting for \$2132 in lost income per family. Average direct charges for NAM totaled \$12 290 for the hospital, physician, and device costs. Over the entire study period, the cumulative direct cost of NAM separate from the surgical repair of the lip was \$970 910. ECLR patients underwent lip repair 80 days earlier than NAM patients (mean of 33 vs 113 days, respectively, P < .001). Following the introduction of ECLR as an alternative to NAM with standard lip repair, NAM usage decreased by 48% (52-27 patients) and unilateral cleft lip patients undergoing NAM decreased by 86% (35 to 5 patients). In the study period offering ECLR, 26 patients were diverted from NAM to ECLR resulting in a health-care cost burden of \$319 540 less (\$96 830 per year).

Conclusions: ECLR without NAM is more cost-effective and results in excellent outcomes. NAM as an adjunct to wide cleft lip and nasal repair is no longer the most cost-effective option at our institution. We believe that ECLR has the potential to decrease the burden of healthcare costs in the United States.

#### 361. Requirement of Wnt Modulator R-Spondin3 in Craniofacial Ossification and Bone Homeostasis

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Background/Purpose: Orofacial cleft (OFC) pathogenesis has a strong genetic predisposition that is influenced by environmental factors. The transcription factor Interferon regulatory factor 6 (IRF6) is one of the most important genes implicated in orofacial clefts, Van der Woude syndrome, and nonsyndromic OFC. We identified R-spondin 3 (rspo3) to be a transcriptional target of IRF6. We hypothesize that since IRF6 is a key gene that regulates craniofacial development, investigation of rspo3 functions in the IRF6 regulatory pathway will provide additional mechanistically insight to understand clinically relevant craniofacial biology that underlie OFC pathogenesis.

Methods/Description: Using ChIPseq and RNAseq, we identified rspo3 as an IRF6 target gene. In zebrafish, rescue of IRF6 mutant was sufficient to rescue rspo3 gene expression, suggesting that rspo3 acts downstream of IRF6. Single-cell gene detection technology (RNAscope) was used to delineate rspo3 expression in the craniofacial region during development. CRISPR gene editing was used to generate rspo3 mutant, and detailed phenotype analysis was carried out to analyze ossification and bone density of the mutants.

Results: Using RT-qPCR, rspo3 mRNA was reduced in IRF6 mutant. However, injection of IRF6 mRNA was able to rescue the expression of rspo3. RNAscope showed co-expression of IRF6 and rspo3 mRNA

in the pharyngeal arches indicating its importance in zebrafish craniofacial development. RNAscope technique identified a unique expression of rspo3 in the perichondrium of the trabecula as well as in Meckel's cartilage. CRISPR-targeted mutagenesis successfully generated rspo3 mutants. The embryonic palate and Meckel's cartilage morphology were very modestly disrupted in the rspo3 mutant. In contrast, as the cartilage elements underwent ossification and the animals matured, the mutant craniofacial skeleton exhibited dramatic defects in bone density, dysmorphology of maxillary and mandibular elements, disorganized relationship between bones, and pathogenic fractures. Formation, maturation, and morphology of the dentition were also disrupted.

Conclusions: We discovered that rspo3 is a target gene of IRF6 and is expressed in perichondrial cells that are juxtaposed to the oral epithelium and cartilage structures. We show that rspo3 function is required in a non-cell autonomous way to regulate osteogenesis of the chondrogenic cells, and that rspo3 is also required for bone homeostasis. In contrast to the many genes known to affect embryonic craniofacial development, this study highlights the key function of rspo3 and Wnt signaling in bone and tooth homeostasis. We are now interrogating the genetic interaction between rspo3 with Wnt genes such as wls, wnt9a, and gpcs. Taken together, this work uncovered the transcriptional activation of rspo3 by IRF6 in craniofacial morphogenesis and integrates rspo3 function and Wnt signaling during palatogenesis.

# 362. Outcomes Assessment of Recombinant Human Bone Morphogenic Protein-2/Alveolar Bone Grafting in Patients With Cleft Lip/Palate: A 10 Year Prospective Analysis

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Background/Purpose: Alveolar clefts are maxillary chasms located between the lateral incisor and canine and are attributed to genetic and environmental factors. Currently, the gold standard of treatment for alveolar clefts is a secondary bone graft using iliac crest cancellous bone to minimize maxillary growth disturbance. Primary bone grafting with rhBMP-2 using minimal incision technique, however, mitigates donor-site morbidity and overcomes shortcomings of interrupted maxillary growth. Recent studies have shown that rhBMP-2 has comparable graft take compared to autogenous iliac crest grafts using computed tomography volumes, Bergland, or Chelsea scoring. Literature also shows that rhBMP-2 is more cost-effective than iliac crest bone grafting. Despite these studies, there remains a paucity of long-term data. This study aims to assess long-term clinical and radiographic outcomes of nonsyndromic cleft lip/palate patients who underwent primary reconstruction of alveolar clefts using rhBMP-2.

Methods/Description: After institutional review board approval was obtained, a retrospective review of all patients who underwent primary alveolar bone grafting with rhBMP-2 at UC San Diego/Rady's Children Hospital from December of 2006 to January of 2010 was conducted. Inclusion criteria included a diagnosis of nonsyndromic cleft lip/palate, primary alveolar bone grafting with rhBMP-2, a minimum of 8-year follow-up and documented imaging analysis with either cone beam computed tomography (CBCT) or panorex imaging. Exclusion criteria included secondary bone grafting and patients with syndromic cleft lip/palate. A Chelsea scoring scale was used to assess long-term outcomes of primary alveolar bone grafting. Skeletal occlusion as well as the presence of a cross bite was used to analyze maxillary growth. All statistical analysis was performed using IBM SPSS.

Results: A total of 18 patients who had undergone primary alveolar bone grafting with rhBMP-2 were identified. Of those 18 patients, 5 patients were lost to follow-up and the remaining 13 patients had a cumulative average postoperative follow-up of 8.3 years. Eighty-five percent of those patients identified had class III skeletal occlusion, 15% patients had class I skeletal occlusion. Of the patients with class I skeletal occlusion, 50% had an anterior cross bite. Of the 13 patients included for analysis, 8 patients had imaging analysis with either a CBCT or panorex at 10-year follow-up. All 8 patients were in the mixed dentition phase. The average Chelsea score was a 6.7/8 with 75% of patients being category A, 12.5% of patients being category B, and 12.5% of patients being category D.

Conclusions: Primary alveolar bone grafting using rhBMP-2 is associated with good graft take and longevity as evidenced by the high Chelsea scores, however, 85% percent of patients had class III skeletal occlusion. This study suggests that use of primary alveolar bone grafting with rhBMP-2 may be associated maxillary growth restriction.

## 363. Does Non-Completion of Nasoalveolar Molding Predict a Poorer Speech Outcome in Cleft Children at 3 and 5 Years of Age?

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Background/Purpose: Nasoalveolar molding (NAM) has been a mainstay of preoperative therapy for cleft lip and palate patients at our institution, however not all families are able to complete NAM despite the numerous known benefits. NAM has been well documented to better reposition the lower nasal cartilages and the cleft alveolar segments in order to achieve improved nasal symmetry, as well as to better align the maxillary alveolar arch form which may reduce the number and complexity of future surgeries. However, NAM completion requires frequent follow-up visits for adjustments of the appliance up until the time of surgery which can be very difficult for families. We sought to investigate whether failure to complete recommended NAM therapy could be used as a surrogate for patients at risk for poorer speech outcomes in the future. The purpose of this study is to examine whether speech outcomes are improved in those patients who completed NAM.

Methods/Description: This study consists of a retrospective review of all patients with cleft lip +/- cleft palate referred for NAM treatment prior to primary cleft lip repair at our multidisciplinary craniofacial clinic from 2008 to 2018. Patients were grouped based on NAM completion versus noncompletion. Patients' charts were reviewed for speech therapy reports at the 3 and/or 5-year age point. Any patient who did not receive a 3- or 5-year speech evaluation at our institution was excluded from the study. All speech therapy visits were conducted by one of the 3 speech-language pathologists on staff at our clinic. Specific speech outcome variables were extracted and compared between the 2 cohorts. Statistical analysis was conducted in Microsoft Excel utilizing Student t test.

Results: A total of 215 patients were referred for NAM and were at least 3 years of age at the time of data collection; 142 (66%) of these patients met inclusion criteria for the study. One hundred thirty-four patients had speech evaluations at 3 years old; 103 (77%) completed NAM prior to surgery while 31 (23%) did not. Eighty-five patients had speech evaluations at 5 years old; 71 (84%) completed NAM prior to surgery while 14 (16%) did not complete NAM. At both the 3-and 5-year visits, there were no statistically significant differences in the

speech outcomes (intelligibility, number of non-air pressure and air pressure sounds, or need for continued speech therapy) when comparing NAM adherence.

Conclusions: Our study found no statistically significant difference observed in speech outcomes between NAM completion and noncompletion patients at 3- and 5-year evaluations. This study is limited, however, based on the lack of speech follow-up (> 60% of patients referred for NAM do not have visits at either 3 or 5 years of age). This implies selection bias in our results as those families who completed NAM served as the majority of those who presented for speech evaluation and therefore may represent more motivated or resource-equipped families to comply with regular recommended speech therapy in the first 3-5 years postoperative.

## 364. Bilateral Cleft Lip Treatment: Two-Staged Vermilion Plasty Using Banked Prolabial Vermilion Island Flap

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Background/Purpose: Soft tissue deficiency in the median tubercle and central vermilion can cause the whistling deformity in bilateral cleft lip patients. This study was conducted to evaluate the outcome of constructing a full median tubercle of bilateral cleft lips in 2 phases: primary repair and preschool surgery. We banked the dry vermilion tissue of the prolabium on the oral side at primary repair, which was subsequently used to correct the deformity. We evaluated the results of this method by comparing the surgical outcomes with the lips of normal children.

Methods/Description: We created a vermilion only with lateral lip elements during the primary repair while banking the dry vermilion tissue of the prolabium on the oral side, following the Mulliken method. The dry vermilion of the prolabium is located on the oral side of the lateral mucosal lips sutured in the midline. The banked prolabial vermilion was subsequently shifted to the outside of the mouth using an island flap in pre-school age to create a full median tubercle. The vermilion was banked on 56 children with a bilateral cleft lip born between 2007 and 2016. Among them, the banked vermillion was put to use in 19 children. Lip measurements of 17 of those patients were obtained and compared with the average lip measurements of 6-yearold Japanese children without lip deformities. We obtained the following 3 antropometric measurements: philtral height(sn-ls), vermilion height of the upper-lip (ls-sto), and total labial height(sn-sto). Each patient's median tubercle was evaluated based on her photographs taken before and after the operation. It was graded on a 5-point scale: Type 1 (whistling), Type 2 (flat), Type 3 (tiny tubercle), Type 4 (enough tubercle and exposed mucosal lip), and Type 5 (enough tubercle and not exposed mucosal lip). The grades were analyzed using the paired sample T test.

Results: Our method produced a natural shape and sufficient volume of the median tubercle without further exposing the mucosal surface to the exterior. The pre- and postoperative photographs showed an average score of 3.8 before the corrective surgery and an average score of 4.4 after the surgery, a statistically significant improvement (P < .05). The average sn-ls was 12.4 cm and did not change postoperation, and there was no significant difference between that of normal children (ave. 11.8 cm). The ave. ls-sto increased from 9.9 cm to 10.6 cm, and the ave. sn-sto changed from 22.3 cm to 23.1 cm after the surgery.

Both measurements were significantly larger than those of normal children (ls-sto ave.: 6.7 cm, sn-sto ave.: 18.5 cm) even before the surgery, and they further increased postoperation.

Conclusions: For patients with cleft lips, the current prevailing surgical technique, which relies solely on the lower lip tissue, poses a challenge as the volume of tissue is insufficient to create a full median tubercle. A 2-step technique using a vermilion tissue that was banked at the time of primary repair is promising in addressing this issue.

## 365. The Effects of Caregiver's Illness Representation of Their Child's Craniofacial Condition on Appointment Compliance in a Craniofacial Center

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Background/Purpose: To ensure full rehabilitation, caregivers of children with a craniofacial condition have the responsibility of adhering to their child's ongoing treatment plan. Craniofacial Centers (CFC) typically consolidate multiple providers into one visit to reduce the burden of care. Despite this, families continue to miss appointments, which disrupts treatment and health-care delivery. Research demonstrates that one's causal beliefs and cognitive representations of an illness or condition (ie, illness representation) are related to health and self-regulation (eg, treatment adherence) behaviors. Given that craniofacial conditions are congenital and have a complex etiology, many caregivers create a "common-sense" understanding of their child's condition based on medical information, cultural beliefs, and/or personal experiences. Literature has shown that changing caregivers' illness representation can improve health and self-regulatory behaviors. This study's purpose is to examine how caregivers' illness representation of their child's craniofacial condition as well as demographic factors impact treatment adherence.

Methods/Description: An anonymous bilingual survey, English and Spanish, was given to caregivers of patients at a Midwest urban CFC. Center's population is estimated to have over 60% Hispanic patients with 40% speaking predominately Spanish and 74% receiving public insurance. The survey measured ethnicity, preferred language, mode of transportation, reasons for missed appointments, and illness representation. A modified version of the Brief Illness Perception Question (IPQ) was used to measure 5 dimensions of cognitive illness representation (causal beliefs, consequences, timeline, personal and treatment control, and identity), emotional representations, and illness comprehensibility.

Results: Sixty-five percent of respondents reported missing appointments due to transportation issues, work demands, and psychosocial stressors. Per the IPQ, Hispanic caregivers indicated cultural-bound causes such as "mal de ojo" (evil eye) and "castigo" (punishment from God) as well as medical causes such as genetics. In addition, African American caregivers reported "mocking other children" as causing their child's condition. Finally, cultural-bound causal beliefs were related to negative illness representation, limited understanding of the condition, and appointment adherence.

Conclusions: Given the cultural and socioeconomic diversity of this CFC population, assessment of caregiver's representation of their child's craniofacial condition can be useful in exploring and resolving caregiver's ambivalence to treatment. This study's implications can assist in increasing appointment compliance by implementing culturally effective coordinated care and education.

## 366. Esthetic, Functional and Psychosocial Outcome on Adolescent Patients With Cleft Lip and/or Palate: Regional Cleft Center Experience

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*Background/Purpose*: This study aims at evaluating esthetic, functional, and psychosocial outcome of adolescent patients with cleft lip and/or palate and their parents' after surgery.

Methods/Description: Patients who had cleft lip and/or palate repair surgery done and aged >= 12 years old and their parents are recruited. An interview-guided questionnaire of 17 questions evaluating esthetic, functional, psychosocial domains, and inclination toward revision surgery was completed. The above study had been approved by Hospital Authority research ethics committee.

Results: Sixty-four patients (aged = 12-31, median = 14) and 47 of their parents were recruited from January 2017 to March 2019. Twenty-five patients had cleft palate (CP), 20 patients had unilateral cleft lip and palate (UCLP), and 19 patients had bilateral CLP (BCLP). The questionnaire showed good internal consistencies; Cronbach α was 0.792 and 0.714 for patient and parents group, respectively. Majority of patient and parents were satisfied with the esthetic (overall facial appearance 92% of patients and 100% of parents had moderate to high satisfaction), functional (overall functional 98% of patient and 97% of parents had moderate to high satisfaction) outcome. Despite the high level of satisfaction, there is a substantial proportion of patients and parents reported to have significant negative psychosocial impact (16% and 21% in patients' and parents' group, respectively). CP were most satisfied with esthetics and functions, follow by BCLP group, and then UCLP; especially for nose, upper lip, and overall facial esthetics (t = 2.045-4.777, P < .05). Patients with higher level of satisfaction of esthetic outcome ( $\rho = -0.284$  to -0.399) and speech outcome ( $\rho = -0.261$  to -0.447) had less negative psychosocial impact.

Conclusions: Most patients after cleft repair surgery are satisfied with esthetic and functional outcome. However, there is a group of patient with significant negative psychosocial impact. Patient has less negative psychosocial impact when they had higher level of satisfaction of esthetic and functional outcome. Further study is needed to look into the factors affecting the psychosocial aspect of patients and their family members.

## 367. Comparative Effects of Furlow Palatoplasty and Modified Two-Flap Palatoplasty on the Jaw Morphology of Cleft Lip and Palate

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Background/Purpose: We have recently been using either Furlow palatoplasty (F procedure) or modified 2-flap palatoplasty (T procedure) for palatal closure in cases of cleft lip and palate. The type of palatoplasty procedure used to treat this condition varies between institutions, and few studies in Japan have focused on the differences between procedures, particularly the comparative effect of the F and T procedures on jaw morphology. In this study, we evaluated the effect

on jaw morphology of differences between these palatoplasty techniques.

Methods/Description: We selected 106 patients (62 boys and 44 girls of mean age 4.44 - 0.35 years) among children with nonsyndromic cleft lip and palate, who underwent palatoplasty and palatal closure in the Department of Orthopedic Surgery of Showa University, were examined in the Department of Orthodontics of Showa University Dental Hospital, and underwent standard lateral cranial X-ray (cephalography). They were divided into 4 groups according to whether they had bilateral cleft lip and palate (BCLP) or unilateral cleft lip and palate (UCLP), and whether they underwent the F procedure or the T procedure. The F and T procedures were performed by different surgeons, but a single surgeon was responsible for each type of procedure. A range of different parameters was measured on cephalograms to assess maxillofacial morphology. Measurements were made using Image J software, and the groups were compared by using a Kruskal-Wallis test for multiple group comparisons and a Mann-Whitney U test for tests between 2 groups. This study was approved by the Ethics Committee of Showa University (DH2017-007).

Results: In distance measurements, multiple comparisons revealed significant differences between Ba-B, NANS, and ANS-PNS in different groups, and in angle measurements, there were significant differences between SNA and ANB. The only significant difference observed in comparisons between 2 groups was Ba-B between patients with UCLP who underwent the F and B procedures, with no significant differences between other groups. There were significant differences in N-ANS, ANS-PNS, SNA, and SNB between patients with UCLP and those with BCLP. These results suggested that although UCLP and BCLP result in differences in jaw morphology, the choice between the F procedure or T procedure did not have a significant effect.

*Conclusions*: The choice of the F or T procedure in UCLP and BCLP made no difference to jaw morphology on cephalometry.

### 368. National Trends in Hospitalization Charges and Utilization of Services for Cleft Alveolar Repair

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Background/Purpose: Patients with cleft lip and palate often receive alveolar cleft reconstruction; however, national utilization and hospitalization charges associated with these procedures are poorly understood. The purpose of this study was to examine national trends in patient utilization and hospitalization charges associated with cleft alveolar repair over multiple years.

Methods/Description: The Healthcare Cost and Utilization Project National Inpatient Sample database was analyzed from January 2007 to December 2014. All patients who were diagnosed with cleft lip or palate who underwent alveoloplasty, bone graft to the face, gingivoplasty or craniofacial fistula closure were included. Variables of interest included demographic data, hospital characteristics, hospitalization data, and total hospital charges. All charges were inflated to May 2019 value with the consumer price index. Univariate and generalized linear models were used to examine associations between various factors of interest and the final adjusted hospitalization charge as well as the change in these factors over the multiple years included.

Results: 62,471 patients were diagnosed with cleft lip or palate during hospitalization between January 2007 and December 2014. Of these patients, 3002 (4.8%) received alveoloplasty, bone graft to the face, gingivoplasty, or craniofacial fistula closure during that hospitalization and were included in the study. Regression analysis showed that there was no significant change in national procedure volume during the period studied (P = .455). High procedure volume (>25 cases/year) was associated with nonprofit private hospitals (P < .001), hospitals with large bedsizes (P < .001), and urban teaching hospitals (P < .001). The mean total hospital charge for these patients was \$28 234.42 (interquartile range: \$16 635.93-\$34 549.42). Hospitalization charges also increased significantly in this time period (P < .001), in conjunction with an average hospital length of stay of 1.12 days that did not change significantly during the study period (P = .125).

Conclusions: Utilization of services for cleft alveolar repair appear to center around large, urban, teaching, and nonprofit private hospitals. While the overall demand for the procedure and the hospital length of stay appears be stable year to year, hospitalization charges associated with the procedure rose significantly during the period studied. Further studies are necessary to examine other factors that could be driving the increased hospitalization charges associated with this procedure in order to improve the health-care burden and patient access to this procedure.

#### 369. Social Media and the Craniofacial Surgeon: Opportunities for Engagement and Ethical Pitfalls

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Background/Purpose: Social media engagement is now virtually mandatory for practicing plastic surgeons. Opportunities for dissemination of information, education, socialization, patient connection, and networking abound. However, the boundaries between posts that are professional/unprofessional and ethical /unethical are often pushed and may be unclear to those habituated to formats where sensationalism is prized. This panel will discuss the variety of ways craniofacial surgeons can and should utilize social media and also discuss how to avoid pitfalls when pursuing the goal of maintaining relevance while upholding the highest ethical and professional standards.