

BRIEF COMMUNICATION

International Confederation for Cleft Lip and Palate and Related Craniofacial Anomalies Task Force Report: Holistic Outcomes

Submitted on behalf of the Holistic Outcomes Task Force by Hillary L. Broder, Ph.D., M.Ed.

Objective: This paper describes the process and outcomes of the 2013 American Cleft Palate–Craniofacial Association task force on Holistic Outcomes. The goals and membership of the task force are presented.

Methods: Using internet communication, the group introduced themselves, shared ideas and information related to holistic assessment and implementation of using a validated holistic measure, the Child Oral Health Impact Profile (COHIP) at participating international sites.

Results: Data from the sites were analyzed using descriptive statistics. Administration of the COHIP was successful. It varied from self-completion as well as verbal presentation due to language differences and a function of the short time period to complete collection. Additionally qualitative comments were reported by the task force site directors.

Conclusions: Future directions for holistic assessment and communication among task force members and sites were discussed at the Congress and are presented in this report.

A series of Task Forces were established by S. T. Lee, chair of the 1997 Eighth Congress of the International Confederation for Cleft Lip and Palate and Related Craniofacial Anomalies in Singapore. The Task Forces for the 2013 12th International Congress in Orlando, Florida, are derived from the “Objectives” of the International Confederation for Cleft Lip and Palate and Related Craniofacial Anomalies as enumerated in Section 2 of its constitution. The Task Force will orally communicate its findings and recommendations not only to the attendees of 12th International Congress on Cleft Lip/Palate and Related Anomalies 2013 but also to a wider audience via the Internet and continue functioning, if indicated, through consecutive Congresses.

ASSIGNED OBJECTIVES OF THE TASK FORCE

To make recommendations for holistic outcome measures for cleft care that are independent of specific culture.

Many health service researchers today embrace a holistic concept of health that goes beyond the absence of disease to include “a complete state of physical, mental, and social well-being” (World Health Organization, 1948). In focusing on health as a multidimensional concept that incorporates symptoms, physical functioning, and emotional and social well-being, these researchers incorporate quality of life (QoL) and oral health-related quality of life (OHRQoL) into their biopsychosocial health model. Quality of life, or

individuals’ “perceptions of their position in life in the context of culture and value systems in which they live, and in relation to their goals, expectations, standards, and concerns” (World Health Organization Quality of Life Assessment, 1994, p. 28), is now recognized as a valid parameter in patient assessment in nearly every area of physical and mental health care, including oral health. Indeed, several instruments now exist to measure OHRQoL, a multidimensional construct that includes a subjective evaluation of an individual’s oral health, functional well-being, emotional well-being, expectations and satisfaction with care, and sense of self (Sischo and Broder, 2011). More specifically, OHRQoL “reflects people’s comfort when eating, sleeping and engaging in social interaction; their self-esteem; and their satisfaction with respect to their oral health” (DHHS, 2000, p. 7). It is the result of an interaction between oral health conditions, social and contextual factors (Locker et al., 2005), and the rest of the body (Atchison et al., 2006).

Measures of OHRQoL, which are now commonly used across health conditions, are important because they have the potential to enhance evaluation, clinical research, and care in a number of ways, including needs assessment of a population or a specific clinical group.

TASK FORCE FUNCTIONING

Membership Acquisition and Demographics

The task force represented multiple disciplines (e.g., speech and language pathology, plastic surgery, oral and maxillofacial surgery, orthodontics, and psychology) with an expressed, common interest in treating individuals with clefts and evaluating cleft outcomes. Membership in the task force was voluntary. Members

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worked in hospitals, universities, or private practice and/or they engaged in international health work. The 19 members included the following: Aziza Aljohar (Saudi Arabia), Alexander Allori (United States), Kristaninta Bangun (Indonesia), Nubia Becerra Imitola (Colombia), Krishnamurthy Bonanthaya (India), Hillary Broder (Chair; United States), David David (Australia), Adham Farouk (Egypt), Christopher R. Forrest (Canada), Tim Goodacre (United Kingdom), Alex Habel (United Kingdom), Lian Ma (China), Agneta Marcusson (Sweden), Michael Mars (United Kingdom), Raigopal Reddy (India), Nichola Rumsey (United Kingdom), Gentur Sudjatmiko (Indonesia), Ashok Utreja (India), and Murthy Jyotsna (India).

Method(s) of Interactivity

Procedures

Team members established an ongoing dialogue regarding the target group, instruments, and task force goals via e-mail from August 2012 through March 2013. Relevant published articles were also disseminated among the task force members.

While several instruments and related articles were reviewed, we opted to select a validated instrument that had been used with our target population. The Child Oral Health Impact Profile–Short Form (COHIP-SF) was sent out for review and critique. The COHIP-SF is a 19-item questionnaire that may be self-administered or read to children between 7 and 20 years old. This QoL assessment includes the following domains: Oral Health, Functional Well-Being, and Socioemotional Well-Being. The COHIP has excellent psychometric properties and discriminates within and across clinical populations in the United States and Canada. The COHIP was pilot tested in both developed and developing countries and has since been forward and reverse translated into several languages. It is currently being used in the six-center ongoing longitudinal National Institutes of Health study, “Quality of Life in Children with Cleft” (principal investigator, Broder). Further, the COHIP is the first children’s OHRQoL instrument to incorporate both positive and negative health impacts, which means it has the potential to measure more than the absence of a condition, including the ability to measure positive attributes or enhanced well-being (e.g., confidence) as a result of care. This aspect is very important given the theoretical orientation that our care aims to enhance the well-being of patients with cleft and related craniofacial conditions. Given that the COHIP properties aligned with the task force goal of finding a holistic outcome measure, it was suggested that pilot testing of the COHIP, if possible, be completed at various sites by the task force members.

Considerations and Issues Addressed by Committee

Initial Issues: Identifying the Usefulness of QoL Measures in Research and Practice

Incorporating QoL measures in patient care creates a shift from traditional medical/dental criteria such as issues of tissue and bone to assessment and care that focuses on a person’s social and emotional experience and physical functioning. Thus, OHRQoL evaluations have the potential to enhance evaluation and clinical research and care in a number of ways from survey research clinical outcomes. QoL assessments are now commonly used across health conditions. While surgeons have been using the rationale that many surgical procedures improve the QoL of patients with cleft, to date there is a dearth of QoL outcome studies in cleft care. Taking QoL impacts into account, however, can differentiate need and help prioritize care for our patients.

Questionnaire development methods, which include input from a variety of disciplines such as psychology and testing specialists, are used to develop reliable, valid assessments. Yet, in addition to achieving sound psychometric properties, issues related to age, culture, and comprehension must also be considered when developing a questionnaire. Item clarity and relevance to the target group warrant consideration and must be pilot tested in different cultural settings. In addition, the length or number of items comprising the assessment along with language and reading skills must also be considered before the assessment is used in any clinical setting. The format of the questionnaire should also be taken into account to avoid participant error and fatigue. Finally, special care must also be taken when designing and implementing questionnaires for use in developing countries. In fact, the open meeting of the task force featured discussion about whether standardized measures were the most appropriate way to measure patient outcomes in less developed countries. In short, the selected instrument must be appropriate for the target population. Ideally, a single instrument will be developed that can be used for all patients (across varying age groups) with clefts, especially given that patients served by cleft and related craniofacial care range from infants to adults.

Achieving these goals requires that questionnaire development be an iterative process that includes a literature review, item generation, face and content validity testing, item-impact analyses, qualitative interviewing, as well as consideration of theoretical issues used to nominate items for inclusion and exclusion.

Age-Related Issues: Targeting Our Population

Specific issues arise when developing OHRQoL measures for children. Because oral health is strongly

age-dependent (John et al., 2004), it is not surprising that differences in OHRQoL have been found between children and adults (Tapsoba et al., 2000). Children's dental, facial, and cognitive development change drastically throughout childhood and adolescence (McGrath et al., 2004), bringing challenges to the development of appropriate measurement tools. Designing instruments specifically for children and adolescents allows researchers to identify and examine OHRQoL factors that are unique to these populations (i.e., self-image, social acceptance, and school environment; Rumsey and Harcourt, 2004; Broder, 2007). Caregiver reports can be used for proxy measures in measuring well-being and family contextual factors in infants and preschool children. To date, most of the research on QoL has involved assessment of school-aged children.

Rumsey reiterated her concern that research among adults and their health-related QoL was limited and warrants further study. She summarized the literature by stating that young adults with cleft report lower QoL than their peers, with the exception of family functioning. Adults with cleft have also been found to have fewer social relationships (Marcusson et al., 2001) and were more likely to use social withdrawal as a coping strategy than their noncleft counterparts (Berger and Dalton, 2011). In short, many questions remain about the OHRQoL and well-being among adults with cleft and related craniofacial conditions. The task force agreed and recognized the dearth of adult QoL research. However, since most patients seeking cleft care are children (infants through adolescents) across locations, the task force identified two age groups for possible questionnaire development and/or pilot testing: caregivers of infants who receive follow-up care/assessment and school-aged children <19 years old. Given the lack of culturally sensitive standard instruments for young children, the consensus among the task force was to assess QoL in youth at this time.

Other Issues Discussed Among Committee Members

Another issue in questionnaire selection is whether to use a generic instrument or one that is condition specific. The generic measure allows for comparison across health conditions. Yet, reportedly, such generic standard measurements may be less sensitive to issues relevant to the target population (Broder et al., 2014). The length of the instrument as well as the time required to complete it is always an issue when conducting clinical research.

Independent of the task force, ongoing semi-structured interviews of caregivers with infants and toddlers with craniofacial conditions as well as clinical observations among the task force members revealed that issues such as parental stress, depression, anxiety, social support, financial/medical concerns, developmental

milestones (e.g., feeding, speaking), socialization (e.g., temperament, interactivity), and self-efficacy (e.g., empowerment) are important considerations in relation to the creation and use of holistic outcome measures. Qualitative research is suggested in countries (not involved in questionnaire development) and/or settings to ensure that relevant cultural-specific factors are not omitted. Such research may also augment generalizability of the findings.

In summary, after careful consideration, the task force chose to use a condition-specific measure, the COHIP-SF, in pilot testing. The COHIP-SF, which has excellent psychometric properties, is a 19-item questionnaire that is self-administered or read to children from 7 years of age (Broder et al., 2012). Further using such subjective reports of children (rather than just caregiver proxy reports) can aid in decision making and helping understand the patient.

Pilot Testing

Procedures. Pilot testing occurred under the direction of task force investigators at eight international sites: Dr. Jyotsna Murthy at Sri Ramachandra University (India); Dr. Aziza Aljohar at King Faisal Specialist Hospital (Saudi Arabia); Nubia Becerra Imitola, psychologist at Operation Smile Colombia (Colombia); Dr. Krishnamurthy Bonanthaya at Mahaveer Jain Hospital (India); Dr. Adham Farouk at Alexandria University (Egypt); Dr. Ashok Utreja at Oral Health Sciences Center, PG Institute of Medical Education and Research (India); Dr. Lian Ma at Peking University, Hospital of Stomatology (China); and Dr. Rajgopal R. Reddy at GSR Institute of Craniofacial and Facial Plastic Surgery (India). The sample size for each site is as follows: site 1: Saudi Arabia (N = 50); site 2: Egypt (N = 50); site 3: China (N = 100); site 4: Colombia (N = 50); site 5: India (N = 36); site 6: India (N = 50); site 7: India (N = 60); site 8: India (N = 49). The COHIP was administered in English at all sites except Colombia, where Spanish forms were used. Administration varied, but at most sites, the questions were verbally read to the participants. Length of administration also varied but averaged 10 to 12 minutes.

Results From Pilot Testing. Descriptive data for the entire sample are presented in Table 1. The average age of youth in the sample was 13 years. The sample was 55% male, and most participants were Asian (62.1%). The vast majority (84.85%) had a previous cleft surgery, and half of the participants were recommended for surgery in the upcoming year.

Table 2 presents domain and total COHIP-SF scores by site. Higher scores represent better QoL. India (sites 5 and 7) had the highest mean scores for both the Oral Health and Social/Emotional Well-Being domains. Little variation existed in the Functional Well-Being total scores, with the exception of Egypt (site 2) and

TABLE 1 Descriptive Statistics

<i>Entire Sample</i>		
<i>Variable</i>	<i>n (%)</i>	<i>Mean (SD)</i>
Age, y*	396	13.31 (6.20)
Diagnosis*		
Cleft lip	57 (14.4)	44.4
Cleft palate only	35 (8.9)	50.03
Cleft lip/palate	299 (75.7)	38.98
Gender*		
Male	218 (55)	
Female	178 (45)	
Race*		
Missing	50 (12.6)	
Asian	246 (62.1)	
Black or African American	5 (1.3)	
White	45 (11.4)	
More than one race	50 (12.6)	
Previous surgery*		
No	60 (15.15)	
Yes	336 (84.85)	
Surgery recommended*		
No	199 (50.25)	
Yes	197 (49.75)	
COHIP-SF	443	46.52 (9.56)
Q20 (oral health)	431	3.23 (0.09)

* Does not include data from one of the sites.

India (site 8). In relation to total COHIP-SF scores, sites 3 (China) and 7 (India) had the highest reported OHRQoL (with mean scores of 58.89 each), while sites 2 (Egypt) and 8 (India) had the lowest (with scores of 36.74 and 38.94, respectively).

In sum, qualitative responses from the sites' task force members revealed that the COHIP-SF was "easily comprehended," "very useful," and "fairly inclusive in relation to treatment concerns." One task force member reported that a couple of questions "may be tainted by the psychological scar that patients with cleft usually have." This response appeared to reflect the social psychological impacts of a cleft as reported in the literature (e.g., socioemotional well-being such as social anxiety; Speltz et al., 1993; Feragen et al., 2009).

CONCLUSIONS OF THE CLEFT 2013 TASK FORCE

Through collaboration and research, the task force arrived at several important conclusions. First, an urgent need exists to improve our understanding of outcomes across the age range in patients served internationally. This

need includes both short- and long-term outcomes relative to QoL, oral health, and education. Second, there was general consensus that patient-reported holistic outcomes are useful and recommended. Factors possibly related to OHRQoL and treatment outcomes (e.g., type of defect, age, social support) may be important to consider as we strive to optimize long-term outcomes such as QoL in our patient populations. To best study these factors, it seems important to use both qualitative and quantitative methodological approaches (e.g., chart reviews, face-to-face interviews with youth and caregivers, and the use of validated standardized instruments such as the COHIP). Implementing such mixed methodology (qualitative and quantitative assessments) will provide the most complete answers to questions related to holistic outcomes. However, because of the variation in health utilization, treatment expectations, socioeconomic status, health beliefs, and professional resources across centers, creating a proper protocol of holistic assessments is a challenging, multi-staged task.

RECOMMENDATIONS FOR CLEFT 2017 TASK FORCE

Following the International Confederation for Cleft Lip and Palate and Related Craniofacial Anomalies (ICC) presentation on holistic outcomes, more than 60 people enthusiastically attended the lunch session. Representation ranged from social work, psychology, health services research, and clinical specialists in cleft care. At this 2013 task force forum, suggestions for the future were as follows:

- Given that the services offered by teams internationally are varied, establishing in-service training may be indicated. This training should range from the basics (e.g., the value of assessing patient-centered [reported] outcomes and psychological factors affecting adjustment to life with cleft) to more sophisticated aspects of research (e.g., the pros and cons of various approaches to measurement).
- Creating a tiered approach to data collection, ranging from a series of simple questions at entry level to more sophisticated standardized measures, is suggested. Relatedly, the creation of a minimum data set (variables needed) regardless of the tools and surveys used to examine QoL in various countries would be useful. However, use of condition-specific standardized measures, such as the COHIP that was created with diverse groups and countries, would allow

TABLE 2 Child Oral Health Impact Profile–Short Form (COHIP-SF) Domain Scores by Site

<i>COHIP-SF Domain</i>	<i>1 Saudi Arabia</i>	<i>2 Egypt</i>	<i>3 China</i>	<i>4 Colombia</i>	<i>5 India</i>	<i>6 India</i>	<i>7 India</i>	<i>8 India</i>
Oral health	11.42	9.18	13.1	12.86	15.09	13.82	16.99	10.76
Social/emotional well-being	22.14	20.94	24.74	23.5	25.06	22.98	30.96	20.99
Functional well-being	9.24	6.62	10.14	10.62	9.92	10.12	10.92	7.2
Total (SD)	42.80 (9.42)	36.74 (6.43)	58.89 (4.01)	46.99 (5.58)	50.05 (3.17)	46.92 (7.96)	58.89 (4.01)	38.94 (6.44)

comparisons across sites by specific variables (e.g., cleft type, age)

- Measuring QoL status and change in a prospective longitudinal design is recommended. Providing educational video conferencing or other mechanisms to enhance education and promote health among our patients, their caregivers, health care providers, as well as professional personnel, such as school professionals (e.g., administrators, counselors), is warranted.
- Identifying the number of adults who have never undergone care but are seeking psychological or other cleft-related care is essential for measuring issues of concern and documenting holistic outcomes in these patients.
- While cross-sectional data reveal the current status of patients' QoL, longitudinal outcomes (e.g., >5 years) are recommended to determine holistic changes in patients over time.
- Socioemotional issues are particularly relevant in this patient population, as reported in the literature and the pilot testing results on the COHIP.
- Issues related to socioeconomic status, age, health insurance payer type (if applicable), and cleft type and severity, as well as whether the cleft is associated with other birth defects or syndromes, are critical to successfully measure known and unknown factors of importance in treatment utilization and access to care.
- Evidence-based foundations for care provision and interventions are needed to establish commonalities and differences across international settings and to facilitate the feasibility of using standardized measures in less developed settings.
- Implementing QoL population-based studies, including cleft and craniofacial registries, is suggested. Birth defects surveillance program data collected in collaboration with cleft and craniofacial teams may be useful.

Critique (Positive and Negative) of the Task Force Process

In general, the task force process was a successful collaboration of a range of specialty areas. Most members had never worked with one another. A diverse group of clinicians and researchers also attended the open meetings and expressed interest in patient-reported outcomes. There was an overwhelming consensus that this task force is vital, and the opportunity to create dialogue by exchanging ideas was welcomed.

On the negative side, there was inadequate time for a full discussion at the meeting, which made it difficult for everyone present to contribute and/or fully express their views. We suggest having breakout groups in future meetings so that everyone has the opportunity to contribute to the discussion. The small groups can then report back the discussion highlights to the entire group.

Finally, we suggest at future meetings having a 1- to 2-minute update from individuals on their research findings, current projects, future projects, and so forth. This information could be collated before the meeting and discussed in person.

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