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Quality-of-Life in Children with Orofacial Clefts and Caregiver Well-being

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Abstract

Quality of life is a valid patient-reported parameter that provides an assessment of treatment need or outcomes complementary to standard clinical measures. Such patient-reported assessments are particularly salient when examining chronic conditions with prolonged treatment trajectories, such as cleft lip and palate. This critical review identifies key questions related to ongoing research on the oral health–related quality of life (OHRQoL) in children with cleft and caregiver well-being. Details of the design and results from 2 longitudinal multicenter studies are presented. This article also provides an update on recent published reports regarding OHRQoL in individuals with cleft. Methodological issues in OHRQoL research are discussed, including condition-specific versus generic instruments, incorporating positive items in OHRQoL instruments, calculating minimally important differences in OHRQoL, implementing mixed methods design, and utilizing validated short assessment forms in OHRQoL research. Finally, new directions for research in cleft as a chronic condition are identified and discussed.

Keywords: cleft lip, cleft palate, resilience, patient-reported outcomes, family relations, longitudinal studies

Introduction

Clefting is the most common facial birth defect worldwide (Shaw 2004). Cleft care in developed countries is delivered by a multidisciplinary team due to the special needs associated with cleft, including speech/language challenges, facial differences, atypical dental development, malocclusion, learning disabilities, chronic ear infections, and associated psychosocial sequelae. Cleft specialists (e.g., plastic surgeons, dentists, psychologists, social workers, speech/language therapists) provide ongoing evaluations and interventions from infancy to late adolescence or young adulthood. In addition to dental and adjunct therapies prior to treatment completion, cleft habilitation can include up to 20 operations through adolescence. Despite the issuance of the American Cleft Palate-Craniofacial Association's Parameters of Care, teams often vary on care implementation, including treatment activism and timing of care (Sitzman et al. 2008). Variations notwithstanding, internationally it is recognized that the overall goal of cleft treatment is to improve patients' quality of life (QoL)—or individuals' "perceptions of their position in life in the context of the culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns" (World Health Organization 1995). Oral health–related QoL (OHRQoL) refers to the effect of an oral disease or condition and its treatment on QoL (Sischo and Broder 2011) and includes functional (e.g., speaking), social (e.g., peer interactions), and psychological well-being (e.g., emotional states; Slade and Reisine 2007).

Since the majority of cleft patients are children, this review focuses on children's OHRQoL. Patient-oriented outcomes such as OHRQoL represent an alternative to the biomedical model encompassing a subjective, comprehensive assessment of treatment outcomes. According to an eminent QoL theoretical framework (Wilson and Cleary 1995) that has been adapted to children with cleft (Broder, Wilson-Genderson, and Sischo 2014), OHRQoL includes social-emotional and functional well-being, oral health, and self-image domains with contextual factors such as family characteristics. Within this framework, cleft-related sequelae (e.g., facial differences, speech deficiencies, social anxiety) are shown to significantly affect the child.

Clefting has an impact on not only the patient but also the larger family unit; in fact, treatment adherence for children with a chronic condition is intrinsically linked to family functioning and treatment perceptions (Thibodeaux and Deatrick 2007). The self-perceptions of affected children are influenced by feedback from others, particularly caregivers, who have a profound effect on their children's adjustment and QoL (Drotar 1997). Likewise, caregivers' well-being is influenced by their children's health and reactions to their children's cleft and cleft treatment (Sischo, Broder, and Phillips 2015).

Children with chronic conditions, who are connected to and reciprocally affected by their caregivers and family environment, are also embedded within the medical system, which can affect their development and well-being (Bronfenbrenner 1999). Since cleft care can continue into young adulthood and beyond, it is paramount to understand the effect of ongoing cleft treatment at different developmental stages. Therefore, the goals of this paper are to summarize recent findings related to caregiver well-being and the OHRQoL of children with cleft from 2 longitudinal multicenter studies, present an updated

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literature review of OHRQoL, identify methodological issues in OHRQoL research, and provide insight into future directions for cleft research.

What Do We Know about Caregiver Responses to Early Cleft Care?

Initial cleft treatment includes primary lip and palate surgery, which typically occur prior to 1 y of age. Some infants may also undergo presurgical nasoalveolar molding (NAM), which is a relatively new, controversial cleft treatment designed to improve cosmetic and functional outcomes among those with cleft lip only or cleft lip and palate. Research is just beginning to explore the impact of these early treatments and hospitalizations on the patient's and caregiver's well-being. A multidisciplinary team composed of 6 American Cleft Palate-Craniofacial Association–endorsed centers recently completed the first prospective mixed methods study of early cleft treatment with and without NAM (National Institutes of Health [NIH] / National Institute of Dental and Craniofacial Research [NIDCR] R21 DE021853; Broder, principal investigator [PI]).

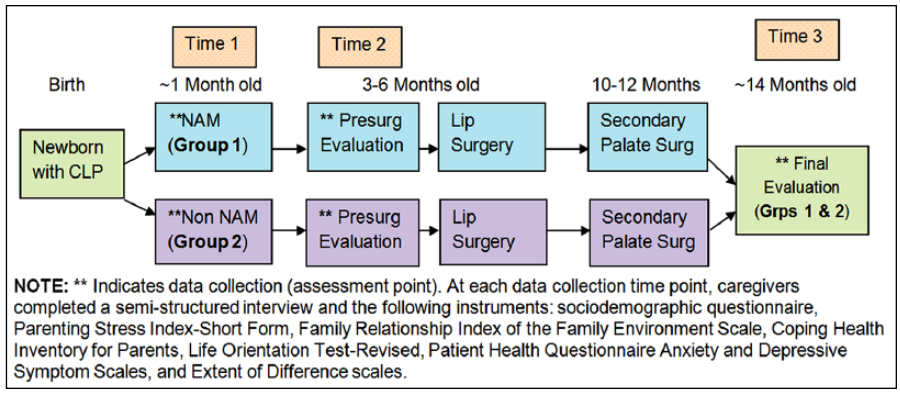

Figure 1. Caregiver responses to nasoalveolar molding (NAM) and early cleft care data collection flowchart. CLP, cleft/lip palate.

The purpose of NAM is to reduce the cleft deformity to allow the surgeon to achieve an improved clinical outcome from the primary lip surgery. NAM uses an intraoral molding plate and a nasal stent to improve nasal symmetry. Proponents argue that NAM reduces the need for future surgery and financial costs over time (Patel et al. 2015), while opponents argue that it places an unnecessary burden (e.g., weekly appointments, appliance cleaning) on caregivers (Hathaway and Long 2014). No prior study had systematically examined this burden; thus, our recent study sought to compare caregiver responses to their infants' early cleft treatment with and without NAM, focusing on caregiver stress and well-being, family functioning, and perceptions of treatment outcomes.

For this research, caregivers (n = 118) of infants (<7 wk old with nonsyndromic cleft lip only or cleft lip/palate [CLP]) completed questionnaires in English or Spanish and one-on-one semi-structured interviews, and standardized photos of the infants were taken at 3 data collection time points. The no-NAM group had primary and secondary palate surgery only, while the NAM group had NAM in addition to these operations. Treatment selection (i.e., group assignment) was made by caregivers. Figure 1 illustrates the data collection flowchart.

Qualitative analyses of semistructured interview data suggested that active participation in infants' cleft habilitation through NAM functioned as a problem-focused coping strategy for most caregivers (Sischo, Broder, and Phillips 2015). Many NAM caregivers experienced increased self-efficacy, positive identity formation, and empowerment. Observing a visible change in the size of the child's cleft and forging relationships with other NAM caregivers at the clinics were often reported as positive experiences.

Mixed methods (i.e., quantitative and qualitative) analyses revealed that NAM caregivers experienced better psychosocial outcomes than no-NAM caregivers despite the greater "burden" of care associated with NAM (Sischo, Clouston, et al. 2015). In particular, NAM caregivers had more rapid declines in depressive symptoms and anxiety and higher levels of medical coping over time than no-NAM caregivers. Mixed methods analyses found that NAM families' functioning improved as their stress levels decreased over the course of treatment, which supported the Family Adjustment and Adaptation Response Model (Patterson 1988).

Caregivers and an expert plastic surgeon assessed treatment outcomes using the Extent of Difference Scale (Mercado et al. 2011). The surgeon, masked to treatment group and phase, rated standardized de-identified photographs of infants. Although NAM infants had more severe cleft ratings than no-NAM infants prior to treatment, the groups had similar professional ratings after treatment (Broder et al. 2016). NAM caregivers perceived better treatment outcomes in their infants versus no-NAM caregivers.

What Do We Know about the OHRQoL of School-Age Children with Cleft?

Understanding caregiver responses to early cleft care begs the question: How do these findings relate to the OHRQoL of children as they age? School-age years encompass another important developmental period for cleft treatment (e.g., alveolar bone graft, secondary revision surgery). Cleft professionals assume that the sequential operations improve patient well-being, although scant empirical evidence is available to support that contention. Does OHRQoL change over time for school-age children with cleft? Does having cleft-related surgery impact well-being? These questions, which can have important implications for cleft treatment decision making and protocol implementation, served as the rationale for the first longitudinal study of OHRQoL in children with cleft.

The research team designed the largest prospective multicenter study of school-age children with CLP to increase understanding of patient-oriented outcomes, such as OHRQoL and health-related QoL (HRQoL) in school-age children with CLP

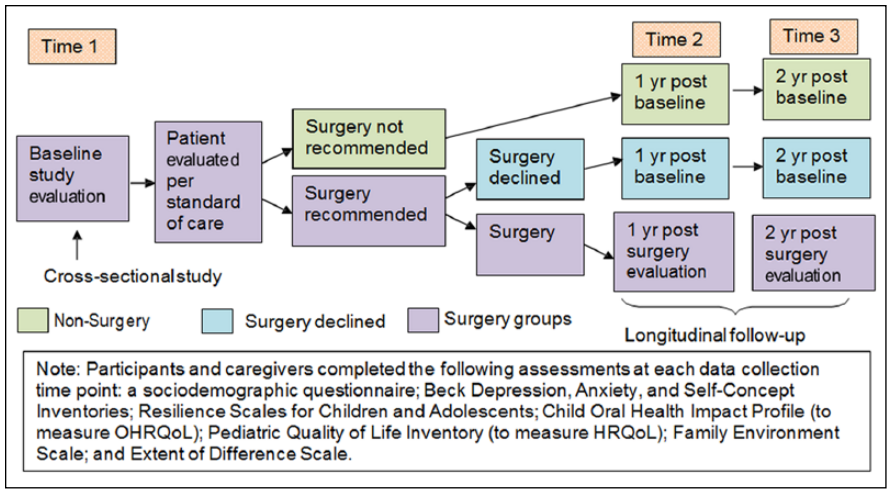

Figure 2. Quality-of-life study flowchart.

and cleft palate only (NIH/NIDCR R01 DE018729; Broder, PI). OHRQoL was measured with the Child Oral Health Impact Profile (COHIP), which was previously developed and validated by the study team (Broder and Wilson-Genderson 2007). The study's theoretical underpinning incorporated biological, social, psychological, and cultural factors into the conceptualization of health, oral health, and QoL (Broder, Wilson-Genderson, and Sischo 2014). The model examined links among clinical variables (cleft diagnosis, health history), functional status (speech, dentition), facial appearance, psychological status, OHRQoL, and overall QoL.

This 6-y study recruited an ethnically diverse sample of children seeking care for cleft from 6 geographically diverse U.S. sites (n = 1,200; 90% enrollment rate). Participants included English- and/or Spanish-speaking children with CLP or cleft palate only who were 7.5 to 18.5 y old at baseline and their caregivers. Child-caregiver dyads were assessed at baseline and up to 3 postbaseline assessments in 1-y intervals (see Fig. 2). Further detail on study design can be found elsewhere (Broder, Wilson-Genderson, Sischo, and Norman 2014).

Cross-sectional Findings

Structural equation modeling demonstrated associations among medical status, sociocultural characteristics, psychological factors, and the mediating role of surgical needs among these children (Broder, Wilson-Genderson, and Sischo 2014). Children with a surgical recommendation at baseline had lower OHRQoL scores on the COHIP than children without a surgical recommendation (Broder, Wilson-Genderson, Sischo, and Norman 2014). Likewise, older females had lower OHRQoL scores than males. Results also identified health disparities as individuals without health insurance and racial/ethnic minorities were more likely than Whites and individuals with private insurance to have lower OHRQoL and a higher rate of surgical recommendations (a proxy for unmet needs; Broder et al. 2012a). These findings imply heightened risk for lower OHRQoL and unmet needs among vulnerable children with clefts. However, results also demonstrated that self-concept was positively associated with OHRQoL and that resilience (i.e., ability to overcome adversity) functioned as a buffer against lower OHRQoL among children with CLP (Broder, Wilson-Genderson, and Sischo 2014).

This study also examined family functioning with the Family Environment Scale. While family functioning was within the normal range, specific areas (e.g., race, ethnicity, type of insurance, and clinical need) were related to 2 domains of family functioning: cohesion (commitment and support within families) and expressiveness (expression of feelings within families; Crerand et al. 2015). Specifically, non-Hispanic and/or White families and those with private insurance reported greater cohesion and expressiveness than African American and Latino families and those with public insurance, respectively. These findings are consistent with prior research on the relationship between adjustment among parents and children with cleft (Berger and Dalton 2011). This research suggests that families play a crucial role in shaping children's experience with cleft treatment, and it highlights the importance of understanding the association between family functioning and children's and caregivers' well-being—an underdeveloped area of cleft research (Baker et al. 2009).

Longitudinal Findings

Longitudinal analyses from this study provided important information regarding the psychosocial well-being of school-age children with cleft postsurgery. While not related to anxiety, depression, and self-concept, receipt of surgery was negatively associated with relatedness (i.e., trust, support, tolerance) and mastery (i.e., optimism, self-efficacy, adaptability) scores in the short term (Ruff et al. 2016b). Sex and race/ethnic differences in psychosocial outcomes were also found: females had higher anxiety, mastery, and relatedness than males; Hispanic children had significantly higher anxiety and depression than White children; Hispanic and Asian children had significantly lower self-concept, relatedness, and mastery than Whites; and Black children had lower relatedness when compared with Whites (Ruff et al. 2016b).

Surgery also affected OHRQoL. Children who received surgery had significantly improved functional and emotional well-being, self-esteem, and overall COHIP scores over time (Broder, Wilson-Genderson, and Sischo 2017). Being male, <12 y of age, and having private insurance were associated with better OHRQoL when compared with being female, >12 y old, and having nonprivate insurance, respectively. Sex differences were congruent with prior appearance research placing girls at risk for body image dissatisfaction (Crerand et al. 2017). Furthermore, results supported prior oral health research that not having private insurance and being a racial/ethnic minority

are associated with less positive health outcomes (U.S. Department of Health and Human Services 2000). Notably, a negative relationship was found between number of prior operations and self- and caregiver-rated OHRQoL. This finding suggests that participants with a greater number of previous operations may have had unrealistic expectations and more severe cleft-related defects, and it might reveal a point of diminishing returns for cleft surgery. Results underscore that patient-oriented outcomes are responsive to change over time and provide an alternative perspective to traditional clinical outcomes, including surgeon ratings of appearance and/or functional status.

Update on OHRQoL Literature

While these 2 studies provide important insights into cleft care in the United States, it is also important to identify the existence and contributions of other cleft-related OHRQoL research. Several systematic reviews have been published on the QoL and OHRQoL of individuals with cleft (Eckstein et al. 2011; Klassen et al. 2012; Antonarakis et al. 2013; Queiroz Herkrath et al. 2015). To provide an update on OHRQoL, we conducted a literature review based on Queiroz Herkrath et al. (2015). Beginning from December 31, 2012 (the date that Queiroz Herkrath's search ended), we completed a PubMed and Web of Science search on studies of OHRQoL among individuals with cleft using the same eligibility criteria, search terms, and instruments as Queiroz Herkrath. Besides the Broder studies cited so far, we identified 9 articles that fit the eligibility criteria. Most of the studies were conducted outside the United States on school-age children with cleft. With some exceptions, results indicated that individuals with cleft had reduced OHRQoL when compared with control groups. See Table 1 for study information and findings.

Despite the existing literature, unanswered questions remain about long-term trends in OHRQoL and psychosocial well-being for individuals with cleft. Continued follow-up for the Broder study on school-age participants is recommended, as half of the sample was ≤12 y old and ~25% had pending treatment recommendations at study completion (Broder, Wilson-Genderson, and Sischo 2017). Such cohort follow-up will provide a more complete understanding of the interrelationships among treatment history, extent of difference, and personal (e.g., type of payer), age, and psychological factors associated with long-term outcomes. Efforts such as continuing to follow this cohort and designing new studies will benefit from examining the current status of statistical and methodological advancements.

Methodological Issues in Cleft and OHRQoL Research

Should You Use Generic or Condition-Specific Measures?

Although patient-oriented assessments based on HRQoL or OHRQoL are valid parameters (Sischo and Broder 2011), important considerations must be weighed when choosing an assessment tool, including whether it is generic or condition specific. While generic HRQoL measures often include normative data that allow comparisons across other health conditions, they also often lack specificity when compared with condition-specific measures (Lee et al. 2010). Examples of generic HRQoL instruments for children include the Pediatric Quality of Life Inventory, Child Health Questionnaire, and the Child Health and Illness Profile. However, the COHIP, the Oral Health Impact Profile (Slade and Spencer 1994), and the Child Perception Questionnaire (Jokovic et al. 2004) are examples of condition-specific validated OHRQoL measures that have been utilized in cleft research.

Do Negative Items Tell the Whole Story?

Traditionally, HRQoL and OHRQoL measures have focused on measuring the negative impact of conditions or disease. Yet, understanding psychological states requires examining both negative and positive psychosocial indicators, since "a high rating of negative influence is not necessarily indicative of an absence of positive effect and vice versa" (Cochrane and Slade 1999). HRQoL experts now recognize that positive attributes are essential in QoL assessment, and positive indicators of well-being are an area of growing interest in health psychology (Yates and Masten 2004). Since the majority of OHRQoL assessments are geared toward the school-age child and focus on negative impact, instruments such as the COHIP that contain positive and negative impacts elucidate the most robust research outcomes.

Are Statistical Tests the Best Way to Detect Clinically Meaningful Difference over Time?

Tests of statistical significance alone may not adequately assess whether a clinical intervention has a meaningful impact on a patient. Utilizing the minimally important difference (MID) to assess clinically meaningful change is a subjective tool for clinical assessment. The MID, which refers to "the smallest difference in score in the domain of interest which participants perceive as beneficial" (Jaeschke et al. 1989), is notably underused in dental research (Masood et al. 2014). MID estimates were calculated for the COHIP in our study of school-age children with cleft (Ruff et al. 2016a). The MIDs of children who did not undergo cleft-related surgery during the study by subscale were as follows: 0.16 (oral health), 0.12 (functional), 0.22 (social-emotional), 0.21 (school environment), and 0.19 (self-image). Future analyses will investigate whether MIDs differ by age or initial cleft severity. These MIDs may provide a valuable tool for identifying the optimal timing of surgery in relation to OHRQoL and for interpreting clinically meaningful change in children's OHRQoL following cleft surgery.

Can a Mixed Methods Research Design Enhance Study Findings?

A mixed methods design, which utilizes qualitative and quantitative assessments, is particularly useful for research on new

Table 1. Updated Literature Review of OHRQoL in Individuals with Cleft.

Authors	Origin	Age, y [a]	Cleft Type	*n*	Instrument	Major Findings
Eslami et al. (2013)	Iran	8 to 15 (N/A)	CLP	50	COHIP	OHRQoL did not change with age; no differences found for unilateral and bilateral clefts; girls had lower OHRQoL than boys.
Ward et al. (2013)	U.S.	8 to 18 (13.0)	CLP, CLO, CPO	75	COHIP	Children with cleft had lower OHRQoL than the control group; negative impact was greater among 15- to 18-y-olds than younger groups.
Antoun et al. (2015)	New Zealand	12.6 (2.8)	CLP, CLO, CPO	24	OHIP	Compared with adults with severe skeletal discrepancies and adolescents with severe malocclusions, adolescents with cleft had the least improvement in OHRQoL after orthodontic treatment.
Konan et al. (2015)	Thailand	8 to 11; 12 to 15 (11.7 ± 2.3)	CLP, CLO, CPO	140	COHIP	Children CLP had generally good OHRQoL. Children with CLO had more positive OHRQoL than children with CLP.
Papi et al. (2015)	Italy	21 to 53 (34.9 ± 7.0)	CLP	63	OHIP	Patients with implant-supported dentures had better OHRQoL than those with fixed partial dentures and removable partial dentures.
Beluci and Genaro (2016)	Brazil	N/A (24)	CLP	50	OHIP, WHOQOL-BREF	Surgery was associated with improved OHRQoL.
Kortelainen et al. (2016)	Finland	11 to 14 (N/A)	CLP	26	CPQ-11-14	Children with cleft had lower OHRQoL than a noncleft control group.
Aravena et al. (2017)	Chile	8 to 15 (11.3)	CLO	48	COHIP	Children with cleft had similar OHRQoL scores as the control group, with the exception of items associated with speech and being understood by other people.
Stelzle et al. (2017)	Germany	12 to 37 (21.9 ± 5.3)	CLP	36	OHIP	OHRQoL was most improved in patients whose own teeth were integrated into cleft.

CLO, cleft lip only; CLP, cleft lip/palate; COHIP, Child Oral Health Impact Profile; CPO, cleft palate only; CPQ-11-14, Child Perception Questionnaire (for 11-14-y-olds); N/A, not available; OHIP, Oral Health Impact Profile; OHRQoL, oral health–related quality of life; WHOQOL-BREF, World Health Organization Quality of Life–BREF (brief version).
[a]Range (mean ± SD).

patient populations and understudied OHRQoL issues (e.g., satisfaction with care, family functioning). Mixed methods can contribute to designing and evaluating interventions to improve the OHRQoL and psychosocial functioning of patients and their caregivers (Rumsey and Stock 2013).

What Is the Role of Short-Form Assessments?

Time restraints associated with a research protocol are critical to consider, especially for patients with conditions requiring multidisciplinary assessments (e.g., cleft). In light of this concern, the COHIP-SF (short form) was created to streamline data collection. The COHIP was reduced from 34 to 19 items in 3 subscales: Oral Health, Functional Well-being, and Socioemotional Well-being (Broder et al. 2012b). Other measures of OHRQoL (e.g., Oral Health Impact Profile) and HRQoL (e.g., SF-36) have also been shortened. Although health service researchers seek shorter assessment tools to address concerns about participant burden and the costs and efficiency of data collection, the psychometric properties of short forms must be demonstrated.

To this end, recent analyses reveal that the COHIP-SF corresponds well to the original COHIP. Table 2 presents a comparison of the long and short forms for the child and caregiver scores, respectively; these data were derived from Broder's study of school-age children with cleft. Like the original, the child COHIP-SF scores improve over time. The correlation between the short and long forms is uniformly high, exceeding 0.96 at each data collection time point.

Similarly, Table 3 presents the COHIP and COHIP-SF scores for surgical and nonsurgical participants and caregiver proxy ratings. Children who had cleft-related surgery had lower self- and caregiver-rated OHRQoL over time when compared with children who did not have surgery and their caregivers. These findings correspond to the relative scores achieved with the long-form COHIP. In sum, the COHIP-SF provides OHRQoL scores comparable to the original COHIP for children with cleft and their caregivers.

New Directions in Cleft and OHRQoL Research

Key Areas for Future Inquiry

Although key results are reported in both Broder longitudinal studies, the empirical thread across developmental strata is lacking. Specifically, the preschool years and young adulthood have been largely ignored in the cleft literature. The preschool years, a critical psychosocial period when children strive for autonomy and have increased social interactions (Campbell 1995), are characterized by significant cognitive, speech/linguistic, social, and behavioral development (Hardin-Jones and Chapman 2011). Preschool children with cleft may have

Table 2. COHIP and COHIP-SF Overall Scores by Visit.

	Baseline		First Follow-up		Second Follow-up		Third Follow-up	
	Mean	*SD*	Mean	*SD*	Mean	*SD*	Mean	*SD*
Child								
COHIP	96.9	18.7	100.3	18.4	102.4	17.2	99.9	18.4
COHIP-SF	53.3	11.3	56.1	10.9	57.9	10.2	57.4	11.3
Pearson correlation	.96		.96		.96		.97	
Caregiver								
COHIP	93.8	19.7	95.6	19.9	97.1	18.7	91.8	17.9
COHIP-SF	51.6	11.6	53.5	11.8	54.5	11.2	51.0	11.2
Pearson correlation	.97		.97		.97		.97	

COHIP, Child Oral Health Impact Profile; COHIP-SF, Child Oral Health Impact Profile–Short Form.

appearance differences and functional limitations (e.g., speech problems, abnormal dental development, hearing loss) that are often a source of significant parental concern, particularly with respect to their children's social interactions and heightened vulnerability to teasing. More research into this developmental period is essential to examine the longer-term associations of treatment during infancy (NAM vs. no-NAM), the effects of treatment on child OHRQoL (e.g., some children have surgery during the preschool period), and whether the disparities and trends observed in school-age children with cleft exist during the preschool years. Given the noted concerns about parental responses to care, family dynamics, and access to care, understanding OHRQoL during the preschool years is vital.

On the other end of the youth developmental spectrum, it is now observed that treatment-seeking for revision surgery is increasing among young adults (Stock et al. 2015). Due to the challenges of systematically collecting standard data among patients who have aged out of children's hospitals (which varies by center) and must then seek treatment elsewhere, who have experienced treatment burnout, and/or have relocated, long-term outcomes regarding patient satisfaction with cleft treatment and OHRQoL in young adulthood remain largely unknown. For example, orthognathic surgery is scheduled when facial growth is completed, which can occur during young adulthood, especially for males; relatedly, lip/nose revisions are scheduled by the surgeon (not the team) after patients have healed from orthognathic procedures. Cleft professionals are beginning to recognize the importance of understanding the effect of cleft and cleft treatment over the life course as patients' experiences and perceived clinical needs, treatment choices, and access to care may change over time (Alansari et al. 2014).

Yet, research on preschool children and young adults with cleft would be remiss without attention to the interrelatedness between family (e.g., perceptions, dynamics) and OHRQoL. Limited evidence suggests that how caregivers cope with their children's conditions may be dependent on cleft type. Caregivers having children with a visible difference (e.g., CLP) have more difficulty coping than do caregivers of children with an invisible difference (e.g., cleft palate only; Kramer et al. 2008). Since caregivers' functioning may be a critical component of children's OHRQoL, future research is recommended that targets family contextual factors related to OHRQoL, treatment adherence and utilization, and health outcomes.

Finally, anxiety regarding medical coverage and/or costs of care may be an important factor in treatment decision making and adherence. An additional burden for the family is the existence of a health record in a world where such records may be purposefully hidden. Therefore, comprehensive patient-oriented cleft research should take social, economic, and family context into account when designing future studies and interpreting findings. Implementing multicenter longitudinal designs is crucial to account for multiple factors within a complex theoretical framework.

Methodological Advancements

Few OHRQoL instruments exist for preschool children, and none have been developed for children having special needs (i.e., CLP). Thus, the COHIP-Preschool was recently validated with data collected from parents of 4 patient samples 3 to 5 y old: children with cleft, children seeking speech therapy, children seeking dental care, and healthy community children (Ruff et al. 2017). Results yielded a final 11-item instrument across 4 domains: oral health, functional well-being, socioemotional well-being, and self-image. The COHIP-Preschool can now be used in clinical research with preschool patients across a variety of oral conditions, including community samples to facilitate family-centered care.

In this era of evidence-based medicine, clinicians require a comprehensive range of well-designed studies to prescribe patient-centered management. Although randomized controlled trials have been considered the research gold standard, such studies are often impractical and ethically unsound, particularly in relation to research measuring the impact of specific, elective, and/or experimental treatments on OHRQoL and psychosocial and clinical outcomes with vulnerable populations (Broder, Crerand, et al. 2017). In recent years, data from observational studies have become an increasingly important source of evidence due to methodological improvements (e.g., electronic deployment of study protocols) and advances in statistical analysis (e.g., mixed models that can control for nesting).

The unique value of observational research can be seen via the systematic observation of parent-child interactions to ascertain physical and psychological developmentally essential milestones and identify the existence of conflicts or lack of attachment (Ainsworth et al. 1956). Indeed, research has

Table 3. COHIP and COHIP-SF Overall Scores by Surgery and Visit.

	First Follow-up				Second Follow-up				Third Follow-up			
	Surgery		No Surgery		Surgery		No Surgery		Surgery		No Surgery	
	Mean	*SD*	Mean	*SD*	Mean	*SD*	Mean	*SD*	Mean	*SD*	Mean	*SD*
Child												
COHIP	97.6	19.2	99.3	18.3	102.2	17.1	103.2	17.7	102.9	18.4	111.5	10.8
COHIP-SF	51.7	11.3	55.1	11.1	53.8	10.8	58.3	10.7	56.0	11.4	64.2	6.1
Caregiver												
COHIP	94.8	26.2	95.9	19.9	97.1	20.2	98.2	19.3	99.1	19.8	106.5	7.04
COHIP-SF	49.2	11.2	52.1	11.9	50.6	11.8	53.9	11.6	52.2	11.2	61.3	4.8

COHIP, Child Oral Health Impact Profile; COHIP-SF, Child Oral Health Impact Profile–Short Form.

illustrated the importance of nurturing, responsive interactions between children and their parents for building infant attachment and for later social and emotional development (Feil et al. 2008). To aid in this type of data collection, the Infant Cleft Observer Outcomes Questionnaire is being developed at the University of Washington (R01DE024986; Edwards and Heike, PIs) for parent-reported observations of the child's feeding, vocalizing, sleeping, hearing, breathing, and activity. Interventions can be designed to mitigate existing or potential problems for at-risk families based on the data collected via this measure. Observational studies can allow researchers to systematically study parent-child interaction to examine intervention effectiveness.

Summary and Conclusions

While treatment varies across chronic conditions, clinical evaluations, therapy, and hospitalizations can be stressful and time-consuming for caregivers and patients. As clinicians strive to improve oral/facial functionality, appearance, and speech production, ultimately measuring patient-oriented outcomes is crucial. Predicting positive OHRQoL and resilience is a challenge for the research team. A chronic condition such as cleft is a diagnosis, but it is not who a person is—indeed, people move differently to the rhythm of circumstance. Therefore, examining OHRQoL among individuals with cleft, as well as caregiver and family well-being, including contextual factors during different development time points, is crucial for understanding change over time, particularly in relation to receipt of surgery. In sum, while existing research has made impressive advances in understanding the OHRQoL of children with cleft, it is vital to continue moving forward by developing novel holistic research paradigms such as psychologically based interventions that integrate OHRQoL findings.

Author Contributions

L. Sischo, contributed to data acquisition and interpretation, drafted and critically revised the manuscript; M. Wilson-Genderson, contributed to conception, design, data analysis, and interpretation, critically revised the manuscript; H. L. Broder, contributed to conception, design, data acquisition, analysis, and interpretation, drafted and critically revised the manuscript. All authors gave final approval and agree to be accountable for all aspects of the work.

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