Achieving consensus in the measurement of psychological adjustment to cleft lip and/or palate

Abstract

Background: Psychological adjustment to cleft lip/palate is multifaceted, and can fluctuate over time and across different situations. Consequently, a comprehensive understanding of adjustment is difficult to capture, and the challenge of achieving consensus among researchers and clinicians regarding key constructs and processes is considerable. Numerous measures have been used in research and clinical audit, resulting in conflicting findings and difficulties in evidencing the value of psychological intervention. The launch of the world's largest cleft lip/palate cohort study has provided an opportunity to standardise data collection across the UK.

Objective: To describe the collaborative process used to achieve consensus in the academic and clinical measurement of psychological adjustment to cleft lip/palate.

Results: Extensive work based on existing literature and clinical experience has resulted in a conceptual framework comprising six key domains of adjustment and corresponding risk/protective factors, measureable across key developmental time points. Driven by this framework, a core pack of standardised measures has been selected according to psychometric properties, clinical utility and pragmatic considerations.

Conclusions: To date, these measures have been implemented within a UK-wide longitudinal cohort study (at diagnosis, 18 months, 3 years, 5 years and 8 years) and adopted into the national routine clinical audit protocol for cleft lip/palate at age five. Further data collection points will follow as the cohorts age. Over time, consistency in data collection will allow researchers to address some of the key unanswered questions in relation to psychological adjustment to cleft lip/palate.

Key words: cleft, psychology, adjustment, measurement

Introduction

Psychological adjustment to a cleft lip and/or palate (CL/P) is multifaceted, involving many different and interrelated constructs (see Rumsey and Stock, 2013; Kapp-Simon, 2006; Hunt et al., 2005). In addition, adjustment is labile and can fluctuate over time and across different situations (Appearance Research Collaboration; ARC, 2009). Many of the factors and life events which may affect adjustment to CL/P also apply to the general population, and without clinical cut-off scores and appropriate normative data it can be challenging to distinguish between 'normal' development and the psychological impact of CL/P (see Stock et al., in press 2015a; Rumsey and Harcourt, 2012). Consequently, a comprehensive understanding of adjustment is difficult to capture, and achieving consensus among researchers and clinicians regarding the key constructs and processes is a significant challenge. A plethora of approaches and measures have been utilised within research and audit as a consequence. To date, this diversity in the measurement of adjustment to CL/P has resulted in conflicting research findings and difficulties in evidencing the value of psychological intervention (Norman et al., in press; Rumsey and Stock, 2013; Klassen et al., 2012). In addition, the quality of the evidence-base is hampered by the challenges of obtaining large, representative samples and an over-reliance on cross-sectional and/or retrospective data (Rumsey and Stock, 2013; Hunt et al., 2005). In order to combat many of these methodological challenges and to create a resource to address some of the key research questions important to all stakeholders (also see Petit-Zeman and Cowan, 2013), a national CL/P research programme was launched in 2012.

The Cleft Collective is the world's largest CL/P research programme to date and is funded by a pioneering UK national fundraising charity, the Healing Foundation (www.thehealingfoundation.org.uk). An integral part of this initiative is the establishment of a UK-wide longitudinal cohort study, designed to collect biological samples and data pertaining to family environment, medical history, treatment experience, educational achievement and psychological wellbeing from families affected by CL/P over time. The study intends to address three key questions that parents often ask following a diagnosis of CL/P in their child: 1) What caused my child's cleft? 2) What are the best treatments for my child? 3) Will my child be OK, both now and in the long term? For further information about The Cleft Collective, see Stock et al., in press 2015b, or visit www.cleftcollective.org.uk.

The focus on psychological adjustment within The Cleft Collective research programme resulted in an imperative to achieve consensus regarding key constructs and associated measures between the clinical psychologists delivering cleft care and the research team. The aims of this paper are to describe the collaborative process used to achieve consensus in the academic and clinical measurement of psychological adjustment to CL/P, and to consider the associated challenges and potential future opportunities.

Achieving consensus

A review of the evidence base

The process began in 2010 with a thorough review of the existing literature in the field of psychological adjustment to CL/P (see Rumsey and Stock, 2013 for an updated summary). This review identified a number of emerging areas of consensus, alongside several gaps in the knowledge base. In addition, the review found more than 60 different measures being employed across tenuous age groups in attempts to capture various aspects of adjustment, rendering findings almost impossible to collate and compare. The review drew into sharp focus the lack of consensus regarding what constitutes a positive psychosocial outcome for the CL/P population, and how and when to measure the factors contributing to this outcome.

Clinical experience

The findings of the review were presented to the Psychology Special Interest Group (SIG) of the Craniofacial Society of Great Britain and Ireland in 2011, along with the proposal that the clinical psychologists and the research team should collaborate to address the challenges identified. Subsequently, we began working together to identify a desired 'end' point (i.e. 'what characteristics might a well-adjusted adult born with CL/P possess?'). In attempting to categorise the issues we believed to be important, derived from both the literature review and clinical discussions, we identified six key domains of adjustment (social functioning, world view, appearance, vocational milestones, psychological wellbeing and condition-specific factors), along with several corresponding risk/protective factors contributing to each domain (see Table 1). The chosen domains were encompassing of each factor we had identified, and were comparable to those detailed in previous literature reviews in the field (e.g. Hunt et al., 2005) and to work conducted in the wider field of visible difference (e.g.

ARC, 2009). These domains have also been corroborated by subsequent qualitative work (e.g. Stock et al., in press 2015a).

We were then able to work through the lifespan to ascertain the key developmental time points at which these risk/protective factors may play an important role. These extensive structured discussions over the course of one year allowed us to develop a conceptual framework of psychological adjustment to CL/P (see Table 1).

Choosing measures

In order to choose measures which mapped onto the conceptual framework, we used the findings of the literature review to identify all of the existing measures known to have been used in research or clinical practice in the measurement of psychological adjustment to CL/P and other similar health conditions. This provided us with an extensive collection of measures which we subsequently evaluated in relation to psychometric properties (e.g. validity and reliability figures, normative data and clinical cut-off scores), clinical utility (e.g. applicability to the patient, clinically relevant information gained) and pragmatic considerations (e.g. cost and availability, time taken to complete and score). This resulted in a core pack of standardised, age-appropriate measures, aimed at eliciting data applicable to both clinicians and researchers.

Patient and Public Involvement

The collaborative process was also informed by a number of qualitative investigations, aimed at gauging the feasibility of implementing The Cleft Collective Cohort Studies and informing the conceptual basis of the framework (for example, see Stock et al., in press 2015a; Stock and Rumsey, 2015; Williams et al., 2012).

In order to test how acceptable the chosen measures would be to the families completing them, we asked a panel of eight parent representatives to review the pack of measures and provide feedback during a specially held PPI workshop. These representatives also completed the pack of psychology measures, indicating an average completion time of 20-25 minutes. Overall, representatives reported the length and content of the pack to be acceptable and relevant to their experiences, and expressed that they would be prepared to complete the pack of measures at regular intervals throughout their child's developmental trajectory.

Achievements to date

To the authors' knowledge, this has been the most thorough examination of both generic and cleft-specific constructs and of the corresponding measures available to date. This unique collaboration between clinicians with experience of working with families throughout the CL/P treatment pathway, and researchers with expertise in outcome measurement and applied research in clinical settings, has ensured the measures pack has both face validity and scientific rigour. This collaborative method has also increased the 'buy-in' from clinical staff in relation to taking part in this process. The authors recommended this approach as being highly beneficial.

Measures have thus far been agreed for several key developmental time points (see Table 2 for a summary of the measures chosen to date and Table 3 for a timeline of these assessments and their stage of development). Data obtained from these measures are already being collected successfully from parents enrolled in The Cleft Collective Cohort Studies shortly after the birth of their child, and when their child is 18 months old and five years old. Additional data collection from parents has already been agreed around the time of an antenatal diagnosis, at three years and at eight years.

The measures agreed for use for when the child is aged five years have been integrated into the existing routine clinics carried out as part of the national audit, cementing the link between research data, audit data and clinical practice. This new protocol has provided clinicians and researchers with joint access to the same data, minimising the burden of questionnaire completion on families and standardising the five-year-old psychology audit across the UK.

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 collaborative meetings over the course of three years (accurate at the time of writing). Initially, the group consisted of more than 30 clinicians and researchers with a special interest in CL/P. Working in such a large group was useful to begin with, although

later became less workable as the task became more intricate. A CL/P psychology research 'sub-group' was subsequently formed, consisting of two research psychologists, one outreach counsellor and three clinical psychologists (authors of the current paper) from different cleft teams across the UK, allowing for the management of several different representative viewpoints.

Having selected key developmental stages, measures were selected to capture parent-reported data pertaining to both parental wellbeing and child development. The measures pack includes a combination of generic and condition-specific measures, so as to capture both 'normative' experiences as well as the intricacies of the condition itself. In finalising the pack of measures, a key priority was to avoid placing unnecessary burden on the patients and families completing them, while maximising clinical utility, and thus our choices regarding measure inclusion needed to be stringent. In the absence of a condition-specific measure meeting our inclusion criteria was available at the time (also see Klassen et al., 2012), we chose to design our own brief measure to tap into key aspects of psychological adjustment to CL/P. This measure was based on points of consensus identified by the literature review and the considerable combined experience of the clinical psychologists. As with all of the questionnaires, this measure was also piloted with age-appropriate parent representatives and adjusted according to the feedback received.

The logistics of integrating the resulting pack of measures into the national audit at age five have been challenging to negotiate and have required significant changes in the way audit data are collected and acted upon. Although little time is required from professionals to administer the questionnaire pack, it is currently taking cleft teams a long time to score each questionnaire and input the data electronically. It is hoped that this will be reduced over time as cleft teams become more familiar with the new audit protocol. A generic database has recently been designed to support data entry and the interpretation of data in a clinically meaningful way. Despite early teething problems, this change in protocol has made more complete datasets available for audit purposes and has already allowed psychologists to identify and address clinically significant levels of reported difficulties which are unlikely to have been identified prior to the measures being implemented (for example, high levels of parental stress). The process has also allowed for the psychology audit to be harmonised across cleft teams, increasing the opportunities for comprehensive and consistent data collection across the UK.

Finally, the benefits of involving parent and patient representatives from the beginning of a project are now well recognised (INVOLVE, 2013). It is acknowledged that the involvement of parents and patients in designing the conceptual framework was not as comprehensive as the authors would have liked, and the process would have been enhanced had the level of parent and patient involvement been augmented. Although neither clinical experience nor the findings of qualitative research with patients and parents should replace active parent and patient participation, great care was taken to incorporate the patient view and to encompass the concepts and time points important and relevant to those affected by CL/P and their families.

Future ambitions and opportunities

Meetings are currently being held to discuss data collection at future time points within The Cleft Collective Cohort Studies. Provisionally, these time points will include age ten, age 12, age 15, age 18 and age 25. After careful consideration, ethical approval is currently being sought to elicit self-reported data from those children with CL/P who are enrolled in the Cohort Studies from the age of eight years onwards.

Over time, The Cleft Collective Cohort Studies will generate one of the largest CL/P data banks in the world. If successful, this longitudinal resource will offer a unique opportunity to address some of the key unanswered questions in CL/P research. These data will be available to clinicians and researchers both within and outside of the UK to undertake ethically approved research projects to address clinical questions. For example, what are the short and long term impacts of having a child with CL/P on family functioning? What is the impact of CL/P on parent-infant interactions? How does early development impact upon education achievement? How are social relationships affected by CL/P? What is the overlap between disciplines (such as psychology and speech)? What types of psychological intervention, and at what time, are helpful for individuals with CL/P and their families? Ultimately, the research findings will be shared with all stakeholders, and healthcare providers will be supported to incorporate these findings into clinical practice around the world.

The new national audit protocol at age five is one example of how research and clinical practice may be integrated. In the future, it is hoped that a similar process can be applied to other existing UK audit points. The collection of consistent and standardised data which can be mapped onto a conceptual framework and utilised

within and between cleft teams will help to demonstrate the need for, and accomplishments of, psychological input within CL/P services.

In order to support this process, as well as the execution of clinically relevant and applied research within healthcare settings more generally, it is acknowledged that additional research staff must be employed to enrol patients and families into studies and to enable data collection. Additional support to facilitate research within clinical settings is crucial, since existing clinical staff are severely stretched. In the future, it is hoped that taking part in research and/or comprehensive audit will be incorporated into the routine treatment pathway, making the collection of large, longitudinal datasets a standard part of the treatment process.

In the UK, clinicians from other disciplines involved in the treatment of patients born with CL/P are also collaborating with researchers from The Cleft Collective team to achieve consensus in outcome measurement. Further afield, this knowledge and experience will be shared with clinicians and researchers in both Europe and globally, in the hope that this approach may provide a foundation for the integration of standardised patient-centred outcome measurement around the world, following appropriate adaptations to reflect differences in culture and resource. As a first step towards this goal, the conceptual framework and agreed measures are currently being incorporated into a tiered approach to capturing patient-centred data, varying from a series of straightforward questions (level 1, appropriate for cleft teams with few resources) to a comprehensive set of measures (level 4). This framework will be offered to members of international cleft collaborations for discussion.

Summary and conclusion

The launch of the world's largest cleft lip/palate research programme, The Cleft Collective, has provided the platform for clinicians and researchers to achieve consensus in the measurement of psychological adjustment to CL/P. Driven by a comprehensive conceptual framework comprising six key domains of adjustment, this unique collaboration has produced a core pack of standardised measures which are applicable across the lifespan and in accordance with appropriate psychometric properties, clinical utility and pragmatic considerations. To date, these measures have been implemented within a UK-wide longitudinal cohort study

and adopted into the national routine audit protocol for CL/P at age five. Although not without its challenges, it is anticipated that this approach will allow researchers and clinicians to address some of the key unanswered questions in relation to psychological adjustment in the future, as well as support the ongoing collection of consistent audit data across UK cleft teams, providing a foundation for the integration of standardised patient-centred outcome measurement in countries around the world.

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