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# Optimizing the Psychosocial Function Measures in the International Consortium for Health Outcomes Measurement Standard Set for Cleft

Inge Apon, MD, MHS<sup>1</sup>Nikki van Leeuwen, PhD<sup>2</sup>Maarten J. Koudstaal, MD,  
DMD, PhD<sup>1</sup>Alexander C. Allori, MD, MPH<sup>3</sup>Carolyn R. Rogers-Vizena, MD<sup>4</sup>Eppo B. Wolvius, MD, DMD,  
PhD<sup>1</sup>Anne F. Klassen, DPhil<sup>5</sup>Sarah L. Versnel, MD, PhD<sup>6</sup>Rotterdam, The Netherlands;  
Durham, NC; Boston, MA; and  
Hamilton, Ontario, CanadaPatient-reported  
Health

**Background:** To ensure the feasibility of implementing PROMs in clinical practice, they must be continually appraised for undue burden placed on patients and clinicians and their usefulness for decision-making. This study assesses correlations between the CLEFT-Q psychosocial scales in the International Consortium for Health Outcomes Measurement Standard Set for cleft and explores their associations with patient characteristics and psychosocial care referral.

**Methods:** Spearman correlation coefficients were calculated for CLEFT-Q psychological function, social function, school function, face, speech function, and speech-related distress scales. Logistic regressions were used to assess the association of cleft phenotype, syndrome, sex, and adoption status on scale scores and clinical referral to psychosocial care for further evaluation and management.

**Results:** Data were obtained from 3067 patients with cleft lip and/or palate at three centers. Strong correlations were observed between social function and psychological function ( $r > 0.69$ ) and school function ( $r > 0.78$ ) scales. Correlation between school function and psychological function scales was lower ( $r = 0.59$  to  $0.68$ ). Genetic syndrome (OR, 2.37; 95% CI, 1.04 to 5.41), psychological function (OR, 0.92; 95% CI, 0.88 to 0.97), school function (OR, 0.94; 95% CI, 0.90 to 0.98), and face (OR, 0.96; 95% CI, 0.94 to 0.98) were significant predictors for referral to psychosocial care.

**Conclusions:** Because social function as measured by the CLEFT-Q showed strong correlations with both school and psychological function, its additional value for measuring psychosocial function within the Standard Set is limited, and it is reasonable to consider removing this scale from the International Consortium for Health Outcomes Measurement Standard Set for cleft. (*Plast. Reconstr. Surg.* 151: 274e, 2023.)

The treatment of cleft lip and palate remains complex and differs between treatment centers, resulting in varying

quality of care worldwide.<sup>1-4</sup> Recognizing the need for uniform outcome measurement in cleft care, the International Consortium for Health Outcomes Measurement (ICHOM) published its Standard Set for the comprehensive appraisal of cleft lip and palate, based on

From the Departments of <sup>1</sup>Oral and Maxillofacial Surgery, <sup>2</sup>Public Health, and <sup>6</sup>Plastic and Reconstructive Surgery, Erasmus University Medical Center; <sup>3</sup>Department of Plastic, Maxillofacial and Oral Surgery, Duke University Hospital; <sup>4</sup>Department of Plastic and Oral Surgery, Boston Children's Hospital; and <sup>5</sup>Department of Pediatrics, McMaster University.

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consensus recommendations of a large, international, multidisciplinary working group.<sup>5-7</sup> The objective of the Standard Set was to provide a starting point for all cleft teams to measure the same outcome domains, using the same methods and instruments, at the same time points, and to record those data in the same structured format. The ultimate goal is to apply these outcomes toward improving patient-centered care.

The Standard Set includes not only traditional, clinician-reported outcomes and clinical variables, but also condition-specific parent- and patient-reported outcome measures (PROMs) in the domains of speech, facial appearance, and psychosocial function.<sup>8</sup> The rationale for including these scales is intuitive: facial appearance and speech are major outcomes domains in the treatment of cleft lip and/or palate, and poor outcomes in these domains may contribute to psychosocial distress. In fact, psychosocial impairment has been commonly reported in patients with cleft lip and/or palate, with main contributing factors including patients being teased or bullied, dissatisfaction with appearance, and dissatisfaction with speech.<sup>9-14</sup> Therefore, it is important for cleft teams to identify psychosocial problems early to provide timely and appropriate care within the team or, if indicated, by referral to a psychologist or psychiatrist for further evaluation and management. To this end, the ICHOM Standard Set includes three psychosocial CLEFT-Q scales: psychological function, social function, and school function.<sup>15-17</sup>

To ensure the feasibility of implementing these scales in clinical practice and sustaining that implementation over time, the ICHOM Standard Set itself must be continually appraised for undue burden placed on patients and clinicians. At the same time, it is important to confirm that scales included in the ICHOM Standard Set manifest useful information that can be used to inform clinical decision-making. The corollary to this statement is that scales deemed suboptimal, statistically invalid, redundant, or otherwise uninformative should be deimplemented. To see whether the psychosocial scales in the ICHOM Standard Set prove their worth or whether inclusion in the Standard Set should be reconsidered, this study assesses correlations between the psychosocial scales and explores their associations with patient characteristics and referrals to psychosocial care.

## PATIENTS AND METHODS

### Participants and Recruitment

Three cleft teams participated in this study (ie, Boston Children's Hospital, Duke Children's Hospital, and Erasmus University Medical Center). Patients with unilateral or bilateral cleft lip, cleft palate, cleft lip and palate, or cleft lip and alveolus, aged 8 to 22 years, were eligible for the measurement of CLEFT-Q psychosocial function scales. Data were prospectively collected according to the guidelines of the ICHOM Standard Set for Cleft Lip and Palate between November of 2015 and April of 2019. CLEFT-Q psychological and school function outcome data were collected at time 12 (age 10 to 13 years), and data for the CLEFT-Q social function scale were collected at time 8 (age 8 to 9 years) and time final (age 20 to 22 years), as recommended by the ICHOM Standard Set.<sup>18</sup> Because the authors felt that there is a large measurement gap between the ages of 12 and 22 years, two time points (age 14 to 16 years and age 17 to 19 years) were added for research purposes. These additional data were collected as part of the CLEFT-Q development and validation project organized by McMaster University. Information on this project, its recruitment, and its data-collection procedures have been described previously.<sup>15,16,19</sup> Institutional review board approval was obtained at every participating site (MEC-2016-156; IRB-P00030776; IRB-Pro00067808; REB Project no. 10-651).

### PROMs

The primary outcomes were the scores of the CLEFT-Q psychological function, social function, and school function scales. These scales focus on the following themes, respectively: "How do you feel?" "How is your social life?" and "How is your school life?" Each scale consists of 10 items, with four possible responses of "never," "sometimes," "often," and "always."

In addition, it was hypothesized that psychosocial function is influenced by facial difference or speech dysfunction. Therefore, the CLEFT-Q face, speech function, and speech-related distress scales, which are also prescribed by the ICHOM Standard Set, were also evaluated. The CLEFT-Q face scale focuses on the theme, "how much do you like how your face looks?" and includes nine items with possible responses "not at all," "a little bit," "quite a bit," and "very much." The CLEFT-Q speech function (12 items) and speech-related distress (10 items) scales assess, respectively, "how is your speech?" and "how do you feel

about speaking?” with possible responses “always,” “sometimes,” and “never.” For each scale, a raw score was transformed to a scale ranging from 0 to 100, where higher scores represent better functioning.

### Additional Variables

The ICHOM Standard Set also includes the collection of various patient characteristics (also known as case-mix variables or predictors), including sex, age (grouped according to Standard Set “time points”), cleft phenotypic group, the presence of a genetic syndrome, and whether or not a child has been adopted. For a subgroup analysis of the patients from The Netherlands, Dutch socioeconomic status scores of 2017 were added as an additional variable to explore its association with outcomes. These scores are based on postal codes, and higher scores represent higher socioeconomic status.<sup>20</sup> Also, information on referral status of the patient to any type of psychosocial care (psychiatrist, psychologist, social care) was gathered retrospectively from the patient’s medical files.

### Statistical Analysis

All analyses were performed using IBM SPSS Version 25.0 (IBM Corp.) For the correlational analyses, Spearman correlation coefficients ( $r$ ) were calculated for every relationship between the different PROM scores per time point, because the PROM scores were not normally distributed based on the histograms. A priori, it was defined that a coefficient above 0.7 was to illustrate a strong correlation, whereas between 0.4 and 0.7 was considered moderate, and coefficients below 0.4 were considered weak. Stronger correlations between scales would indicate similar constructs are measured. Univariable linear regression was performed to assess the influence of the time points on the psychosocial outcome scores.

Subgroup analysis was performed on the Dutch sample and included univariable linear regression to investigate the influence of patient characteristics on the psychosocial function scores, and logistic regression to explore the associations of patient characteristics and psychosocial scores with referral to psychosocial care. All analyses were performed based on complete cases. The two-tailed significance level was set at  $P < 0.05$ .

## RESULTS

The complete data set included 3067 patients who provided a total of 3103 assessments. The

majority of patients were diagnosed with cleft lip and palate [ $n = 1773$  (58%)], and 1714 patients (56%) were male patients. In 1080 cases (35%), the PROMs were completed around the age of 12 (Tables 1 and 2).

Strong correlations were found between psychological and social scales ( $r = 0.74$  to  $0.76$ ) at all measured time points, except at age 8 to 9 years ( $r = 0.69$ ). The correlations between social and school function scales were between 0.78 and 0.85. This correlation could not be computed for those aged 20 to 22 years because the school scale was not completed at this time point. The correlations between psychological and school function scales varied between 0.59 and 0.68. (See Figure, Supplemental Digital Content 1, which shows Spearman correlation plots for the psychosocial scales at age 10 to 13 years, <http://links.lww.com/PRS/F578>.) The face scale was moderately correlated ( $r = 0.37$

**Table 1. Patient Characteristics and Variables**

Characteristic	Complete Sample (%)	Subset (%)
No. of patients	3067	353
Sex		
Male	1714 (56)	200 (57)
Female	1353 (44)	153 (43)
Cleft type		
Cleft lip	301 (10)	34 (10)
Cleft palate	718 (23)	117 (33)
Cleft lip and palate	1773 (58)	172 (49)
Cleft lip and alveolus	275 (9)	30 (8)
Adoption		
No	—	292 (83)
Yes	—	61 (17)
Unknown	—	—
Genetic syndrome		
No	—	303 (86)
Yes	—	50 (14)
Socioeconomic status		
Mean	—	0.06
Range	—	-3.63 to 2.31

**Table 2. Patient Characteristics and Variables**

Characteristic	No. of Measurements	
	Complete Sample (%)	Subset (%)
No. of assessments	3103	365
Timing of PROMs		
8 yr ( $t_8$ )	735 (24)	134 (37)
12 yr ( $t_{12}$ )	1080 (35)	154 (42)
15 yr ( $t_{15}$ )	593 (19)	—
17 yr ( $t_{17}$ )	386 (12)	—
22 yr ( $t_{final}$ )	309 (10)	77 (21)
Psychosocial care referral		
No	—	330 (90)
Yes, after PROM scores	—	18 (5)
Yes, other reason than PROM scores	—	17 (5)

$t_8$ , age 8–9 years;  $t_{12}$ , age 10–13 years;  $t_{15}$ , age 14–16 years;  $t_{17}$ , age 17–19 years;  $t_{final}$ , age 20–22 years.

to 0.65) and the speech-related scales had low to moderate correlations ( $r = 0.14$  to  $0.53$ ) with all three psychosocial scales (Table 3). Similar findings were found for each of the four cleft phenotypic groups. (See Table, Supplemental Digital Content 2, which shows Spearman correlation coefficients. *N/C*, could not be computed, because of too few observations; *CL*, cleft lip; *CP*, cleft palate; *CLAP*, cleft lip and palate; *CLA*, cleft lip and alveolus. \*Statistically significant, <http://links.lww.com/PRS/F579>.)

Linear regression revealed a negative significant association between time points and outcome scores of the psychological function scale; a higher age group was associated with lower scores: age 10 to 13 years,  $\beta = -3.30$  (95% CI,  $-5.33$  to  $-1.56$ ); age 14 to 16 years,  $\beta = -6.70$  (95% CI,  $-8.99$  to  $-4.41$ ); age 17 to 19 years,  $\beta = -8.87$  (95% CI,  $-11.44$  to  $-6.29$ ); and age 20 to 22 years,  $\beta = -8.71$  (95% CI,  $-11.70$  to  $-5.72$ ). No significant associations between time points and the social and school scales were found (Table 4).

The Dutch subset included 353 patients who provided 365 measurements. The phenotypic group of cleft lip and palate included 172 patients

(49%), 200 (57%) were male, and PROMs were mostly completed around the age of 12 [ $n = 154$  (42%)] (Tables 1 and 2). No statistically significant associations were found between patient characteristics and PROM scores (Table 5).

In total, 35 patients (10%) were referred to psychosocial care, of which 18 patients (5%) were referred based on the result of a PROM score and 17 (5%) were referred for another reason (Tables 1 and 2). The majority of the referred patients were diagnosed with cleft lip and palate [ $n = 21$  (60%)], were male [ $n = 23$  (66%)], and were approximately 8 years of age [ $n = 17$  (49%)]. Concerns about appearance or speech were detected by the PROMs, whereas non-PROM-related reasons for referral consisted of anxiety, behavioral, and coping problems. Patients referred because of PROM scores were most likely to score low on the psychological and face scales, with mean scores of 26 (range, 19 to 32) and 40 (range, 0 to 59), respectively. (See Table, Supplemental Digital Content 3, which shows characteristics of patients referred to psychosocial care, <http://links.lww.com/PRS/F580>. See Table, Supplemental Digital Content 4, which shows reasons for referral to psychosocial

**Table 3. Spearman Rho Correlation Coefficient**

	$t_8$		$t_{12}$		$t_{15}$		$t_{17}$		$t_{final}$	
	Correlation	<i>P</i>	Correlation	<i>P</i>	Correlation	<i>P</i>	Correlation	<i>P</i>	Correlation	<i>P</i>
Psychological–social	0.69	0.00 <sup>a</sup>	0.74	0.00 <sup>a</sup>	0.76	0.00 <sup>a</sup>	0.75	0.00 <sup>a</sup>	0.75	0.00 <sup>a</sup>
Psychological–school	0.59	0.00 <sup>a</sup>	0.68	0.00 <sup>a</sup>	0.66	0.00 <sup>a</sup>	0.67	0.00 <sup>a</sup>	N/C	
Social–school	0.80	0.00 <sup>a</sup>	0.85	0.00 <sup>a</sup>	0.82	0.00 <sup>a</sup>	0.78	0.00 <sup>a</sup>	N/C	
Psychological–face	0.60	0.00 <sup>a</sup>	0.61	0.00 <sup>a</sup>	0.59	0.00 <sup>a</sup>	0.61	0.00 <sup>a</sup>	0.65	0.00 <sup>a</sup>
Social–face	0.46	0.00 <sup>a</sup>	0.55	0.00 <sup>a</sup>	0.48	0.00 <sup>a</sup>	0.51	0.00 <sup>a</sup>	0.58	0.00 <sup>a</sup>
School–face	0.37	0.00 <sup>a</sup>	0.45	0.00 <sup>a</sup>	0.40	0.00 <sup>a</sup>	0.49	0.00 <sup>a</sup>	N/C	
Psychological–speech distress	0.24	0.00 <sup>a</sup>	0.35	0.00 <sup>a</sup>	0.34	0.00 <sup>a</sup>	0.39	0.03 <sup>a</sup>	0.28	0.00 <sup>a</sup>
Psychological–speech function	0.22	0.00 <sup>a</sup>	0.27	0.00 <sup>a</sup>	0.26	0.00 <sup>a</sup>	0.14	0.00 <sup>a</sup>	0.14	0.09
Social–speech distress	0.42	0.00 <sup>a</sup>	0.49	0.00 <sup>a</sup>	0.46	0.00 <sup>a</sup>	0.53	0.00 <sup>a</sup>	0.46	0.00 <sup>a</sup>
Social–speech function	0.37	0.00 <sup>a</sup>	0.41	0.00 <sup>a</sup>	0.40	0.00 <sup>a</sup>	0.30	0.00 <sup>a</sup>	0.26	0.00 <sup>a</sup>
School–speech distress	0.36	0.00 <sup>a</sup>	0.41	0.00 <sup>a</sup>	0.39	0.00 <sup>a</sup>	0.46	0.00 <sup>a</sup>	N/C	
School–speech function	0.33	0.00 <sup>a</sup>	0.32	0.00 <sup>a</sup>	0.30	0.00 <sup>a</sup>	0.31	0.00 <sup>a</sup>	N/C	

$t_8$ , age 8–9 years;  $t_{12}$ , age 10–13 years;  $t_{15}$ , age 14–16 years;  $t_{17}$ , age 17–19 years;  $t_{final}$ , age 20–22 years; N/C, could not be computed, because of too few observations in school function scale at  $t_{final}$ .

<sup>a</sup>Statistically significant.

**Table 4. Linear Univariable Regression Analysis for Different Time Points per Psychosocial Function Scale**

Time Point	CLEFT-Q Psychological Function			CLEFT-Q Social Function			CLEFT-Q School Function		
	$\beta$	95% CI	<i>P</i>	$\beta$	95% CI	<i>P</i>	$\beta$	95% CI	<i>P</i>
$t_8$ (ref)	78.48			73.04			75.18		
$t_{12}$	-3.30	-5.33 to -1.56	0.00 <sup>a</sup>	1.27	-0.58 to 3.13	0.18	-0.06	-2.04 to 1.93	0.96
$t_{15}$	-6.70	-8.99 to -4.41	0.00 <sup>a</sup>	-0.04	-2.07 to 1.98	0.97	0.83	-1.43 to 3.08	0.47
$t_{17}$	-8.87	-11.44 to -6.29	0.00 <sup>a</sup>	-0.45	-2.75 to 1.86	0.71	-0.37	-3.80 to 3.06	0.83
$t_{final}$	-8.71	-11.70 to -5.72	0.00 <sup>a</sup>	-1.71	-4.25 to 0.83	0.19	-7.18	-32.08 to 17.72	0.57

$\beta$ , regression coefficient;  $t_8$ , age 8–9 years; ref, reference group;  $t_{12}$ , age 10–13 years;  $t_{15}$ , age 14–16 years;  $t_{17}$ , age 17–19 years;  $t_{final}$ , age 20–22 yr. <sup>a</sup>Statistically significant.

**Table 5. Univariable Linear Analysis for Psychological Function, Social Function, and School Function Outcomes Based on Data Collected at Erasmus University Medical Center**

Variable	CLEFT-Q Psychological Function			CLEFT-Q Social Function			CLEFT-Q School Function		
	$\beta$	95% CI	<i>P</i>	$\beta$	95% CI	<i>P</i>	$\beta$	95% CI	<i>P</i>
Sex									
Male (ref)									
Female	-0.65	-6.86 to 5.57	0.84	0.40	-6.56 to 7.36	0.91	-0.47	-6.38 to 5.45	0.88
Cleft type									
Cleft lip and palate (ref)									
Cleft lip	-4.99	-18.12 to 8.14	0.45	0.71	-10.29 to 11.71	0.90	-2.97	-15.56 to 9.62	0.64
Cleft palate	2.78	-3.98 to 9.54	0.42	-1.91	-10.30 to 6.47	0.65	1.53	-4.95 to 8.01	0.64
Cleft lip and alveolus	-4.57	-15.24 to 6.09	0.40	3.63	-14.36 to 21.62	0.69	-0.78	-11.00 to 9.45	0.88
Adoption									
No (ref)									
Yes	-2.17	-10.08 to 5.74	0.59	0.71	-8.49 to 9.90	0.88	1.88	-5.65 to 9.40	0.62
Genetic syndrome									
No (ref)									
Yes	1.98	-6.91 to 10.87	0.66	-3.20	-13.80 to 7.43	0.55	0.99	-7.47 to 9.45	0.82
Socioeconomic status	0.01	-2.64 to 2.66	0.99	2.95	-0.61 to 6.50	0.10	0.91	-1.61 to 3.43	0.48

ref, reference group.

care, collected by themes, <http://links.lww.com/PRS/F581>.)

The score for the psychological function scale was significantly associated with referral to psychosocial care (OR, 0.92; 95% CI, 0.88 to 0.97). Similar effects were found for the school (OR, 0.94; 95% CI, 0.90 to 0.98) and face (OR, 0.96; 95% CI, 0.94 to 0.98) scale scores. The presence of a genetic syndrome was significantly associated with referrals (OR, 2.37; 95% CI, 1.04 to 5.41), whereas other patient characteristics were not (Table 6).

## DISCUSSION

This study evaluated the correlations between the CLEFT-Q psychosocial scales that are recommended by the ICHOM Standard Set to determine whether each scale measures a unique construct or overlaps other scales. The CLEFT-Q social function scale measures a construct very similar to the CLEFT-Q school function scale and also has significant common ground with the psychological function scale, whereas the correlation between school function and psychological function was more modest. This suggests that the school function scale addresses a particular aspect of psychosocial function that the other instruments do not capture, namely, aspects related to the social environment at school. In contrast, the social function scale does not contribute much unique information, as there is great overlap with the school and psychological function scales. In other words, the Standard Set might be limited to administering only the CLEFT-Q psychological and school function scales, without losing any relevant information. Dropping the social function scale will reduce the number of questions by 10,

helping to reduce the burden for both patient and clinical team and making the outcomes measurement project more sustainable in the long run. In situations where a child does not attend school, the CLEFT-Q social function scale may serve as a reasonable alternative.

The finding of moderate correlations between the CLEFT-Q face and psychosocial scales suggests that a patient's psychosocial functioning is

**Table 6. Univariable Logistic Analysis with Odds Ratios for Referral to Psychosocial Care after Completion of PROMs**

Variables	Referral to Psychosocial Work		
	OR	95% CI	<i>P</i>
Sex			
Male (ref)			
Female	0.65	0.31–1.35	0.25
Cleft type			
Cleft lip and palate (ref)			
Cleft lip	0.96	0.31–2.99	0.94
Cleft palate	0.45	0.19–1.10	0.08
Cleft lip and alveolus	0.83	0.23–2.96	0.77
Adoption			
No (ref)			
Yes	1.22	0.51–2.94	0.65
Genetic syndrome			
No (ref)			
Yes	2.37	1.04–5.41	0.04 <sup>a</sup>
Timing of PROMs			
$t_8$ (ref)			
$t_{12}$	0.69	0.33–1.46	0.33
$t_{final}$	0.38	0.12–1.17	0.09
CLEFT-Q scale			
Psychological function	0.92	0.88–0.97	0.00 <sup>a</sup>
Social function	0.96	0.92–1.00	0.06
School function	0.94	0.90–0.98	0.01 <sup>a</sup>
Face	0.96	0.94–0.98	0.00 <sup>a</sup>
Speech distress	0.98	0.95–1.01	0.12
Speech function	0.99	0.97–1.02	0.56
Socioeconomic status	0.83	0.61–1.13	0.24

ref, reference group;  $t_8$ , age 8–9 years;  $t_{12}$ , age 10–13 years;  $t_{final}$ , age 20–22 years.

<sup>a</sup>Statistically significant.

influenced by a patient's subjective appraisal of facial appearance. This finding was confirmed in the subgroup analysis performed on the Dutch dataset where patients with a visible cleft lip achieve lower scores on the psychological and school function scales than patients with cleft palate. The weak correlations between the two speech-related scales and the three psychosocial scales suggests that a patient is able to achieve high scores for psychosocial function while experiencing speech problems. This finding is in concordance with a large study on CLEFT-Q normative scores where only small differences in mean scores of psychological, school function, and social function scales between patients with a clinically moderate to severe speech problem and patients with mild or no speech problems were found.<sup>19</sup>

### Addressing the Measurement Gap during Teenage Years

The ICHOM Standard Set presently has a measurement gap, as there are no assessments performed on patients between the ages of 12 and 22 years. The teenage years are very important, because young people undergo puberty and experience many changes in their physical and psychosocial development. To improve the possibilities for longitudinal follow-up and future benchmarking projects, administering the psychological and school function scales at the age of 15 and 17 years of age would provide important additional information about psychosocial adjustment. Importantly, the CLEFT-Q psychological function scale could provide useful information regarding a patient's functioning around young adolescence, because regression analysis showed a decreasing trend in outcome score over time, suggesting that this scale is most sufficient to intervene on.

### Influence of Patient and Demographic Characteristics on Psychosocial Functioning

The second part of this study performed on the Dutch subset of data, exploring associations between patients' clinical and demographic characteristics and the PROM scales, did not find any statistically significant associations. This finding may be attributable to limitations of the demographic and clinical information collected according to the Standard Set. A recently published study on language proficiency of parents from children with craniofacial anomalies, including patients with cleft lip and palate and cleft palate,

demonstrated that parental limited English proficiency was a risk factor for the development of psychosocial distress in terms of higher anger, anxiety, and depression, and poor peer relationships.<sup>21</sup> Other variables that could be thought to be of influence are family composition (such as siblings or divorce), parental income, and level of education or the child's educational performance. The limitations of the present exploratory project precluded us from including these variables, but we recommend that future prospective studies dealing with psychosocial well-being or functioning take them into account.<sup>13,22</sup>

For the subgroup analysis of the patients from The Netherlands, the socioeconomic status score was added to explore its value as a patient characteristic, because little is known about its influence on patient-reported outcome scores in patients with a cleft. A nonsignificant trend showed that children with lower socioeconomic status scores reported lower scores on the psychosocial function scales and were more likely to be referred to psychosocial care. Unfortunately, for international use in benchmarking projects, the generalizability of this finding is limited, because these status scores are specific for Dutch regions and therefore not directly transferrable to other countries. Education, income, and profession are three other indicators for socioeconomic status.<sup>23</sup> Collecting these data in future research could provide more generalizable insights.

### The Use of PROMs in Referring Patients to Psychosocial Care

Patients who reported poorer outcomes on the psychological function, school function, and/or face scales were more likely to have been referred for psychosocial evaluation and management. The cleft team actively used the scales to review symptoms. A poor score on one of these psychosocial scales could prompt the clinician to investigate further and make the appropriate referral when concerned about the patient's psychosocial health. Patients who were referred because of reasons other than PROM scores mainly experienced anxiety or behavioral problems. Previous literature showed that social anxiety disorder is more prevalent in children with cleft lip and palate compared to a healthy control group.<sup>24</sup> Also, higher levels of social anxiety were found in adults compared to adolescents with cleft lip and palate, whereas dental treatment anxiety was highest in children aged 4 to 6 years.<sup>25,26</sup> Therefore, it could be taken into

consideration to include a valid screening tool—parent-reported at the young age and patient-reported from the age of 8 years—to measure anxiety problems.

### Strengths and Limitations

A strength of this study is the large international sample for the correlational analyses, although we recognize that the second part of this study was limited to a smaller population recruited at one university hospital. Even though patient demographics were comparable between these two data sets, the smaller sample size could have resulted in less power to detect clinically relevant differences when evaluating associative relationships. When closing data collection for this study, the ICHOM Standard Set was implemented for 4 years. This has resulted in a cross-sectional study design, in which the maximum possible follow-up period between two measurements was 4 years (age 8 to 9 years and age 10 to 13 years) and very few longitudinal data were gathered. Therefore, results should be interpreted on a group level rather than on an individual patient level and do not reflect a patient's psychosocial well-being in the long term.

### CONCLUSIONS

This is the first study to explore the psychosocial domain within the ICHOM Standard Set and specifically six CLEFT-Q scales of psychological function, social function, school function, face, speech function, and speech-related distress. Because the CLEFT-Q social function scale showed strong overlap with both psychological function and school function scales, its value is limited, and inclusion in the Standard Set should be reconsidered. Including only the CLEFT-Q psychological function and school function scales is recommended. Further recommendations are expansion of required time points to include the teenage years (eg, 15 and 17 years of age) and addition of expanded demographic and socioeconomic variables.

**Inge Apon, MD, MHS**

Erasmus University Medical Center  
Dr. Molewaterplein 40  
3015 GD Rotterdam, The Netherlands  
i.apon@erasmusmc.nl

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