Headlines
Craniofacial Support

PSYCHOSOCIAL ADJUSTMENT AND SUPPORT NEEDS OF INDIVIDUALS WITH CRANIOSYNOSTOSIS AND THEIR FAMILIES

VTCTF SMALL GRANT 2019/2020
FULL REPORT SEPTEMBER 2020

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Registered charity no. 1058461

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Summary

Background:

Craniosynostosis is a relatively rare and complex craniofacial condition. Craniosynostosis and its treatment is expected to bring many challenges, and the physical and psychological wellbeing of those born with craniosynostosis and their families can be impacted. Access to appropriate information and support throughout this journey is therefore crucial for families, children, young people, and adults. Headlines Craniofacial Support is a UK-based charity dedicated to improving the lives of people affected by all types of craniosynostosis. Unfortunately, little research about the physical and psychological effects of craniosynostosis exists. This has meant that knowledge about craniosynostosis and how to best support those affected is limited.

Aims of the Project:

In 2017, Headlines published a list of the top ten priority questions for future research. In 2019, Headlines partnered with the Centre for Appearance Research, based at the University of the West of England, to apply for funding for Headlines' first major research project. A small grant was awarded to Headlines by the VTCT Foundation to address two of their priority research questions:

- **What are the long-term physical and psychological effects for individuals with syndromic and non-syndromic craniosynostosis?**
- **Are individuals with craniosynostosis likely to suffer from mental health difficulties, or are they more resilient?**

Due to the known importance of the health of the family as a whole, this project also aimed to gain a better understanding of the psychological **wellbeing of parents of children born with craniosynostosis.**

Study Design:

An online survey was designed. The survey consisted of standardised parent- and self-reported outcome measures and free-text boxes. Topics for parents of children born with craniosynostosis included: Experiences of their child's treatment, their child's wellbeing and development, their own emotional wellbeing and relationships, and experiences of support services. Topics for adults with craniosynostosis included: Emotional wellbeing, relationships, starting a family, education and employment, satisfaction with appearance, physical health, experiences of treatment, and experiences of support services. The survey was based on previous research (where available) and was reviewed by members of the specialist NHS craniofacial teams and by families and adults affected by craniosynostosis at a Stakeholder Workshop in September 2019. The survey was launched in October 2019 and remained open until April 2020.

Findings:

Parents

Parents' psychological wellbeing and relationships were considerably impacted by their child's diagnosis and ongoing treatment. Parents reported higher levels of stress and anxiety than the general population and described symptoms of medical traumatic stress. Parents reported lower levels of optimism and resilience than the general population, although many also described positive outcomes of their child's condition.

Children and Young People

Parents reported their children to have more behavioural difficulties, more emotional problems, more difficulties with peers, and more difficulties overall in comparison to the general population. However, these scores were not considered to be of immediate

clinical concern. Parents also reported their children to have many physical complaints, such as hearing, vision, speech, and movement difficulties.

Adults

Adults with craniosynostosis reported higher levels of anxiety and appearance concerns than the general population and were less comfortable in adult relationships. Others lacked experience of dating altogether and felt that having craniosynostosis posed a challenge to forming meaningful social connections. Starting a family was a cause of concern for many, due to the increased risk of future generations being affected. Physical health, care coordination, making treatment decisions, and gaining employment were also highlighted as ongoing concerns in adulthood. Adults' levels of optimism and resilience were lower than the general population, yet adults also described positive aspects to their condition and their self-esteem did not appear to be affected overall.

Syndromic and Non-Syndromic Conditions

Few differences were found between individuals with syndromic and non-syndromic craniosynostosis on standardised measures, but a greater impact of having a syndrome was suggested in parents' and adults' free-text responses.

Psychological Support

Only a minority of parents and adults had received psychological support for challenges associated with craniosynostosis or had directly engaged with Headlines' existing services.

Recommendations:

There is a clear need for routine psychological screening and support for individuals and families throughout childhood and into adulthood. Routine physical assessments are also needed to identify troubling symptoms and prevent them from worsening. While the provision of support early on may increase resilience and prevent long-term distress for those currently engaged in services, ways of supporting 'older' adults requires further consideration. Education for non-specialist health professionals (such as midwives and General Practitioners) is of high importance to prevent delayed diagnosis and upsetting interactions, and to improve access to care. Going forward, Headlines could consider an evaluation of their current services against the support needs described in this report to highlight any gaps. Future research with young people, other family members (such as grandparents and siblings), and minority groups is suggested. Above all, continued collaboration between Headlines, its members, researchers, the specialist craniofacial teams, and other charities is needed wherever possible to carry out further high-quality research and to continue to improve support and long-term outcomes for individuals and families.

Acknowledgments:

It has been a pleasure and a privilege to work with Headlines Craniofacial Support on this project and we are highly motivated to continue our collaboration. We are grateful to the VTCT Foundation for funding this project and to everyone who contributed to the development of the study, including those who attended the stakeholder workshop, the Clinical Psychologists from the four highly specialist craniofacial teams, and in particular Dr Natasha Rooney from Great Ormond Street Hospital. We are especially grateful to all study participants for candidly sharing their experiences with us.

Introduction

Craniosynostosis

Craniosynostosis occurs when two or more of the cranial plates fuse prematurely and affects approximately one in every 2,000 live births (McCarthy et al., 2012). Syndromic forms of craniosynostosis (including Apert, Pfeiffer, Muenke, and Crouzon syndromes) are much rarer. Depending on the severity of the condition, surgical intervention may be necessary. Despite ongoing multidisciplinary treatment throughout childhood and into adulthood, craniosynostosis can leave the affected individual with a visible facial difference, alongside other medical challenges.

Psychosocial Impacts of Craniosynostosis

For new parents, a diagnosis of a craniofacial condition in their child can evoke feelings of shock, guilt, and grief, as well as concerns for their child's future (Nelson et al., 2012). Families must come to terms with their child's condition, process a wealth of medical information, and make difficult decisions regarding surgical intervention (Feragen & Stock, 2017). As the child grows older and enters school, potential concerns centre on appearance-related teasing and bullying, behavioural conduct, medical absences, speech and communication, and educational difficulties (Feragen & Stock, 2017). Periods of transition may be particularly challenging for individuals with a craniofacial condition, such as the move from primary to secondary school, and the transfer from child to adult services (Stock et al., 2015; Stock & Ridley, 2018). Access to appropriate information and support across the lifespan is therefore vital to facilitate psychosocial adjustment among families, children, young people, and adults affected by all forms of craniosynostosis and associated conditions (Feragen & Stock, 2017).

Headlines Craniofacial Support

Headlines Craniofacial Support is a UK-based charitable organisation run by a board of voluntary trustees, supported by two part-time members of staff. The charity was established by a group of parents in 1993 and became a registered charity in 1996 (#1058461). In 2018, Headlines undertook a strategic review and set out a new vision: a world in which the physical, psychological, and social impacts of craniosynostosis and other rare craniofacial conditions no longer exist. A new mission was also introduced to:

1. Raise awareness and educate people to improve public understanding of craniosynostosis and other rare craniofacial conditions;
2. Support people with craniosynostosis and other rare craniofacial conditions throughout their lives to overcome the physical, psychological, and social impacts of living with these conditions; and
3. Support research that seeks to advance understanding, ensures the provision of quality care, and identifies the best treatments for craniosynostosis and other rare craniofacial conditions.

Headlines currently provides a wide range of support services. These include, but are not limited to: a helpline, Welfare Fund, information leaflets, an e-newsletter, fundraising activities, family days out, a Residential Family Weekend, and a biennial conference.

Existing Evidence Base

Headlines is extremely committed to supporting research and to offering those affected by craniosynostosis and rare craniofacial conditions evidence-based information and interventions. However, little research currently exists, limiting understanding of craniosynostosis and the

types of support that may be beneficial (Feragen & Stock, 2017). In 2017, Headlines carried out a comprehensive consultation with their members, as well as health professionals working in the field, to produce a list of the Top Ten Priority Research Questions **(www.headlines.org.uk/Top-10-Research-Questions1.pdf).** While many of these questions relate to medical outcomes and aetiology and therefore require more intensive investigation, several others can be addressed through the direct exploration of individuals' and families' psychosocial and healthcare experiences. A better understanding of the issues which are important to individuals and their families, in addition to a greater awareness of the information and support that is needed at different time points, would provide Headlines with clear direction for service development and delivery.

Current Project Aims

The aim of the current project was to address Priority Research Questions 1 and 10, and to set recommendations for future research and service development and delivery. Specifically, the questions this project sought to address were:

- **What are the long-term physical and psychological effects for individuals with syndromic and non-syndromic craniosynostosis?**
- **Are individuals with craniosynostosis likely to suffer from mental health difficulties, or are they more resilient?**

In addition, and due to the known importance of the wellbeing of the family for psychological adjustment, this project aimed to gain a better understanding of the psychological wellbeing of parents of children born with craniosynostosis.

VTCT Foundation Small Grant 2019/20

The project was funded by a Small Grant awarded to Headlines Craniofacial Support by the Vocational Training Charitable Trust Foundation (VTCTF). The total award of the grant was £20,644. The project ran from 1st July 2019 to the 30th June 2020. The principal research investigator was Dr Nicola Stock, based at the Centre for Appearance Research (CAR), University of the West of England (UWE). Ms Bruna Costa (CAR) led the project while Dr Stock was on a period of leave from 1st December 2019 to 31st May 2020. An Interim Progress Report was produced in June 2020.

Aims of the Report

This report outlines the findings of the project in full, as well as recommendations for future research and service development and delivery in the area of craniosynostosis. Further details regarding the survey design and data analysis are available on request.

Methodology

Research Design

This study utilised an online survey design. The survey consisted of standardised adult- and parent-reported outcome measures to assess participants' psychological wellbeing, alongside a series of open-ended qualitative questions regarding participants' healthcare experiences and support needs.

Patient and Public Involvement

A Stakeholder Workshop was held in London on 7th September 2019. Attendees included 10 adults born with craniosynostosis, 8 family members, and 1 Consultant Clinical Psychologist, in addition to two members of staff from Headlines Craniofacial Support and two members of staff from CAR. Attendees provided valuable feedback on the proposed study design, study materials, and survey questions. All materials were revised according to the feedback received and attendees received a summary describing how their feedback had been addressed.

Ethical Approval

Ethical approval to conduct the study was provided by the Research Ethics Committee of the Faculty of Health and Applied Sciences at UWE (Ref: HAS.19.07.218).

Procedure

Participants were recruited through Headlines Craniofacial Support (including social media pages, website, and other member communications including emails and newsletters) and CAR (social media pages and Participant Pool communications). Participants were self-selecting. Eligibility criteria included 1) aged 16 years+, 2) living in the UK, and 3) an adequate understanding of written English. The survey was launched in October 2019 and remained open to responses until April 2020.

Main Outcome Measures

Standardised outcome measures were selected according to several key factors: 1) their ability to address one or more of the Headlines research priority questions, 2) their research properties, such as the existence of general population norms and the length of time taken to complete, and 3) their previous use in other craniofacial and related populations, including the VTCTF-funded collaborative grant between the Cleft Lip and Palate Association and CAR (the CLAPA Adult Services Project), so that findings could be compared across conditions.

All Participants	Adults only	Total 2015–16
Revised Life Orientation Test 10-item self-report measure of optimism and pessimism LOT-R; Scheier et al., 1994	**Perceived Stress Scale** 10-item self-report measure of perceived stress during the past month PSS; Cohen et al., 1983	**Body Esteem Scale for Adolescents and Adults (Appearance Evaluation subscale)** 10-item self-report measure to evaluate subjective satisfaction with overall appearance BES-AA; Mendelson et al., 2001
Hospital Anxiety and Depression Scale 14-item self-report measure of common 'symptoms' during the past month HADS; Zigmond & Snaith, 1983	**Strengths and Difficulties Questionnaire** 25-item parent-reported measure of their child's emotional problems, conduct problems, hyperactivity, peer problems, and prosocial behaviours SDQ; Goodman, 2001	**Harter Self-Perception Profile for Adults** Series of self-report measures of global self-worth (6-items), social competence (4-items), intimacy (4-items), and job competence (4-items) SPP-AA; Messer & Harter, 2012
Relationship Satisfaction Scale 10-item self-report measure of subjective satisfaction with their current relationship RS10; Røysamb et al., 2014		**Fear of Negative Appearance Evaluation Scale** 6-item self-report measure of concern that others will judge them negatively due to their appearance FNAE; Lundgren et al., 2004
Connor-Davidson Resilience Scale 10-item self-report measure of an individual's level of resilience CD-RISC10; Connor & Davidson, 2003		**Cleft Hearing, Appearance and Speech Questionnaire** 15-item self-report measure of satisfaction with facial appearance, speech and hearing CHASQ; Psychology Clinical Excellence Network, 2014
		Mini-Social Phobia Inventory 3-item self-report measure of symptoms of social anxiety Mini-SPIN; Connor et al., 2001
		Perceived Sociocultural Pressures Scale 5-item self-report measure of perceived sociocultural pressures PSPS; Diedrichs et al., 2015

Additional Measures

As well as the standardised measures listed above, additional demographic data and other single-item questions were collated. These included questions about: relationships with family and friends; mental health; the experience of having a child with craniosynostosis (where applicable); ongoing physical and functional difficulties; medical and surgical history; and experiences of engaging with Headlines Craniofacial Support. All questions were optional and therefore percentages have been adjusted throughout.

Qualitative Data

Qualitative data were collected using open-ended free-text boxes throughout the survey.

Analysis

Analyses aimed to determine, where possible:

- **How parents' scores on measures of relationships, depression, anxiety, optimism, resilience, and their child's development compare to scores taken from the general population;**
- **How adults' scores on measures of appearance satisfaction, self-worth, social interactions, relationships, employment, depression, anxiety, optimism, and resilience compare to scores taken from the general population;**
- **How adults' and parents' scores compare to related studies of other craniofacial or appearance-altering conditions;**
- **Any differences in scores between syndromic and non-syndromic diagnoses; and**
- **Adults' and parents' self-reported qualitative experiences of craniosynostosis, its impact, and its treatment.**

Results

Participant Characteristics

Parents

A total of 111 parents of children born with craniosynostosis took part in the study. Parents had a mean age of 41 years (range 20-80), were mostly female (91%, n=101), and married (75.7%, n=84). Most of the parents were White (94.6%, n=105) and had been born in the UK (94.6%, n=105). Parents largely reported either being qualified to a pre-graduate standard (23.4%, n=26), graduate standard (31.5%, n=35), or post-graduate standard (27.5%, n=11), and over half were employed at the time of completing the survey (54.1%, n=60). A large proportion of the sample did not report a family history of craniosynostosis (89.1%, n=90).

Children and Young People

Only children born in the UK were included in the sample, due to known differences in healthcare systems around the world. Most of the parents' children were male (64.9%, n=72) and White (93.7%, n=104). The children's ages varied from 3 months to 49 years (mean = 10 years). The most common diagnosis was single suture craniosynostosis (62.2%, n=69). While some children had been diagnosed at the antenatal scan (3.6%, n=4), shortly after birth (28.8%, n=32), or less than one month after birth (15.3%, n=17), the majority of parents had not received a diagnosis until more than one month after birth (41.4%, n=46) or more than one year after birth 10.8% (n=12).

Adults

Only adults born in the UK were included in the sample for the same reason described above. A total of 36 adults (aged 16+) with craniosynostosis took part in the study.

Adults had a mean age of 31 years (range 16-55), were mostly female (69.4%, n=25), and single (62.9%, n=22). Most adults were White (91.7%, n=33). Adults largely reported either being qualified to a pre-graduate standard (33.3%, n=12), graduate standard (16.7%, n=6), or post-graduate standard (27.8%, n=10), and over half were employed at the time of completing the survey (63.9%, n=23). A large proportion of the sample did not report a family history of craniosynostosis (80.6%, n=29). Diagnoses included Crouzon Syndrome (27.8%, n=10), Single Suture Craniosynostosis (25.0%, n=9), Apert Syndrome (16.7%, n=6) and Saethre-Chotzen Syndrome (5.6%, n=2).

Comparisons to Other Data

A review, verification, and validation of the database was undertaken prior to analysis. Due to the broad age range of participating parents' children, a one-way ANOVA was performed to see if there were any differences in outcomes across age groups (0-3 years, 4-11 years, 12-17 years, 18+ years). No significant differences were found and therefore the sample was analysed as a whole.

Sample means were generated for standardised adult- and parent-reported outcome measures. Where available, sample means were compared with other published data using independent samples t-tests. This allowed us to assess the relative psychological adjustment of our sample to the general population and to other studies in the craniofacial and/or visible difference field (where available). Cohen's d was calculated to provide an indication of the size of the difference between our sample and our chosen comparison groups. Cohen's d values between 0.2 and 0.5 represent a small effect, values between 0.5 and 0.8 represent a medium effect, and values of more than 0.8 represent a large effect (Cohen, 1988). Results are described below.

Parents

In comparison to the general population, the results showed that parents of children born with craniosynostosis reported:

- **Lower levels of resilience (large effect)**
- **Lower levels of optimism (large effect)**
- **Less relationship satisfaction (large effect)**
- **Higher levels of stress (large effect)**
- **More symptoms of anxiety (medium effect)**
- **More symptoms of depression (medium effect). However, depression scores were still within the 'normal' range and would therefore not be considered a cause for concern.**

Compared to parents of children with other craniofacial conditions (most notably cleft lip and palate), parents of children with craniosynotosis reported similar scores on the majority of outcome measures. However, parents of children with craniosynostosis did report:

- **Higher levels of stress (small effect)**
- **More symptoms of anxiety (medium effect)**
- **More symptoms of depression (medium effect). However, as before, depression scores were still found to be within the 'normal' range.**

Children and Young People

Parents also completed the Strengths and Difficulties Questionnaire (SDQ) about their children. The SDQ scores children separately according to age (2-4 years and 4-17 years).

Compared to children in the general population, results suggested that 2-4-year-old children with craniosynostosis may have:

- **More emotional problems (large effect)**
- **More behavioural difficulties (medium effect)**
- **More difficulties with peers (medium effects)**
- **More difficulties overall (large effect)**

Similarly, compared to children in the general population, results suggested that 4-17-year-old children with craniosynostosis may have:

- **More emotional problems (medium effect)**
- **More difficulties with peers (medium effect)**
- **More difficulties overall (large effect)**

The SDQ also offers a way of categorising children to indicate whether they should be referred to a specialist for further support. When looking at these categories, 2-4-year-old children scored within the 'normal' range on all subscales, indicating no cause for concern. However, the overall difficulties score was slightly raised, indicating a need for further observation. Similarly, 4-17-year-old children scored within the 'normal' range on most subscales, with some exceptions, suggesting a need for further observation in relation to difficulties with peers and overall functioning.

Adults

In comparison to the general population, results showed that adults with craniosynostosis reported:

- **More fear of negative appearance evaluation (large effect)**
- **Poorer body esteem (large effect)**
- **Lower levels of optimism (large effect)**
- **Lower levels of resilience (large effect)**
- **Less relationship satisfaction (large effect)**
- **Less secure attachment styles in adult relationships (large effect)**
- **More general anxiety (medium effect)**
- **More pressure to conform to appearance 'ideals' (small effect)**
- **A higher self-perceived ability to initiate and maintain Intimate Relationships, including friendships (small effect)**
- **Similar levels of depression, social anxiety, and self-perceived global self-worth, social competence, and job competence.**

Compared to published studies of adults born with other craniofacial conditions, adults with craniosynostosis reported similar scores on the majority of outcome measures. However, adults with craniosynostosis did report:

- **Higher self-worth (large effect)**
- **Higher levels of self-perceived social competence (large effect)**
- **Higher levels of self-perceived competence in intimate relationships (large effect)**
- **Poorer body esteem (large effect)**
- **More symptoms of anxiety (medium effect)**
- **A more anxious attachment style in adult relationships (medium effect)**
- **More fear of negative appearance evaluation (small effect)**

Comparisons between Syndromic and Non-Syndromic Cases

To assess the impact of a syndromic diagnosis on psychological outcomes, data were analysed separately according to syndromic vs. non-syndromic craniosynostosis and the two groups were compared using independent samples t-tests. For statistically significant results, Cohen's d was calculated as described above.

Of the parents who participated in the survey, 70 reported their children to have non-syndromic craniosynostosis and 37 reported a diagnosis of an associated syndrome in their child. In four cases, the syndromic status of the condition could not be determined and these data were excluded from the following analyses. Of the adults who participated in the survey, 10 reported having non-syndromic craniosynostosis, while 20 reported a diagnosis of an associated syndrome. Five stated they did not know if their condition was syndromic, while the syndromic status of one other participant could not be determined. As above, cases whereby the syndromic status could not be determined were excluded from the following analyses.

The results showed that as a whole the two groups did not significantly differ in relation to the majority of scores on the outcome measures used. However, parents of 4-17-year-olds with syndromic craniosynostosis reported their children to have significantly better social skills (large effect) and less behavioural problems (medium effect) than parents of children with non-syndromic craniosynostosis. Adults with syndromic craniosynostosis scored significantly more favourably regarding their self-perceived ability to initiate and maintain Intimate Relationships.

Additional Data

Additional demographic data and other single-item questions were analysed using descriptive statistics and adjusted percentages.

Parenting a Child with Craniosynostosis: Experience of Diagnosis

A large proportion of parents of children with craniosynostosis reported feeling upset (92.8%, n=103) when their child was born, in addition to feelings of shock (73.9%, n=82) and worry (99.1%, n=110). Seventy-six parents reported feeling guilty (68.4%) upon diagnosis and a small proportion (10.8%, n=12) reported feeling blamed by others.

Of the adults who participated in the study, nine reported having had children who were also born with craniosynostosis. Seven adults responded to the following questions and all indicated they had felt upset (100%, n=7), worried (100%, n=7), and guilty (100%, n=7). Some also reported feeling shocked at their child's diagnosis (71.4%, n=5). Most (71.5%, n=5) felt their own experiences of craniosynostosis had made it more difficult to cope with their child's diagnosis, while one adult (14.3%) felt their own experiences had made them better equipped to cope with their child's condition.

Medical History and Ongoing Physical Complaints

The most common medical procedures undergone by children according to

participating parents were: surgery to reshape the skull, genetic testing, speech and language therapy, and surgery to relieve pressure on the brain. The most common procedures reported by adults included: orthodontic treatment, surgery to reshape the skull, genetic testing, surgery to relieve pressure on the brain, and surgery to improve appearance.

A number of ongoing physical difficulties were reported. The most common physical complaints made by parents on behalf of their children were: speech, hearing, vision, movement, and eating and drinking. The most common complaints made by adults included: hearing, fatigue, vision, teeth, and migraine.

Psychological Challenges and Support

Of the participating adults, five (13.9%) reported having an eating disorder. Four (11.1%) reported drug misuse and two (5.6%) reported alcohol misuse. Four (11.1%) participants reported having previously self-harmed and 12 (33.3%) reported experiencing thoughts about self-harm or suicide. When compared to figures obtained from UK-wide NHS surveys (McManu et al., 2007; 2016), reports of drug (22.2-35.4%) and alcohol (16.6%) misuse are lower in the craniosynostosis sample compared to the general population. However, adults with craniosynostosis reported higher rates compared to the general population in respect to eating disorders (6.4%), self-harm (6.4%), and suicide ideation (5.4%).

Only 27 percent (n=30) of parents reported having received psychological support from their craniofacial team (for them or their child). Similarly, only 16.7 percent (n=5) of adults reported having received psychological support from their craniofacial team while growing up, and only one participant (3.2%) had received psychological support as an adult.

Engagement with Headlines Craniofacial Support

Most participating parents (87.7%, n=64) were members of Headlines. Although the majority had not previously volunteered for Headlines (88.6%, n=70), a large number said that they would consider volunteering in the future (67.1%, n=53). Similarly, while only 26.6% (n=21) of parents had previously attended a Headlines event, may stated that they would consider doing so in the future (79.8%, n=63). A large proportion of parents had either sought support or information from Headlines and found it helpful (48.1%, n=38) or would consider seeking support or information from Headlines in the future (46.8%, n=37).

Most participating adults (82.1%, n=23) were members of Headlines. Again, though most had not volunteered for Headlines in the past (89.6%, n=26), a large proportion said that they would consider doing so in the future (89.6%, n=26). Just under half of adults had previously attended a Headlines event (44.8%, n=13) and 86.2% (n=25) stated they would consider doing so in the future. Finally, only seven adults (26.7%) had sought support or information from Headlines in the past, but 60% (n=18) said they would consider doing so in the future.

Qualitative Data

Qualitative data were collected using free-text boxes and were analysed using inductive content analysis (Hsieh & Shannon, 2005). Two researchers coded the data independently and the findings were then cross-checked for any discrepancies. Several key themes were identified in the data. These are summarised below.

Parents

Experiences of birth and diagnosis

Many parents reported a traumatic birth. A delayed diagnosis was also very common (ranging from several months to several years), with a large number of parents reporting that non-specialist health professionals had missed the signs and/or been dismissive of parents' concerns. Some parents stated that the hospital staff had suspected something but had failed to communicate this clearly to the parents or refer the family to a specialist. Many parents had needed to advocate for their child persistently to achieve a diagnosis and had carried out their own research online. However,

other parents reported a timely diagnosis and praised non-specialist healthcare professionals for their efficiency. Once diagnosed, several parents reported a long wait (up to two months) before being seen by a specialist. Parents described a range of difficult emotions upon diagnosis, with some reporting symptoms of medical traumatic stress. Several parents had believed their child would die and many stated they were still processing the emotions years later.

Experiences of their child's treatment

Almost all parents had found their child's surgeries extremely challenging and some reported symptoms of medical traumatic stress, yet the majority also praised the craniofacial teams for their expertise. Nonetheless, some parents described receiving conflicting opinions from specialists over whether certain surgeries were necessary, particularly in the case of aesthetic surgery. Parents described an ongoing burden of care related to the frequency of appointments, the distance travelled, the amount of time spent away from work and school, and a perceived lack of coordination between services. A large number of parents commented on the lack of awareness of the condition among non-specialists and how accessing treatment and support could be slow.

The impact of craniosynostosis on families

Parents described feeling anxious, isolated, stressed, frightened, panicked, grief-stricken, exhausted, depressed, angry, devastated, and traumatised. Many stated that the first year was the most difficult time of their lives and that the experience overall had been lifechanging. Some parents reported that the condition and its treatment had put the family under enormous strain and had considerably impacted marital and familial relationships. Parents also worried about the impact on the child's siblings and grandparents. Some had also lost friendships or felt disconnected from others. However, some parents described feeling stronger and more resilient as a result of the adversity they had experienced, and some felt their experiences had brought their family closer together.

The impact of craniosynostosis on affected children

Parents believed that having craniosynostosis had negatively impacted their child's wellbeing and development. Parents commented specifically on the impact of their child's scarring, facial appearance, speech difficulties, and physical health, and felt their child was sometimes bullied, excluded from activities, and/or underestimated at school. Some parents reported a number of behavioural challenges exhibited by their child. Parents were aware that staring and comments from members of the public upset their child, and some felt the condition had affected their child's ability to build meaningful relationships. Time out of school and the volume of hospital appointments and operations were also concerns for some. Parents whose children also had learning disabilities were particularly worried about their child's wellbeing. Parents also reported a number of concerns for their child's future, including their ability to live independently, gain employment, and engage in romantic relationships. Some were worried about the impact of further surgeries, yet also whether their child would be able to access the treatment and support they might need in the future. Despite these difficulties, several parents believed their child to be well adjusted and happy, and that having craniosynostosis had not held their child back in life overall. Some parents stated that having craniosynostosis had helped their child develop a strong character and a positive outlook. Many parents commented they were proud of their child for how they had coped with the challenges they had experienced.

Support needs and advice for others

A strong theme throughout the parents' experiences was the need for ongoing psychological support, for both the parents themselves and their children. Equally, parents felt that education for non-specialist health professionals (such as midwives, health visitors, and General Practitioners) was essential to improve care coordination, the accuracy of information, empathy, and timely referrals.

Parents also wanted support with medical decision-making and help to understand the purpose of various treatments. Many parents reported that Headlines was a fantastic and essential organisation, yet felt the charity could do more to proactively reach new parents. Some parents commented that activities and events in the community were predominantly aimed at specific groups, such as young families or those with syndromic conditions. Some also commented that events were normally located far away and that an alternative option was needed for those who lived more remotely or who found social events difficult. A number of parents highlighted a need for online support and a stronger social media presence. Advice to others included talking things through, asking for help and information, trusting their instincts, joining peer support groups, taking one day at a time, focusing on their child's strengths, taking time to enjoy their child, and accepting that although the journey would be difficult there would be many good times too.

Adults

The impact of craniosynostosis on emotional wellbeing

Adults reported ongoing emotional challenges related to craniosynostosis, many of which interfered with everyday life. These included difficulties regulating emotions, generalised anxiety, low self-worth, depression, and a lack of self-confidence. Some adults reported thoughts of self-harm and suicide. Coping mechanisms included self-compassion, self-acceptance, spirituality, psychoactive medications, and illicit drug use. A minority of adults had pursued counselling and/or psychiatric treatment outside of the craniofacial team. Some adults reported they were happy with the way they are and felt their experiences had helped them become more resilient.

The role of appearance concerns

Adults reported having been bullied about their appearance as a child. Almost all adults had ongoing appearance concerns, including facial asymmetry, scarring, and skull shape, as well as specific facial features (particularly their eyes). Some stated they felt unattractive and wanted to change their appearance through surgery. Adults reported that they often felt self-conscious about their appearance, particularly in the presence of people they didn't know well. Many covered up the features they disliked when in public and some stated they were frightened of going bald as it would emphasise their skull shape and scarring. Appearance concerns had a negative impact on adults' self-esteem, relationships, and life engagement. However, some adults reported they had worked hard to accept their appearance and had adjusted to looking different over time.

The impact of craniosynostosis on interpersonal relationships

Although some adults reported a close and open relationship with their family growing up, others commented that their condition had not been spoken about and that they believed their parents had struggled to cope. Some adults reported that they felt different to other people and didn't feel they fit in. Many stated they had experienced difficulties in initiating close friendships and/or romantic relationships and had not engaged in these types of relationships as a result. These difficulties included a lack of confidence or self-esteem, feeling unattractive, social anxiety, difficulties explaining their condition to others, a fear of having to reveal their syndrome, a negative online dating culture, and difficulties finding someone who is accepting of their condition. Some adults had recently ended a long-term relationship and stated that the prospect of starting again was daunting. While some reported having had difficult relationships, others stated their experience of relationships had been largely positive.

Experiences of stigma and discrimination

Adults reported that they often experienced social anxiety when in public and felt negatively judged by others. Some had experienced staring and rude comments from members of the public, while others reported being excluded, laughed at, intimidated, or patronised. A proportion of adults reported their experiences with the public had been largely positive or uneventful, while others stated it depended largely on the situation and how they were

feeling on a given day. Some adults had taken a proactive role in initiating and facilitating conversations which they felt helped to overcome social barriers, while others stated that communication difficulties (such as speech) meant they were often misunderstood. Those adults who were employed believed that being at work had provided them with opportunities to overcome their social anxiety and build confidence. However, many adults had struggled to gain employment, often getting to interview stage before being rejected. These adults believed other people's perceptions of their condition prevented them from being successful, while others did not believe that having craniosynostosis had held them back in life.

Physical health problems associated with craniosynostosis

While some adults felt they were able to maintain a normal life, many felt that the physical aspects of craniosynostosis prevented them from engaging in a range of activities, including exercise, social events, driving, and work. Some stated they needed to visit their General Practitioner frequently and many reported finding it difficult to self-manage their symptoms.

Experiences of treatment

A common experience among this group of adults was not having received a formal diagnosis. As a result, they and their families had lacked information and appropriate treatment. Others had received a formal diagnosis yet still felt the information had been poor. Some adults recounted traumatic memories of treatment while growing up, such as not understanding what was being done or what the outcome would be, not being able to speak or move after surgery, and medical complications and pain following surgery. Some had only had minor surgery as a child and couldn't recall any other treatment. Many adults felt their psychological needs had not been taken into account. Some adults commented that the transition to adult care had also been poor. During adulthood, decision-making around further treatment was often complex and treatments were not always successful. Adults commented on the burden of treatment (e.g. time out of study or work) once they had committed to it and some had reexperienced trauma symptoms. Adults reported challenges trying to access specialist care through their General Practitioner and had needed to advocate for themselves repeatedly. Some adults felt they had a good relationship with their craniofacial team and felt confident to seek information and support in future if needed. Others had been unaware that formal treatment and support existed.

Starting a family

Many adults were concerned that future generations of their family would be affected by craniosynostosis. For some, this impacted their views on having their own children, stating that they wouldn't want their children to experience some of the things they had been through. Some had pursued genetic testing, IVF, or adoption in attempts to avoid this outcome. Others believed that the heritability risk wouldn't prevent them from having children and that with the right support having a child with craniosynostosis would be OK. Some adults had received conflicting opinions as to whether their condition was or was not hereditary.

Support needs and advice for others

Adults felt that more psychological and peer support for those with craniosynostosis and their families was crucial. In particular, they felt support for adolescents and adults was rare and that most information was tailored to young children and parents. Adults also strongly advocated for increased awareness of craniosynostosis among non-specialist health professionals and felt that local and specialist health services needed to be better connected. Advice to others included not being defined by the condition, asking for help, self-acceptance, self-compassion, finding others who are accepting, being determined, and identifying the gifts that come with being different.

Discussion

The aim of this VTCTF-funded Small Grant was to address two of the Priority Research Questions identified by Headlines Craniofacial Support, and to set recommendations for future research and service development and delivery.

- **What are the long-term physical and psychological effects for individuals with syndromic and non-syndromic craniosynostosis?**
- **Are individuals with craniosynostosis likely to suffer from mental health difficulties, or are they more resilient?**

The project utilised an online survey to collect quantitative data using standardised outcome measures, alongside participants' more detailed qualitative reports of their experiences. In addition to examining the physical and psychological impact of craniosynostosis on children (parent-report) and adults (self-report) born with the condition, this project aimed to gain a better understanding of the psychological impact on parents of children born with craniosynostosis. The results are discussed below, with reference to existing literature (where available).

Summary of Findings in Relation to Previous Literature

Parents

The psychological wellbeing of participating parents was impacted considerably by their child's birth, diagnosis, and subsequent treatment. Shock, guilt, grief, and concern for the future have also been reported by parents of children born with cleft lip and palate (Nelson et al., 2012) and craniofacial microsomia (Johns et al., 2019), and these experiences therefore seem to be common across craniofacial diagnoses. However, parents participating in the current study also reported higher levels of stress, and more symptoms of anxiety and depression than parents of children born with cleft lip and palate (Stock et al., 2020). Further, symptoms of medical traumatic stress were evident in parents' qualitative reports, which seem to be more prevalent in parents whose children are born with a rare and/or complex condition (Feragen et al., 2019). This appears to be due largely to a relative lack of understanding of craniosynostosis (Zerpe et al., 2020), resulting in delayed diagnosis, challenging interactions with non-specialist health professionals, and difficulties accessing appropriate care, in addition to surgery being more complex and medically invasive. Parents also reported less satisfaction with their relationship with their partner than the general population and described a considerable impact on their relationships and the wider family in their qualitative accounts. Having a child born with a craniofacial condition has been shown to create challenges in couples' relationships, as well as having the potential to bring the family closer together (Zeytinoglu et al., 2017; Feragen et al., 2019). Parents described an ongoing impact of their child's condition and treatment on their psychological health, which remained evident in parents of older children. Although some parents described positive outcomes of their child's condition in their qualitative accounts, the parent group as a whole reported lower levels of optimism and resilience than the general population. Only a minority of parents reported having received psychological support for concerns related to craniosynostosis and few had directly engaged with Headlines' existing support services.

Children and Young People

According to parent reports, children and young people with craniosynostosis exhibited more behavioural difficulties, more emotional problems, more difficulties with peers, and more difficulties overall, compared to the general population. Although these problems were not necessarily of immediate clinical concern, the findings indicate that ongoing psychological and developmental observation may be warranted. Prior studies in this area have suggested that children with craniosynostosis may be at risk of behavioural problems (Becker et al., 2005; Kelleher et al., 2006) and emotional (Bolthauser et

al., 2003) and social (Kapp-Simon et al., 2012) difficulties, particularly in the case of syndromic diagnoses (Sarimski, 2001; Bannink et al., 2011; Maliepaard et al., 2014). However, in the current study, parents of 4-17-year-olds with syndromic craniosynostosis also reported their children to have significantly better social skills that those with non-syndromic craniosynostosis. Prior studies have suggested that young people with syndromic craniosynostoses may be more proactive in initiating social interactions in order to overcome any initial disadvantage (Stavropoulos et al., 2011; Raposo-Amaral et al., 2012). Parents' and adults' qualitative reports also highlighted specific challenges associated with craniosynostosis that are pertinent for young people with craniofacial conditions during the school years (Stock & Ridley, 2018), including appearance concerns, staring, forming meaningful friendships, bullying, exclusion from activities, and burden of care, in addition to many ongoing physical complaints.

Adults

Participating adults reported a considerable and ongoing impact of craniosynostosis on several domains of life. In their qualitative accounts, adults described traumatic memories of early treatment, and difficult interactions with health professionals, peers, and members of the public, which they felt had shaped their emotional development. Concurrently, adults reported more general anxiety, as well as more fear of negative appearance evaluation, poorer body esteem, and more pressure to conform to appearance 'ideals' than the general population. Ongoing appearance concerns in adulthood were also prominent in adults' qualitative reports. Further, rates of reported eating disorders, self-harm, and suicidal ideation were higher than in the general population, as has been found in adults with cleft lip and palate (Ardouin et al., 2020). However, social anxiety and depression scores were not elevated. Adults demonstrated a less secure attachment style in adult relationships and less satisfaction with their relationship with their partner (where applicable). Others lacked experience of romantic relationships altogether and felt that having craniosynostosis posed many challenges to forming long-term and meaningful connections. Similar findings have been reported among adults with a range of craniofacial conditions and have demonstrated a close link between appearance concerns and social and emotional wellbeing (Roberts & Mathias, 2012; Stock & Feragen, 2016). In the current study, the issue of starting a family was a significant cause for concern for many, as is the case in cleft lip and palate (Stock & Rumsey, 2015), due to the risk of future generations being affected. Those adults who had gone on to have children born with craniosynostosis described the additional impact of this reality. Physical health, care coordination, and treatment decision-making were also highlighted as ongoing concerns in adulthood, in addition to challenges related to gaining employment for some. Although few significant differences were found between syndromic and non-syndromic cases on standardised outcome measures, adults described the additional impact of having a syndrome in their qualitative reports. Although previous research is limited, a greater overall impact of syndromic craniofacial conditions in adulthood has been tentatively observed (Tovetjärn et al., 2012; Roberts & Mathias, 2013; Fischer et al., 2014). While many adults qualitatively described positive aspects of having been born with craniosynostosis, levels of optimism and resilience were lower than the general population overall. However, participating adults reported similar scores to the general population on measures of global self-worth, and self-perceived social and job competence, indicating that although they struggled in some areas of life, their self-esteem had not been affected overall. Only a few adults had ever received psychological support for challenges related to craniosynostosis, and in all cases this support had been sought outside of the craniofacial team. Similarly, few adults had directly engaged with Headlines' existing support services.

Recommendations for Future Service Development and Delivery

This study has identified a range of pressing support needs for both individuals with craniosynostosis and their families. All participants reported concerns across one or more domains of adjustment, which were prominent across all age groups. While some individuals and families will require a more intensive level of support, others are likely to be content with a lower level of intervention (Clarke et al., 2013). Similarly, the need for support is likely to fluctuate over time, as well as interact with various life events and the craniosynostosis treatment pathway (Clarke et al., 2013). Providing different levels of clinical and community support across the lifespan which encompasses key concerns and points of risk is therefore key.

Implications for Specialist Craniofacial Teams

The following recommendations are made for clinical practice:

- **Routine psychological screening to identify individuals and family members who may be at risk of psychological distress**
- **Early psychological intervention to reduce distress, increase resilience, and prevent difficulties persisting into adulthood**
- **Psychological support for couples to encourage relative adjustment, increase coping skills, and improve interpersonal relationships**
- **Routine physical assessments to identify symptoms and prevent them from worsening**
- **Increased engagement with regional services and schools to promote a more cohesive health service and support network where possible**
- **A review of current patient and parent literature and identification of any gaps in information provision**
- **Continued collaborative working with CAR and Headlines Craniofacial Support to enhance the research agenda and develop complementary support services**

Implications for Headlines Craniofacial Support

The following recommendations are made for the development and delivery of Headlines' support services:

- **Conduct an audit of the services that are currently offered against the support needs described in this report, to highlight any gaps as well as areas of strength**
- **Consider the location and format of events and activities and ways of including groups who do not currently feel represented**
- **Consider opportunities to engage current members in more volunteering and/or fundraising activities**
- **Consider ways of reaching and supporting adults who may not be aware of the support and treatment options available to them**
- **Continued collaborative working with CAR and the four specialist craniofacial teams to enhance the research agenda and develop complementary support services**

Finally, although minor differences were observed between the data collected in this study and comparable published data collected in other craniofacial populations, the findings overall suggest that similar challenges and points of risk apply across conditions. This indicates that similar services may be of interest to those with a range of craniofacial conditions, such as craniosynostosis, cleft lip and palate, and craniofacial microsomia/microtia.

Our team will:

- **Consider the feasibility of collaborative working across the craniofacial Appearance Collective charities to ensure services are more aligned and more financially sustainable.**

Recommendations for Future Research

This study has collected a considerable amount of information from an understudied population affected by a rare craniofacial condition. While the project has contributed new knowledge to the field and has provided a starting point for further investigation, the samples are relatively small and heterogenous, and therefore results need to be interpreted and disseminated with caution. If research in this area is to overcome these common methodological challenges and to begin to unpick the complexities of living with a rare condition, larger and more representative studies are necessary.

Our team will:

- **Meet with the Clinical Psychologists from the four specialist craniofacial units to discuss ways in which this could be achieved.**

The qualitative data collected in this survey provided depth of insight not achieved through standardised measures alone, particularly in the case of adults, whose experiences are lifelong and individual.

Our team will:

- **Consider the possibility of engaging in additional qualitative research to further elicit the life narratives of adults born with craniosynostosis.**

This study has strongly highlighted the need for improved education for non-specialist health professionals with regard to craniosynostosis, its impacts, and its treatment. Similar studies carried out by CAR have also identified this need in the areas of cleft lip and palate and craniofacial microsomia/microtia (e.g. Johns et al., 2018; Stock & Costa, 2020).

Our team will:

- **Consider the opportunity to work with the other craniofacial Appearance Collective charities to develop appropriate training materials for non-specialist professionals.**

As was the case in the current study due to ethical and practical reasons, the majority of existing research has relied on parent-reported outcomes to assess the impact of craniosynostosis on children and young people. Understanding of craniosynostosis from the perspective of children and young people is therefore scarce. CAR is currently in the process of establishing a UK-wide study focused specifically on the self-reported psychological adjustment of children and young people affected by a visible difference, which will seek to address this issue.

Our team will:

- **Share this report with the CAR team to ensure this gap in knowledge is addressed for those with craniosynostosis.**

As has been identified in the area of cleft lip and palate, an impact of craniosynostosis on the wider family, including siblings and grandparents, was highlighted.

Our team will:

- **Consider opportunities to involve the wider family in future research endeavours where possible, in order to better capture this impact and any additional support needs.**

Finally, participants in the current study were predominantly white, relatively well educated, and born in the UK. As such, those from black and other ethnic minority communities, as well as those with lower socioeconomic status and non-UK nationals, were underrepresented. While not uncommon in research, this becomes problematic when considering that minority groups may experience poorer outcomes (Stock & Feragen, 2016).

Our team will:

- **Share this report with the CAR team to support ongoing efforts to ensure our research, as well as the services offered by the Appearance Collective charities, is more representative of and relevant to minority groups going forward.**

Next Steps

The findings of this VTCTF Small Grant will be shared with the Clinical Psychologists from the four specialist craniofacial centres, and with the individuals and parents who attended the Stakeholder Workshop in September 2019 for comment. A summary of the findings will be made available on the Headlines website. The findings will also be shared at the Headlines (virtual) conference and via a UK-wide press release in October 2020. The results of this study will be written for publication in a leading academic journal(s).

Concluding Statements and Acknowledgements

It has been a pleasure and a privilege to work with Headlines Craniofacial Support on this VTCTF-funded Small Grant. We believe there is much more work to do in this area and hope to continue our partnership to improve the experiences of those born with craniosynostosis and their families. We are grateful to the VTCTF for funding this project and to everyone who contributed to the development of the study, including all stakeholder workshop participants and Dr Natasha Rooney from Great Ormond Street Hospital. We are especially grateful to all study participants for candidly sharing their experiences with us.

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Summary

Background:

Craniosynostosis is a relatively rare and complex craniofacial condition. Craniosynostosis and its treatment is expected to bring many challenges, and the physical and psychological wellbeing of those born with craniosynostosis and their families can be impacted. Access to appropriate information and support throughout this journey is therefore crucial for families, children, young people, and adults. Headlines Craniofacial Support is a UK-based charity dedicated to improving the lives of people affected by all types of craniosynostosis. Unfortunately, little research about the physical and psychological effects of craniosynostosis exists. This has meant that knowledge about craniosynostosis and how to best support those affected is limited.

Aims of the Project:

In 2017, Headlines published a list of the top ten priority questions for future research. In 2019, Headlines partnered with the Centre for Appearance Research, based at the University of the West of England, to apply for funding for Headlines' first major research project. A small grant was awarded to Headlines by the VTCT Foundation to address two of their priority research questions:

- **What are the long-term physical and psychological effects for individuals with syndromic and non-syndromic craniosynostosis?**
- **Are individuals with craniosynostosis likely to suffer from mental health difficulties, or are they more resilient?**

Due to the known importance of the health of the family as a whole, this project also aimed to gain a better understanding of the psychological **wellbeing of parents of children born with craniosynostosis.**

Study Design:

An online survey was designed. The survey consisted of standardised parent- and self-reported outcome measures and free-text boxes. Topics for parents of children born with craniosynostosis included: Experiences of their child's treatment, their child's wellbeing and development, their own emotional wellbeing and relationships, and experiences of support services. Topics for adults with craniosynostosis included: Emotional wellbeing, relationships, starting a family, education and employment, satisfaction with appearance, physical health, experiences of treatment, and experiences of support services. The survey was based on previous research (where available) and was reviewed by members of the specialist NHS craniofacial teams and by families and adults affected by craniosynostosis at a Stakeholder Workshop in September 2019. The survey was launched in October 2019 and remained open until April 2020.

Findings:

Parents

Parents' psychological wellbeing and relationships were considerably impacted by their child's diagnosis and ongoing treatment. Parents reported higher levels of stress and anxiety than the general population and described symptoms of medical traumatic stress. Parents reported lower levels of optimism and resilience than the general population, although many also described positive outcomes of their child's condition.

Children and Young People

Parents reported their children to have more behavioural difficulties, more emotional problems, more difficulties with peers, and more difficulties overall in comparison to the general population. However, these scores were not considered to be of immediate

clinical concern. Parents also reported their children to have many physical complaints, such as hearing, vision, speech, and movement difficulties.

Adults

Adults with craniosynostosis reported higher levels of anxiety and appearance concerns than the general population and were less comfortable in adult relationships. Others lacked experience of dating altogether and felt that having craniosynostosis posed a challenge to forming meaningful social connections. Starting a family was a cause of concern for many, due to the increased risk of future generations being affected. Physical health, care coordination, making treatment decisions, and gaining employment were also highlighted as ongoing concerns in adulthood. Adults' levels of optimism and resilience were lower than the general population, yet adults also described positive aspects to their condition and their self-esteem did not appear to be affected overall.

Syndromic and Non-Syndromic Conditions

Few differences were found between individuals with syndromic and non-syndromic craniosynostosis on standardised measures, but a greater impact of having a syndrome was suggested in parents' and adults' free-text responses.

Psychological Support

Only a minority of parents and adults had received psychological support for challenges associated with craniosynostosis or had directly engaged with Headlines' existing services.

Recommendations:

There is a clear need for routine psychological screening and support for individuals and families throughout childhood and into adulthood. Routine physical assessments are also needed to identify troubling symptoms and prevent them from worsening. While the provision of support early on may increase resilience and prevent long-term distress for those currently engaged in services, ways of supporting 'older' adults requires further consideration. Education for non-specialist health professionals (such as midwives and General Practitioners) is of high importance to prevent delayed diagnosis and upsetting interactions, and to improve access to care. Going forward, Headlines could consider an evaluation of their current services against the support needs described in this report to highlight any gaps. Future research with young people, other family members (such as grandparents and siblings), and minority groups is suggested. Above all, continued collaboration between Headlines, its members, researchers, the specialist craniofacial teams, and other charities is needed wherever possible to carry out further high-quality research and to continue to improve support and long-term outcomes for individuals and families.

Acknowledgments:

It has been a pleasure and a privilege to work with Headlines Craniofacial Support on this project and we are highly motivated to continue our collaboration. We are grateful to the VTCT Foundation for funding this project and to everyone who contributed to the development of the study, including those who attended the stakeholder workshop, the Clinical Psychologists from the four highly specialist craniofacial teams, and in particular Dr Natasha Rooney from Great Ormond Street Hospital. We are especially grateful to all study participants for candidly sharing their experiences with us.