Abstract

While cleft lip and/or palate (CL/P) has been a focus for psychological research for a number of years, investigation of adjustment to other, rarer craniofacial anomalies (CFAs) has been scarce. Yet, many features of the journey experienced by patients with CL/P could also have relevance for those affected by other CFAs, and vice versa. This brief article summarises the literature pertaining to psychological adjustment across both craniofacial groups, as represented by several prominent review articles. Similarities across the two patient groups in relation to key domains of psychological adjustment and corresponding factors are identified. Consequently, a standardised approach to measurement across all congenital craniofacial conditions is proposed, alongside suggestions for the potential application of similar intervention frameworks and clinical approaches.

Key words: cleft, craniofacial, measurement, intervention, psychological adjustment
Historically, research investigating psychological adjustment to congenital craniofacial conditions has focused predominantly on those affected by cleft lip and/or palate (CL/P), while other craniofacial anomalies (CFAs), such as syndromic and non-syndromic forms of craniosynostosis, craniofacial microsomia, and Treacher Collins Syndrome, have received much less attention. Nonetheless, many features of the patient journey for those affected by CL/P could be thought to be broadly generalisable to those experienced by patients with other CFAs, and vice versa. This brief article will outline the main research findings captured by several prominent reviews of psychological adjustment to CL/P and other CFAs, with a view to identifying any similarities and discrepancies across the two patient groups in relation to key psychological domains.

Review articles were identified using a range of key search terms (see Table 1 and 2) contained within the PsycInfo, MEDLINE, Science Direct, Web of Science and CINAHL databases. In the absence of conventionally agreed domains and definitions of adjustment, the choice of domains was guided by recent literature (e.g. see Klassen et al., 2012; Stock et al., 2016). Domains include social interaction, emotional wellbeing, behaviour, cognitive development, appearance, treatment experiences, and overall adjustment and quality of life. The potential implications of these findings for outcome measurement and psychological intervention will be discussed.

Social integration

The birth of a child with a craniofacial anomaly can be a distressing time for parents (Nelson et al., 2012), and literature in the area of CL/P has thus investigated the impact of this condition on parent-infant interactions and attachment. While several reviews of the literature have found few difficulties overall, attachment representations among children with CL/P do appear to be less stable over time (Hunt et al., 2005; Collett and Speltz, 2007; Stock and Feragen, 2016). Attachment has not yet been investigated in the context of other CFAs, and thus research in this area is needed. During childhood, studies have suggested that those with CL/P appear socially inhibited when compared to their unaffected peers, demonstrating a less responsive and less assertive pattern of communication (see Lockhart, 2003; Collett and Speltz, 2007; Stock and Feragen, 2016). In the case of other CFAs, affected children have also been judged to be less socially competent than their peers (see Feragen and Stock, 2017). In adolescence, reviews have described those affected by CL/P to have interpersonal difficulties, fewer friends, and to be less likely to participate in social activities (Hunt et al., 2005; Collett and Speltz, 2007; Richman et al., 2012). Adolescents with CL/P have also reported unwanted reactions
and teasing in relation to appearance and speech, as well as challenges in initiating romantic relationships (see Turner et al., 1998; Lockhart, 2003; Stock and Feragen, 2016). There is evidence of similar findings among adolescents with other CFAs, with patients reporting lower ratings of social acceptance, perceived stigma, teasing, and concerns related to finding a life partner who is accepting of their condition (see Feragen and Stock, 2017). In both those with CL/P and those with a CFA, reviews have concluded that a subgroup of affected adults may experience social anxiety and avoidance (Turner et al., 1998; Richman et al., 2012; Feragen and Stock, 2017), and may be less likely to get married or to have children of their own (Hunt et al., 2005; Wehby and Cassell, 2010; Feragen and Stock, 2017). Recent research with both groups has also provided evidence of good social functioning overall, as well as factors which may protect individuals from psychosocial distress, such as close friendships, support from family members, and meeting others with the same condition (see Stock and Feragen, 2016; Feragen and Stock, 2017).

Emotional wellbeing
While some studies have identified higher levels of anxiety and depression among individuals with CL/P, others have found few differences when comparing those with CL/P to their same-aged peers (see Lockhart et al., 2003; Hunt et al., 2005; Stock and Feragen, 2016). In addition, some reviews have found self-esteem to be good overall (Hunt et al., 2005), while others have concluded that confidence can be significantly impacted by the condition (Lockhart, 2003). A similar pattern is true for research in the case of other CFAs (see Feragen and Stock, 2017), with positive findings in both patient groups attributed to the use of effective coping strategies in a number of studies (see Stock and Feragen, 2016; Feragen and Stock, 2017).

Behaviour
Although children with CL/P have been suggested to have increased levels of internalising and/or externalising behaviours when compared to their unaffected peers or siblings, reviews of the literature have presented these findings as being largely inconclusive (Collett and Speltz, 2007; Klassen et al., 2012; Stock and Feragen, 2016). Similarly, some research within the area of CFAs has identified a considerable increase in the prevalence of behavioural difficulties, while other studies report similar behavioural scores to that of control groups (see Speltz et al., 2004; Knight et al., 2014; Feragen and Stock, 2017). Several studies investigating CFAs have
suggested that behavioural problems may in fact be a result of underlying cognitive impairments (see Feragen and Stock, 2017), and given the known frequency of such impairments among individuals with CL/P (see Richman et al., 2012; Stock and Feragen, 2016), the same could also be true for CL/P.

**Cognitive development and educational achievement**

In relation to both CL/P and CFAs, global IQ has been found to be within the normal range in some reviews (Collett and Speltz, 2007; Kapp-Simon et al., 2007; Knight et al., 2014). Nonetheless, a considerable percentage of those affected are reported to have at least one additional condition, such as a learning disability, language impairment, or developmental delay (for an overview see Speltz et al., 2004; Kapp-Simon et al., 2007; Richman et al., 2012; Stock and Feragen, 2016). A neurobiological foundation for cognitive difficulties has been suggested across both patient groups (see Collett and Speltz, 2007; Kapp-Simon et al., 2007; Richman et al., 2012). Educational attendance, academic performance and job salary were found to be affected in a number of reviews of CL/P and other CFAs (Kapp-Simon et al., 2007; Richman et al., 2012; Knight et al., 2014), while others have concluded that individuals with CL/P have a comparable educational and employment status to that of their unaffected peers, and report a high level of job satisfaction (Turner et al., 1998; Lockhart, 2003).

**Appearance**

Review articles in both research areas have described the challenges of living with an unusual appearance, and have discussed the possible impact of a visible condition on appraisals of appearance, particularly during adolescence (Turner et al., 1998; Lockhart, 2003; Collett and Speltz, 2007; Feragen and Stock, 2017). Nonetheless, a number of reviews have concluded that subjective satisfaction with appearance and body image among those with CL/P and with CFAs is positive overall (Hunt et al., 2005; Stock and Feragen, 2016; Feragen and Stock, 2017), while also recognising some dissatisfaction with the facial features directly affected by the condition (Klassen et al., 2012; Plomp et al., 2016; Stock and Feragen, 2016). Within the CL/P literature, dissatisfaction with appearance has been linked to a desire for further surgery (see Hunt et al., 2005; de Sousa et al., 2009; Stock and Feragen, 2016), which may be problematic if the patient has unrealistic expectations of the possible outcomes (see Turner et al., 1998). The additional challenge of adjusting to an altered appearance
following surgery has been identified in patients with CL/P and with other CFAs (see Collett and Speltz, 2007; de Sousa et al., 2009; Feragen and Stock, 2017).

**Treatment experiences**

Reviews of the literature pertaining to patients born with CL/P have discussed the importance of patient autonomy within the treatment pathway (Lockhart, 2003; Collett and Speltz, 2007; Stock and Feragen, 2016). This is particularly pertinent during adolescence, when the patient becomes more able to discuss treatment decisions with their parents and medical staff (see Collett and Speltz, 2007); a finding also identified by research with other CFAs (see Feragen and Stock, 2017). Both research areas have reported that treatment can be intimidating, demanding, and not always perceived as worthwhile by the patient (Collett and Speltz, 2007; Stock and Feragen, 2016; Feragen and Stock, in press). In the case of CFAs, (Feragen and Stock, 2017) identified a paucity of studies measuring patients’ perceptions of treatment decisions and outcomes, and suggested that more research in this area was necessary.

**Overall adjustment and quality of life**

A number of recent reviews have concluded that overall adjustment seems to be within the normal range for those affected by a range of craniofacial conditions, and report those affected by CL/P and other CFAs to have positive perceptions of their quality of life (Hunt et al., 2005; Stock and Feragen, 2016; Feragen and Stock, 2017). Nonetheless, dissatisfaction with appearance, poor physical health, functional difficulties, unwanted social reactions, distressing treatment experiences, and the presence of an additional condition all have the potential to impact upon patients’ quality of life within both groups (see Wehby and Cassell, 2010; Klassen et al., 2012; Stock and Feragen, 2016; Feragen and Stock, 2017). These associations may be further complicated by additional factors, such as condition subtypes (e.g. syndromic cases versus non-syndromic cases), gender, and extrinsic factors, as demonstrated in reviews of CL/P (Hunt et al., 2005; Richman et al., 2012; Stock and Feragen, 2016) and of other CFAs (Kapp-Simon et al., 2007; Knight et al., 2014; Feragen and Stock, 2017).

**Summary of the literature**
The comparison of review articles investigating psychological adjustment to CL/P and other CFAs has identified many broad similarities between these two patient groups, across a range of psychological domains. In particular, patterns of adjustment were comparable in relation to interpersonal relationships, perceived social acceptance, prevalence and impact of additional conditions affecting cognitive development, self-perceptions of appearance, treatment autonomy and decision-making, overall adjustment and quality of life, and a number of interposing factors. Research findings appeared more inconclusive across both research areas in regard to emotional wellbeing, behavioural conduct, educational achievement and vocational outcomes. Factors that have been explored in relation to CL/P, but require further investigation among those affected by other CFAs include parent-infant interactions and attachment representations, self-perceptions of appearance, and patients’ satisfaction with treatment decisions and outcomes.

**Implications for measurement**

While there are considerably fewer studies examining psychological adjustment to CFAs when compared to the CL/P literature, the various domains of psychological adjustment and the corresponding factors appear to be broadly similar within both groups. This observation suggests that similar approaches and outcome measures could be utilised across the two research areas. Working toward a consensus in the measurement of adjustment could help to standardize the way these groups are investigated, and could lead to an overall increase in knowledge of how patients manage the challenges of living with a congenital craniofacial condition. Although some studies and review articles have already sought to combine data collected from individuals born with CL/P and other CFAs, it is questionable whether we currently know enough about the individual diagnoses and subtypes to be able to combine this data meaningfully. One well-known methodological concern within the current literature is the difficulty of comparing findings across studies when researchers employ different measures, in spite of investigating the same domains of adjustment. Utilising the same measures with all patients affected by CL/P and other CFAs would yield a better understanding of the similarities between conditions, without the risk of losing any condition-specific variations, and could support the decision as to whether samples could indeed be combined in the future. In order to guide the measurement selection process, evaluation of psychometric properties and clinical utility, as well as a consideration of the measures that have been used elsewhere, is necessary.
Implications for intervention

If the broad domains of psychological adjustment and corresponding factors are thought to be comparable across CL/P and other CFAs, then similar intervention frameworks and clinical approaches may also be applicable to both patient groups. In relation to improving social experiences, self-perceptions of appearance, and emotional wellbeing, work in the field of visible difference has shown promise, and is already inclusive of a broad range of appearance-altering conditions (for examples, see Clarke et al., 2013; Williamson et al., 2016). Complementary information, support and referrals provided by relevant lay-led organisations could also be beneficial for patients and families affected by a range of conditions (e.g. Partridge, 2010). Interventions aimed at improving communication between patients and health professionals, as well as patient satisfaction with treatment decisions and outcomes, have recently undergone feasibility trials in other fields of health research (e.g. Harcourt et al., 2016), and could be assessed for use with patients with CL/P and other CFAs. In the area of congenital craniofacial conditions more specifically, youth camps and positive exposure techniques have been shown to improve psychological wellbeing and perceived social acceptance among patients with CL/P and other CFAs (Tiemens et al., 2006; Loewenstein et al., 2008; Heike et al., 2010), and thus warrant further evaluation. In addition, previous reviews of adjustment to congenital craniofacial conditions have highlighted the need to introduce routine cognitive screening, in order to identify those at risk of developmental difficulties (Collett and Speltz, 2007; Richman et al., 2012); an area of intervention which requires further development. In a recent systematic review of the literature relating to individuals affected by CL/P and their families, no convincing evidence was found to support the effectiveness of any existing interventions (Norman et al., 2014), and although no comparable review exists in regard to other CFAs, it could be assumed that similar conclusions would be drawn. It is clear that much more work is needed in order to offer a range of interventions that could be beneficial for those affected by CL/P and other CFAs alike.

Methodological considerations

While this article advocates for more collaboration across all craniofacial conditions, there is also a need for further investigation into aspects of adjustment which may be condition-specific. In this article, all ‘other’ CFAs were grouped together, due to an overall lack of studies investigating individual diagnoses. It is clear
that research into rare conditions is challenging, due to the difficulty in acquiring large samples. Yet, grouping all diagnoses together at this early stage may be detrimental to understanding condition-specific nuances. Carrying out collaborative research into rare CFAs across centres and across countries could help to overcome this challenge, so long as researchers and clinicians are able to work toward a consensus in regard to consistent study design and measurement. In future, it is hoped that considerably more research of this type will be conducted, so that more can be understood about these conditions, and so that more meaningful comparisons can be made.

Conclusions

This brief article has examined the available body of literature pertaining to psychological adjustment to CL/P and other CFAs, as presented by several prominent review articles. A number of similarities in the research findings have been identified across both patient groups, suggesting that similar domains of adjustment may be applicable irrespective of the type of craniofacial condition. Consequently, a standardised approach to measurement across all congenital craniofacial conditions is proposed, alongside suggestions for the potential application of similar intervention frameworks and clinical approaches.

References


*NB. One reference has been removed to protect author anonymity.*