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Original Article

Core outcomes for orofacial clefts: reconciling traditional and ICHOM minimum datasets

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Summary

Objective/Design/Setting: This retrospective study sought voluntary participation from leading cleft centres from Europe and Brazil regarding core outcome measures. The results of this study would inform the debate on core outcome consensus pertaining to the European Reference Network for rare diseases (ERN CRANIO) and achieve a core outcome set for cleft care providers worldwide.

Intervention/Method: Five orofacial cleft (OFC) disciplines were identified, within which all of the International Consortium of Health Outcomes Measurement (ICHOM) outcomes fall. One questionnaire was designed for each discipline and comprised 1. the relevant ICHOM's outcomes within that discipline, and 2. a series of questions targeted to clinicians. What core outcomes are currently measured and when, did these align with the ICHOM minimum, if not how did they differ, and would they recommend modified or additional outcomes?.

Results: For some disciplines participants agreed with the ICHOM minimums but urged for earlier and more frequent intervention. Some clinicians felt that some of the ICHOM standards were compatible but that different ages were preferred and for others the ICHOM standards were acceptable but developmental stages should be preferred to absolute time points.

Conclusion/Implications: Core outcomes for OFC were supported in principle but there are differences between the ICHOM recommendations and the 2002 WHO global consensus. The latter are established in many centres with historical archives of OFC outcome data, and it was concluded that with some modifications ICHOM could be moulded into useful core outcomes data for inter-centre comparisons worldwide.

Introduction

Cleft care is multidisciplinary comprising a range of healthcare specialties including speech/language therapy, otolaryngology, surgery, orthodontics, dentistry, and psychology, and the success of this care can be objectively determined by measurable outcomes at specific stages and time points within each discipline. The creation of a 'core outcome set' is regarded as an extremely important element of clinical care, clinical effectiveness, quality of care, and research aimed at health improvement, and in the field of orofacial clefts (OFCs) there is a history of attempting to achieve global consensus on core outcomes.

The standardization of these outcomes at a global level is an important goal to achieve for a more coordinated approach towards equality for cleft care internationally and universally, ultimately benefiting patients wherever they are in the world. To this end, in 2002, the World Health Organisation (WHO) published a report of global strategies to reduce the healthcare burden of craniofacial anomalies following a series of consensus meetings on International Collaborative Research held in Geneva (2000) and Utah (2001) (1). This report details a set of minimum records which should be taken for each individual patient for the purposes of treatment planning, monitoring, outcome evaluation, and further research/quality improvement (1). They further encourage taking additional records depending on each team's unique clinical protocols, infrastructure, capacity, and desires.

The International Consortium of Health Outcomes Measurement (ICHOM) is a non-profit organization that is currently regarded as internationally accredited for standardization of health outcomes in a range of medical and dental conditions. In 2017, ICHOM published a set of standards for the holistic appraisal of the treatment for cleft lip and/or palate (CL/P) patients. This ICHOM Standard Set

recommends a series of outcomes which should be measured and recorded as a minimum requirement, and encourages cleft teams worldwide to adopt their protocol (2). However, ICHOM has recommended clinical records to be taken at stages and time points that are different to those recommended by the WHO. This ICHOM Standard Set may therefore not necessarily reflect the collective view of all the established cleft organizations around the world and the WHO.

The European Reference Networks (ERNs) for rare diseases is a virtual network of healthcare providers across Europe that aims to encourage cross-border collaboration and has specifically sought to improve the standards of care for EU patients afflicted with rare diseases. In March 2017, 24 ERNs were established for various rare diseases; of these 24 ERNs, one known as the ERN CRANIO includes a spectrum of healthcare providers (HCPs) with expertise in providing multidisciplinary care for patients born with CL/P. They too recognize the need for standardization in outcome measures, as this is important with respect to both audit and research aimed towards the improvement of standards of care. It can therefore be seen that reputable centres of expertise which have historical databases and established protocols have a vested interest in engaging with ICHOM. This study sought views on ICHOM's Standard Set of minimum outcomes and to help achieve a consensus applicable for all healthcare providers worldwide. In the spirit of mutual collaboration, it should therefore be relevant and beneficial to include the perspectives of these organizations and institutions.

The purpose of this study therefore was to gather professional opinion on ICHOM's outcomes for CL/P from cleft centres within Europe by sending out an invitation to the ERN HCPs requesting participation. Four centres volunteered to participate in the study. These were the Smile House in Italy, the University of Amsterdam's University Medical Centre (UMC) and the University Medical Centre Utrecht (UMCU) in the Netherlands and the University of Gothenburg in Sweden. It was felt that it might be valuable to include the Hospital for Rehabilitation of Craniofacial Anomalies (HRAC) in Brazil, one of the largest and most reputable cleft centres in the world, with a view to asking about their current outcome measures and whether they might assist in reaching a consensus on universal outcomes for cleft care.

Aims/objectives

The overall aim of this study is to determine whether the Brazilian and European cleft teams consider the ICHOM 'minimum' outcomes sufficiently and adequately serves as a minimal recommendation. The four objectives are as follows:

1. To obtain the opinions of the various multidisciplinary teams regarding the recent ICHOM Standard Sets for CL/P.
2. To identify alignment or difference in opinion between these centres and ICHOM regarding the outcome measures and timing of their collection.
3. To identify convergence or divergence between ICHOM Standard Sets and current practice at each centre.
4. To determine if ICHOM Standard Set requires revision or if the Brazilian and European cleft teams can adopt the ICHOM recommendations for outcome measurements in all disciplines.

Methodology

The ICHOM Standard Set (2) which appraises cleft care was analysed and five main disciplines, within which all outcome criteria fall under, were identified for further investigation. These five disciplines are speech/language therapy, otolaryngology, dentistry/orthodontics, surgery, and psychology (Table 1). One discipline-specific questionnaire was then designed for each discipline. These consisted of the ICHOM minimum outcomes for that discipline in the preamble, and a series of questions targeted to relevant consultants and clinicians. These questions aimed to find out what outcomes the cleft teams measure in their own centres/disciplines, their opinions about the suitability of ICHOM minimum outcomes, and whether they are comfortable with the ICHOM outcomes being recommended as a minimum standard worldwide. In dentistry/orthodontics, additional questions on alveolar bone grafting (ABG) were included to find out whether they consider this to be an important outcome to measure, what features and instruments were considered as appropriate for outcome measures, and the optimum timing for when should these outcomes be collected.

All healthcare providers of multidisciplinary CL/P care within ERN CRANIO (Europe) were invited to participate in discussions on outcome measures and compatibility with ICHOM and were issued with information regarding the ICHOM outcome set (Table 1) plus a set of questions related to these (Tables 2 and 3). Five agreed to participate and of these three returned their questionnaires by the agreed deadline. As part of a student summer elective project the same questionnaires were brought to one of the largest and most respected craniofacial centres in the world—HRAC in Bauru, Brazil. HRAC has been involved in the multidisciplinary care for CL/P patients for over 20 years, and are seen as leaders in the field, and they were willing to participate.

From the feedback received, it was apparent that some teams were unable to meet the time deadline for all disciplines to complete the questionnaires, and we agreed that to improve the response rate we would simplify the process using what we described as 'Phase 2' questionnaires.

It was agreed that this Phase 2 questionnaire should be as simple and non-burdensome as possible (Table 4) and it comprised three questions and referred to two tables, the first outlining the proposed ICHOM outcomes and the second suggesting modifications suggested by the centres that had responded to the original questionnaire.

The results of this study were therefore based on 1. comprehensive multidisciplinary feedback (Phase 1) surrounding the five main disciplines from these four CL/P centres, three of which were in Europe and the other from Brazil, and 2. opinions from a further six centres on the comparison between ICHOM core outcomes and outcomes derived from Phase 1 via what was described as a Phase 2 questionnaire (Table 4).

Results

Outcome measures in OFC are defined by the disciplines that comprise the multidisciplinary teams, so the results of the questionnaire are presented according to the participating disciplines.

Table 1. International Consortium of Health Outcomes Measurement (ICHOM) recommendations for cleft lip and palate outcomes (with modifications)

Discipline	Outcome	Age*	Instrument	Reported by
SLT	Articulation	*3*, 5, 12, End of tx	Percent Consonants Correct	C**
	Velopharyngeal competence	*3*, 5, 12, End of tx	VPC graded rating	C
	Documetation (appraising language)	*3*, 5, 12, End of tx	Recording of a standarised speech/ language sample	C
	Intelligibility	*3*, 5, 12	Itelligibility-in-Context	P**
	Overall speech	12, End of tx	CLEFT-Q Speech and Speaking	P
ENT	Hearing	*2*, 5,12	Puretone average	C
	Otologic health	*2*, 5,12	Otologic health screening questions	C
	Nasal breathing	8, 12	NOSE questionnaire	P
Dentistry and Orthodontics	Dental health	5, *10*	dmft and DMFT	C
	Occlusion 1	5, *10*, End of tx	Overjet assessment	C
	Occlusion 2	*Pre-ABG*, End of tx	Lateral cephalogram	C
	Oral health	8, *10*	COHIP	P
	Mastication	8, *10*, End of tx	CLEFT-Q Eating and Drinking	P
Surgery	Documentation (appraising appearance)	5, 12, End of tx	Facial photographs	C
	Nasolabial appearance	8, 12, End of tx	CELFT-Q Face	P
	Smile	8, 12, End of tx	CLEFT-Q Dental	P
	Facial profile	12, End of tx	CLEFT-Q Jaw	P
Psychology	Sociometrics 1	8, End of tx	CLEFT-Q Social Life	P
	Sociometrics 2	12	CLEFT-Q School Life	P
	Psychometrics	12	CLEFT-Q Feelings	P

The above table shows the outcome domains as identified by ICHOM, the age at which measurements are recommended to be taken, the instrument/ tool recommended to use and the person responsible for reporting this outcome. These outcome domains are organized by discipline and presented to each participating cleft team/discipline in questionnaires. Text in italics = suggested modifications.
*All ages are in years.
**C = clinician, P = patient.

Table 2. Questionnaire issued to healthcare professionals in all five disciplines involved in cleft care

Core questions
What outcomes do you measure in your discipline and at what age do you measure them?
What is your opinion of the above minimum outcomes and corresponding time/age protocols?
Are you comfortable with the above minimum outcomes and corresponding time/age protocols? YES/NO. If not, why?

Speech and language therapy

Brazil's HRAC measures articulation and velopharyngeal competence using their own unique protocol and documentation using a speech audio recording. The ages and time periods used by HRAC is 1 year after primary palatal repair (which translates to around 2 years of age given HRAC's surgical protocol), 4–6 years, 8–10 years, 10–12 years, and end of treatment. In their opinion, they believe that the ICHOM outcomes for speech and language/communication could be started earlier in life, and have suggested around the ages of 2 years consistent with their protocol. In addition, it was mentioned that 'Percent Consonants Correct', which is a tool recommended by ICHOM for assessment of articulation, is a difficult one to use before the ages of 5–6 years. They therefore maintained that the ICHOM minimum outcomes are insufficient and suggested that further measurement of speech/language outcomes are necessary to enable appropriate interventions.

Milan's The Smile House measures speech and phonation skills using the Great Ormond Street Speech Assessment (GOS.SP.ASS revised) (3) perceptive evaluation tool at ages 3 years, 5 years, 10 years, and end of treatment. While the Italian team agrees with the ICHOM minimum outcome time/age protocols, they would have preferred measurements at 5 years, 10 years, and end of treatment. This is justified by the Italian school system where student cohorts enter primary around age 5 years and get promoted to secondary school at the age of 10 years. These age groups present as the perfect ages for them to measure speech outcomes and identify any discomfort in the voice during the transition from primary to secondary education. In addition, the Italian team would have preferred a speech and language assessment (phonation and articulation) at 3 years of age and has said that is it better to evaluate velopharyngeal competence before 5 years of age in order to manage them as soon as possible.

Amsterdam's UMC measures intelligibility and articulation (consonant production) using the GOS.SP.ASS'.98 (Dutch version) and velopharyngeal competence using scales different to those recommended by ICHOM, namely resonance, nasal emission, and nasal turbulence. In terms of age groups, they

Table 3. Additional questions posed to selected disciplines of participating cleft care teams

Additional questions	
For dentistry/orthodontics:	- ICHOM does not include alveolar bone grafting (ABG) as an outcome measure. Do you feel this is important? YES/NO - If ABG outcome is regarded as important, what do you feel is an appropriate outcome? [e.g. bone level (at a specified time post-op), incisor/canine eruption, etc.] - If bone level is regarded as important, how long after the ABG procedure should this be done?
For surgery:	- Do you use or plan to use 3D and 4D measurements in future outcome measurement of facial aesthetics and asymmetry? YES/NO. If not, why?
For psychology:	- Would you support use of 3D and 4D measurements in future outcome measurement of facial aesthetics and asymmetry? YES/NO. If not, why?

Table 4. The Phase 2 questionnaire: the six additional European Reference Network (ERN Cranio) Healthcare Providers (HCPs)

1. Do you feel that the ICHOM core outcomes are in alignment with your thoughts on what the minimum set of outcomes for CLP should be? For ease of reference I have appended Table 1 below. You can merely answer YES/NO
2. Table 2 suggests a few modifications (in italics) to timing etc that align better to the thoughts of the current authors (not yet submitted). Do you prefer this set of outcomes? You can answer YES/NO
3. Do you wish to make comments on outcomes that you feel could be included in this core set that may not be in accordance with those in the table?

begin measurements from 2 years regularly until 6 to 7 years. It is their opinion that the ICHOM recommended minimum outcomes, while acceptable for the patients, are very limited in the clinicians' perspectives and does not say anything about the quality of the speech in time, and are therefore not comfortable with the minimum outcomes. It is also mentioned that the gap between 5 and 12 years as suggested by ICHOM is too big a gap, where there is huge development occurring. They go further to suggest measurement of speech and language development from a younger age (3 years) and to record presence or absence of any speech therapy or other intervention and for how long this therapy was given.

Utrecht's UMCU measures articulation, velopharyngeal competence, intelligibility, and overall speech (which are similar parameters to the ICHOM standard) as well as phonology and language observation. These are performed at ages 2 years, 3.5 years, 5 years, 12 years, and 17 years with a margin of 6 months. In addition, when secondary operation is required for velopharyngeal insufficiency (VPI), an extra evaluation is done 1 year after surgery. It is their opinion that the first speech evaluation at the age of 5 years is too late, and they prefer the age of 3.5 years because of the timing of the VPI surgery intervention and the start of speech therapy. UMCU has reported not being fully comfortable with the ICHOM minimum, and suggests some additional tools and measurements: evaluation of feeding skills after birth and surgery, using CAPS-A (4) as a validated instrument and nasometry (at age 4 years and above).

Otolaryngology (ENT)

Brazil's HRAC measures hearing of the patient every year until the age of 10. In their opinion, earlier measurements of the ENT outcomes before 5 years of age is more important and justify that hearing and language skills development is more important at younger ages, indicating that 5 years is too late for measuring otologic health and hearing. Therefore, they are not comfortable with the ICHOM minimum outcomes for ENT.

Utrecht's UMCU observes for sleeping problems at ages 8 years and 12 years, and screens for breathing problems pre- and post-operatively in relation to the surgical closure of the palate. They described the use of a palatal obturator with soft palate extension and hospital admission pre-operatively to measure breathing and oxygen saturation with the obturator *in situ*, and then 6 weeks post-pharyngoplasty ask about breathing problems with the possibility of hospital admission for one night of observation and measurement. While they did not explicitly report measuring any of the ICHOM outcomes of hearing and otologic health, they commented that audiometry at ages 5 and 12 years seems to be minimal and suggested that hearing outcomes be tested at ages 1 and 2 years with visual reinforcement audiometry.

Dentistry and orthodontics

In Brazil, HRAC measures occlusion and growth in UCLP using the Five Years Old Index (Atack index (5)) at age 5–6 years and the GOSLON Yardstick (6) (in the mixed dentition/ permanent dentition phase and end of growth) and 'Bilateral yardstick' for BCLP patients (7). Study casts and clinical photographs (facial and intraoral) are also done at all intermediate stages of treatment, namely before lip repair, 5 to 6 years, 8 years, before and after ABG, usually this phase vary between 8 and 12 years, and at the end of treatment. Because 8 years of age represents the ideal orthodontic/pre-bone grafting phase for most patients, they feel that the ICHOM minimum outcomes are insufficient as they lack this stage, and have recommended the inclusion of occlusal assessments at 8 years using either the GOSLON Yardstick or Atack index and lateral cephalogram. Considering that it is a country with continental dimensions and many social, economic, and geographic challenges, we are not always able to apply the ideal protocols, resulting in many cases with delays in therapeutic procedures (lip/palate surgery, ABG), as well as in obtaining documentation in standardized phases.

With regards to ABG and measuring its outcomes, HRAC considers it to be important as the procedure itself offers

excellent rehabilitation for patients with clefts of the lip and palate. They recommend the measurement of bone level and bone thickness using radiographs (periapical and/or CBCT for some specific cases) at the following stages: pre-operative (before ABG), at 2 months, 6 months, and 1 year post-ABG. It is also suggested that the Bergland and Clelsea scale (8) could be used to assess bone grafts levels, and that plaque indices should be included more frequently to monitor dental health as cleft treatment (orthodontic and surgical) proceeds.

Milan's the Smile House measures occlusion and growth at 5 years, 10 years, 15 years, and 20 years with intra and extraoral photographs (in order to do photographic GOSLON assessments, validated in 2011, by Dogan *et al*. (9), thus reducing the need for plaster casts, which need space and increase costs) and lateral cephalograms. The reason why Milan has kept the original 10-year-old assessment, as proposed by the Eurocleft mimimum standards, is, first, because at 10 years of age records do not need to be separated between females and males, as is needed at 12, given the differential pubertal onset, second, because a vast majority of centres have collected data at 10 years and inter-centre comparisons with centres who now have long term data. They are in favour of using 10 years of age rather than 12 years (as recommended by ICHOM). Because of the different time points used by the Italian team, they are not comfortable with the recommendations of ICHOM to measure outcomes at 5 years, 12 years, and end of treatment. It is their opinion that end of treatment often does not equate to end of growth, therefore, records at 19–20 are generally more reliable than record taken at 17–18 years. With regards to ABG and measuring its outcomes, the Italian team regards it as important and recommends measurement of bone levels using CT radiographs after 1 year post-ABG, and periodontal assessment after full canine eruption.

The University of Gothenburg in Sweden records lateral cephalograms, study casts, and clinical photographs at the ages of 5, 7, 10, 13, 16, and 19 years. Panoramic radiographs are also taken at 7 and 10 years as additional radiographs depending on individual patient indications. At 5 years, the Atack index is assessed for unilateral cleft lip and palate (UCLP), while the modified Huddart and Bodenham (MHB) scoring system (10,11) is used for all other types of clefts. At 10 years, 16 years, and 19 years (or before orthognathic surgery), the GOSLON yardstick is used for UCLP and the MHB scoring system remains the same for all other types of clefts. After orthognathic surgery, the GOSLON yardstick is used for UCLP and the MHB scoring system is used for all other cleft types, and a lateral cephalogram is also taken. The Swedish team differ from ICHOM in recommending end of growth as opposed to end of treatment, and maintain that measurements of growth should take priority. Furthermore, they believe that measuring overjet alone is insufficient in monitoring occlusion as it leaves other dimensions such as antero-posterior skeletal pattern and transverse relationships unassessed. With regards to ABG and measuring its outcomes, the Swedish team certainly regards ABG and the measurement of its outcomes as very important. They are in the midst of discussing within their own country what ABG outcomes should be measured, but are currently using the Bergland scale (12) for assessing ABG outcomes. For the majority (80%) of their cases, occlusal radiographs are also used which sufficiently measure ABG outcomes, while in minority of cases CBCT is used as an adjunct. They measure these ABG outcomes at 3 months post-operatively, at 10 years and 16 years of age.

Utrecht's UMCU assesses for oligodontia, supernumerary or deformed teeth for the primary dentition at age 5 years, and for the permanent dentition at ages 8 and 12 years. It is their opinion that the end of treatment in terms of dentistry cannot be evaluated before the age of 25 years as implant treatments might be needed after that age, and they are not comfortable with the minimum ICHOM outcomes. UMCU considers ABG outcomes to be very important, however, they acknowledge that many current methods for measuring pre- and post-operative alveolar bone/cleft defect volume in 3D are largely extremely inaccurate. They therefore suggested to assess ABG using clinical parameters such as the continuity of the alveolar process, residual oronasal communication, eruption of the canine/lateral incisor into the reconstructed cleft and periodontal status of teeth in the cleft region, and further augment the results with a Bergland (2D) radiographic scale or a Bergland scale derived from a CBCT. They measure the above ABG outcomes at least after 1 year post-operatively, as they point to current literature suggesting that the 6-month post-operative time point shows almost unequivocally bone or fibro-osseous tissue that is still undergoing remodelling. In addition, they suggested distinguishing between dental occlusion and the orthognathic outcome (dished-in profile) and discussing the difference with patients before commencing orthodontic treatment as a requirement for consent within this discipline.

Surgery

Brazil's HRAC measures the surgical outcomes namely nasolabial appearance, facial profile at 5–6 years, 9–10 years, and at the end of growth, using facial photographs and velopharyngeal function at same phase. In addition, they believe alveolar arch study models and evaluation of facial and nasolabial photographs to be important, and would include ages 5–6 years, 8 years, 15 years, and 20 years. They advocate that documentation is very important and urge for the standardization of photographs, ages, measurement of clefts during operation, and use of plaster study models or intraoral digital models in order to compare the results of protocols, surgeons, and techniques between centres.

Amsterdam's UMC review their patients at the ages of 1, 6, 9, 12, 15, and 18 years and take lateral cephalograms, dental pantomograms (DPT), study casts, and clinical facial photographs at these time points. In addition, aesthetic outcomes of unilateral clefts are measured at 6 and 18 years using the Cleft Aesthetic Rating Scale (13). In their opinion, 8 years of age is an inconvenient moment as many patients with clefts of the palate and alveolus are not yet indicated for a bone graft. They are therefore not comfortable with the current time points and intervals recommended by ICHOM and would be keen to give their input and propose changes to these protocols.

Utrecht's UMCU appraises the appearance by using facial photographs in 2D and 3D, as well as lateral cephalograms (in 2D) and/or CBCT (in 3D). However, they did not describe any specific ages/time points for when they would measure these outcomes. They suggested that the current ICHOM recommendations lack a validated instrument to measure the final aesthetic outcome at age 18 years, and said that there should be an agreement on what kind of tools (pictures/photographs) a team should collect at age 18 years, both in 2D and 3D. As per the dental/orthodontic disciplines, they suggested distinguishing between dental

occlusion and the orthognathic outcome (dished-in profile) and discussing the difference with patients before commencing orthodontic treatment as a requirement for consent within this discipline.

Psychology

Milan's the Smile House measures psychological well-being and family relations during infancy but do not routinely measure outcomes from 8 years of age. While they think the ICHOM minimum outcomes for psychology are coherent and useful, they are not fully comfortable with the recommendations as it is not part of clinical routine. They suggest the Italian version of CLEFT-Q (14) to be useful and could be introduced in routine clinical practice.

Amsterdam's UMC measures the psychosocial functions of CL/P patients using the following CLEFT-Q tools at the corresponding ages (in years):

-ENT & otologic health questionnaire: 5, 12
  -Nasal obstructing nose scale:8, 12, 15, 22
  -COHIP:8, 12, 15
  -CLEFT-Q Speech/Speaking:12, 22
  -CLEFT-Q Eating/Drinking:8, 12, 22
  -CLEFT-Q Face:8, 12, 15, 22
  -CLEFT-Q Dental:8, 12, 22
  -CLEFT-Q Jaw:12, 15, 22
  -CLEFT-Q Social Life:12, 22
  -CLEFT-Q School Life:8, 12
  -CLEFT-Q Feelings:12, 15

In addition, they measure psychological outcomes under 8 years of age by asking parents to fill in questionnaires on the quality of life for their child using TAPQOL from 0 to 1 years and PedsQL from 2 to 7 years. They feel that the ICHOM minimum outcomes are not entirely appropriate to the age of children and adolescents, highlighting that the CLEFT-Q Social Life is not suitable for children aged 8 years. For this reason, UMC considers the importance of the feelings of adolescents by measuring CLEFT-Q Social Life between ages 12 and 22 years (which is regarded as the end of treatment) and CLEFT-Q Feelings at ages 12 and 15 years. They are therefore not comfortable with the ages that ICHOM recommends for psychosocial function measurement.

Results from the Phase 2 questionnaires

The Phase 2 questionnaires presented in Table 5 were designed to obtain information from the teams without the need to complete the Phase 1 questionnaires for each discipline. This resulted in the receipt of six other teams the agreed protocols that were used by their MDT (multidisciplinary team) for collection of core outcomes, and a response to a direct question about whether they felt that the timing of the ICHOM recommendation for minimum or core record collection was acceptable. The Phase 2 message enabled the evaluation of preferred outcomes from the following six centres. These were from Germany, France, Portugal, Hungary, Finland, and Austria (Table 5).

All of these centres in response to Q3 of the questionnaire provided further information about their centre protocol related to the question of core outcomes. As a result we are able to present responses from a OFC specialist HCP from every participant country involved in the ERN Cranio with only one exception Spain. In addition to this the for UK healthcare providers that where formerly participants in the ERN until Brexit excluded their participation from November 2020 their consensus agreed protocol for outcome measures across all UK CLP HCPs is recorded in the CRANE registry, and this is mentioned in the discussion. The responses for the six additional centres in Phase 2 are presented in Table 5 with the names of those who provided the responses on behalf of their CLP teams.

Discussion

Speech and language therapy

There is a general trend amongst the Brazilian, Italian, and Dutch teams to begin speech and language assessments at the age of 2–3.5 years, as described in their own protocols. Furthermore, these teams routinely assess the progress between the ages of 5 and 12 years due to the huge potential for development and the need for surgery during this period. The Dutch team has suggested that this period is too big a gap of time for not having routine measurements. The consensus is that outcomes should be recorded earlier and more regularly as a minimum requirement. In summary, while the ICHOM 5-year, 12-year, and end of treatment timings are compatible, HRAC (Brazil) and all the European centres

Table 5. Responses from the six additional European Reference Network (ERN Cranio) Healthcare Providers (HCPs)

Centre	Country	ICHOM acceptance	Comment summarizing their outcomes
Berlin	Germany	No	Oral health and mastication should be assessed by 10 years of age (*Siegmar Reinert, Tuebingen*)
Strasbourg	France	No	Responded 'yes' to the question relating to the modified table and provided detail of internal consensus study confirming MDT outcomes at ages 6, 10, and 15 years most popular. (*Bruno Grollemund*)
Lisbon	Portugal	No	Indicated preference to have dental health, occlusal, and COHIP completed at 10 years (*Maria Joao Alves de Castro*)
PECS	Hungary	No	Yes, I agree with the modifications of Maria Joao Alves de Castro (*Katalin Vajda*)
Helsinki	Finland	No	For speech and orthodontic outcomes 10 years is better than 12 years (*Arja Heliovaara*)
Salzburg	Austria	No	Routine records/outcomes-concerning SLT, ENT, orthodontics, and OMFS surgery are collected at 5, 10, and 15 years (final outcomes at 20 years). For SLT and ENT outcomes are routinely collected at 2 and 3 years *(Peter Schachner)*

which participated felt that there must be an earlier intervention and that more frequent outcomes are required to support optimum patient care.

Otolaryngology (ENT)

The Brazilian ENT specialist team from HRAC and Utrecht's UMCU participated in this questionnaire and the consensus gained for this discipline was that the ICHOM Standard Set is insufficient and starting records at 5 years of age is too late. It is recommended that ICHOM should review their outcomes within this domain/discipline and include earlier timings and more regular timings as a minimum requirement. In summary, it was felt that an earlier hearing assessment would be important to enable earlier intervention for infants who are living with clefts and have a hearing deficit.

Dentistry and orthodontics

There is a variety of tools used by the Brazilian, Italian, Dutch, and Swedish teams with regards to measuring dental and orthodontic outcomes but all have recorded outcomes between 8 and 10 years, which ICHOM does not include. The tools used by these teams include the Atack index (used at 5 years), GOSLON yardstick (used at and after 10 years), intra- and extra-oral radiographs, and photographs in order to fully assess the dental and occlusal development. ICHOM however only recommends measurement of DMFT, overjet and use of lateral cephalograms, which some have suggested that this is insufficient in three dimensions. Furthermore, there is unanimous support for the need to measure outcomes on ABG and that some institutions have made use of the Bergland Scale and radiographs to do this. ABG outcomes have been excluded by ICHOM.

While the ICHOM minimum outcomes for dentistry and orthodontics were largely compatible with the desires of the clinicians in all centres who responded, all felt the need to measure outcomes at other ages and have different instruments to facilitate optimal patient care. For example, all participants regard the assessment of ABG outcomes as being crucially important for CLP, the use of occlusal assessment apart from overjet is necessary and that dmft measurement for dental health at 5 years is much too late. As such, the dental and orthodontic components of the ICHOM Standard Set have been considered to be insufficient as a minimum requirement and it is recommended for further revision.

Surgery

The surgical teams from Brazil, Amsterdam, and Utrecht participated in this questionnaire and their protocols generally aligned with the ICHOM recommendations in measuring nasolabial aesthetics by use of clinical facial photographs. However, they also measure additional outcomes such as velopharyngeal function (in HRAC) and use other tools such as cephalograms, pantomograms, study casts, and the Cleft Aesthetic Rating Scale (in Amsterdam and Utrecht), which ICHOM have not included in their Standard Set recommendations. It should be borne in mind that in addition to nasolabial aesthetics, the orthodontic outcomes with respect to facial growth and the psychology outcomes for face, jaw, and teeth (and the CLEFT-Q measurement for these) are also surgical outcomes. Surgeons clearly believe that while the ICHOM recommendation of 5 and 12 years would be important, there is less rationale for measurements at strictly 8 years; this should instead be a developmental stage that is pre-ABG which can range from 8 to 11 years. Facial photographs and short questionnaires are relatively non-invasive and could be used to good effect to support optimum treatment. Comments were made regarding 3D facial photographs and intra-oral scanning for digital study models becoming routine in cleft units and in the future facial animation might be a useful tool.

In summary, the centres who responded agree that the surgical outcomes should be measured as per ICHOM recommendations at 5 years, 12 years, and end of treatment, but also at other times as part of the minimum outcome set.

Psychology

Between Milan and Amsterdam, there is quite a bit of variation in opinion about the use of the various psychosocial instruments and varying timings. Both centres have adopted the CLEFT-Q systems that ICHOM recommend, but Milan query whether all should be regarded as 'routine' or 'core', and they have shorter CLEFT-Q versions of their own. Amsterdam prefer earlier psychosocial assessment and query the 8-year CLEFT-Q Social Life. The consensus for psychology is that, while most of the ICHOM recommendations are coherent and useful, some areas require revision. These include involving parents during psychological assessments under the age of 8 years, and also that the use of CLEFT-Q questionnaires should be age-appropriate. The Dutch team have suggested that an 8-year-old might not be able to adequately and reliably complete certain CLEFT-Q questionnaires.

Experts in psychology accept that there are differences of opinion together with variability in patient needs; in combination with that, there is some room for flexibility around the ICHOM recommended timings. However, the key importance when implementing psychological assessments is also to obtain social determinants of health variables. This is to ensure that the data collected accurately represent the whole population and enables stratification.

Overall summary of suggested modifications

An extremely important principle in cleft care all over the world is standardized outcomes, and a core outcome set which should be looked upon as essential for audit/ clinical governance but also for inter-centre research in pursuit of quality improvement. In the field of CLP this has been used to good effect in the past as for decades Europe has demonstrated leadership and were participants in a previous attempt by the WHO in 2000 to achieve global consensus on an optimum core outcome set for OFC and this was adopted by teams all over the world including Europe. The implication of this is that many teams across the world have adopted the WHO core outcomes and future research on quality improvement is dependent on a continuum with past research and it is important that the legacy of decades of previous research data is sustained for the sake of all infants that are born with CLP.

The principle behind core outcomes measures is that care for the patient can be optimized via inter-centre comparisons. If a measure that could potentially improve patient care (by detection and intervention) is missing from the core outcomes this could potentially affect the achievement of optimal care and clearly should be addressed.

The ERNs would be an appropriate forum to obtain consensus as outcome measures and standards of care are integral elements of the ERNs and for each of these the objectives

are to progress the development of 1. standardized core outcome measures for all conditions and 2. to ensure that the core outcomes are simultaneously useful for clinical and research purposes, both being integral to the principles of setting up the ERNs.

Focussing on the areas of consensus that have emerged from this survey of opinion on core outcomes for orofacial clefts in general and ICHOM in particular, there appears to be sufficient common ground for agreement on a number of modifications that would enable overall consensus on the core outcomes for OFC that would improve patient care.

1. Speech and language therapy assessments at 3 years. Rationale: amenable to SLT intervention to improve clinical outcomes and be compatible with historic data.
2. Occlusal outcomes at 10 as opposed to 12 years. Rationale: amenable to Orthodontic intervention to improve clinical outcomes and be compatible with historic data.
3. Measurement of ABG outcomes as a developmental stage (e.g. pre-ABG) rather than a chronological time point. Rationale: ABG essential for optimum outcomes and intervention variable according to developmental status of canines and lateral incisors. This will range between 8 and 11 in terms of age, and is compatible with WHO recommendations and current practice.
4. Psychology Cleft-Q questionnaires (or modifications of Cleft-Q) based on individualized needs rather than rigid chronology and in the core outcome table could perhaps indicate that the measure should be recorded 'by the age of ….'.

It is noteworthy that none of the ERN HCP is who reported back have endorsed the ICHOM core outcome set as it stands at the moment. It is clear also that many of the European centres were signed up to the Eurocleft protocols before the establishment of the ERNs in 2015, and these outcomes were also endorsed by the WHO, and as a result are also being recommended in many countries beyond Europe. As long-established outcome protocols have been used in OFC governance and research over many years, there is understandably a reluctance to exchange established, tried, and tested protocols for a new non-validated set of core outcomes. There is however a clear willingness and commitment to the establishment of a consensus set of core outcomes as this is regarded as good governance within cleft care, and furthermore it is pragmatic that core outcomes are regarded as serving a dual purpose of being used for both clinical governance (audit) and research. This was a principle of the WHO consensus core outcome set and de-burdens both the patients and the MDT personnel and is regarded as being of overall benefit for everyone.

The overall findings of this survey therefore show that for OFC some aspects of ICHOM are compatible with current practice, but some are not. Clinicians are obliged to do the best for the patients under their care and therefore for recording patient outcomes that can optimize beneficial interventions across all specialties. Table 1 highlights the areas where modifications should be considered.

Conclusions

Practitioners with expertise in the cleft and craniofacial disciplines in the newly established ERN CRANIO have discussed and debated issues relating to OFC outcomes and in the context of that, the ICHOM protocols. All agree the importance of a set of core outcome measures, as this in turn influences standards of care, and the issue is whether ICHOM would be the chosen tool for outcome measurement or whether the ERN should seek modification or alternatives. This study is a step towards resolving these issues, and in a critical appraisal of the methodology there are some strengths and weaknesses.

This study is the most comprehensive study of OFC core outcomes for the last 2 decades. Being conducted via the ERN CRANIO Network where all participating centres have extensive experience in multidisciplinary cleft care, including a large centre outside of Europe (HRAC, Brazil) with a global reputation and an interest in OFC outcomes. It was a condition of participation that opinions be sought/obtained from the entire spectrum of care disciplines including speech/language therapy, otolaryngology, paediatric dentistry and orthodontics, surgery, and psychology. Thirdly, the study was able to capture and record a variety of different methodologies and care protocols from each participating centre for the measurement of outcomes.

However, this study is not without limitations, and the Phase 1 responses were limited to five participating centres, which was felt to be because the initial entry criteria for teams were extremely demanding. This prompted a Phase 2 questionnaire which brought input from a further six ERN Craino HCPs. This study has been largely confined to Europe, with only one centre outside of Europe (HRAC, Brazil) and none from the developing world. This contrasts with the 2002 WHO consensus statements that were approved in a WHO meeting in Geneva with representation from every continent.

Despite the wide range of views on CL/P outcomes collected, some elements of consensus have emerged from this study. For example, all participating centres agree that in speech and language therapy, there should be an earlier, and thereafter more regular, assessment of outcomes before 5 years as per ICHOM recommendations. Another consensus within dentistry and orthodontics considered the measurement of ABG outcomes as crucial in CL/P care, and all agree that 8-year outcomes should instead be revised as a developmental stage (e.g. pre-ABG) rather than a chronological time point, and by consensus 10 years was preferred to 12 years for outcome measures. In otolaryngology, agreement was less clear and therefore no absolute consensus in terms of chronology could be identified, and in psychology Cleft-Q questionnaires were well received by most with some favouring shorter questionnaires, and the principles of individualized needs and flexibility within the system seem to be important. However, this provides optimism and hope that overall consensus on core outcomes can be achieved with continued open discussions among all interested parties in CL/P care.

The overall findings therefore are that ICHOM is considered insufficient for recording patient outcomes beneficially across all specialties in its current form and version. The aim must be to be in a position to compare data between cleft centres worldwide as the WHO consensus statements had agreed, and while this study shows that some ICHOM measures are agreeable in some specialties, there was consensus that some other ICHOM proposals are unacceptable or inappropriate. The ERNs could be an appropriate forum to obtain consensus as outcome measures and standards of care are integral elements of their strategy, and standardized

core outcome measures for all conditions are accepted as being essential.

Finally, it is acknowledged in the field that dialogue on outcome measures should be a dynamic as opposed to a static process as surgical techniques, multidisciplinary care protocols and technology evolves, and advances in research provide new approaches to achieving optimal outcomes. Therefore the dialogue on core outcomes and the technology used to measure and record them for OFC should be ongoing within and between all disciplines involved in patient care.

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Conflicts of interest

The authors declare that we have no conflict of interest with regard to this paper and the information it contains.

Data availability

The authors confirm that the data supporting the findings of this study are available within the article and its supplementary materials (Tables 1–5).

Statement on ethics

Ethics approval was not required for this as there is no patient data, and no requirement for anonymization. All of the information obtained through the questionnaires issued to the clinical teams is included in this manuscript, and in the event of publication all of the information is freely available and in the public domain.

Supplementary material

Supplementary material is available at *European Journal of Orthodontics* online.

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