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ORIGINAL ARTICLE

A Standard Set of Outcome Measures for the Comprehensive Appraisal of Cleft Care

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Care of the patient with cleft lip and/or palate remains complex. Prior attempts at aggregating data to study the effectiveness of specific interventions or overall treatment protocols have been hindered by a lack of data standards. There exists a critical need to better define the outcomes—particularly those that matter most to patients and their families—and to standardize the methods by which these outcomes will be measured. This report summarizes the recommendations of an international, multidisciplinary working group with regard to which outcomes a typical cleft team could track, how those outcomes could be measured and recorded, and what strategies may be employed to sustainably implement a system for prospective data collection. It is only by agreeing on a common, standard set of outcome measures for the comprehensive appraisal of cleft care that intercenter comparisons can become possible. This is important for quality-improvement endeavors, comparative effectiveness research, and value-based healthcare reform.

KEY WORDS: *cleft lip, cleft palate, continuous quality improvement, learning health care, outcomes, patient-reported outcomes, value-based health care*

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Cleft lip and/or palate (**CL/P**) is the second most common congenital structural anomaly and affects one baby born in the United States every 2 hours. Children with CL/P may be functionally disabled with regard to eating, drinking, speaking, breathing, and hearing, and they may carry the visible stigma of being different unless provided appropriate care. Treatment of CL/P has been the subject of innumerable studies in the surgical, medical, and allied health literature. A Medline search for CL/P yields more than 20,000 results that cover all aspects of care. Several significant efforts have studied and continue to study cleft outcomes; among these are the pioneering Eurocleft, Clinical Standards Advisory Group (**CSAG**), Americleft, and Scandcleft. (Cf. Eurocleft: Asher-McDade et al., 1992; Mars et al., 1992; Mølsted et al., 1992; Shaw, Asher-McDade et al., 1992; Shaw, Dahl et al., 1992; Shaw et al., 2001; Brattström et al., 2005; Mølsted et al., 2005; Semb, Brattström, Mølsted, Prahl-Andersen, Shaw, 2005; Semb, Brattström, Mølsted, Prahl-Andersen, Zuurbier, et al. 2005; Shaw et al., 2005. CSAG: Bearn et al., 2001; Sandy et al., 2001; Sell et al., 2001; Williams et al., 2001. Americleft: Daskalogiannakis et al., 2011; Hathaway et al., 2011; Long et al., 2011; Mercado et al., 2011; Russell et al., 2011; Chapman et al., 2016. Scandcleft: Bannister et al., 2013; Heliövaara et al., 2013; Karsten et al., 2013; Lohmander et al., 2013; Persson et al., 2013; Rautio et al., 2013; Semb et al., 2013; Willadsen et al., 2013.)

There is an opportunity to further build on these efforts by enabling the systematic measurement of a standardized set of outcomes agreed on by global consensus and by focusing on outcomes that truly matter most to patients with orofacial clefts. Standardized outcome measures will help us answer challenging questions about the comparative effectiveness of particular interventions and treatment protocols (World Health Organization [**WHO**], 2002). The importance of standardizing and systematizing outcome measurements in cleft care was recognized by the WHO (2002), the American Cleft Palate–Craniofacial Association (**ACPA**; Fitzsimons, 2011), Eurocleft, Americleft, and others; yet international standards remain undefined.

Outcomes research in cleft care is difficult because of its inherent complexity (Sitzman et al., 2014): A child with CL/P is treated over time, from birth to young adulthood. All the while, he or she is growing and developing physically, cognitively, and psychosocially. Multidisciplinary care is administered at different stages by a range of specialists, each of whom has his or her own particular focus and perspective. These practical challenges obfuscate the process of objective outcomes assessment in cleft care.

Even if these can be overcome, a team must then decide several important questions: Which outcomes are important? How are these outcomes defined? At what time points should they be assessed? In what manner should they be measured? Using which tools or methods? And from whose perspective are they best considered? Although these questions may seem intuitive, disagreement on these issues has been a major impediment to outcomes research and intercenter comparisons in the past (Shaw, Dahl, et al., 1992; Bearn et al., 2001; Shaw et al., 2001; Lohmander and Olsson, 2004; Sell, 2005; Shaw et al., 2005; Fitzsimons, 2011; Russell et al., 2011; Allori, 2012; Sitzman et al., 2014; Stock et al., in press).

Outcomes assessment is not only problematic for collaborative research studies but also it is a considerable challenge for the individual cleft team. Every team wants to know how well it is doing, and every clinician would like to identify ways in which he or she might improve. However, without standardized methods, monitoring the performance of a particular team may differ year to year as techniques, particular emphases, and leadership or personnel changes occur.

Recognizing the need for standardized outcome measurement in cleft care, the International Consortium for Health Outcomes Measurement (**ICHOM**; www.ichom.org) convened an international, multidisciplinary working group to develop a parsimonious, standard set of outcome measures for the comprehensive appraisal of cleft care that would reflect the complexity of cleft care and respect what matters most to individual patients with orofacial clefts.

In this article, we present the resulting standard set for CL/P. It is hoped that this set will empower cleft teams worldwide with a standard core set of meaningful metrics by which to measure their performance and the value they provide to patients (Porter and Teisberg, 2007; Porter, 2010; Kaplan et al., 2011; Black, 2013; Kim et al., 2013; Coulter et al., 2014; Porter et al., 2016).

It is only by measuring and reporting patient outcomes in a standardized way that the global community can truly transform cleft care.

Methods

Objectives

The primary objective for this initiative was to reach multidisciplinary and international consensus for a standard set of outcomes in cleft care. This standard set must be comprehensive enough to cover the full breadth of cleft care, yet practical enough for sustainable implementation. This would permit teams around the world to measure their own performance in a consistent fashion from year to year.

The secondary objective was to identify a standard set of variables that would enable risk adjustment and case-mix adjustment. This would support longitudinal and cross-sectional comparisons of outcomes among centers that serve slightly different populations in different environments.

TABLE 1 Working Group Members

Name	*Location*	*Clinical Discipline*	*Additional Perspective†*
Asteria Albert, MD	Barcelona, Spain	Pediatric Surgery	
Alexander C. Allori, MD, MPH*	Durham, NC, USA	Plastic Surgery Craniofacial Surgery	• Americleft Surgery Working Group • NSQIP–Pediatric • ACPA Data Standards Committee
Krishnamurthy Bonanthaya, MBBS, MDS	Bangalore, India	Oral & Maxillofacial Surgery	
Kathy Chapman, SLP, PhD	Salt Lake City, UT, USA	Speech-Language Pathology	• Americleft Speech Working Group
Michael Cunningham, MD, PhD	Seattle, WA; USA	Pediatrics	
John Daskalagiannakis, DDS, MSc	Toronto, Ontario; Canada	Dentistry Orthodontics	• Americleft Task Force
Henrietta de Gier	Rotterdam, Netherlands	Otolaryngology	
Cindy Guernsey, RN, BScN	Toronto, Ontario, Canada	Nursing	• Cleft team coordinator
Andrew Heggie, MBBS, MDSc	Melbourne, Australia	Oral & Maxillofacial Surgery	
Cristina Hernandez, RN, BSN, MSHI	Houston, TX, USA	Nursing	• Outcomes & Impact Service
Oksana Jackson, MD	Philadelphia, PA, USA	Plastic Surgery	
Yin Jones	United Kingdom	Parent	• Cleft Lip & Palate Association (CLAPA)
Loshan Kangesu, MBBS, MS	London, UK	Plastic Surgery Craniofacial Surgery	
Thomas Kelley, MD, MBA*	Boston, MA, USA	Medicine	• Group methods • Delphi process • Scale development
Maarten J. Koudstaal, MD	Rotterdam, Netherlands	Plastic Surgery Craniofacial Surgery	• ICHOM† Craniofacial Microsomia working group
Rajiv Kuchhal	Bangalore, India	Parent	
Anette Lohmander, SLP, PhD	Stockholm, Sweden	Speech-Language Pathology	• Scandcleft
Ross E. Long, Jr., DMD, MS, PhD	Lancaster, PA, USA	Dentistry Orthodontics	• Americleft
Leanne Magee, PhD	Philadelphia, PA, USA	Psychiatry Behavioral Studies	
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Laura Monson, MD	Houston, TX, USA	Plastic Surgery Craniofacial Surgery	
Elizabeth Rose, MD	Melbourne, Australia	Otolaryngology	
Thomas Sitzman, MD, MPH	Cincinnati, OH, USA	Plastic Surgery	• Americleft Surgery Working Group • ACPA Data Standards Committee
Jesse Taylor, MD	Philadelphia, PA, USA	Plastic Surgery Craniofacial Surgery	
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John Wirthlin, DDS	Houston, TX	Dentistry Orthodontics	
Karen Wong, MD, MSc	Toronto, Ontario, Canada	Plastic Surgery	• CLEFT-Q project

* Core project leaders.
† NSQIP = National Surgical Quality Improvement Program; American Cleft Palate–Craniofacial Association; ICHOM = International Consortium for Health Outcomes Measurement.

Composition of Working Group

A core project team consisted of a clinical lead (J.G.M.), a research fellow (A.C.A.), and a project manager (T.A.K.). The core project team had both subject matter expertise and experience in knowledge engineering and group methods.

The working group was composed of 28 internationally recognized clinicians and academicians (Table 1). These members represented eight countries on four continents. Clinical disciplines represented included pediatrics, nursing, speech-language pathology, otolaryngology, dentistry, craniofacial orthodontics, oral and craniomaxillofacial surgery, and plastic surgery. Several of these experts were members of other pioneering cleft-related projects, including Americleft, Eurocleft, Scandcleft, and the CLEFT-Q. The working group also included one young adult with repaired unilateral cleft lip, alveolus, and palate (from the United Kingdom) and two parents of young children: one with isolated cleft palate (from the United Kingdom) and one with bilateral cleft lip, alveolus, and palate (from India). These patient and family representatives attended every teleconference. Their personal perspectives, as patient and caregivers, gave important context to all issues

being discussed and helped clarify which outcomes mattered most to patients and families.

External advisors provided additional expertise in the following disciplines: audiology; social work; team administration; public and private health-system management; epidemiology; public registries; medical informatics, data analytics, and data visualization; patient-centered outcomes research; outcomes measurement; performance improvement and patient safety; learning health care; and patient advocacy. These advisors did not attend each teleconference, but they assisted in informing development of the standard set whenever the working group required specific domain expertise.

Process

The core project team conducted a preliminary fact-finding mission in two phases. First, a representative review of major peer-reviewed and unpublished outcomes-assessments efforts (i.e., white and gray literature review) was conducted. Because of the volume of cleft literature, this review was representative of major efforts at outcomes assessment but was not systematic and inclusive of all outcomes articles. Based on this review, the core project team cataloged the main focuses for outcomes assessment and the methods used by each project. Second, structured interviews were conducted of experts from several respected cleft teams, hospitals, and government agencies. Nominal group technique and thematic content analysis were used to identify, organize, and understand the outcomes-assessment needs from the differing perspectives of each stakeholder. The results of this fact-finding mission were reported at the 12th International Congress of Cleft Lip & Palate and Craniofacial Anomalies (Allori, 2013).

Major influences for understanding the clinical perspective included prior cleft-related research and quality-improvement groups, principally Eurocleft, CSAG, Americleft, Scandcleft, Great Ormond Street Hospital, the ACPA Data Standards Committee, and the American College of Surgeons National Surgical Quality Improvement Program (**NSQIP**). An initial understanding of patient concerns and priorities was obtained through literature review and discussion with experts who are presently working on the development of patient-reported outcome measures for patients with CL/P. Most influential of these inputs were the CDC conference on unmet research needs (Yazdy et al., 2007), the James Lind Alliance's priority-setting partnership with patients with CL/P (Petit-Zeman and Cowan, 2013), the CLEFT-Q project (Klassen et al., 2012; Wong, 2012; Wong et al., 2013, 2014), the Children's Oral Health Impact Profile (**COHIP**) project (Broder and Wilson-Genderson, 2007; Broder et al., 2012; Broder, 2014), and others (e.g., Eckstein et al., 2011; Gosain and Chim, 2011; Sell et al., 2012; Ranganathan et al., 2015). Additional influences included established partnerships for continuous quality improvement, such as the NSQIP (Bruny et al., 2013; Paine, Paliga, et al., 2016; Paine, Tahiri, et al., 2016). These data helped to preliminarily define the scope and depth required for our standard set. Moreover, these data served as a starting point for our working group discussions and helped organize the sequence of teleconferences.

Next, between May 2013 and December 2014, the working group participated in seven large-group teleconferences. A modified Delphi process was used to reach consensus for all major decision areas, including the scope of the population to be covered, the "minimum required" outcome set (what outcomes the working group felt that all cleft teams should be measuring), and other variables required for case-mix adjustment. In planning each teleconference, preparatory discussions were held with key representatives of each clinical and academic discipline relevant to the outcome domains being considered. The core project team developed an agenda, listed key proposals, and summarized relevant evidence from the literature. Working group members were given time to review these documents in advance of each teleconference. Conferences were held for 2 hours each, beginning at 08:30 Eastern Standard Time (14:30 Greenwich Mean Time) to better accommodate the wide array of global time zones from our working group participants. Each conference was consistently attended by more than three-quarters of the working group members.

The teleconferences followed a structured sequence:

- Teleconference 1: Working Group process launched and scope of work defined.
- Teleconference 2: Identification of the outcome domains to include in the set.
- Teleconferences 3 and 4: Definition of the outcome domains.
- Teleconference 5: Identification and definition of the perioperative events.
- Teleconference 6: Identification and definition of case-mix adjustment variables.
- Teleconference 7: Review and finalization of the draft standard set.

Outcomes considered during each teleconference included major long-term outcomes, short-term outcomes, and perioperative events. *Outcomes* were specifically defined as attainment or lack of attainment of the specified goal of treatment. The working group selected outcomes based on the following four criteria: (1) the frequency of the outcome, (2) its impact on the patient, (3) the potential to modify the outcome, and (4) the feasibility of capturing the outcome in clinical practice. Whenever possible, complementary clinician-reported and patient-reported outcomes were identified. In addition to defining outcomes, corresponding time

points for data collection were selected for each outcome. Case-mix-adjustment variables were selected based on the following four criteria: (1) the potential relevance (strength of the causal linkage between the risk factor and the outcome), (2) the risk factor independency, and (3) feasibility of measurement.

Postconference surveys were prepared and administered to working group members using Qualtrics (Provo, UT). We consistently had >90% response rate on all questionnaires. Following each survey, the core project team circulated detailed minutes from the teleconference and a summary of the results from the postconference survey. Decisions were finalized when more than three-quarters of the working group members concurred; in cases where consensus was not reached, individual discussions were held with specialty leaders and stakeholders, and the advisory board was consulted as needed. The final standard set was approved unanimously by members of the working group.

Results

The Standard Set

The following is a summary of the ICHOM standard set for the comprehensive appraisal of cleft care. A detailed reference guide may be downloaded directly from ICHOM at www.ichom.org/medical-conditions/cleft-lip-palate. Because the standard set is intended to be iteratively improved over time, please be sure to consult the official data dictionary that will be maintained at this site.

Conceptual Model

Conceptually, the standard set is a multitiered framework that establishes the following:

1. An agreed-on method of classifying, describing, and grouping patients with CL/P;
2. Clear definitions of each outcome and other variables;
3. Specifications of the methods by which these outcomes and variables are to be measured;
4. Data standards describing how these data are to be recorded; and
5. Recommended time points and protocols for data collection.

The standard set is designed to be extensible. Use of the standard set does not preclude any team from collecting and reporting additional measures according to its particular interests and abilities. Of course, any such additional measures should extend, not replace, the methods prescribed by the standard set.

TABLE 2 Phenotypic Categories*

	Definition	
Category	*Anatomy Clefted*	*Anatomy Intact*
CL	Cleft lip	Intact alveolus, primary palate, and secondary palate
CL+A	Cleft lip, alveolus, and possibly primary (preforaminal) palate	Intact secondary (postforaminal) hard and soft palate
CP	Cleft secondary (postforaminal) hard and/or soft palate	Intact lip, alveolus, and primary (preforaminal) palate
CL+P	Cleft lip and secondary palate	The state of the alveolus is not specified in this category

* Note additional aggregate groupings: CL±A = cleft lip with/without cleft alveolus and primary palate but intact secondary palate, CL±P = cleft lip with/without cleft palate, CL/P = cleft lip and/or cleft palate. Note that different methods of notation or coding may be maintained by each team according to their conventions as long as these categories remain discernable.

Classification and Nomenclature

Outcome measures were designed to be relevant for all patients diagnosed with isolated and nonisolated CL/P. Specifically, this includes the following phenotypic groups, which are denoted according to the conventions of *Cleft Palate–Craniofacial Journal*: cleft lip, cleft lip with cleft alveolus, cleft palate (**CP**), and cleft lip with cleft palate (Table 2; Allori, Mulliken, et al., in press). Severity, laterality, and morphology of the lip, alveolus, and palate are also specified (Table 3). Teams may continue to use the particular methods of documentation (e.g., striped-Y diagrams, LAHSHAL notation, CLAP notation, etc.) and/or coding (ICD-9- and ICD-10-based systems, SNOMED-CT, etc.) that are their custom, as long as the aforementioned phenotypic groups and descriptors are easily discernible (Allori, Cragan, et al., in press; Allori, Mulliken, et al., in press). The major phenotypic groupings and phenotypic descriptors chosen for the standard set are clinically meaningful and will enable future subgroup analyses and outcome comparisons.

Treatment Approaches

In construction of the standard set, the working group considered all operative and nonoperative interventions that comprise contemporary practice for the management of CL/P. These included the following:

- Presurgical (dentofacial) orthopedics
- Labial (nasolabial) adhesion
- Premaxillary setback
- Gingivoperiosteoplasty
- Primary labial (nasolabial) repair
- Primary palatoplasty
- Pharyngoplasty, sphincteroplasty, and veloplasty for correction of velopharyngeal insufficiency
- Alveolar bone grafting
- Secondary labial revisions

TABLE 3 Phenotypic Description

Parameter	*Descriptor*	*Definition**
Lip		
Severity	*Complete*	Cleft violating vermilion, cutaneous lip, and nasal floor. A cutaneous bridge ("Simonart's band") is a complete defect.
	Incomplete	Cleft violating vermilion and partial cutaneous lip. The nasal base is typically widened, but the nasal floor is not clefted.
	Lesser-form	Cleft limited predominantly to the vermilion and white roll. Optionally, may be more precisely classified as *minor-form*, *microform*, and *mini-microform*.
	Asymmetric	For bilateral cleft lip, asymmetric severity of the right and left sides. The severity of each side may be specified (e.g., complete with incomplete, complete with lesser-form, etc.).
Laterality	*Unilateral*	Cleft on either *right* or *left* of the premaxilla, caused by failure of fusion of the ipsilateral maxillary prominence with the premaxillary segment during embryogenesis.
	Bilateral	Cleft on both sides of the prolabium due to failure of fusion of both right and left maxillary prominences with the premaxillary segment.
	Median	Midline prolabial cleft resulting from incomplete fusion of the medial nasal prominences to form the premaxillary segment.
Alveolus		
Severity	*Complete*	Cleft violating the entirety of the alveolar process through maxilla and into pyriform aperture. In unilateral cases, this creates a *greater* and *lesser* alveolar segment. In bilateral cases, this creates a "horseshoe" defect surrounding the premaxilla.
	Incomplete	Cleft violating more than one-third of the vertical height of the alveolar arch but with preservation of the maxilla and pyriform aperture.
	Notched	Cleft violating less than one-third of the vertical height of the alveolar process; alternatively, a cleft in which the vertical height is preserved, but the anteroposterior thickness of the arch is reduced.
Laterality	*Unilateral*	Cleft of either *right* or *left* alveolar processes.
	Bilateral	Cleft of both right and left alveolar processes.
	Median	Midline premaxillary cleft.
Palate		
Morphology	*Submucous*	Intact oral and nasal mucosa with a submucous cleft violating hard palate and/or velar musculature. Optionally, may be more precisely described as *overt* or *occult*.
	Veau-I	Midline cleft of the soft palate; the posterior hard palate may be notched but otherwise is intact.
	Veau-II	Midline cleft of the secondary (postforaminal) hard and soft palate. Optionally, may be further characterized as *complete* (cleft extending to the incisive foramen) or *incomplete* (cleft terminating posterior to the incisive foramen).
	Veau-III	Unilateral cleft extending through secondary soft and hard palate, through right or left primary palate, and into the alveolar process. The vomer is attached to the greater segment. Veau-III CP typically accompanies an ipsilateral CL.
	Veau-IV	Bilateral cleft extending through secondary soft and hard palate, through bilateral primary palate, and through bilateral alveolar processes. The vomer is attached to the premaxillary segment but not the premaxillary shelves. Veau-IV CP typically accompanies bilateral CL.

* CP = cleft secondary (postforaminal) hard and/or soft palate; CL = cleft lip.

- Secondary nasal revisions
- Secondary palatal revisions, including repair of oronasal fistulae
- Dental and orthodontic management
- Orthognathic correction of maxillary constriction and/or retrusion
- Functional and aesthetic rhinoplasty

Where relevant, specific techniques for each intervention were also explored; however, the ultimate standard set is agnostic to specific treatment modality. Outcome domains assessed by the standard set are universally applicable and relevant to all patients with CL/P, irrespective of operative technique or treatment protocol.

Time Points

Three predominant factors contributed to the selection of time points for the standardized data collection: (1) typical treatment periods, (2) stages of growth and development, and (3) potential burden of data collection on a team (Table 4).

Although treatments may be administered at any time (based on the unique needs of the patient or according to particular team-based practices), they may be clustered into three general periods of treatment: primary repair in infancy (e.g., labial and palatal repair); secondary, or intermediate, treatment in early childhood and adolescence (e.g., pharyngoplasty, alveolar bone grafting, and orthodontics); and final treatment in the latter years (orthognathic surgery and rhinoplasty). Members of the working group felt the final treatment period in young

TABLE 4 Timeline for Data Collection

Time Point	*Target Age*	*Defined Age Window*
t_0	Baseline	First encounter
t_{3m}	3 Months	2.5–3.5 months
t_5	5 Years	4–6 years
t_8	8 Years	8–9 years
t_{12}	12 Years	10–12.5 years
t_F	Final (young adult at end of treatment)	End of treatment or 22 years of age, whichever is soonest.

TABLE 5 Outcome Domains

Outcome Domain	*Included Outcomes*	*Instruments Used**	*Data Source**	*Time Points*
Eating and drinking	Body weight	Growth chart	Clinician (P/N)	t_0, t_{3m}
	Change in weight centile	Growth chart	Clinician (P/N)	t_{3m}
	Eating and drinking	CLEFT-Q Eating-and-Drinking subscale	Patient	t_8, t_{12}, t_F
Dental and oral health	Dental health	dmft and DMFT scores	Clinician (D)	t_5, t_{12}
	Oral health	COHIP Oral Symptoms subscale†	Patient	t_8, t_{12}
	Occlusion	Overjet assessment	Clinician (D)	t_5, t_{12}, t_F
		Lateral Cephalogram		t_F
	Mastication	CLEFT-Q Eating-and-Drinking subscale†	Patient	t_8, t_{12}, t_F
Speech/Communication	Intelligibility	Intelligibility-in-Context scale†	Family	t_5, t_{12}
	Articulation	Percent Consonants Correct scale†	Clinician (SLP)	t_5, t_{12}, t_F
	Velopharyngeal competence	VPC graded rating scale†	Clinician (SLP)	t_5, t_{12}, t_F
	Overall speech	CLEFT-Q Speech and Speaking subscales†	Patient	t_{12}, t_F
	Documentation	Standardized speech and language sample	n/a	t_5, t_{12}, t_F
Otologic health	Hearing	Puretone average	Clinician (A)	t_5, t_{12}
	Otologic health	Otologic health screening questions	Clinician (A/O) Family	t_5, t_{12}
Breathing	Nasal breathing	NOSE questionnaire†	Patient	t_8, t_{12}
Appearance	Nasolabial appearance	CLEFT-Q Face subscale†	Patient	t_8, t_{12}, t_F
	Facial profile	CLEFT-Q Jaw subscale†	Patient	t_{12}, t_F
	Smile	CLEFT-Q Dental subscale†	Patient	t_8, t_{12}, t_F
	Documentation	Standardized series of facial photographs	n/a	t_5, t_{12}, t_F
Psychosocial development	Sociometrics	CLEFT-Q Social Life subscale†	Patient	t_8, t_F
		CLEFT-Q School Life subscale†	Patient	t_{12}
	Psychometrics	CLEFT-Q Feelings subscale†	Patient	t_{12}
Burden of care	Total number of interventions requiring anesthesia	Medical record	Admin (T)	t_F

* dmft = ;DMFT = ; COHIP = Children's Oral Health Impact Profile; VPC = ; NOSE = Nasal Obstruction Symptom Evaluation; n/a = ; A = audiologist; D = dentist/orthodontist; N = nurse; O = otolaryngologist; P = pediatrician; S = surgeon; SLP = speech/language pathologist; T = team coordinator, social work, and administrative support personnel.
† Validated instruments.

adulthood to be of particular interest and importance because most prior research efforts have focused on outcomes in childhood (Stock, Feragen, et al., 2015). The outcomes-assessment framework should consider the specific goals of each of these treatment periods.

In addition, during the course of 2 decades of care, the child is growing physically, cognitively, and psychosocially. Therefore, it is critically important that the outcomes-assessment framework also consider the psychosocial impact of CL/P during infancy, early childhood, preschool, preadolescence, adolescence, and young adulthood.

Finally, the burden of data collection on a team must also be considered. In designing the standard set, our working group identified critical major time points that reflected treatment periods, stages of development, and team practices. Each time point was designated a corresponding window of acceptable ages to respect existing scheduling practices and other clinical constraints. Table 4 summarizes these major time points. Briefly, they include data collection at baseline (first encounter), ~3 months (t_{3m}), ~5 years (t_5), ~8 years (t_8), ~12 years (t_{12}), and a final point (t_F, at the end of treatment or 22 years of age, whichever comes first).† Additional postoperative adverse events are captured within 30 days of intervention.

Data Sources

Data obtained at each time point is reported by clinicians, administrative personnel, family members, and patients. (Patient-reported outcome measures are only obtained for patients 8 years of age or older.) We attempted to establish a balance of complementary clinician-reported and patient-reported outcomes wherever possible.

Major Outcome Domains

Eight major outcome domains and 22 subdomains were chosen (Table 5). These domains include eating and drinking, dental and oral health, speech, otologic health, breathing, appearance, emotional and psychosocial development, and aspects related to process of care or burden of treatment. To the best degree possible, all outcome domains and subdomains were structured according to the WHO International Classification of Function, Disability, and Health (e.g., function, activity, participation; WHO, 2001).

Eating and Drinking

Body weight was considered an important measurable outcome that reflected the nutritional status and well-

† The major time points chosen reflect the desire of the working group to respect existing team protocols and workflows while minimizing the burden of data collection. Individual teams may choose to collect data more frequently if desired and if time and resources allow as long as they also adhere to the major time points established by the standard set.

being of the infant as well as the effectiveness of a team's program for feeding education and support. The working group selected clinician-reported body weight (kg) at the t_{3m} time point (~3 months of age) as well as change in weight percentile since birth. For the older child, eating and drinking may also have a psychological and sociological impact. Therefore, our group elected to collect patient-reported outcome measures at time points t_8, t_{12}, and t_F using the CLEFT-Q Eating-and-Drinking subscale (Wong et al., 2014).

Dental and Oral Health

The working group recommended measures reflecting dental health, oral (periodontal) health, occlusion, and mastication. The **DMFT** index was chosen as a clinician-reported summary of dental decay (caries), missing teeth, and filled teeth for both deciduous and permanent dentition (**dmft** and DMFT, respectively; Anaise, 1984). The COHIP Oral Symptoms subscale was chosen as a patient-reported reflection of periodontal health, including gingivitis (Broder, 2014). Building off the experience of Eurocleft, Americleft, and others, occlusion is assessed at time points t_5, t_{12}, and t_F. We elected to use a clinical assessment of overjet based on the GOSLON scale because it was determined that this would be most practical for a minimum dataset.[‡] Mastication is the functional corollary to occlusion and is assessed in part of the patient-reported CLEFT-Q Eating-and-Drinking scale at time points t_8, t_{12}, and t_F. Note that the early time point (t_8) differs from that for clinical assessment of occlusion (t_5). Working group members considered it important to assess clinical occlusion at about 5 years of age, but patient-reported outcome measures are not employable until 8 years of age or older. At the other two time points, t_{12} and t_F, both clinician-reported and patient-reported outcomes are obtained synchronously.

Speech and Communication

The working group identified intelligibility, articulation, and velopharyngeal competence as specific aspects for consideration. Intelligibility is measured at t_5 and t_{12} using the family-reported Intelligibility-in-Context Scale (McLeod, 2012). Articulatory proficiency is assessed via the clinician-reported Percent Consonants Correct (Shriberg and Kwiatkowski, 1982).[§] Because languages differ in the occurrence and frequency of different phonemes and in the phonetic characteristics of consonants, the potential impact a cleft palate may have on speech varies between languages (Hutters and Henningsson, 2004). A restricted number of phonetically similar speech sounds enhances the validity of speech outcomes reported from different languages if the assessment is made primarily on speech sounds that are highly vulnerable to the cleft condition, that is, pressure consonants (Henningsson et al., 2008). Speech material designed in this way will be short but sufficient for reporting speech outcome following surgical treatment (Lohmander et al., 2009). Embedding the target sound in single-word contexts seems to enhance the reliability between raters (Klintö et al., 2011). With regard to velopharyngeal closure, several alternative methods of assessment were considered; these included both perceptual rating scales as well as diagnostic evaluation, such as videofluoroscopy, nasoendoscopy, and nasometry. Although each of these methods had its advantages and disadvantages, overall most were deemed impractical for sustainable implementation by teams because of logistical complexity. For these reasons, a simple but reliable clinician-reported three-tier rating scale was chosen as described by Lohmander et al. (2009): 0 = competent (no velopharyngeal dysfunction), −1 = marginally competent (evidence of minor problems suggesting borderline closure), −2 = incompetent (evidence of significant problems, usually requiring surgical management). To obtain the patient-reported perspective regarding speech, the working group selected the CLEFT-Q Speech subscale, as well as the Speaking subscale. Although the former subscale targets patient perception related to the mechanics of speaking, the latter subscale focuses more on the psychosocial effects of the speaking process. Of note, given the international and multicultural audience for the standard set, our working group decided it was not practicable for the standard set to recommend specific methods for the appraisal language. However, a recording of a standardized speech/language sample is

[‡] It is noteworthy that there was much discussion within our working group regarding the optimal method for the assessment of occlusion. Several experts argued that blinded ratings of dental models was the gold standard, others argued that grading dental photographs was more practical and just as valid, and still others felt that bedside clinical grading at the time of the clinical encounter was appropriate and sufficient. Ultimately, the working group accepted a simple measure of the clinical grading of overjet performed during the clinical encounter because of the convenience for assessment, data collection, and data storage. The group recognizes that there is limited evidence for validity and reliability of this method, but in this case the working group prioritized simplicity, implementability, and sustainability. This is an example of one outcome measure that is likely to be iteratively improved in the future.

[§] The measure of percentage consonants correct was originally developed by Shriberg and Kwiatkowski (1982). They measured the proportion of correctly articulated consonants in phonetic transcriptions of conversational speech to assess the severity of involvement. Single-word material may be used provided that results are not to be related to severity of involvement (Shriberg et al., 1997). Calculating the percentage of consonants correct in single word samples has been used to assess articulation skills in children born with cleft palate (Lohmander and Persson, 2008; Scherer et al., 2008; Klintö et al., 2014).

recommended (but not yet required) to enable independent review for research or intercenter comparisons. Project leaders in each country may consult the references listed previously and the CLISPI instructions for the creation of representative, language-specific word banks and language samples.

Otologic Health

The working group recommended measured pure-tone average to be recorded at t_5 and t_{12}. Although no validated instruments exist for summarizing audiologic function or otologic health, five health screening questions were constructed for completion by both clinicians and family members. These screening questions relate to the use of hearing aids, frequency and chronicity of otitis media, use of tympanostomy tubes, and development of complications such as cholesteatoma, ossicular chain disruption, and mastoiditis.

Breathing

An underappreciated component of CL/P is the functional aspect of the cleft lip nasal deformity and palatal deformity that leads to airway obstruction. In the standard set, nasal breathing is appraised by way of the Nasal Obstruction Symptom Evaluation (**NOSE**) scale (Stewart et al., 2004). This six-question scale was originally validated in adult patients undergoing septoplasty as a preoperative screen for nasal obstruction and as a postoperative measure of degree of symptom improvement. Although not specifically validated in the cleft population, the NOSE scale has been employed by several cleft teams to assess the effectiveness of cleft rhinoplasty (Marcus et al., 2015). Thus, in the absence of any superior pediatric-specific or cleft-specific instrument, the working group agreed to include the NOSE scale as an essential component of the standard set. To the contrary, in the case of obstructive sleep apnea (**OSA**), no appropriate screening or diagnostic instrument could be identified. Many OSA screening tools and semiquantitative rating tools exist, but these were felt to be too generic or too focused on typical adult OSA. That is, critical aspects relating to pediatric and cleft populations were not adequately considered. It is the desire of the working group that a cleft-specific OSA grading scale should be developed and validated.

Appearance

The aesthetic result is by its very nature subjective. Methods for the semiquantitative grading of aesthetic results have been proposed. Some successful and reliable methods rely on averaged ratings by a review panel (Asher-McDade et al., 1991; Tobiasen et al., 1991; Mercado et al., 2011), whereas others advocate the use of computational analysis of symmetry and form (Bearn et al., 2002a and 2002b; Fisher et al., 2008; Pigott and Pigott, 2010). The working group agreed that although each of these methods was worthy of further consideration, the burden that performing such analyses would impose on teams would be considerable. Thus, at this time, no clinician-reported rating of aesthetic result is presently required by the standard set. Fortunately, several options existed for the appraisal of aesthetic result from a patient-reported perspective. The working group included the CLEFT-Q Face subscale, Jaw subscale, and Dental (Smile) subscale as required parts of the standard set. Teams may also choose to administer the CLEFT-Q Lip subscale and Nose subscale if very specific information regarding appearance of the lip and nose is desired, although these two highly specific subscales are not required by the standard set. Future development of a complementary clinical scale for nasolabial aesthetics was identified by our working group as a top priority. Such a scale is presently being developed by the Americleft Surgery Working Group, which includes two members from our CL/P standard-set working group (T.J.S. and A.C.A.). It is our intention to propose inclusion of the Americleft nasolabial aesthetic rating scale in the ICHOM standard set once its development is complete and after it has been validated through pilot tests. To become a required part of the standard set, the new instrument must be supported by the ICHOM CL/P steering committee and tested through the implementation community (see the following sections).

Psychosocial Outcomes

The need for psychometric and sociometric outcomes was made readily apparent from precedent research as well as by the patient and family representatives in our working group (Semb et al., 2005; Yazdy et al., 2007; Porter, 2010; Eckstein et al., 2011; Gosain and Chim, 2011; Allori et al., 2012; Klassen et al., 2012; Sell et al., 2012; Petit-Zeman and Cowan, 2013; Wong et al., 2013; Broder, 2014; Sitzman et al., 2014; Stock and Rumsey, 2015; Stock et al., 2015, 2016, in press). Sociologic concerns begin with the neonatal period and specifically with parent-child bonding and the doctor-family relationship (Stock and Rumsey, 2015). The working group had initially considered the Measure of Processes of Care-20 instrument as a measure of these issues. However, the 20 questions required by this instrument were thought to be too generic and too onerous for sustainable implementation. Consequently, although we do feel that sociologic aspects related to parent-child bonding and the doctor-family relationship are important issues, no appropriate and practical measure was identified for inclusion in the standard set. In later life, psychological and sociological issues abound. The

working group selected the CLEFT-Q Social Life subscale for time points t_8 and t_F; at t_{12} (middle school years), the more specific CLEFT-Q School Life subscale is employed. These are paired with the CLEFT-Q Feelings subscale.

Postoperative Events (Safety Data and Process Measures)

In addition to the major outcomes described previously, the working group also identified postoperative events that could have significant impact on patients' lives. These adverse events include bleeding, infection, wound-healing problems, respiratory distress, and death (from any cause). In addition, the working group voted to capture several postoperative process measures, including the number of hospitalized days following a procedure and readmission (for any cause) within 30 days of discharge following an operation.

Risk Adjustment and Case-Mix Adjustment

The secondary objective of the working group was to identify a set of candidate variables for risk and case-mix adjustment based on the potential impact of these variables on the outcomes in the standard set. These variables include the following: gender, race and ethnicity, language spoken at home, level of parental education, insurance status, geographic residence, phenotypic classification and severity, genetic diagnoses, comorbidities, age at first encounter with the team, loss to follow-up, transferal of care into or away from a team, adoption status, and distance from treatment team.

To our knowledge, no risk-adjustment or case-mix-adjustment models exist for CL/P. Development of risk-adjustment and case-mix-adjustment models is planned as an ongoing part of this work.

Special Populations

The standard set is appropriate for the measurement of outcomes in all patients with CL/P, whether isolated or nonisolated, syndromic or nonsyndromic (Watkins et al., 2014; Aylsworth et al., 2015); however, the standard set does not encompass the noncleft issues that may be of great importance for such conditions. In addition, the cleft-related outcomes in these groups may be influenced (positively or negatively) by other noncleft issues. For that reason, nonisolated cases, syndromic cases, and other special populations are flagged so that they can be excluded from aggregated data analysis. Subgroup analysis for these special populations is still possible, but exclusion from aggregated statistics ensures that these data will not distort the data for typical CL/P cases. (For example, a child with CP as part of Robin sequence could still participate in data collection, and those data would help a team understand their cleft-related outcomes for patients with Robin sequence; but this child's data would not be included in aggregated data analysis for the generic "CP" cohort.)

The standard set specifies the following flags for exclusion from aggregated analysis:

- Median cleft lip (premaxillary cleft)
- Atypical (Tessier) facial clefts
- Robin sequence
- 22q11 deletion
- CL/P associated with other craniofacial anomalies
- CL/P associated with other congenital anomalies
- CL/P associated with significant comorbidities (see reference guide)
- Late entry or transferral of care to the team from another institution
- Incomplete care or transferral of care from the team to another institution

Discussion

The purpose of our working group was to answer the following questions: What outcomes could the average cleft team track at each stage of treatment that would reflect what matters most to their patients? How could those data be collected and evaluated? The ICHOM standard set of outcome measures for cleft care is the fruit of many months of intense preparation, discussion, collaboration, and testing among multidisciplinary clinicians, academicians, administrators, and patients from around the world.

The resulting set has many strengths. (1) The standard set is holistic and meaningful: It focuses on "true" outcome measures that reflect the quality of care and value to many stakeholders, allowing for continuous quality improvement and "learning health care." (2) The standard set is patient-centric: Because the standard set was constructed with input from patients and family members and because it employs patient-reported outcome measures alongside complementary clinical measures, the standard set is inherently patient-centered. (3) The standard set is robust: It has been constructed according to accepted methods of ontology development, uses mixed methods, and includes validated measures with multilingual translations. (4) The standard set is practical: Outcome measures have been carefully selected to be comprehensive yet manageable for all teams to sustainably implement. It also considers teams in resource-rich and resource-poor environments. (5) The standard set is universal: Because centers using the standard set are collecting the same outcomes measured and recorded in the same way, data become interchangeable. This allows for intercenter comparisons, multisite research, and pragmatic research.

All projects of this magnitude have limitations. First, although comprehensive, the standard set is not exhaus-

tive—it is intentionally designed as a minimum standard set of outcome measures that we propose all teams collect. Some teams will desire to do more, whether for research or quality-improvement purposes, and this is laudable. Any extensions will be complementary to—not competitive with—the standard set.

Second, although our working group tried its best to compile a thorough and clear data dictionary with specific definitions and data standards, inevitably some items will be identified as warranting revision. Such issues may be identified during the early phases of implementation. A project steering committee has been established to periodically reappraise the set and publish interval updates to the reference guide.

Despite such real-world concerns, we encourage all teams to consider adopting this standard set to measure outcomes within their teams. We are optimistic that implementation is feasible and sustainable, and we are also confident that the outcome data will be of great interest to clinicians, patients/families, payers, and policy makers.

Next Steps: Dissemination and Implementation

The standard set is open source and may be freely implemented by any interested team. Each team is urged to adhere to the definitions and protocols described in the official reference guide. Implementation may occur by way of paper forms, local databases, custom-designed screens in electronic health records, and so on. A pilot implementation is presently underway in several centers. A detailed description of this implementation trial is beyond the scope of the present manuscript, but in this field test, we will appraise implementation according to RE-AIM, PRECIS, and quality-assurance models (Glasgow et al., 1999; Bakken et al., 2009; Gaglio et al., 2014). This trial includes testing of a public registry that may one day serve as a secure central repository for CL/P data from participating centers.

To encourage and support broader intercenter collaboration, ICHOM has established an implementation community that is a voluntary network of international teams that have implemented (or intend to implement) the standard set. Each team is led by a local site director who coordinates and oversees local activities. Although each team must obtain its own institutional approval and support for the implementation, ICHOM is able to provide guidance and assistance with the process (e.g., templates for institutional review board protocols). Regular web-based teleconferences connect each team with peers around the world so that they can share experiences, discuss challenges, and develop strategies for sustainable implementation.

For institutions needing substantial development of information technology systems, the ICHOM website also lists certified providers that may be hired on a contractual basis. These companies have experience in implementing standard sets but are not otherwise affiliated with ICHOM or with the CL/P working group.

Feasibility, Acceptability, and Validation

The standard set was assembled with previously validated instruments; however, it is important to conduct and report the reliability and acceptability of these outcome measures used in concert. Because the data-measurement burden can be high for some teams, it is also important to review feasibility in different team contexts (e.g., resource-rich and resource-poor environments). A critical evaluation of the ICHOM standard set in comparison with other relevant initiatives is planned to enable potential adopters to more fully evaluate their options.

Iterative Improvement

The standard set is intended to establish a sound foundation for a "learning health-care system" for continuous quality improvement in cleft care (Institute of Medicine, 2013; National Research Council, 2013; Abernethy, 2014). As such, the standard set itself will be subject to periodic internal and external review under direction of the project steering committee. Iterative improvements to the standard set may become necessary to clarify definitions, to respond to advances in methods of outcomes assessment, and to reflect changes in capacity for data collection. Three examples of possible modifications expected in upcoming years are (1) shorten many of the patient-reported outcome scales as the CLEFT-Q project completes its broader phase of field testing, (2) inclusion of a clinical nasolabial aesthetic rating scale, and (3) refinement of the clinical measure for occlusion. Each of these proposals will be evaluated by the steering committee and evaluated at sites in the implementation community prior to official endorsement and incorporation into the standard set.

All official changes will be published on the website accompanied by a changelog that details the revision history. Notification of updates will be distributed throughout the implementation community by e-mail and by way of social media.

Great effort has been extended in future-proofing the standard set such that improvements may be confidently made, whenever necessary, while maximizing compatibility of the data collected before and after these modifications.

Conclusion

There would be numerous advantages to having a documented set of agreed data fields and data types available for many areas of cleft and craniofacial care .... [T]he included information could be drawn upon ... by all the ACPA members for their own clinical, quality

improvement, research and/or audit efforts. ACPA Data Standards Committee (Fitzsimons, 2011)

Through the efforts reported in this paper, we have defined a relatively simple, easily implemented set of outcomes that we believe can, and should, be measured by the typical cleft team. This is a first step in an effort to drive what we hope are meaningful and significant improvements in the care of children with CL/P.

If all cleft teams begin measuring the same outcomes in the same way, we—as a global cleft-care community—can learn from each other's outcomes and ultimately improve the quality and value to patients. It is the sincerest hope of our working group that the standard set presented herein may assist many cleft teams worldwide in reaching this goal. Teams interested in learning more about the standard set are encouraged to download the full reference manual at www.ichom.org/medical-conditions/cleft-lip-palate.

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The International Consortium for Health Outcomes Measurement (ICHOM) is a nonprofit organization founded by the Institute for Strategy and Competitiveness at Harvard Business School, The Boston Consulting Group, and the Karolinska Institutet. ICHOM believes that healthcare systems worldwide can be transformed by measuring and reporting outcomes in a standardized way. ICHOM organizes global teams of clinician leaders, outcomes and health services researchers, and patient advocates to define standard sets of outcomes for each condition. ICHOM also drives adoption and facilitates implementation across institutions globally, enabling health care providers to compare, learn, and improve. The goals of these efforts include improving healthcare quality, reducing healthcare costs, and supporting informed decision making. Readers interested in learning more about ICHOM may visit www.ichom.org. Information specific to the Standard Set for CL/P is accessible at www.ichom.org/medical-conditions/cleft-lip-palate.

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