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Research Paper
Cleft Lip and Palate

Evaluation and recommendations of the oral health, oral function, and orofacial aesthetics-related measures of the ICHOM Standard Set for Cleft Lip and Palate☆

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Abstract. This study was performed to evaluate the efficacy of outcome measures for the orofacial domain included in the International Consortium for Health Outcomes Measurement Standard Set for Cleft Lip and Palate (ICHOM-SCS). In this multicentre study involving two cleft centres, suggestions to optimize the type and timing of outcome measures were made based on data and clinical experience. Patient-reported outcome measures (PROMs) (CLEFT-Q Jaw, Teeth, Eating/Drinking; Child Oral Health Impact Profile—Oral Symptoms Scale (COHIP-OSS)) and clinical outcome measures (caries experience and dental occlusion) data were collected retrospectively for age 5, 8, 10, 12, 19, and 22 years. The data were categorized by cleft type and analysed within and between age groups using Spearman correlation, the distribution of responses per item, a two-sample test for equality of proportions, and effect plots. Most correlations between PROMs and clinical outcome measures were weak ($r < 0.5$), suggesting PROMs and clinical outcome measures complement each other. The COHIP-OSS and CLEFT-Q Eating/Drinking barely detected problems in any patient category and are no longer recommended. A suitable

Keywords: Quality of life; Patient outcomes assessment; Cleft lip; Cleft palate; Oral health; Dental esthetics.

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alternative appears complex to find; outcomes of this study and the recent literature doubt an added value. Similar problems were found in the CLEFT-Q Jaw at time-point 12 years. Therefore, time-points 15 and 17 years are currently suggested.

Cleft lip and/or palate (CL/P) is one of the most common congenital malformations worldwide[1,2]. Due to functional and aesthetic differences, children with CL/P face many difficulties and undergo multiple treatments from birth to adulthood[3]. A frequent problem is impaired oral health[4]. Hypodontia, microdontia, dilacerations, agenesis, and supernumerary teeth are reported. Furthermore, malformations in tooth shape or enamel hypoplasia, often combined with suboptimal oral hygiene, lead to a high caries risk in patients with CL/P[5–8]. Moreover, impaired growth of the maxilla due to a cleft palate can lead to an altered facial appearance and additional functional problems[9,10].

The many factors that can affect the orofacial domain in patients with CL/P may result in lower quality of life (QoL)[11–13]. Given the complex and multifactorial nature of the impact on QoL, it is essential to systematically monitor oral health, oral function, and orofacial aesthetics[14].

Monitoring the orofacial domain in a meaningful way poses several challenges[15]. First, selecting the types of outcome measure to cover the many aspects of the domain appropriately is important. These should be scientifically sound and provide reliable follow-up to detect problems in a timely manner. They should also accurately represent the true findings of the individual and be valid for the patient group. Second, the timing of assessment is essential to determine when clinical interventions are most effective and when problems are likely to occur. Finally, to improve cleft care (inter) nationally, outcome measures should enable inter-patient and inter-centre comparison.

In 2015, the International Consortium for Health Outcomes Measurement (ICHOM) developed the Standard Set for Cleft Lip and Palate (ICHOM-SCS), which meets all of the criteria mentioned[14]. The ICHOM-SCS was designed by a global group of clinicians in cleft care, in collaboration with patients, parents, and patient representatives, and includes clinical outcome measures and patient-reported outcome measures (PROMs) covering function, aesthetics, and psychosocial well-being[14,16]. Through inter-patient comparison and benchmarking between cleft teams, ICHOM aims to improve the quality of care, reduce costs, and support shared decision-making in health care[17].

The ICHOM-SCS was first implemented in 2015 and is nowadays used in numerous hospitals worldwide. However, whether the included outcome measures cover all factors of the orofacial domain appropriately and at the right moment, and whether the included outcome measures complement each other or if they collect overlapping information, has not been examined so far. Therefore, the Erasmus Medical Center (EMC) in Rotterdam, the Netherlands and the Karolinska University Hospital (KUH) in Solna, Sweden, both of which have implemented the ICHOM-SCS, established an international collaboration to evaluate these issues based on data and clinical experience.

The aim of this study was to evaluate the types and timing of the outcome measures for the orofacial domain in the ICHOM-SCS, consider other measures that are not currently included, and make recommendations for improvement.

Materials and methods

EMC and KUH have both implemented ICHOM-SCS as part of regular clinical care. Both centres obtained medical ethical approval and signed a data-sharing agreement; the data were anonymized at the respective centres prior to analysis.

Patient population

All patients treated for CL/P who were assessed according to the ICHOM-SCS (age range 5–22 years) at EMC (since 2015) and at KUH (since 2017) were included in the cross-sectional analysis. An overview of the outcome measures assessed and the measurement time points at the two centres is given Table 1.

The patients were categorized into four cleft type groups: cleft lip (CL), cleft palate (CP), cleft lip and alveolus (CLA), and cleft lip and palate (CLAP).

Assessments took place at different time points (EMC: 5, 8, 12, and 22 years; KUH: 10 and 19 years) and within age ranges according to the ICHOM protocol[18,19].

Outcome measures

The PROMs included the CLEFT-Q Jaw and Teeth scales, CLEFT-Q Eating/Drinking (CLEFT-Q ED) checklist, and Child Oral Health Impact Profile—Oral Symptoms Scale (COHIP-OSS).

The CLEFT-Q Jaw and Teeth scales consist of 7 and 8 questions, respectively, regarding the aesthetics of the jaw and teeth. Responses are based on a four-point Likert scale (1 = not at all, to 4 = very much). The CLEFT-Q ED checklist comprises 9 questions about oral functioning related to food intake. Responses are based on a four-point Likert scale (1 = always, to 4 = never). The COHIP-OSS measures the patient's perceived oral health with five questions, answered on a five-point Likert scale (0 = never, to 4 = constantly). It is a validated subscale of the COHIP for the measurement of oral health-related QoL[10].

Items of the CLEFT-Q Jaw and Teeth are positively worded, with higher scores indicating higher satisfaction. Items of the CLEFT-Q ED and COHIP-OSS are negatively worded, with higher scores indicating lower satisfaction. The CLEFT-Q Jaw and Teeth scales have been proven valid and reliable, and sum scores or logit scores can be collected for analysis. The CLEFT-Q ED and COHIP-OSS should be used as problem checklists, meaning each item should be interpreted individually[20,21].

Clinical outcome measures included the DMFT/dmft and occlusion overjet (OCC). The DMFT is a clinician-reported tool, used for quantitative evaluation of caries experience. The number of teeth (T = teeth) with caries lesions (D = decayed), missing due to caries (M = missing), and with restorations due to

Table 1. Measurement time points for all orodental-related outcome measures, per cleft type and at both centres.

Outcome measures[a]	Time points of measurement EMC	Time points of measurement KUH	Type of cleft[b]
Dental-related clinical outcomes			
DMFT/dmft	Age 5, 12	N/A	CP, CLA, CLAP
OCC	Age 5, 12, 22	N/A	CP, CLA, CLAP
Oral function-related PROMs			
CLEFT-Q ED	Age 8, 12, 22	Age 10, 19	CP, CLA, CLAP
COHIP-OSS	Age 8, 12	N/A	CP, CLA, CLAP
Oral aesthetics-related PROMs			
CLEFT-Q Jaw	Age 12, 22	Age 10, 19	CL, CP, CLA, CLAP
CLEFT-Q Teeth	Age 8, 12, 22	Age 10, 19	CL, CP, CLA, CLAP

EMC, Erasmus Medical Center; KUH, Karolinska University Hospital; N/A, not applicable; PROMs, patient-reported outcome measures.

[a]DMFT (permanent dentition)/dmft (primary dentition), number of teeth (T) decayed (D), missing (M), with restorations due to caries (F); OCC, occlusion overjet; CLEFT-Q ED, CLEFT-Q Eating/Drinking; COHIP-OSS, Child Oral Health Impact Profile—Oral Symptoms Scale.

[b]CL, cleft lip; CP, cleft palate; CLA, cleft lip and alveolus; CLAP, cleft lip, alveolus, and palate.

caries (F = filled) are calculated. Lowercase letters reflect the primary dentition (dmft), while uppercase letters reflect the permanent dentition (DMFT)[22,23]. The OCC is determined on a five-point scale: 1 = positive overjet > 3 mm, 2 = positive overjet 1–3 mm, 3 = edge to edge bite, 4 = negative overjet 1–3 mm, and 5 = negative overjet > 3 mm. A score of 2 indicates a neutral and optimal relationship.

Data collection procedure

The data were collected retrospectively. Baseline characteristics included sex, type of cleft, and age at the time of assessment.

The patients completed the PROMs prior to the cleft team visit, by email or in the outpatient clinic. Patients with severe cognitive impairments or language barriers were excluded from the study.

Clinical outcome measures (DMFT/dmft and OCC) were scored after intraoral inspection by a paediatric dentist, maxillofacial surgeon, or orthodontist during a follow-up visit.

Data analyses

The data were analysed using R statistical software version 4.2.0 (R Foundation for Statistical Computing, Vienna, Austria). Sample characteristics were analysed first, then analyses of the outcome measures per time point were performed. Spearman's correlation was used to examine the correlations between the clinical outcome measures and PROMs logit scores (if applicable). Correlations with $r < 0.5$ were considered weak. In the case of moderate ($r > 0.5$) or strong ($r > 0.7$) correlations, subgroup analyses by cleft type were conducted, while applying the Bonferroni correction (starting with a regular significance level of 0.05). Strong correlations between scales were further examined at an individual item level.

Furthermore, the distribution of the responses per item were visualized for the PROMs at each time point. Clinical outcome measures were presented as tables for the available time points. A two-sided, two-sample test for equality of proportions (without continuity correction) was used to examine differences in outcome scores of the clinical outcome measures between the age groups.

The scores of the CLEFT-Q Teeth were checked for normality, and the average score over the treatment period was estimated using a linear regression model. Data from both centres were used to fit the model and to adjust for cleft type. The effect of age was included as a non-linear variable using splines (df = 2). The average score over time was estimated for each cleft subtype allowing an interaction between the two variables. The fitted model was visualized using effect plots.

Results

Sample characteristics

A total of 1157 patients were analysed; slightly more were male (56.0%) than female (41.6%). The largest proportion of the included patients had CLAP (46.8%), followed by CP (35.1%), CLA (11.3%), and CL (6.8%). A total of 1460 measurements were done (Table 2).

Results at the different time-points (by age category)

Time point 5 years

Mean age was 4 years and 10 months (standard deviation (SD) 11.2 months). A weak correlation ($r = 0.13$, $P = 0.029$) was found between OCC and DMFT/dmft.

The lowest DMFT/dmft outcomes, and thus the best scores, were found in the CP group, with 71.3% having a score of zero points (Supplementary Material Table S1). In contrast, in the CLAP group, 57.0% scored zero and 25.0% had a DMFT/dmft score higher than 3. Concerning OCC, a neutral position of the jaw (OCC 1–3 mm) was most often found, especially in the CLA patients (80.0%, Supplementary Material Table S2). In the CLAP group, only 46.2% had a neutral position of the jaw, and the most altered jawlines were categorized as 'negative overjet' (25%) or 'edge to edge bite' (22.1%, Supplementary Material Table S2).

Time point 8 years

Mean age was 9 years and 3 months (SD 5.0 months). Correlations between items of the COHIP-OSS, CLEFT-Q Teeth, and CLEFT-Q ED were all weak ($r < 0.50$, Supplementary Material Fig. S1).

The majority of the respondents answered 'never' to all items of the COHIP-OSS and CLEFT-Q ED. An exception was item 3 of the COHIP-OSS ("Have you had crooked teeth or spaces between your teeth?"), where the given answers were more distributed across the options. Responses to the items of the CLEFT-Q Teeth were more divided (Supplementary Material Fig. S2).

Time point 12 years

Mean age was 12 years and 2 months (SD 3.8 months). A moderate correlation ($r = 0.50$, $P < 0.001$) was found between CLEFT-Q Jaw and CLEFT-Q Teeth.

Moderate correlations were found between item 3 of the COHIP-OSS and items 3 ($r = -0.55$, $P < 0.001$) and 5 ($r = -0.66$, $P < 0.001$) of the CLEFT-Q

Table 2. Patient characteristics, presented per time point.

	Time point for measurements (age in years)						
	5	8	10	12	19	22	Total
Number	282	293	259	300	153	173	1460
Age (years), mean (range)	4.80 (3.5–6.7)	9.22 (7.2–10.6)	10.1 (9.4–12.9)	12.2 (10.4–13.2)	19.1 (17.9–20.3)	22.2 (21.7–23.1)	-
Sex, *n* (%)							
Male	174 (61.7)	179 (61.1)	139 (53.7)	177 (59)	76 (49.7)	74 (42.8)	819 (56.1)
Female	106 (37.6)	110 (37.5)	119 (45.9)	123 (41)	77 (50.3)	74 (42.8)	609 (41.7)
Not available	2 (0.7)	4 (1.4)	1 (0.4)	0 (0)	0 (0)	25 (14.5)	32 (2.2)
Cleft type[a], *n* (%)							
CL	1 (0.4)	27 (9.2)	16 (6.2)	22 (7.3)	19 (12.4)	14 (8.1)	99 (6.8)
CP	128 (45.4)	108 (36.9)	84 (32.4)	104 (34.7)	53 (34.6)	45 (26.0)	522 (35.8)
CLA	47 (16.7)	27 (9.2)	19 (7.3)	33 (11)	10 (6.5)	22 (12.7)	158 (10.8)
CLAP	106 (37.6)	131 (44.7)	140 (54.1)	141 (47)	71 (46.4)	92 (53.2)	681 (46.6)
Centre, *n* (%)							
EMC	282 (100)	293 (100)	N/A	300 (100)	N/A	173 (100)	1048 (71.8)
KUH	N/A	N/A	259 (100)	N/A	153 (100)	N/A	412 (28.2)

EMC, Erasmus Medical Center; KUH, Karolinska University Hospital; N/A, not applicable.
[a]CL, cleft lip; CP, cleft palate; CLA, cleft lip and alveolus; CLAP, cleft lip, alveolus, and palate.

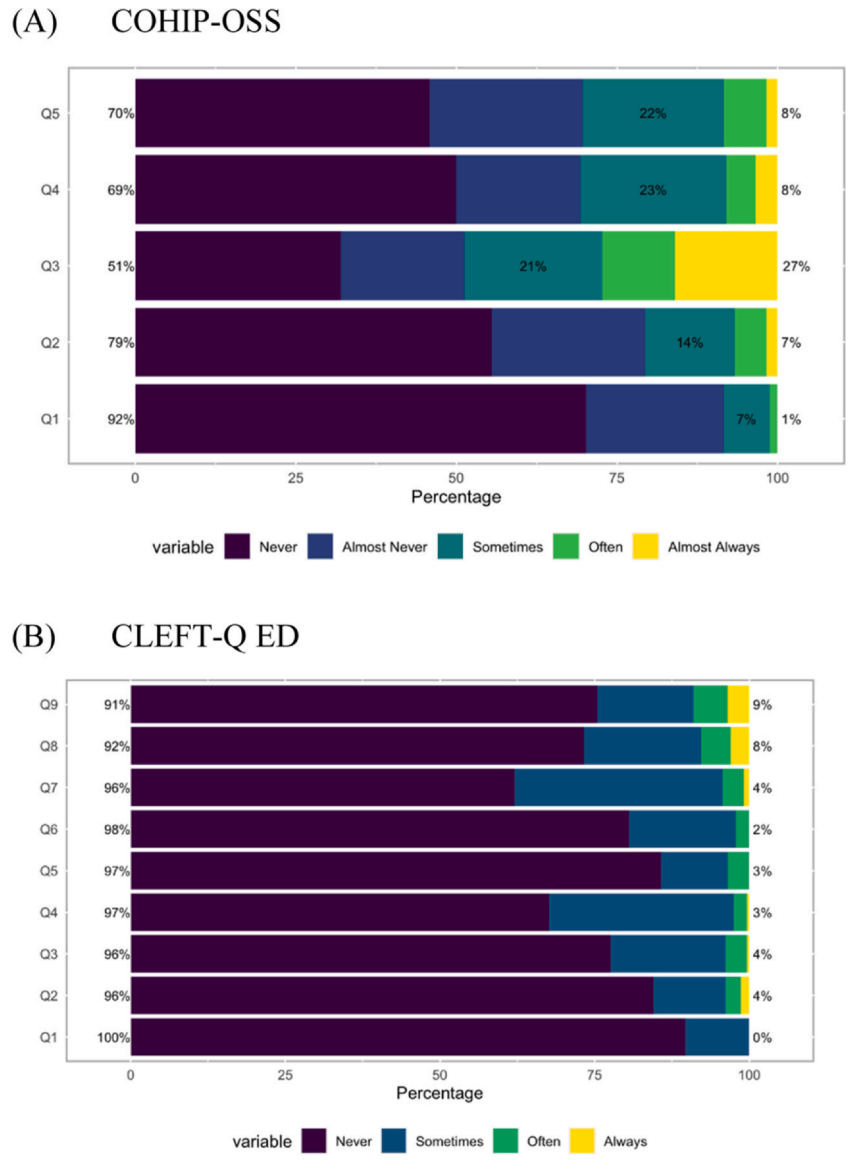

Fig. 1. Distribution of item responses for (A) COHIP-OSS, (B) CLEFT-Q ED, (C) CLEFT-Q Jaw, and (D) CLEFT-Q Teeth, at age 12 years.

Teeth. After subgroup analyses, correlations remained significant in CP ($r = -0.79$, $P < 0.001$; $r = -0.68$, $P < 0.001$) and CLAP ($r = -0.57$, $P < 0.001$; $r = -0.50$, $P < 0.001$). Correlations between the other items of the PROMs were all weak ($r < 0.5$, Supplementary Material Fig. S3).

The distribution of item responses for the COHIP-OSS and CLEFT-Q ED were comparable to the results at 8 years (Fig. 1). Regarding the CLEFT-Q Jaw, the items were answered with 'never' or 'sometimes' by 89–94% of the patients, indicating that they experienced hardly any problems in the appearance of their jaw (Fig. 1). Item responses for the CLEFT-Q Teeth were more divided.

DMFT/dmft at age 12 years significantly more often showed a score between 1 and 3 ($P < 0.001$) and significantly less often a score > 3 ($P = 0.005$) when compared to the results in the 5-year-olds. The frequency of a score of zero in the different cleft groups was similar to that in the 5-year-olds (Supplementary Material Table S1). The OCC in 12-year-olds was mostly scored as neutral (1–3 mm positive overjet), and no significant difference was detected when compared to the 5-year-old group (Supplementary Material Table S2). Fig. 2 shows the correlations between the outcome measures at age 12 years.

Time point 22 years

Mean age was 22 years and 2 months (SD 3.5 months). A moderate correlation was found between the CLEFT-Q Teeth and Jaw scales (Supplementary Material Fig. S4; $r = 0.60$, $P < 0.001$). Subgroup analyses showed similar

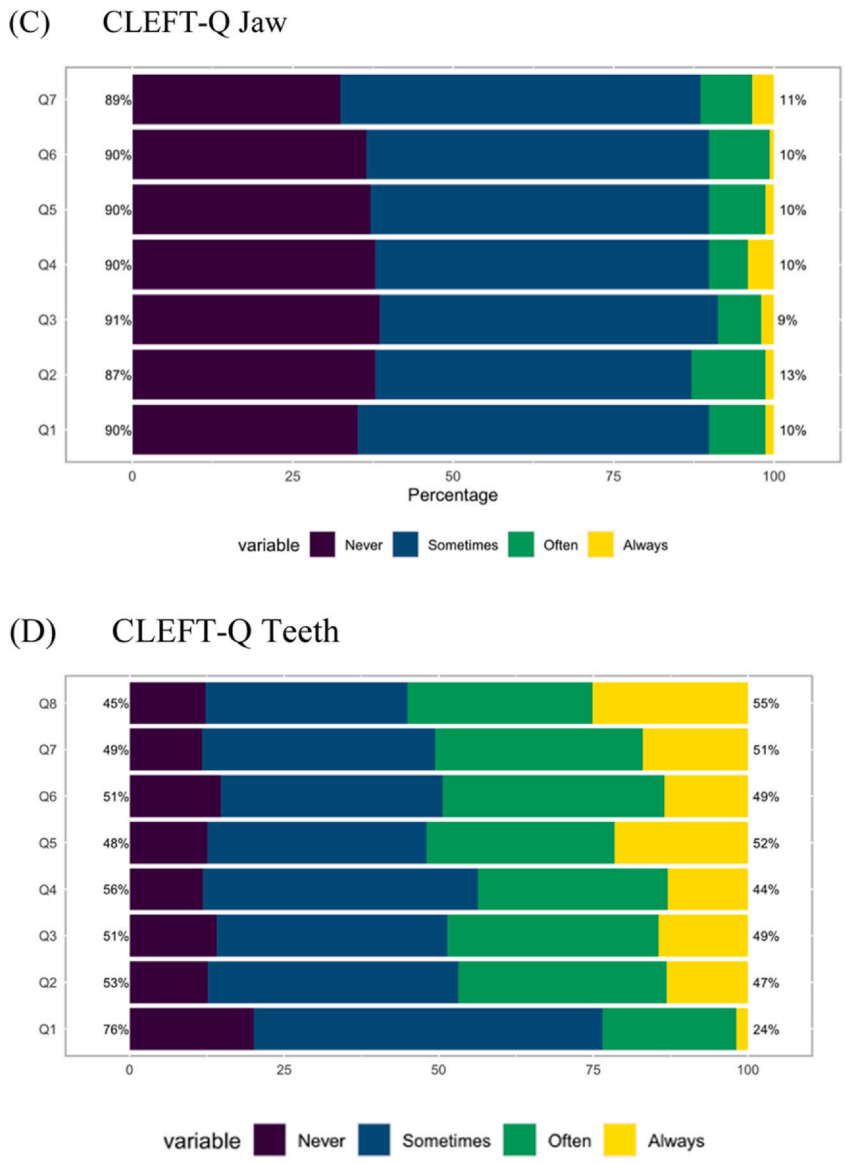

Fig. 1. (*continued*)

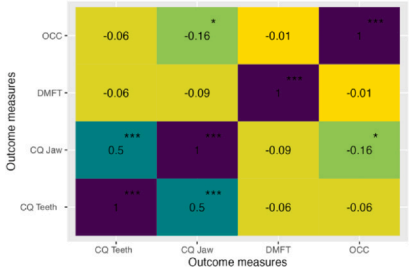

Fig. 2. Correlations between the outcome measures at age 12 years.

correlation values in all cleft types ($P < 0.001$, Supplementary Material Fig. S4). Correlations between the other outcome measures were weak (Supplementary Material Fig. S5).

Concerning the item distributions of the CLEFT-Q ED and CLEFT-Q Jaw, no item showed a high detection of problems that happen 'often' or 'always'. Responses to the CLEFT-Q Teeth were more divided (Supplementary Material Fig. S6).

The OCC was scored neutral in 74.1% of the 22-year-olds, which is a higher percentage than in the lower age groups (Supplementary Material Table S2). In comparison with the 12-year-old group, significantly more patients had an edge-to-edge bite, and significantly fewer patients had a negative overjet > 3 mm (Supplementary Material Table S2).

CLEFT-Q Teeth outcomes over time

Outcomes of the CLEFT-Q Teeth were normally distributed. The estimated overall average score was linear over age and ranged between 47.6 and 70.5. The effect plots showed a slightly upward trend in all cleft types (Fig. 3). Averages per cleft type were similar, and the confidence intervals overlap. Regression coefficients are given in Supplementary Material Table S3.

Discussion

This study is novel in performing a data-based evaluation of the orofacial outcome measures of the ICHOM-SCS. The goal was to improve the set in a meaningful and efficient manner.

No correlations were found between the PROMs and clinical outcome measures at any of the time points. The weak correlation between the CLEFT-Q Jaw and OCC is remarkable, as a non-neutral occlusion overjet was expected to affect the patient's perception of the appearance of the jaw. The same finding accounts for the weak correlation between the CLEFT-Q Teeth and its clinical counterpart, the DMFT/dmft.

The absence of strong correlations between the clinical outcome measures and PROMs indicates that the PROMs capture information other than objective clinical outcome measures. The PROMs have the ability to examine the more nuanced, complex ideas that patients experience about their own health and appearance[24,25]. With the additional information from the PROMs, orofacial problems are detected and explored more comprehensively, and help to clarify whether additional treatment might be indicated.

The outcomes of the CLEFT-Q Teeth varied considerably among all of the patients, suggesting that the scale detects and differentiates relevant problems adequately. Given its lack of correlation with any clinical outcome measure, no adjustments are recommended for this scale.

The outcomes of the CLEFT-Q Jaw were less variable and showed that the scale barely detected problems in this patient population at the specific time points. Furthermore, correlations with the CLEFT-Q Teeth were moderate to strong in the implemented age groups. These findings are comparable to those of a previous study that examined the appearance scales of the CLEFT-Q questionnaire in another cohort with the same age groups[26].

The CLEFT-Q ED did not correlate strongly with any clinical outcome measure, or with another PROM. However, both the CLEFT-Q ED and

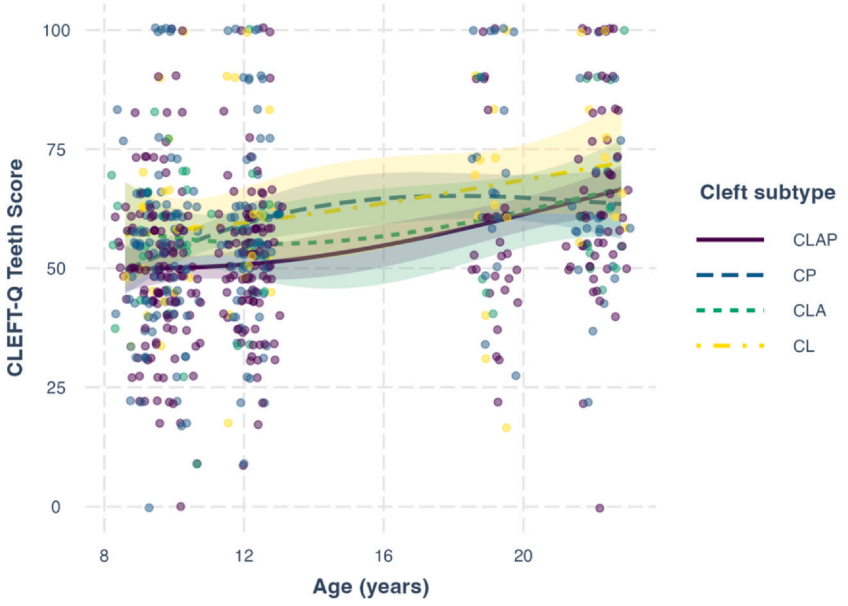

Fig. 3. Effect plot of the CLEFT-Q Teeth over time.

COHIP-OSS did not seem to detect any problems in the study patient group, with the exception of item 3 of the COHIP-OSS. This item correlated strongly with two items of the CLEFT-Q Teeth, which all concern crooked teeth. Moreover, the clinicians experienced that some items also caused confusion among patients. For example, 'food gets stuck in a hole in my mouth' (item 3, CLEFT-Q ED), referring to the occurrence of oronasal fistula, was answered positively by patients during their mixed dentition phase, who thought the item refers to empty spaces caused by missing teeth. Problems with braces and tooth mobility were also referred to by patients through the checklists. Therefore, by removing the COHIP-OSS and CLEFT-Q ED from the ICHOM-SCS, the patient burden will be reduced without compromising on the information collected from the patient perspective.

A large number of patients with CL/P have high DMFT/dmft scores (> 3). Compared to the Dutch normative sample, the population with CL/P from this study showed fewer patients with a DMFT/dmft score between 1 and 3, and more patients with a score > 3[27]. This indicates that patients with CL/P who have dental health problems are often confronted with more severe problems than their peers without CL/P. The collection of DMFT/dmft values therefore seems relevant, especially given the weak correlations with other data collected in the ICHOM-SCS.

Non-neutral OCC scores were found in all age groups and in all cleft types. The prevalence of patients with a neutral overjet was strongly reduced in the study patients when compared to international normative samples (80–85%)[28,29].

Overall, patients with an altered OCC were most often scored as having an edge-to-edge bite or a negative overjet, which is a well-known phenomenon in patients with a cleft. Moreover, correlations of the OCC with other collected data in the ICHOM-SCS were weak, meaning that the OCC provided clinicians and patients with unique and meaningful information. Therefore, assessment of the OCC seems valuable.

Based on the current findings, the CLEFT-Q Teeth, OCC, and DMFT/dmft collect relevant information at the currently implemented time points that are also suitable from a clinical perspective. They provide information on the primary and permanent dentition.

Time points 19 and 22 years of age reflect the results at the end of treatment at each centre, and the decision regarding secondary corrections before ending the standard follow-up pathway. This last moment of measurement should be implemented according to the protocol at each individual centre, although the time window suggested is between 20 and 22 years to enable inter-centre comparison of outcomes. This is in line with previously presented results concerning the PROMs assessing facial appearance in the ICHOM-SCS[19,26].

Twelve-year-old patients had difficulties completing the CLEFT-Q Jaw scale and showed limited awareness of their jawline. For instance, patients frequently asked for a mirror to observe their jawline and forwarded the questions towards the clinicians and parents. This could explain why barely any problems were detected through the scale in this age group.

In line with the previous findings and recommendations of Ombashi et al.[26], assessment of the CLEFT-Q Jaw at an older age could be more suitable, when patients might have developed awareness of separate aspects of their face. Ages 15, 17, and 22 years are suggested instead of 12 and 22 years. These time points would be clinically relevant too: at age 17 years, maxillary and mandibular growth are finished, and aesthetic corrections like a Le Fort I osteotomy are possible. Therefore, time points 17 and 22 years of age could collect pre- and postoperative results.

Data of the ICHOM-SCS could not yet be analysed longitudinally, as the number of years of data collection is too few. In the future, longitudinal analyses will add information on the dynamic character of patient-reported outcomes and their relationship to clinical outcomes.

Experiences of the cleft teams at the two study institutions were added to ensure clinical relevance of the data-driven outcomes. However, experiences were not collected in a systematic manner (i.e., qualitative interviews or focus groups). Patient and parent opinions were not included either. Although more generic interviews with cleft experts have been described in a previous study[30], research regarding patient and parent opinions has not yet been conducted. The patient burden and their experiences with both PROMs and clinical outcome measures could direct future improvements to the ICHOM-SCS.

Further possible improvements of the ICHOM-SCS concern the evaluation of all other domains and the assessment of appearance-related clinical outcome measures that assess the entire facial appearance instead of only the orodental part. A suggestion for adding a reliable scoring system to the ICHOM-SCS was made in another study by Ombashi et al.[26], where the Cleft Aesthetic Rating Scale (CARS) was discussed.

Another recommendation for improving the ICHOM-SCS involves proper assessment of oral functioning. As COHIP-OSS and CLEFT-Q ED do

not seem to detect oral functioning problems adequately in patients with CL/P, an alternative measure could be considered. Another available tool is the Child Perceptions Questionnaire on Oral Health-related Quality of Life for children from 11 to 14 years old (CPQ-11–14)[31,32]. However, the CPQ-11–14 appears to run into similar problems as the COHIP-OSS and CLEFT-Q ED: despite validation through classical test theory, Rasch analyses showed that the CPQ-11–14 lacks items at the better end of the scale to detect problems adequately[31–33]. As no outcome measure detects any (severe) problems in orofacial functioning, the severity and occurrence of it should be reconsidered. Taking patient burden into consideration, there might be no need to systematically assess orofacial dysfunction in patients with regular follow-up and an adequate treatment plan.

In conclusion, patient-reported outcome measures and clinical outcome measures seem to complement each other. The clinical outcome measures seem to detect problems in the populations of the two study centres adequately, and no recommendations for adjustment are made for the overjet assessment and decayed/missing/filled teeth index. Concerning the patient-reported outcome measures in the International Consortium for Health Outcomes Measurement Standard Set for Cleft Lip and Palate, the assessment of CLEFT-Q Jaw shows no added value, and implementation is now recommended at 15, 17, and 22 (instead of 12 and 22) years of age. The assessment of self-reported oral health and nutritional problems brings a burden that seems to outweigh the benefit. Hence, further use of the Child Oral Health Impact Profile—Oral Symptoms Scale and CLEFT-Q Eating/Drinking is not encouraged. As similar findings are reported with possible alternatives like the Child Perceptions Questionnaire, the relevance of measuring these functional problems should be reconsidered.

Ethical approval

Obtained from the Erasmus University Medical Center Medical Ethics Review Committee (MEC-2016-156 355) .

Patient consent

Not applicable.

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Competing interests

None.

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Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.ijom.2024.01.001.

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