Achieving consensus in the measurement of psychological adjustment to cleft lip and/or palate at age 8 years+

Abstract

Background: Consensus regarding optimal outcome measurement has been identified as one of the most important, yet most challenging developments for the future of cleft lip and/or palate (CL/P) services. In 2011, a process began to adopt a shared conceptual framework and to identify a set of core outcome measures for the comprehensive assessment of psychological adjustment.

Objectives: The aim of the current article is to outline the collaborative process used to achieve consensus in the academic and clinical measurement of psychological adjustment to CL/P from the age of eight years onwards.

Results: A conceptual framework and corresponding parent- and self-reported outcome measures for use at ages 8, 10, 12, 15, 18, 20, and 25 years have been agreed upon by clinicians, researchers, and patient and parent representatives. All measures have been evaluated according to their psychometric properties, clinical utility, ability to produce meaningful longitudinal data, and a range of pragmatic considerations.

Conclusions: Although the collaborative process has been challenging and has required ongoing dedication from multiple stakeholders, consistency in data collection over time will allow for key research questions in CL/P to be addressed, both in the UK and internationally. The process has also demonstrated the clinical utility of the measures and the potential for the gradual integration of the measures into clinical practice. UK progress has sparked global interest, and the adaptation of the framework and its corresponding measures for worldwide use is now a prominent focus.

Keywords: cleft lip and palate, psychological wellbeing, outcome measurement; collaboration; adjustment

Introduction

Consensus among clinicians, researchers, and patients regarding optimal outcome measurement has been identified as one of the most important developments for the future of cleft lip and/or palate (CL/P) services (Klassen et al., 2012; Stock et al., 2018). This is a particularly critical goal for the field of psychosocial adjustment, which has received relatively little attention despite its importance in the delivery of patient-centred care across all disciplines. Due to the multifactorial and fluctuating nature of psychosocial adjustment, a comprehensive assessment is difficult to achieve, and consensus as to the most appropriate outcome measurements has previously been difficult to establish (Stock et al., 2018).

The Cleft Collective Cohort Studies, an initiative of the Scar Free Foundation (www.scarfree.org.uk), were established in the United Kingdom (UK) in 2012 (see Stock, Humphries et al., 2016). In order to build a dataset that would have the potential to address the key research questions important to all stakeholders (Petit-Zeman & Cowan, 2013), it was crucial to achieve consensus in data collection among all project partners, including clinicians from UK CL/P teams, researchers, and individuals born with CL/P and their families. In response to this challenge, the researchers reviewed the existing literature and potential measures extensively, drew upon the clinical experience of those working in the field of CL/P, and consulted with patient and parent representatives. In the case of psychosocial adjustment, a research subgroup comprising two researchers and four clinically based psychosocial specialists was also established. A paper outlining the subgroup's initial progress in relation to the design of a conceptual framework and the identification of appropriate parent-reported measures for use during the first years of the child's life was published in 2016 (Stock, Hammond et al., 2016).

Building on this previous work, the research subgroup recruited additional members and set out to develop of a set of psychological outcome measures for use at age eight years and above, including measures that could be completed by the patient themselves. The present article outlines the collaborative process used to achieve consensus in the academic and clinical measurement of

psychological adjustment to CL/P from the age of eight years onwards. In addition, this article will consider the associated challenges of this process, as well as potential future opportunities.

Achieving Consensus

A Review and Extension of the Evidence Base

An up-to-date comprehensive review of the existing literature was carried out in order to clarify the key factors thought to contribute to psychological adjustment during late childhood, adolescence, and early adulthood (Stock & Feragen, 2016). In some cases, knowledge was found to be scarce, and therefore a number of additional studies were conducted (e.g. Stock, Feragen et al., 2016; Stock & Ridley, 2018; Stock et al., 2018; *unpublished thesis*). These combined activities confirmed the components previously included in the conceptual framework (Table 1) and identified a series of potential outcome measures for consideration.

Choosing Measures

All potential measures were evaluated according to their psychometric properties, clinical utility, and a range of pragmatic considerations (also see Tables 2 and 3). To be considered for inclusion, measures needed to possess at least 'acceptable' levels of validity and reliability, and to have age-comparable USA/European normative data and/or cut-off scores available. Wherever possible, freely available measures were chosen. Where there were cost implications, measures with no fee for hospitals were preferred. Existing literature and clinical experience were used to assess the anticipated amount of clinically relevant information to be gained from each measure. The number of items and ease of scoring were also considered, both for individual measures and the set as a whole. Where available, measures with 10 items or less were favoured. Finally, measures were evaluated according to their ability to produce longitudinal data capable of underpinning meaningful comparisons across age groups. In the case that not all measures met all the inclusion criteria, the

relative contributions of each measure to the overall set were weighed and the ability of the overall set to meet the requirements of the conceptual framework were assessed.

To determine the age at which self-report was appropriate, current guidance surrounding research with children was reviewed (e.g. Barnard et al., 2012; Modi et al., 2014). This included practical and ethical factors (e.g. the length of time involved in questionnaire completion and the potentially emotive content of the measures), children's cognitive ability, and the anticipated utility of the data collected. It was subsequently agreed among stakeholders that self-report measures would be recommended for use with children from the age of eight years. The group also concluded that parent-report measures would not be recommended after their child reached the age of 18 years.

Once the subgroup had selected the measures, progress was shared with the wider Psychology Clinical Excellence Network of the Craniofacial Society of Great Britain and Ireland to gather additional feedback.

Public Involvement

The next stage was to assess the acceptability of the chosen measures from the perspectives of patient and parent representatives. A multidisciplinary workshop was held in September 2017 in collaboration with the Cleft Lip and Palate Association (www.clapa.com). At this workshop, parents/caregivers and young adults born with CL/P provided their feedback on the conceptual framework and the proposed outcome measures, as well as on the future research agenda of the Cleft Collective Cohort Studies more broadly. First, the researchers gave presentations on the work carried out to date and outlined future goals. Delegates then participated in an hour-long group discussion on the pros, cons, suitability, and practicalities of pursuing the research agenda in the proposed ways. Next, delegates received copies of the draft framework and outcome measures and discussed these materials in smaller focus groups lasting approximately 90 minutes. All discussions were audio recorded with delegates' permission. Following the workshop, the audio recordings were transcribed verbatim and key points were extracted and actioned. Workshop delegates were also sent a lay

summary of the workshop outcomes and invited to comment further if they wished. Once all amendments had been made, a teleconference was held with parents/caregivers and young adults born with CL/P to confirm the suitability of the final version.

Achievements to Date

This process involved a thorough examination of generic and condition-specific constructs, as well as the corresponding outcome measures currently available. This unique collaboration between clinicians with experience of working with patients and families throughout the CL/P treatment pathway and researchers with expertise in outcome measurement and applied research in clinical settings has ensured that the measures set has both face validity and scientific rigor (see Tables 2 and 3). This method of working has also increased the buy-in from clinical staff to participating in The Cleft Collective Cohort Studies and other research projects. The authors recommend this collaborative approach as being highly beneficial for all stakeholders and in the interest of advancing the field as a whole.

Data obtained from the measures are already being collected successfully from parents and children enrolled in The Cleft Collective Cohort Studies. Analysis of the data collected from parents during the first 18 months has successfully demonstrated the research and clinical utility of the initial set of measures recommended for use from the point of diagnosis (see Stock, Hammond et al., 2016). Specifically, the measures set has been used to assess parental wellbeing and child development compared to the general population, to evaluate parents' satisfaction with healthcare, and to identify the risk and protective factors that contribute to psychological distress in parents (Stock et al., 2019; *manuscript in press; manuscript under review*). From the age of eight years onward, the recommended set also includes self-report measures for completion by individuals born with CL/P.

The Clinical Psychologists working in UK CL/P teams have begun to integrate measures for use at ages 5 and 10 years into the routine national audit process, cementing the link between research data,

audit data, and clinical practice. While not without its challenges, this new protocol has provided clinicians and researchers with joint access to the same data, thereby minimising the burden of questionnaire completion on families and boosting efforts to achieve a standardised, comprehensive psychology audit across the UK. The further use of these measures at additional time points within clinical practice to assess psychological wellbeing and treatment outcomes, as well as the impact of psychological interventions, is currently being discussed.

Finally, and given recent work demonstrating that the various craniofacial conditions may have many aspects in common (Stock & Feragen, 2019), the measures are now being used in a series of other large-scale investigations of previously under-studied craniofacial populations, including adults with CL/P, patients and families affected by craniosynostosis, and patients and families affected by craniofacial microsomia. The expansion of this work to other craniofacial conditions will be crucial in advancing knowledge in these areas, as well as providing insight into the overlapping and distinct aspects of the various craniofacial diagnoses.

Challenges and Considerations

The challenges involved in achieving consensus among various stakeholders in relation to a complex task such as outcome measurement should not be underestimated. This process constituted a significant undertaking, presenting the group with a number of challenges and requiring regular telephone and in-person collaborative meetings over a total of eight years.

Once key developmental stages had been established, measures were selected to capture parent-reported data pertaining to parental wellbeing and child development. The measures set includes a combination of generic and condition-specific measures so as to capture normative experiences and access to population norms, as well as the intricacies of the condition itself. In finalising the set of measures, a key priority was to avoid placing unnecessary burden on the patients and families

completing them while maximising clinical utility; thus, choices regarding inclusion needed to be highly stringent.

In the absence of a condition-specific measure that met the inclusion criteria at the time (also see Klassen et al., 2012), the group chose to design a brief measure to tap into key aspects of psychological adjustment not measured elsewhere in the set. This measure was based on points of consensus identified by the literature review, the additional qualitative studies, and the combined experience of the clinical psychologists and patient/parent representatives. Since this work was completed, two additional measures for specific use with the CL/P population have been proposed. The Psychosocial Assessment Tool-Craniofacial Version (PAT-CV; Crerand et al., 2018) is a brief screen of psychosocial risk in eight domains and was adapted for use with craniofacial populations. The Cleft-Q (Wong-Riff et al., 2018) is designed to evaluate outcomes related to an individual's satisfaction with appearance, health-related quality of life, and facial function. Both measures are currently deemed too long to include in the set, but as more information on these measures becomes available (Klassen et al., 2018; Kapa et al., 2019), individual subscales may be considered for inclusion in the future.

Future Ambitions and Opportunities

The Global Task Force for Holistic Outcomes is an initiative of the American Cleft Palate-Craniofacial Association. In 2013, the Task Force set out to promote a shared framework for the global measurement of patient-centred psychosocial outcomes in craniofacial care, inclusive of differing healthcare systems and levels of resource. Building on the work described above, the first step was to carry out consultation with health professionals around the world to assess whether the UK conceptual framework could also be applied in other countries. Health professionals from more than 20 countries representing a range of disciplinary backgrounds participated, and the exercise confirmed the global applicability of the UK framework and the corresponding outcome measures.

The Task Force subsequently designed a tiered outcome measurement system (Tier 1 = low resource; Tier 2 = medium resource; Tier 3 = high resource) to allow for all countries to participate in data collection at their chosen level. The tiered system has been successfully trialled in a number of countries, with participating clinicians reporting the set to be relatively easy to integrate into clinical practice, to improve communication between clinicians and patients, and to provide clinically informative data capable of aiding treatment decision-making. This tiered outcome measures set, alongside supplementary materials and training modules will soon be made available via a specifically designed website[1]. In the interests of continued collaboration and advancing the field, these materials will be available free of charge. A future ambition of the Global Task Force includes the translation of the materials into several languages. If widely adopted, this approach could not only increase awareness of psychosocial issues and how to address them among craniofacial teams around the world, but could help the community to build a large dataset that can be directly compared across countries, cultures, conditions, and healthcare systems.

Conclusions

Driven by a comprehensive conceptual framework comprising six key domains of adjustment, this unique collaboration has produced a core set of standardised measures that are applicable across age groups and meet stringent criteria in regard to psychometric properties, clinical utility, and pragmatic considerations. Although this process has been challenging and has required ongoing dedication from multiple stakeholders, consistency in data collection over time will allow for key research questions in CL/P to be addressed and will support a foundation for integrating standardised patient-centred outcome measurement in both the UK and internationally.

[1]Please contact the corresponding author for further information.

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