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The Americleft Psychosocial Outcomes Project: A Multicenter Approach to Advancing Psychosocial Outcomes for Youth With Cleft Lip and Palate

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Abstract

Cleft lip and/or palate (CL/P) are among the most common of all birth defects. Habilitation requires multiple surgeries and other therapies throughout childhood and adolescence. While

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multidisciplinary care is recommended, there is a great deal of variation in treatment protocols for this condition. There is ample evidence that children with CL/P are at risk for psychosocial problems. However, to date, few studies have systematically investigated parent and patient self-reported psychosocial and quality of life (QOL) outcomes for children with CL/P as they relate to variations in treatment protocols. The Americleft Outcomes project was initiated to demonstrate and document outcomes to be expected with team care, and to define the key features or characteristics of various team treatment protocols and procedures that are associated with more or less favorable/desirable outcomes. This article will describe the psychosocial component of the Americleft Outcomes project that is aimed at developing a protocol that will allow cross team assessment of psychosocial outcomes for children with CL/P in relationship to the treatments they received. The protocol will be detailed along with a description of the process and considerations that were instrumental in the development of the project. Stakeholder input about the project's perceived relevance to families of children with CL/P will be reported. The paper concludes with a discussion of the challenges encountered with this project, clinical implications, and future directions.

Keywords

cleft lip and palate; quality of life; adjustment; treatment outcomes

Cleft lip with or without cleft palate (CL/P) has been estimated to affect one out of every 940 live births, making it the second most common birth defect in the U.S. (Parker et al., 2010). CL/P can result in a range of complex functional and aesthetic problems that can have a profound impact on physical and psychosocial functioning and quality of life (QOL; Hunt, Burden, Hepper, & Johnston, 2005). For example, CL/P can result in speech, hearing, and dental problems, bone and soft tissue deformities, facial asymmetries, and scarring from surgeries (Losee & Kirschner, 2016).

Children with CL/P are typically treated by multidisciplinary teams of specialists beginning at birth and continuing through early adulthood (American Cleft Palate-Craniofacial Association, 2009; Losee & Kirschner, 2016). Children may have multiple reconstructive surgeries, typically performed beginning in infancy and continuing through young adulthood to correct form and function of the mouth and face. Additionally, children with CL/P typically require evaluation and treatment from a range of specialists including geneticists, speech and language pathologists, dentists, orthodontists, otolaryngologists, audiologists, social workers, psychologists, and other disciplines as indicated (American Cleft Palate-Craniofacial Association, 2009). The lifetime cost of treating children with CL/P is estimated at $697 million (U.S. Department of Health and Human Services, 2000). Surgeries, hospitalizations, and ongoing medical monitoring and interventions by specialists can also result in significant time and financial burdens for affected families (Long, 2016).

Data suggest that tremendous variation exists in the total number of surgeries children have over time. Semb et al. (2005) looked at data from five European centers (Clinical Standards Advisory Group [CSAG]) for cleft treatment. They reported that patients with unilateral cleft of the lip and palate had an average number of 3.5 to 6 surgeries by age 17 with additional

surgeries anticipated. This study likely underestimated the total amount of surgical intervention typically received because surgeries were limited to plastic surgeries on the cleft and did not include other cleft-related problems such as otitis media (Weckwerth et al., 2009). It is not uncommon for children treated in the U.S. to have 10–15 cleft-related surgeries before they are young adults. These include primary closure of the cleft lip and palate, lip/nasal revisions, secondary procedures on the palate to address velopharyngeal insufficiency or residual palatal fistula, and jaw surgery (Losee & Kirschner, 2016). These same children may also receive other interventions including nonsurgical molding before initial lip closure (collectively called “pre-surgical orthopedics”), extended or repeated orthodontic treatment, speech therapy, audiometric assessment, and surgical insertion of pressure equalization ear tubes (Losee & Kirschner, 2016).

The psychosocial consequences of CL/P are significant and include increased risk for depression, social problems, anxiety, learning deficits, poor QOL, and low self-esteem (Hunt et al., 2005; Kapp-Simon, 2017). Learning disabilities, most frequently manifesting as a reading disability, occur in 30–40% of children (Conrad, McCoy, DeVolder, Richman, & Nopoulos, 2014) and can affect school achievement and overall adjustment (Feragen, Særvold, Aukner, & Stock, in press). Teasing, because of facial differences and/or speech production, affects as many as 65% of patients (Semb et al., 2005).

Studies have identified differences in psychosocial problems based on type of cleft (e.g., cleft lip and palate [CLP] vs. cleft palate only [CPO]), sex, race, and ethnicity. For example, children with CPO have been found to have more cognitive difficulties compared with non-affected children and children with CLP (Richman, McCoy, Conrad, &, Nopoulos, 2012). Children with visible clefts (e.g., cleft lip only [CLO] or CLP) frequently report greater dissatisfaction with appearance compared with those with invisible clefts (e.g., CPO; Broder, Smith, & Strauss, 1994). Sex differences have also been observed with girls reporting more emotional problems (Feragen & Stock, 2016) and greater appearance concerns (Crerand, Sarwer, Kazak, Clarke, & Rumsey, 2016) whereas boys report more behavior, attention, and peer problems (Feragen & Stock, 2016). Differences have also been observed by type of rater (e.g., parent vs. self-report; Feragen & Stock, 2016). Studies have documented that African American and mixed ethnicity youth with CL/P report lower QOL compared with Caucasian youth (Broder, Wilson-Genderson, & Sischo, 2012).

While numerous studies examined aspects of psychosocial functioning in children with CL/P, there are limited data on the relationship between psychosocial outcomes and variations in treatment protocols. This lack of outcome data related to treatment protocols is not insignificant. Treatment protocols differ in the number, types, and timing of interventions patients receive, all of which can impact the demands placed on children and their families (Long, 2016). Despite the fact that children with CL/P are candidates for multiple interventions throughout their lives, few randomized, controlled trials (RCTs) have been conducted on cleft treatment. Cochrane reviews regarding C/LP treatment are limited to infant feeding, secondary alveolar bone graft, management of submucous cleft palate, and interventions for articulation disorders in children with CP (de Ladeira & Alonso, 2012). The absence of a sound evidence base for the selection of treatment protocols was demonstrated by a striking diversity of practices across Europe for surgical care of just one

cleft subtype, unilateral CLP. Of 201 teams performing initial cleft lip repair, 194 different protocols were being practiced (Grollemund et al., 2012; Shaw et al., 2001).

Few studies investigating psychological or QOL outcomes for children with CL/P link those outcomes to variations in treatment. One exception is the report by Hentges et al. (2011) in which age of lip repair was found to be associated with cognitive development and school achievement at age 7, such that children receiving earlier lip repair (neonatal) performed better than children whose lips were repaired at a later age (3–4 months). Results from the CSAG and Eurocleft studies (Semb et al., 2005; Williams et al., 2001) suggest patient satisfaction was generally high for adolescents regardless of treatment outcomes, but the authors did not assess psychological adjustment or QOL. No difference was found in frequency of patient-reported peer teasing for children ages 8–11 among the treatment teams, suggesting that teasing was not related to treatment protocols (Semb et al., 2005). Feragen and Borge (2010) found that self-reported peer teasing was associated with self-reported appearance satisfaction for 10 year olds with CLP, which suggests a stronger relationship with patient perceptions of outcome and teasing than with the types of outcomes measured by professionals. However, this study did not investigate outcomes related to differences in treatment protocols.

In the United States, there are currently 165 cleft teams (Cleft Palate Foundation, 2014). While the exact number of protocols has not been reported, clinical experience suggests that there is considerable variation within and across teams. For example, some teams offer presurgical orthopedics whereas others do not offer these interventions, preferring to use traditional surgical repair at 3 months of age. Little is known about how variations in the type and timing of interventions impact psychosocial outcomes such as appearance satisfaction and peer victimization. Similar questions pertain to the type of palate repair received as this can have implications for speech and related social functioning and ultimately, QOL. These types of questions are critical to address to ensure that all children with CL/P receive treatment that promotes desired psychosocial outcomes. Furthermore, in the current era of patient-centered care, it is imperative that outcomes assessment incorporates the perspectives of patients and their families (Wong et al., 2013), particularly given that many cleft-related surgeries are performed with the intent of improving QOL and psychosocial functioning (e.g., normalizing appearance to reduce stigmatization, improve body image).

The Americleft Outcomes project, which is sponsored jointly by the American Cleft Palate-Craniofacial Association (ACPA) and The Cleft Palate Foundation (CPF) has been assigned the goal of documenting outcomes of cleft care across centers with the precise aim being “to demonstrate and document outcomes to be expected with team care, and to define the key features or characteristics of various team treatment protocols and procedures that are associated with more or less favorable/desirable outcomes” (American Cleft Palate-Craniofacial Association, n.d., p. 5). The Americleft Outcomes project was initiated to address the lack of outcome data for CL/P treatment in North America, a concern shared by professionals representing the multiple disciplines involved in cleft care (e.g., orthodontics, speech pathology). The Americleft Psychosocial Group (APG) partnered with the Craniofacial Special Interest Group (SIG) of the Society of Pediatric Psychology (Division

54 of the American Psychological Association) to develop a protocol that will allow cross-team assessment of psychosocial outcomes for children with CL/P in relation to the protocol under which they received treatment and the number of surgeries received.

This article will detail the development of the APGs protocol to assess psychosocial outcomes in youth with CL/P, including data obtained from caregivers of children with CL/P about the relevance of the study. Challenges encountered during the process of developing the protocol will be detailed, and implications for clinical care and future directions for outcomes assessment in this patient population will be described.

Americleft Psychosocial Outcomes Project Protocol Development

Group Formation and Process

The APG was established in 2012. Initial membership included six psychologists and social workers from six sites across North America including: Shriner's Hospitals for Children, Chicago, IL; University of Iowa Children's Hospital, Iowa City, IA; Nationwide Children's Hospital, Columbus, OH; Children's Health/Children's Medical Center, Dallas, TX; The Hospital for Sick Children, Toronto, Ontario, Canada; and Lancaster Cleft Palate Clinic, Lancaster, PA. APG members were invited to participate from centers such that there was representation of large and small teams, variation in treatment protocols, and staff with research experience with cleft populations. Both social workers and psychologists were included as the ACPAs Parameters of Care require that craniofacial teams include members from either discipline. Several of these individuals were also founding members of the Craniofacial SIG. Members communicated regularly through email and conference calls, and in-person meetings were conducted twice a year to facilitate protocol development. Protocol feedback was sought from members of other disciplines within Americleft and through discussion and presentations at the ACPA annual meeting. Each member of APG discussed strategies within their own team. Several iterations of the protocol occurred in relationship to these discussions.

Through the synthesis of the APG and Craniofacial SIG, some unique opportunities were encountered for multidisciplinary collaboration and exploration of systemic challenges associated with developing a multisite outcomes assessment protocol. The Craniofacial SIG annual meeting is often held during the ACPA meeting to capture the largest number of SIG members, as many attend ACPA. Because the SIG meeting is typically scheduled immediately after the psychosocial forum discussion at ACPA, social workers, parent advocates, nurse coordinators, and professionals from other disciplines are typially present at the SIG meeting and contribute to discussion of outreach opportunities and evidence-based practice. Contact with these professionals through SIG meetings was one of the mechanisms through which the APG connected with the Ameriface organization, a parent advocacy organization, whose members provided input on the relevance of the protocol to families of children with CL/P. The inclusion of professionals and parent advocates with diverse clinical, research, and personal experiences with CL/P was invaluable to the APG in the development of a multisite protocol that could be used regardless of clinical context.

Rationale and Considerations for Protocol Development

The APGs primary goal was to establish a protocol that would provide a brief, yet comprehensive evaluation of psychosocial outcomes in children with CL/P that could be used by teams for clinical audit and/or research purposes. A cross-sectional, observational design was selected primarily given our interest in assessing feasibility of protocol implementation and funding constraints. Ideally, outcomes assessment would occur longitudinally at varying points across the child's life span (e.g., first year of life, school age, adolescence, and adulthood). However, we agreed that cross-sectional data would be important to obtain as a starting point to inform the development of such longitudinal assessments.

Other considerations related to the feasibility of collecting data across sites given differing levels of resources and known constraints on time for data collection within the context of a multidisciplinary team appointment. The protocol had to be comprehensive enough to address academic, behavioral, emotional, and social issues, brief enough to be administered during regular clinic visits, and simple enough for administration in clinics without dedicated mental health specialists but who desire to conduct a clinical audit of their team outcomes. We agreed that protocol administration should be limited to 30 min or less.

There was much discussion about the age of initial focus among APG members and the broader Americleft Outcomes group. The initial focus of the Americleft group at large was 5 years because many of the objective outcomes of primary treatment could be addressed at that age (speech, dental form, and lip appearance). However, APG members were focused on the importance of both parent and patient feedback because there can be discrepancies in parent versus patient self-report of psychosocial functioning (e.g., Feragen & Stock, 2016). Keeping with the need to focus on the youngest possible group, we choose the limited age range of 8–10 to ensure that youth would be able to complete the questionnaires selected for use in this protocol independently and provide meaningful reports of their own psychosocial functioning. This also enabled us to reduce developmental confounds. Children in this age range are also more likely to be seen consistently across sites for annual team evaluations because of surgical treatment needs.

Finally, considerable time was spent discussing inclusion and exclusion criteria for the project. To minimize confounding variables that could have an impact on psychosocial outcomes (e.g., presence of craniofacial or other syndrome, other significant medical conditions), it was decided that the initial focus would be on children with isolated or nonsyndromic CL/P. Both English and Spanish-speaking families were deemed to be eligible given that some sites (e.g., Texas) have large Hispanic populations. Parents of youth with cognitive disabilities will be eligible to take part (although the child would be excluded from participation).

Assessment of Psychosocial Outcomes

The APG group met several times to identify and achieve consensus about what types of psychosocial outcomes should be measured and how this could be accomplished. The APG members began this process by reviewing the cleft literature and discussing their clinical

experiences working with children with CL/P and their families. Additionally, we consulted with Dr. Nichola Rumsey and her colleagues from the United Kingdom. Cleft Collective to gain insight into the critical domains of adjustment they identified as part of a psychosocial outcomes study in the United Kingdom with children younger than 8 years of age (N. Rumsey, personal communication, June 2012). The constructs identified by the U.K. Cleft Collective included social functioning, worldview, appearance, vocational milestones, psychological well-being, and condition-specific factors (Stock, Hammond et al., 2016). We used these domains as a guideline for the selection of the measures included in this research (see Table 1).

In the cleft literature, over 60 different instruments have been used to assess psychosocial functioning (Stock, Hammond, et al., 2016), thus, a significant amount of time was spent on measure selection. Care was taken to select instruments that could provide assessment of both general psychosocial functioning as well as aspects of functioning that may be specific to the experience of having a cleft (e.g., appearance, stigma; see Table 1). We selected measures available in English and Spanish with established psychometric properties that could be administered across multiple treatment sites and that had the ability to capture differences in medical treatment and diagnostic condition. Cost of the instruments was also considered, and measures that were either free or available at relatively low cost were given greater consideration. Measures included in the protocol also allowed for comparisons between children diagnosed with a cleft-craniofacial condition, a normative population, and children who present with other types of medical diagnoses. Measures selected for the protocol are described below.

CLEFT-Q.—The CLEFT-Q is a newly developed, reliable, and valid patient-reported outcome instrument designed specifically for patients with CL/P ages 8 to 29 years. It has been developed through multiple phases of patient and parent consultation and evaluation in both United States and international samples. It assesses five areas including appearance, speech, social and psychological well-being, and facial function (Wong, 2012; Wong et al., 2013).

Patient reported outcomes measurements information system (PROMIS measures).—Self-report measures from the National Institutes of Health Patient Reported Outcomes Measurements Information System were used to assess self-reported anxiety, depression, and experiences of stigma (PROMIS Scoring Manuals, n.d.). These instruments are valid, reliable, and precise assessments of patient-reported physical and psychosocial functioning (PROMIS Scoring Manuals, n.d.).

Pediatric Quality of Life (PedsQL).—The PedsQL is a reliable and valid 23-item instrument that measures health-related QOL from the perspectives of both children and parents (Varni, Seid, & Kurtin, 2001).

PedsQL Family Impact Scale.—The PedsQL Family Impact Module Scale is a reliable and valid parent-report instrument consisting of 36 items that assess how pediatric health conditions impact parental and family functioning (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004).

Adaptive Behavior Assessment System-Third Edition (ABAS-3) Functional Academics Scale.—The ABAS-3 is a reliable and valid measure of child functioning based on parent and teacher report. The Functional Academics parent-report subscale was selected as a screening measure for school performance (Harrison & Oakland, 2015).

Child Behavior Checklist (CBCL).—The CBCL is a reliable and valid parent report measure rating children's externalizing (e.g., non-compliance, disruptive behavior) and internalizing (e.g., shyness, anxiety) problems (Achenbach & Rescorla, 2001). The CBCL is widely used in both clinical practice and for research purposes.

Assessment of Treatment History

As described previously, treatment protocols vary considerably across cleft teams both in terms of type and timing of interventions. This variation had important implications for the protocol. To make the number of variables manageable and to take into account the major variations in treatment protocols, we identified categories of intervention along which to stratify treatments. This strategy is similar to the approach used by the Eurocleft Studies (Semb et al., 2005). The first category relates to the use or nonuse of presurgical orthopedics, or interventions to improve the relationship of the lip and nasal structures before lip surgery and would be stratified as use of (a) presurgical orthopedics (Clark et al., 2011; Grayson & Cutting, 2001); (b) use of tape or glue to bring the lip segments together before surgery; or (c) bringing the segments together at the time of surgery. Type of palate repair will be stratified as Furlow versus von Langenbeck or other types of repairs (Williams et al., 2011). Other categories include (a) age at lip repair; (b) age at palate repair; (c) need for secondary palate surgery (yes/no); (d) age at alveolar bone graft surgery; and (e) total number of surgeries received.

Recording treatment history was identified as being a potentially laborious task given the complexity of treatment for children with CL/P and the lack of a preexisting measure to use. As such, the APG designed a treatment history assessment to record the variables of interest (i.e., demographics, surgeries received) in a standardized method across sites using a combination of parent interview and medical chart review. Items include: diagnosis, caregiver demographics, child academic information, treatment history (i.e., types and dates for surgeries; orthodontic and hearing-related procedures), and referrals for other services (i.e., speech, psychological therapy).

Stakeholder Input

In line with the U.K. Cleft Collective methodology (Stock, Humphries, et al., 2016) and to ensure that the outcomes selected for assessment would be meaningful to patient stakeholders, an anonymous, online survey was distributed to families with a child with CL/P through Ameriface, a national nonprofit support and advocacy organization (www.ameriface.org). The survey aimed to identify the issues most relevant to caregivers regarding their children's psychosocial adjustment in relation to cleft treatment. Caregivers were asked three questions about specific areas of psychosocial functioning. The questions, which were generated by APG members, included: "How important is this area for research?", "How much has your child's treatment for cleft impacted this area of

functioning?", and "How important is this area of functioning in making treatment decisions for your child?" Key areas of functioning included: "How your child feels about the appearance of his/her face, nose and teeth," "How your child is doing in school," "How clear your child thinks he/she speaks," "How your child is behaving and feeling," and "How your child makes friends and fits in with his/her classmates." Caregivers were also asked "What other topics should be included in this study?" to obtain feedback about other concerns.

When asked about the importance of research in each area, over 84% of participants rated all areas as (very) important. Speech perception (95%), feelings about appearance (94%), and social aspects (94%) were rated the highest. When asked how much impact treatment for cleft had on functioning in each of these areas, feelings about appearance (79%) and speech perception (77%) were rated as being the most impacted. School functioning was rated the lowest, with only 59% of responders rating that treatment had a big impact on school. Finally, when asked what areas of functioning were important when making treatment decisions, speech perception (89%) and feelings about appearance (88%) were rated as (very) important. Again, school functioning was the rated the lowest with 64% of responders rating it as (very) important.

Findings from the survey provided confirmation that the areas under investigation in the Americleft protocol were in fact meaningful to families of children with cleft conditions. Open-ended responses were in line with the topics identified and did not result in additions to the protocol. This information aided the APG in finalizing measure selection for the project.

Procedures

Data collection.—To support project implementation, APG members applied for a small grant from the Cleft Palate Foundation. Funding was awarded in April 2016 to support partial costs of protocol administration for a 3-year period. Enrollment began in July 2016 as sites obtained Human Subjects approval. Screening for eligible subjects is conducted before annual clinic visits. Eligible families are approached for participation by trained research staff during their appointment; interpreters are used for Spanish-speaking families. Records are kept regarding inclusion/exclusion as well as recruitment rates. Informed consent and assent are obtained. Parents and children complete the survey independently, with research staff available to answer questions. Study data are managed using REDCap (Research Electronic Data Capture), a secure, Web based application designed to support data capture for research studies (Harris et al., 2009). The majority of sites are using tablets and REDCap surveys to collect data. Sites without tablets use printed forms and hand-enter responses into the REDCap database. One site (Nationwide Children's Hospital) serves as the data coordinating site and oversees data analyses.

Data analysis plan.—The primary aim of this study is to assess the impact of medical treatment on the psychosocial outcomes of children at ages 8–11 years old. Treatment differences to be evaluated include: use of presurgical orthopedics, taping, or none; Furlow versus von Langenbeck palate repair; age at lip repair; age at palate repair; age at bone grafting; need for secondary palate repair (yes/no); and total number of surgeries. Data

analysis will be conducted through separate linear regressions to assess whether there are differences in behavior, psychosocial, QOL, family functioning, and academic achievement (dependent variables) for each of the treatment predictor variables (i.e., presurgical orthopedics vs. none; Furlow vs. other type of palate repair; age at lip repair; age at palate repair; age at bone graft; need for secondary palate surgery; and total number of surgeries) while controlling for age, sex, race/ethnicity, and socioeconomic status (because of known differences related to these variables). Each analysis will be run first with the group as a whole and then run separately for each cleft type (CPO, CL, and CLP). Given the number of predictor and control variable for the planned regression analyses, a minimum sample size of 788 will be needed to obtain 80% power given an alpha of 5% and conservatively small effect size (.02). Based on patient volume at each site, our goal is to enroll 1,040 parents (as both parents are eligible to take part) and 804 children.

The secondary aim is to determine the feasibility of routinely using standardized outcome measures as quality assurance within cleft clinics. During administration, records will be kept on the time it takes patients and parents to complete the surveys, as well as the time required for staff to manage the data (i.e., administration, scoring, and data entry). Descriptive statistics will be calculated to assess feasibility and administration burdens across sites.

Discussion

The purpose of this article was to present the development of a multisite program for psychosocial outcomes research in craniofacial care that has been fostered through the Craniofacial Special Interest Group. In this vein, we sought to describe the clinical needs of children with CL/P and the development of a shared protocol.

The ultimate goal of this project is to identify which CL/P protocols lead to the best psychosocial outcomes through the use of valid, reliable, and consistent instruments across cleft teams. Additionally, this study will offer families and children a mechanism to express their perceptions about treatment, appearance, psychosocial and academic functioning, and QOL. Currently, parents of children with CL/P make decisions regarding treatments based on provider-driven information that implies “this treatment will provide your child with the best longterm outcome;” however, there is a limited empirical basis for choosing one approach versus another. This study will provide an empirical basis for evaluating the psychosocial outcomes of cleft care.

As detailed above, development of this multisite study protocol required careful consideration of both professional and stakeholder needs. Inclusion of stakeholders in this process was important in the current era of evidence-based care where resource allocation is increasingly dependent upon clinicians’ abilities to document treatment benefits and particularly because the goal of treating CL/P is to optimize function, aesthetic outcomes, and QOL (Wong et al., 2013). As such, their input was vital given our goal of assessing clinically meaningful psychosocial outcomes as they relate to CL/P treatment. Our query of parents through an online survey indicated high interest for the types of data our group is proposing to collect. The majority of parents thought treatment related to CL/P would make

a difference their child's feelings about facial appearance, their ability to make friends, clarity of speech, the child's behavior, and even their child's academic success. Of equal importance, the majority of parents reported that they considered each of these areas when making surgical treatment decisions. Because parents value these outcomes highly, it is critical that teams evaluate the extent to which different treatment protocols actually result in better outcomes in each of these areas. It may be that some treatment approaches result in better outcomes related to appearance and social acceptance, but require more surgeries, which may have a negative impact on child behavior and academic performance. We need to learn from parents and children about the importance of these variables and about how teams can support treatment-related decision-making.

The development and initiation phases of this international, multicenter, committee-based project have taught us several lessons about the benefits and challenges of psychosocial outcomes assessment related to the treatment of a chronic pediatric condition. We attribute our success thus far to several factors, including ongoing, regular communication among the APG members and dedicated in-person meetings; respect for the diversity among APG members, our respective disciplines, and cleft teams; the support of the larger Americleft group which provided guidelines for outcomes assessment as well as the input of our U.K. Cleft Collective colleagues who graciously shared their experience of developing their protocol. We were also fortunate in that our committee formation coincided with the development of the CLEFT-Q that saved us from needing to develop our own patient-reported outcome instrument.

Nonetheless, we encountered some challenges, most notably related to time and resources. None of the APG members receive any salary support or protected time to participate in this initiative, although some support was received from our respective institutions and ACPA/CPF to fund one in-person meeting each year. Combining meeting with the annual ACPA conference helped defray costs and enabled us to meet twice a year. While we were able to successfully obtain some pilot funding, financial support will likely remain a challenge. Our ultimate goal is to develop a practical, cost-efficient protocol that can be implemented even for centers with limited resources. Measure selection was also challenging as a considerable amount of time was spent debating the merits and disadvantages of various instruments. Initially, time was spent modifying a screening instrument, but this was later abandoned in favor of using instruments that have been used in the cleft population previously and the CLEFT-Q.

Currently, the Americleft sites are in the process of implementing the assessment protocol. Although data collection is currently in its infancy, several challenges for protocol implementation have been encountered. While we planned to initiate data collection in July 2016 at all sites, timelines for institutional review board (IRB) approval have varied widely across sites, with some being completed in under three months while others are still under review. The APG utilized a Web based file sharing system to allow all members to access to the study protocol, measures, and other documents necessary for Human Subjects and performance site approval, which helped expedite submission of these documents and minimized systemic delays in approval, to the extent possible. While IRB delays disrupted study initiation at all sites, they offered the opportunity for us to begin enrollment on a

smaller scale and to troubleshoot minor problems with the REDCap survey. Data sharing agreements have also been needed with varying requirements at each institution. Other challenges have involved variations in site resources (e.g., staff time) to actually implement the study. The support of a small grant has enabled sites to hire research staff to support data collection.

While this project has several strengths, several limitations are evident. Because of funding restrictions, only English and Spanish-speaking families are being assessed. In the future, we hope to be more inclusive of diverse populations. The data to be collected are cross-sectional and limited to a restricted age range. While longitudinal outcomes assessment is clearly needed, our study will lay the foundation for these types of investigations and identify important predictors that can be studied prospectively. Finally, while we obtained some stakeholder input, time and financial limitations prohibited more extensive inclusion of both caregivers and children in the project conceptualization (e.g., conducting focus groups). However, we are using the CLEFT-Q which is based on interviews with a large, diverse sample of persons with CL/P, and feedback about the current protocol is being solicited from caregivers and children.

Despite these challenges, we have successfully developed and initiated implementation of a multisite protocol to assess psychosocial outcomes in youth with CL/P. Future directions include expanding outcomes assessment to additional sites and conducting mixed methods, longitudinal assessments to examine how treatment relates to psychosocial functioning over time (e.g., from infancy through adulthood). This research represents a critical step toward advancing our understanding of how cleft treatments impact psychosocial functioning and will aid in the identification of empirically supported approaches to achieve the best outcomes for all children with CL/P. We hope that our experience can serve as a model for successful collaboration across multiple specialty groups in the effort to understand and improve psychosocial outcomes for children with chronic health conditions.

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Table 1

Assessment Domains and Associated Measures

Domain	Measures	Reporter/source	General or cleft specific instrument
Condition-specific factors	PedsQL Family Impact Module	Parent	Both
	Cleft Q		
		Child	
Demographic and treatment history	Family Medical History Interview	Parent, chart review	Cleft specific
Social functioning	Cleft Q	Child	Both
	CBCL	Parent	
Stigma	PROMIS Stigma Scale	Child	General
Appearance	Cleft Q	Child	Cleft specific
School performance	ABAS-3 Functional Academics Subscale	Parent	General
Psychological well-being	CBCL	Parent	Both
	Cleft Q	Child	
	PROMIS Anxiety and Depression Scales	Child	
	PedsQL	Parent and child	

Note. ABAS-3 = Adaptive Behavior Assessment System, 3rd Edition; CBCL = Child Behavior Checklist; PedsQL = Pediatric Quality of Life Inventory; PROMIS = Patient Reported Outcomes Measurements Information System.