An exploration of living with a craniofacial anomaly and experiences of Midface Advancement Surgery.

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Introduction

A congenital craniofacial anomaly (CFA) is a term used to describe abnormalities in the bone or soft tissue of the face or head, comprising of a wide range of conditions with many associated syndromes (Holmbeck & Aspinall, 2015). Some CFAs are relatively common, such as cleft lip/palate, whereas other conditions, such as craniosynostosis and Treacher Collins syndrome are far rarer (Feragen & Stock, 2018). While each condition will affect every individual to differing degrees, CFAs often result in a visible difference, and can impact upon several domains of psychological, emotional and social functioning, such as levels of distress and anxiety (Feragen & Stock, 2016).

A growing body of literature has highlighted that despite the challenges associated with a CFA, many individuals positively adjust to living with a CFA (Rumsey & Stock, 2013; Roberts & Mathias, 2013). Several internal psychological factors, such as sense of self and optimism (Appearance Research Collaboration, 2009; Rumsey & Harcourt, 2004), and factors external to individuals, such as family relationships, treatment autonomy and availability of social and practical support have been shown to play an important role in adjustment (Thompson & Kent, 2001; Rumsey & Harcourt, 2005). A recent review of the literature (Feragen & Stock, 2017) suggests that investigating positive, in addition to negative impacts of CFAs could provide a broader understanding of the complexity of adjustment and useful insight for promoting resilience within clinical practice (Feragen, 2012). However, studies and literature reviews to date, have placed greater emphasis on factors that hinder adjustment to living with a CFA rather than on helpful factors. Studying hindrances or stressors alone provides an incomplete and negative picture of responding to challenges. With previous reviews focusing on the challenges of living with a CFA, the first research chapter provides a novel systematic review of the literature on positive adjustment to living with a CFA.

The second research chapter focuses on a specific type of CFA, craniosynostosis. This condition is under research, despite its widespread impact on individuals and families. Surgical interventions are often recommended to individuals with CFAs, including craniosynostosis to improve physical function, aesthetic outcomes and psychological wellbeing (Feragen, Stock, Sharrat & Kvalem, 2016; Marsh, 2006). Research exploring children and young people's views on motivations for surgery found that whilst for some individuals with CFAs the motivations for surgery were self-derived, others felt external pressure from parents and outside influences, such as societal pressures to undergo surgery (Bemmels, Biesecker, Schmidt, Krokosky, Guidotti, Sutton, 2013).

Whilst many individuals seek reconstructive surgery for various reasons, the potential physical risks of craniofacial procedures are well established. However, there is little information about the potential social and psychological implications of surgery (Asch, 2006). Some studies support the theory that reconstructive surgery for CFAs can increase self-esteem (Kay, Tubbergen, Warschausky & Buchman , 2005), perceived improvement in appearance (Hansen, Kreiter, Rosenbaum, Whitaker, & Arpey, 2003), and enhanced social engagement (Arndt et al., 1987). Others have however, challenged this and there is now much debate in this area as to whether surgery always improves individuals' psychological wellbeing (Marsh, 2006). Research has found that although individuals might be initially satisfied with the outcomes of surgery, the satisfaction often dissipates over time, leaving the children susceptible to future psychosocial difficulties (Arndt et al., 1987; Sarwer, Bartlett, Whitaker, Paige, Pertschuk, & Wadden, 1999; Bruce, 2009).

The lack of long-term studies measuring long-term satisfaction and psychological benefits following surgery (Marsh, 2006), has encouraged some health professionals to reflect critically on whether surgical intervention to prevent predicted social and psychological challenges is the most effective route to take when weighed against the potential risks of surgery (Mouradian et al., 2006).

As the area of craniosynostosis and surgical interventions remains relatively under researched and given the centrality of extensive and long-term treatment for syndromic craniosynostosis conditions, the psychological impact of this treatment on individuals and their families is an important topic (Feragen et al., 2016). The second study therefore employs a qualitative methodology to explore children’s and parents’ experiences of Midface Advancement surgery, one form of surgical intervention offered to individuals with craniosynostosis.

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Chapter one: Positive adjustments to living with craniofacial anomaly: A systematic review

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Abstract

Purpose: A congenital craniofacial anomaly (CFA) is expected to impact upon several domains of emotional, psychological and social functioning. However, research has shown that despite the challenges of a CFA, many individuals positively adjust to living with a CFA. This study therefore aimed to review and synthesise the literature on positive adjustment to living with a CFA.

Method: A systematic literature search was undertaken of four electronic databases. Thirteen relevant studies were identified as appropriate and were quality assessed. A thematic synthesis was used to synthesise the key themes and concepts of the included papers.

Results: The analysis produced six overarching themes regarding positive adjustment to living with a CFA. 1) Dispositional style, 2) Sources of support, 3) Positive self-concept, 4) Transitional stages, 5) Difference in self and others, 6) Surgical interventions.

Conclusion: The review suggests individuals go through periods of adjustment across the lifespan, with adjustment influenced by protective dispositional factors, connections with others and the meanings ascribed to difference. It is important we question the ideas that difference automatically equates to negative experiences or disability and give voice to those who flourish. The review also identified important clinical implications and areas for future research.

Keywords: congenital craniofacial anomaly, visible difference, positive adjustment, qualitative

Introduction

Congenital craniofacial anomaly (CFA) is a broad term used to describe a range of diagnoses, which can be isolated to the face and head or form part of a wider genetic syndrome (Holmbeck & Aspinall, 2015). This includes common CFAs, such as cleft lip and/or palate, in addition to rarer conditions, such as craniosynostosis and Treacher Collins syndrome (Feragen & Stock, 2018) that can result in a visible difference.

Appearance plays a dominant part in individuals’ self-concept. Previous research proposes if appearance has been judged and evaluated negatively, this will have a detrimental impact on overall levels of self-esteem (Rumsey & Harcourt, 2005). Social and cognitive processes in early life, in relation to how others react to an individual’s appearance, will affect their sense of worth, self-esteem and levels of confidence (Rumsey & Harcourt, 2005). As hypothesized by Rogers's (1959) model of personality development, individuals with a visible difference often grow up with an awareness of their difference and their self-concepts can be organised in such a way that appearance information is more important (Moss & Carr, 2004).

Studies on the psychological impact of CFAs have however produced conflicting results. While some studies indicate that those born with a CFA experience more psychosocial difficulties than their peers without a CFA, other research has found few differences between these two groups (Rumsey & Stock, 2013; Roberts & Mathias, 2013). Such findings illustrate the complexity of adjustment to a condition that causes visible differences (Rumsey & Stock, 2013).

A number of factors may help or hinder adjustment to a CFA. These include background factors, such as gender, age, cultural and religious upbringing (Appearance Research Collaboration, 2009; Feragen & Stock, 2014; Thompson & Kent, 2001). Factors external to the individual such as treatment autonomy, availability of practical and social support, family coping and relationships with health professionals have also been shown to play an important role in adjustment (Thompson & Kent, 2001; Rumsey & Harcourt, 2005). Internal psychological factors including sense of self, optimism, expectations of treatment and recognition of positive growth have also been shown to be key to adjustment (Appearance Research Collaboration, 2009; Rumsey & Harcourt, 2004). Thus, psychological adjustment may depend on hierarchies of socioecological structures at the individual, peer and family, school, community and other levels.

Bronfenbrenner ‘s (1977) ecological systems theory offers a possible explanation of how these different structures impact adjustment. The ecological system approach proposes that the inherent qualities of children and their environments interact to influence how they develop. According to this theory, individuals find themselves enmeshed in various ecosystems. The microsystem includes the most intimate home system to the larger school system. Relationships within this system are considered to be bidirectional and the most influential level. Interactions with family and peers will influence how the individual views themselves, such as feeling the same or different to others (Eiserman, 2001; Havstam, Laakso, Ringsbearg, 2011). The microsystem also includes health services. Individuals with health conditions often have frequent contact with these services and it is suggested that the support offered here will influence a child’s progression (Thomson et al., 2001). Beyond this is the most expansive system which includes society and the attitudes and ideologies of the culture around the individual.

Each of these ecological systems inevitably interact with and influence each other across all aspects of the children’s lives. These different systems influence the individual’s development and view of themselves from a young age and in turn can influence their acceptance of a visible difference (Cash, 2012). Developmental stages and life transitions have an influence, too. Evidence has suggested that a person’s age and life stages play a role in their acceptance of visible differences, with individuals becoming more accepting as they mature (Beaune, Forrest & Keith, 2004).

Investigating positive, in addition to negative impacts of CFAs could provide a broader understanding of the complexity of adjustment and useful insight for promoting resilience within clinical practice (Feragen, 2012), suggests a recent review (Feragen & Stock, 2017). To date, however, studies and literature reviews have placed greater emphasis on the factors that hinder adjustment to living with a CFA than on helpful factors. Studying hindrances or stressors alone provides an incomplete and negative picture of responding to challenges. Discussing the growing field of positive psychology, Aspinwall and Staudinger (2003) wrote “… people can draw on these strengths without ignoring or diminishing the negative realities of their situations …” (p. 16). Strauss (2001) described how adults with CFAs encountered positive and growth-enhancing aspects of learning to live with their conditions. This was echoed by Eiserman (2001) who found adults with a CFA experienced significant positive outcomes from living with a CFA. The novelty of these findings demonstrates that to capture the variability and complexity of adjustment across the lifespan, and associations across different domains of adjustment, comprehensive reviews are needed.

It is important to understand the experiences of those with CFA, including positive and negative aspects. Focusing on a contributional perspective of visible difference does not inherently suggest that all or even the majority of the experiences associated with a CFA are positive. It can, however, potentially add a greater balance in understanding the lives of those living with a CFAs. The experiences of those who have positively adjusted to living with CFA can make a significant contribution to shaping practices that will benefit all individuals with CFAs. Research on resilience argues that it is the stories of those who are thriving, that can light the way for developing interventions for those experiencing distress, as well as indicating ways others can continue to flourish (Eiserman, 2001; Ickovics & Park, 1998).

No published systematic review to date has synthesised the findings from qualitative studies exploring individuals' experiences of positively adjusting to CFA. Whilst quantitative methodology is important, it doesn't fully examine young people's thoughts and concerns (Kendall 1997; Varley 2011). Qualitative studies can lead to new insights and improvements in service provision and are integral for good clinical practice and (Department of Health 2015; Varley 2011). Although there remains a paucity of qualitative in comparison to quantitative research, an increasing number of studies have explored individuals' experiences of living with a CFA (Roberts & Shute 2010; Stock, Fergan & Rumsey, 2016). Given the rare nature of these conditions the question of adjustment to CFAs is ideally suited to qualitative methods (Holmbeck & Aspinall, 2015; Knight, Anderson, Spencer-Smith & Da Costa, 2014). A qualitative systematic review may lead to new insights and clinical implications for the support and interventions offered to individuals with a CFA.

Review aims

The present systematic review aims to provide a comprehensive and updated synthesis of the existing literature investigating factors that influence positive adjustment to living with a craniofacial anomaly. In addition, the review aims to critically assess the quality of current evidence.

Method

A protocol was registered with the PROSPERO database for systematic reviews (registration number CRD42020175041). Reporting of this review followed the guidance set out in the Preferred Reporting Items for Systematic reviews and Meta-analyses (PRISMA) statement (Moher, Liberati, Tetzlaff & Altman, 2009).

Search strategy

The following databases were searched: CINAHL, PsychInfo, MEDLINE and EMBASE. The Cochrane Library and PROSPERO were searched to identify existing or anticipated relevant reviews. Attempts to identify grey literature were made by searching EThoS and ProQuest databases. Key journals were hand- searched (The Cleft Palate-Craniofacial Journal, Clinical Child Psychology & Psychiatry, Psychology and Health) cited references in eligible articles and key review articles. Search terms were formulated using the SPIDER tool (Cooke, Smith & Booth 2012) (see Appendix B). Thesaurus tools (such as Medical Subject Headings [MeSH]) were utilised to identify relevant terms within each database, this is in line with the Cochrane Collaboration guidelines (Higgins & Green, 2008). Different controlled vocabulary terms were identified for all four databases. Literature searches took place in November-December 2019.

Inclusion and exclusion criteria

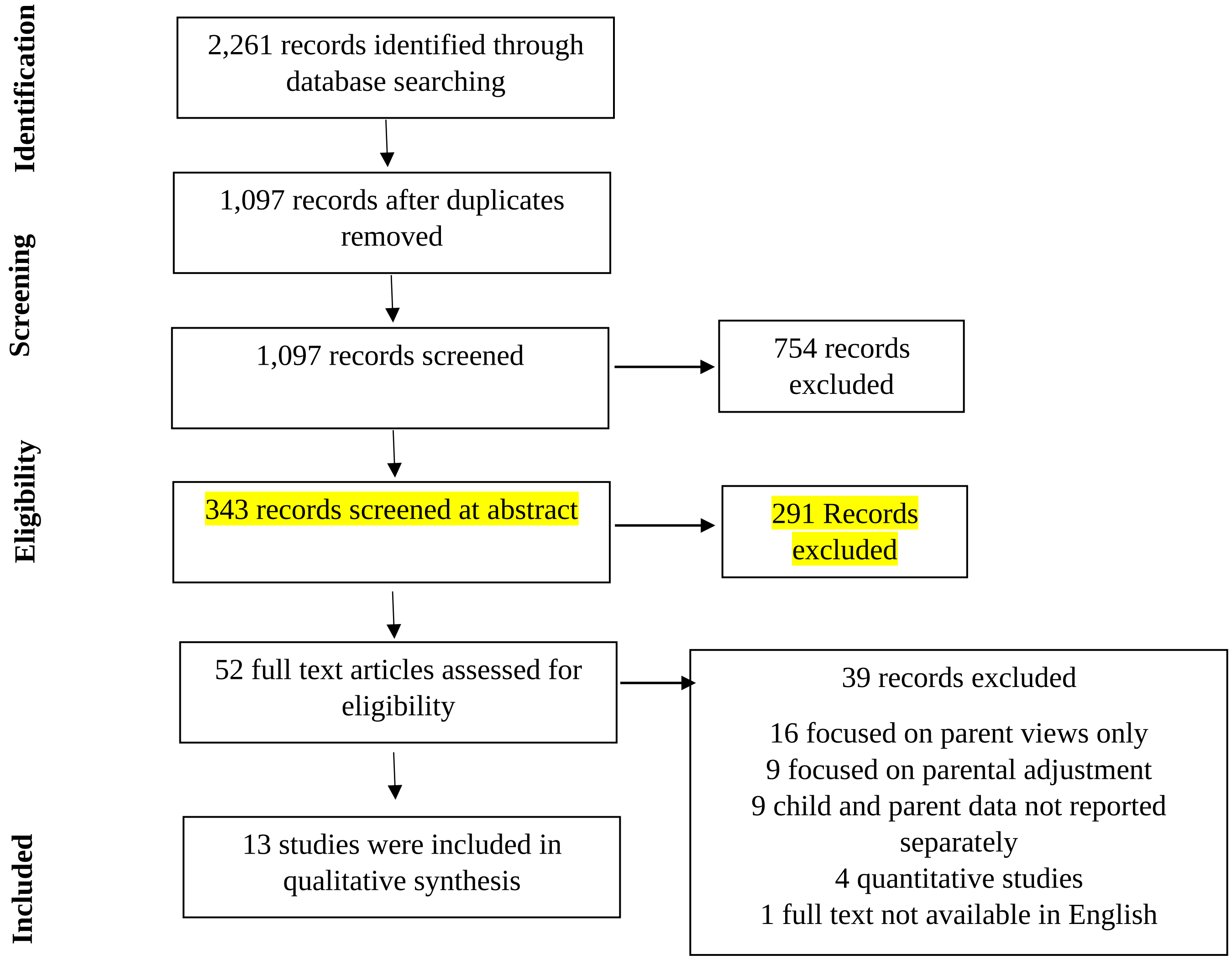
The following inclusion criteria were applied: (a) papers written in or translated into English; (b) papers exploring individual's experiences of adjusting to living with a craniofacial anomaly; (c) papers where participants had a congenital craniofacial anomaly (d) papers utilising qualitative methods of data collection and analysis, including mixed method designs; (e) peer reviewed, primary research, except grey literature; (d) papers published in the last 20 years.

Papers were excluded if individuals' personal narratives could not be distinguished from those of others. For example, cases where there was an acquired craniofacial condition such as from a result from trauma or disease; or being unable to distinguish between dyads (e.g., parents or professionals). Papers were also excluded if they focused exclusively on parental or family adjustment. Unpublished book chapters theses, non-peer reviewed journal articles, reviews, opinion pieces and commentaries were excluded due to a lack of peer-review.

Study selection

The search yielded 2261 articles (EMBASE, 949; CINAHL, 670; MEDLINE, 582; PsychInfo, 60). MO applied the inclusion/exclusion criteria and identified 13 studies that were eligible for synthesis (Figure 1). A colleague (LA) of MO selected 10% of articles at random to screen at both title/abstract and full text stages and applied the inclusion and exclusion criteria to each of the 13 selected full texts to confirm eligibility. There were no disagreements in the decisions made.

Figure 1. *Flow diagram of the systematic search process following PRISMA guidelines* (Moher, Liberati, Telzlaff & Altman, 2010).



Data extraction and synthesis

A data extraction form was created for the purpose of the review. This was first piloted and subsequently used by the first author to extract the following data; demographic, methodological and outcome data.

A thematic approach was used to synthesise the findings (Thomas and Harden 2008). Thematic synthesis (Thomas and Harden 2008), the chosen method for this review, offers a transparent method, and a high level of analytical abstraction from the primary studies to contribute new understandings of participants' experiences. This technique adopts methods from grounded theory and meta-ethnography to develop descriptive themes and the generation of 'analytical themes'. While the development of descriptive themes remains 'close' to the primary sources of data, the analytical themes represent a stage of interpretation whereby the reviewers aim to 'go beyond' the primary sources of data and generate new interpretive explanations, constructs or hypotheses (Thomas et al., 2008).

Within this approach, all text labelled as 'results' or 'findings', was considered data for abstraction. Full-text PDF documents were entered into QSR's NVivo 12 software for qualitative data analysis. The synthesis took the form of three stages which overlapped to some degree. Firstly, all data, whether author-defined codes or participants' quotes was coded line-by-line by MO to capture its meaning and content. Every sentence had at least one code applied, with many categorised using several codes. Concepts were translated from one study to another by either applying existing codes or developing new concepts when necessary. MO examined the data for similarities and differences between the codes to group codes into a hierarchical structure. This organisation of codes aided the construct of descriptive themes. Finally, these descriptive themes were grouped and through discussions with SW and LC, analytical themes were developed.

Results

Study characteristics

Table 1 summarises the characteristics of the 13 studies included in the present review. Studies including specific or mixed CFAs are presented first followed by studies including participants with a cleft/ lip palate.

Table 1. *Study Characteristics*

Author (s) year	Method	Phenomena	Setting	Geography	Culture	Participants & sampling method	Analysis	Conclusions
Beaune, Forrest & Keith 2004	Long interview method; semi structured interviews and peer debriefing techniques	Perspectives on living and growing up with Treacher Collins Syndrome	Hospital	Canada	Developed country	6 participants. Male (5), female (1). Age range 12-18 years. Range of ethnicity Black Canadian (3), Caucasian (2) and other (1) Purposive sampling	McCracken's 1998 qualitative analysis	Good psychosocial adjustment experienced as; acceptance of self, condition, social acceptance and resilient adaptive strategies. Difficulties included social stigma and fitting in in school
Roberts & Shute 2010	Semi-structured interviews	Experiences of living with craniofacial anomalies	Hospital	Australia	Developed country	26 children between ages of 7-19 years. Male (13), Female (13). 28 parents Purposive sampling	Systematic analysis	Four major stress-related themes were found; self-acceptance, responses of others, disabilities and impairments, and treatment. Positive themes included personal qualities and support.
Stavropoulos, Hallberg, Mohlin & Hagberg 2010	Telephone interviews	Living with Crouzon Syndrome	Telephone	Sweden	Developed country	8 participants. Male (6), Female (2). Mean age 25.4 years Caucasian ethnic background Purposive sampling	Grounded theory	The adaptation of successful coping strategies is crucial in the quest of attainment of higher self-esteem. Recognised the importance of using personal coping strategies to enhance well-being.
Riklin, Calandrillo, Blitz, Zuckerberg & Annuziato 2019	Mixed method; qualitative focus groups and quantitative measures	Examining the psychosocial needs of adolescence with a craniofacial condition to	Outpatient clinic	USA	Developed country	14 adolescences. Age range 14-18 years Purposive sampling	Descriptive statistics and inductive approach	Several factors associated with psychological well-being of adolescents with craniofacial diagnoses were identified; stress, bullying but also positive coping strategies. The importance

		inform intervention development						of creating interventions to target specific psychosocial needs was also demonstrated.
Stock, Feragen & Rumsey 2015	Telephone interviews	To explore psychological adjustment and support needs of those with a cleft lip/palate	Telephone	UK	Developed country	52 participants. All adults. White British (48), mixed Caucasian (2), British Asian (2), White Irish (1) Purposive sampling	Thematic analysis	The findings imply that most adults with a cleft adjust well to varies life challenges and report many positive outcomes. For a minority of patients, issues attributed to the cleft may continue to cause distress in adulthood.
Tiemens, Nicholas & Forrest, 2013	Semi-structured interviews	To explore the experiences of adolescent's with CL/P preparing for orthognathic surgery	Tertiary care paediatric hospital	Canada	Developed country	Seven participants. Female (7). Adolescents.; age range 15-20 years. Caucasian (5), Other (2). Purposive sampling; criterion sampling	Phenomenological approach	Adolescent girls can experience strain associated with living with a facial difference; however, they find strategies to cope with the perception of difference. Reconstructive surgery is viewed as a means to increase confidence.
Havstam & Laakso, 2011	Semi-structured interviews	To explore young adults accounts of growing up with a CL/P	Participants workplace, home or researcher's university site	Sweden	Developed country	11 participants. Male (6), Female (7). Mean age 30. Consecutive sampling and theoretical sampling	Grounded theory	Different aspects of the process of developing self-image in relation to the cleft were identified; Shaping one's attitude to the cleft and dealing with being different.

Searle, Neville & Waylen, 2017	Interviews; unstructured followed by semi-structured interview	Psychological growth and well-being in individuals born with a cleft; applying self-determination theory	Participant's home or telephone	UK	Developed country	15 participants. Female (9), Male (6). Age range 17-62. Purposive Sampling	Thematic analysis	A supportive family and social environment can lead to the development of positive psychological growth and well-being in those with CL/P.
Chetpakdeechit, Hallberg, Hagberg, Mohlin, 2009	Open- ended interviews	To explore perceptions and values of young adults with CL/P; main focus around social life	Telephone	Sweden	Developed country	12 participants. Female (6), male (6). Age range 24-33. Consecutive sampling and theoretical sampling	Grounded theory	Young adults with either cleft lip and palate or isolated cleft palate who received recognition from significant others reported increased self-esteem and greater ability to cope with their social lives.
Stock, Fergan & Rumsey, 2016	Telephone interviews	Explore factors and processes contributing to psychological adjustment in those with CL/P	Telephone	UK	Developed country	52 participants. Female (30), male (22). Age range 22-77. White British (48), mixed Caucasia (1), British Asian (2), White Irish (1). Purposive sampling	Thematic analysis	Findings demonstrate various background, external and internal factors that contribute to challenges and adjustment to living with a CL/P. Also illustrates the potential degree of individual variation in perspectives.

As per the inclusion criteria, all studies included participants with a CFA. Participants in seven studies included participants with a cleft lip/ palate. One study included participants with Treacher Collins syndrome (Beaune, Forrest & Keith, 2014), another included participants with Crouzon syndrome, a complex craniosynostosis (Stavropoulos, Hallberg, Mohlin & Hagberg, 2011). Four studies included participants with a variety of craniofacial conditions including hemifacial microsomia, Sturge-weber syndrome, complex craniosynostosis and cleft lip/palate (Eiserman, 2001; Loewenstein, Sutton, Guidotti, Shapiro, Ball, McLean & Biesecker, 2008; Roberts & Shute, 2010; Riklin, Calandrillo, Blitz, Zuckerberg & Annuziato, 2019).

Four studies (Beaune et al., 2004; Riklin et al., 2019; Roberts et al., 2010; Tiemens et al., 2013) included children and adolescents as the sample population. Ages ranged from 12-20 years of age. One study (Loewenstein et al., 2008) included participants ranging from 12-61 years of age. Six studies included adult participants with various age ranges, from 17- 77 years of age.

The majority of studies completed unstructured or semi structured interviews (N=10), four studies utilised a mixed methods design. One study (Eiserman, 2001) used closed and open-ended questionnaires, in addition to unstructured interviews. Another study (Loewenstien et al., 2008) used open-ended questionnaires and quantitative measures. Riklin et al (2019) carried out a qualitative focused group, in addition to quantitative measures.

Qualitative data analysis varied between studies. Four studies used thematic analysis (Kappen, Mirza-Babaei & Nacke, 2019; Searle et al., 2017; Stock et al., 2015; Stock et al., 2016) and three used a grounded theory approach (Chetpakdeenchit et al., 2009; Havstam et al., 2011; Stavropoulos et al., 2010). Other studies utilised systematic analysis (Roberts et al., 2010), content analysis (Eiserman, 2001), interpretive analysis (Loewenstien et al., 2008), inductive analysis (Riklin et al., 2019), a Phenomenological approach (Tiemens et al., 2013) and McCrackens 1998's approach to data analysis (Beaune et al., 2004). Different analyses may have drawn out different themes, impacting on what was reported in study findings. All studies took place in developed countries.

Quality assessment

The Critical Appraisal Skills Programme (CASP, 2015) checklist for qualitative research (see Appendix C) was used to assess the selected final studies for methodological issues, rigour and biases that may impact on the quality of the findings (see Appendix D). The CASP allows for critical appraisal of qualitative studies, using a ten-item protocol. Whilst no formal scoring system is embedded within the CASP, as widely applied in previous studies (Butler, Hall, & Copnell, 2016) a scoring system was applied for this review (item not met=1, item partially met/unsure=2-, item fully met=3). Higher scores reflected greater methodological quality, with a maximum score of 30. MO and a colleague (LA) assessed the studies independently using the CASP and any discrepancies were resolved through discussion, resulting in an overall consensus for each paper.

The value of assessing quality in qualitative research is subject to ongoing debate, with the notion that qualitative research can be methodologically flawed contested. Rather than exclude studies based on their quality rating, the CASP (2015) appraisal criteria was used to critique the included studies to contextualise the thematic synthesis. All studies scored above 22 out of 30. Most studied had a clear statement of aims, research design, used an appropriate method, and grounded their conclusions in the data. Many studies did not discuss ethical implications in detail, despite these being considered core aspects of qualitative research. Although some made reference to gaining ethical approval, many lacked discussions on how consent was gained, how information was presented or considerations for working with children. Only one study commented on researcher reflexivity, despite its influence in qualitative research. To our knowledge no studies included service-user involvement in the design of the research. Across studies there was variation in the detail of description given to each stage of the data collection, with some reporting minimal information, raising the issue of replicability.

Summary of findings

Six overarching themes regarding positive adjustment to living with craniofacial anomalies were identified, as shown in Table 2, with a table of quotes reflecting the sample of analytical and descriptive themes in Appendix E.

Table 2. *Analytical and descriptive themes*

Analytical theme	Descriptive theme
Dispositional style	Optimism Openness Resilience Reappraisal
Source of support	Family Friends Romantic relationships Psychological services
Positive self-concept	Identity, ‘part of me’ Capabilities and potential for success
Transitional stages	
Difference in self and others	Perceptions of difference Family appraisal of difference Difference in others
Surgical intervention	Decision making Realistic expectations

Distribution of these themes across each of the 13 studies are shown in Table 3 with illustrative themes displayed.

Table 3: *Analytical and Descriptive Theme Table*

Analytical and Descriptive Themes	Beaune at al. 2004	Chetpakdeechit et al. 2009	Eiserman et al. 2000	Havstam et al. 2011	Kappen et al. 2019	Loewenstein et al. 2008	Riklin et al. 2019	Roberts et al. 2011	Searle et al. 2017	Stock et al. 2015	Stock et al. 2016	Stavropoulas et al. 2010	Tiemens et al. 2013
Dispositional Style													
Optimism	X		X		X	X	X	X		X	X		
Openness				X			X		X				
Resilience			X		X		X	X			X		
Reappraisal	X				X		X		X				
Sources of support													
Family	X		X	X	X		X	X	X		X		X
Friends	X		X				X						X
Romantic relationships		X			X								
Psychological services					X					X	X	X	
Positive self-concept													
Identity	X		X			X			X	X	X		X
Capabilities and potential for success	X					X			X		X	X	
Transitional stages	X	X		X	X	X			X		X		X
Difference in self and others													
Perceptions of difference	X		X			X					X		
Family Appraisal of difference				X		X	X		X		X		
Difference in others			X	X		X		X		X			
Surgical interventions					X				X		X		X

Theme 1: Dispositional style

The first analytical theme reflected participants' dispositional style. Eight studies cited optimism as a key feature. Participants who identified as having adjusted positively to their visible difference stated they had a positive look on life and a certain resilience against the challenges associated with living with a visible difference. *"In all it [having a facial difference] has affected a lot of stuff and it makes me feel positive and I have certain ways of looking at things. When I say looking at stuff, I mean, as I said in the speech, optimism in my life; it has made me optimistic toward the future."* (Beaune et al., 2004). Along with a positive outlook, many participants stated they believed they appreciated life more than others did and where hopeful for their future.

Havstam et al., (2011), Riklin et al., (2019) and Searle et al., (2017) found that choosing to be open about their visible difference was described by participants as an important way of dealing with experiences and crucial for dealing with other people's reactions. Although differences were found in how much participants wanted to talk about the visible difference, with some describing being more open as they got older, many reported that by being open about the condition and prepared to talk to others, it served to increase relatedness and the support they received.

Reappraisal of events in four studies (Beaune et al., 2004; Kappen et al., 2019; Riklin et al., 2019; Searle et al., 2017) was shown to be important. This related to participants cognitively reframing events and situations which in turn reduced the distress associated with the event. This was found across ages. For example, in a study with adolescents with Treacher Collins syndrome, staring or teasing from younger children was now reconstructed as curiosity, which was experienced as a non- or less harmful interaction (Beaune et al., 2004). *''Yeah, now that I know that little kids are curious about what I have and why..."*

(Beaune et al., 2004). Other participants stated that remarks about appearances were made by people who were not happy in themselves *"When someone makes a nasty remark, it tells me more about that person than anything else."*(Kappen et al., 2019) This led participants to engage in positive and assertive based responses to being stared at or teased, such as initiating a conversation, confronting or educating/informing others about their condition. By moving from negatively-based response strategies to more assertive and positive strategies these participants reported to have reconstructed the meanings of their difference, and as a result experienced an increase in self-acceptance (Beaune et al., 2004).

Five studies (Eiseman, 2000; Kappen et al., 2019; Riklin et al., 2019; Roberts et al., 2010; Stock et al., 2016) noted how participants described how difficult experiences had made them more resilient and able to face further challenges. Many participants described an 'inner strength' that led them to be more determined to succeed and, in some cases, 'prove others wrong': *"It made me stronger, more determined to fight the battles ahead. It gave me guts and I thought, no, I'm going to fight this"* (Roberts et al., 2010)

Theme 2: Sources of support

The second analytical theme reflected participants' views on the importance of various sources of support.

Nine studies found that having encouraging, supportive parents and siblings were factors that participants described as contributing to positive adjustment (Beaune et al., 2004; Eiserman, 2001; Havstam et al., 2011; Kappen et al., 2019; Riklin et al., 2019; Roberts et al., 2011; Searle et al., 2017: Stock et al., 2016; Tiemens et al., 2013) Participants described supportive parents as those who did not treat them as any different to others and valued them as individuals. In these studies, the family context emerged as a 'needs supportive' environment, one that nurtured the whole child, rather than dwelling on their visible difference. Participants also described seeking support and advice from their parents to help understand and cope with adverse responses to being visibly different. The family context provided the building blocks for the development of psychological growth and wellbeing in many of the participants. Many commented that they believed that family support had served to make them more determined individuals: *"So you know, right now I owe everything to my parents, you know, bringing me up to be confident."* (Searle et al., 2017).

Support wasn't limited to family; many participants spoke about the value of support from friends, describing how they felt accepted by their friendship group (Beaune et al., 2004; Eisemen, 2000; Riklin et al., 2019; Tiemens et al., 2013). This created an underlying sense that their friends cared and would support them through emergent difficulties of stigma and adjustment. This included support in the face of negative attitudes or bullying, *"I found my group of friends and they stood up for me. They were really cool about it.''* (Teimens et al., 2013). Accordingly, this social support served as a strong buffer against the stigma some participants experienced. Participants in these studies also spoke about importance of feeling part of a social circle. Three of these studies included children or adolescents as their sample.

Two studies with young adults with cleft lip/palate (Chetpakdeechit et al., 2019; Kappen et al., 2019;) found that being in a relationship and receiving recognition from significant others had enabled participants to focus less on their difference and this contributed to feeling more self- confident, valued, and socially accepted. Chetpakdeechit et al., (2019) also found that young adults who has long-term partners or were married were more satisfied with their facial appearance than participants without partners. A sample of adolescents with Treacher Collins syndrome (Beaune et al., 2004) identified having a partner and a family as part of their social expectations, with one expectation. This is higher than previously reported low rates of dating and projections about marriage in the physical disability literature (Stevens et al., 1996). This difference in findings may be due to differences in methodology of studies. Some previous research has used surveys to gather information about romantic partners (Stevens et al., 2006; Collisson et al., 2020), perhaps unintentionally using leading questions based on stereotypical outdated views.

Participants in four studies (Kappen et al., 2019; Stavropoulous et al., 2010; Stock et al., 2015; Stock et al., 2016) described how receiving formal psychological support from others was valuable in various ways such as managing stressors. Psychological support offered an exploration of how some participants viewed themselves and helped them to prepare psychologically for surgery. Some participants stated that psychological support was not offered but believed it would have been very valuable. Young adults with cleft/lip palate felt it should be available from an early age, and that support should focus on dealing with bullying ,resiliency and preparing for associated stressors (Kappen et al., 2019). *"Some level of counselling, psychological support . . . someone just to be very honest with . . . in my teens especially I definitely would have benefited from that* (Stock et al., 2016).

Theme 3: Positive self-concept

A key theme identified in seven studies was the development of a positive self-concept. Self- concept is active, dynamic, and malleable and can be influenced by social interactions. In this sense, the development of a positive self- concept will be dependent upon, to some degree the support participants have received from others, as discussed in theme two.

Acceptance of their visible difference and viewing it as part of their identity was discussed by many participants (Beaune et al., 2004; Eiserman, 2001; Loewenstein et al., 2008; Stock et al., 2015; Stock et al, 2016; Searle et al., 2017; Tiemens et al., 2011). Those who constructed positive identities were those who conceptualised their difference not as a personal failure, but as a situation that could be resisted and challenged (Stavropoulos et al., 2010). This outlook appeared to play a key role in positively adjusting to living with a visible difference. In a sample of young adults with various craniofacial conditions (Eisermen, 2000) study participants were asked "*if you could live your life again, would you deliberately choose to remove this experience of facial difference from your life?*" More than half of the participants stated they would not choose to remove this experience from their lives. One participant stated: "*While I would give up the hard part in a second, I have a feeling that I would give up a central part of who I am that makes me the person I am if I sacrificed the experience of having a facial difference. I just could not do this in good conscience. If blending in were all that really mattered, then I guess I would remove the experience next time around. But I don't want to blend in. I don't want to be just one more person. I kind of like the unique things I have to offer to others and the world because of this experience. It seems like I add a little piece to the balance of the world because of this"* (Eiserman, 2000).

Participants in five studies (Beaune et al., 2014; Loewenstein et al., 2008; Searle et al., 2017; Stavropoulous et al., 2010; Stock et al., 2016;) reported how they believed that being involved in a variety of activities, and focusing on their strengths, had contributed to their adjustment and increased their wellbeing. These participants focused on strengths rather than 'defects' and saw themselves as capable individuals who are able to make valuable contributions in all areas of life. Some described positive intrapersonal traits that contributed to adjustment and wellbeing, including internal motivation to push themselves to excel in areas such as sports/ hobbies, achieving good grade in school, going on to college or a university or having a good career. In two studies adolescents and young adults described excelling in other areas of life had become a coping strategy which minimised their perceived difference and highlighted their competencies, thus helping them to protect their self-image (Beaune et al., 2004; Stavropoulos, et al., 2010).

Theme 4: Transitional stages

The notion that various transition stages in life impact upon adjustment to living with a visible difference and the view one held of themselves, was detailed in eight of the included studies (Beaune et al., 2004; Chetpakdeechit et al., 2009; Havstam et al., 2011; Loewentstein et al., 2008; Kappen et al., 2019; Tiemens et al., 2011; Searle et al., 2017; Stock et al., 2016). This was evident across ages and diagnosis. Stock et al., (2016) reported adults with cleft lip/palate became more accepting of their cleft over time and developed a more positive self-concept as they got older. Similarly, Teismens et al., (2011) noted a shift with age and maturity from shame and self-consciousness to a reasoned decision to accept themselves and as such be less concerned with others' perception of them and their difference. Other participants believed that the value they placed on appearance reduced over time, while their level of acceptance of their appearance increased (Kappen et al., 2019).

Whilst these studies reported how participants had an increased social confidence over time, this was perhaps also due to other factors such as reappraisal or different transitions in life. Searle et al., (2017) reported how the start of primary school was a difficult time for some, where difference was noted, as participants progressed onto college or employment, peers moved past negative attitudes toward their difference and become more accepting (Beaune et al., 2004; Kappen et al., 2019; Searle et al., 2017). One participant stated: *"I used to value appearance a great deal, but as one grows older, over 30, one starts to look at it differently and it becomes less of an issue. When I finished high school and went to higher education it suddenly seemed all OK. People acted normal, nobody teased. It was a turning point for me."*(Kappen et al., 2019)

Adolescents referred to this as not being so focused on individual differences and acceptance of diversity, leading these individuals to develop more acceptance of themselves

(Beaune et al., 2004). Kappen et al., (2019) and Chetpakdeechit et al., (2009) also reported how finding a romantic partner in adult life had contributed to acceptance of the self and increased esteem.

Theme 5: Difference in self and others

A key finding in some studies was the perception of difference. Adolescents with various craniofacial conditions described how they did not see themselves as different to others and commented how they had a right to be treated as equals to others. Other participants commented that they felt unique or special and enjoyed ‘standing out above the crowd*’* (Beaune et al., 2004; Eiseman, 2000; Loewenstien et al., 2008*)*. One stated *“This makes me unique; I don’t just sit there as another face in the crowd. I stand up and I am proud to stand up”* (Beaune et al., 2004). Similar results were found in a sample of adults with cleft lip/palate (Stock et al., 2016) and this perception of the self as either the same as others, or unique, appeared to contribute to participants identity and acceptance of their identity.

Loewenstein et al., (2008) and Stock et al., (2016) described how families viewed individuals and their visible difference contributed to how the individual perceived difference in general. Some participants reported that their parents felt secure and were comfortable with their visible appearance. Through this interaction with others, participants developed a sense of difference as a unique and positive experience or a sense of sameness and inclusion. As a result, the notion of difference can be seen as extrinsic in origin, a consequence of social context. Similarly, Havstam et al., (2011), Riklin et al., (2019) and Searle et al., (2017) found parental coping style and familial dynamics made an important contribution to the participants own adjustment. A young adult with a cleft lip/palate commented on their experiences as a child “*My parents have never pointed out that I’m different in any way, that* *there are things I can't do … I've always done everything other kids do"(*Havstam et al., 2011).

Participants in five studies spoke about how their experiences had resulted in an increased understanding and acceptance of others with disabilities or differences (Eiserman, 2001; Havstam et al., 2011; Loewesnstein et al., 2008; Roberts et al., 2016; Stock et al., 2015). This included not teasing others, not being prejudiced, standing up for others and having a well-developed understanding of others' feelings and contexts. In addition, participants described themselves as more compassionate, tolerant, and empathic than the wider population. Some participants expressed that their ability to reach out and empower others struggling with stigmatisation and discrimination stemmed from their own personal struggle to overcome experiences of rejection. This appeared to be evidenced in adult samples, perhaps reflecting growth and maturity.

Theme 6: Surgical interventions

With surgical intervention often recommended for CFAs, Searle et al., (2017) and Kappen et al., (2019) discussed the importance of being involved in the decision-making process and having realistic expectations of surgery. An adult with a cleft lip/ palate commented: *"… yeah don't kind of just discuss things with their parents: make sure they're involved in those conversations as well. I was always there for the conversations and kind of knew everything as they knew it, which was good. But if I hadn't known those things I'd be a lot more uncertain about it"* (Searle et al., 2017).

Adults with a cleft lip/palate (Stock et al., 2016) reiterated the value of talking to them and parents to ensure the rationale and expected outcomes of treatment are well understood by all parties. In addition, they found that many adult participants recalled traumatic experiences of hospitals during childhood, which was linked to a lack of autonomy regarding treatment, leaving some feeling disempowered. Other adult participants (Searle et al., 2017;Stock et al., 2016) noted that although medical teams pushed for surgery, they were able to weigh up the potential risks and benefits of surgery and make an informed decision to decline surgery. *"I think doctors and surgeons want to do everything that they can to help, but it doesn't necessarily mean that it's right for you. And I think it's important to focus on the person inside than the person outside. And if you're comfortable with the person inside then it doesn't, you know, matter to have great big implants in your face or …"* (Searle et al., 2017*).*

Despite surgical interventions often being conducted from a young age, only adult participants spoke about different aspects of surgery. Parents in some studies discussed surgical interventions, perhaps reflecting their input surrounding uptake of appearance altering-surgery. However, as stated in inclusion criteria for the review these comments were not included in the analysis.

Discussion

The review set out to examine psychological adjustment, specifically from the point of view of individuals with a CFA, focusing on factors that positively aid adjustment to living with a CFAs. In total 13 studies were included in this review, with individuals with a congenital CFA, across the life span. Several relevant factors that contributed to positive adjustment were identified. The review suggests individuals go through periods of adjustment across the lifespan and that adjustment is multifaceted and multifactorial. The application of the thematic synthesis methodology has enabled new insights to be achieved with key concepts and themes gathered across a body of literature. Although the literature has grown over the past decade, there still remains a paucity of research focusing on a wide range of CFA, with the majority of research focusing on cleft lip/ palate.

Although all the studies examined life as an individual with a CFA, their aims, design, and methodology varied. Only two studies commented on research reflexivity, despite its influence in qualitative research. Data gathering and analysis, can be influenced by the researcher's preconceptions of phenomena and can unintentionally impact the direction of research, perhaps towards a focus of negative outcomes. Some studies did not provide detailed information regarding data analysis, making it difficult to review the process of analysis. Two studies that highlighted many factors that hindered adjustments with minimal focus on positive adjustment, utilised a grounded theory approach. Perhaps, reflecting what was found in early data collection and the influence this had on the focus of further data collection. In addition, some differences were highlighted between samples age ranges, with younger participants reporting friends to be an important factor in adjustment. During these years friends often play an important role in individuals lives, forming a main source of social interaction and support (Taylor & Townsend, 2016). Perhaps as individuals grow, more emphasis is placed on romantic partners or family support networks, explaining why friendship was not raised as an important factor in these studies, compared to family support. Within adult samples accepting difference in others was evidenced, possibly reflecting growth and maturity, with previous research supporting the notion that difference in others is more accepted in early adulthood and beyond (Livneh, 2012).

Factors that influenced positive adjustment were grouped into six themes; dispositional style, positive self-concept, sources of support, transition, perceptions of difference and treatment.

The results of the review suggest that an individual's dispositional style plays an important role in their adjustment to living with a CFA. Several studies found that having a positive outlook on life and a certain resilience against the challenges associated with living with a visible difference acted as a key factor in their psychological adjustment (Beaune et al., 2004). This is consistent with previous research by the Appearance Research Collaboration (2009) and Rumsey and Hardcourt (2004).

The review also found that reappraisal of events was a key component in individuals' adjustment. Where negative comments or staring were reappraised as curiosity or that others were not happy within themselves, this reduced the emotional impact of the event. In addition, it led to a reconstructed meaning of difference resulting in individuals experiencing an increase in self-acceptance (Beaune et al., 2004). This sits in line with Roger's (1959) theory of personality development which suggests that the Self is influenced by personal experiences and their interpretation of those experiences. According to Rogers (1959), individuals want to feel, experience and behave in ways which are consistent with their self-image and which reflects how they would like to be; the ideal self. If a person interprets experiences in line with their ideal self, the more congruent their self-image and ideal self are, resulting in a higher sense of self-worth (Harter, 2012).

Research studies investigating dispositional style and personality attributes and their associations with adjustment to living with a CFA are scarce within the literature. However, in the last few decades, there appears to be shift from searching for pathological personality traits that could be a consequence of living with a visible difference, toward a 'normalising' approach and a search for individual characteristics that may protect individuals against negative experiences. Research findings of Eiserman, (2001), Havstam et al., (2011) and Stock et al., (2016) for example, should be examined further in future research, to identify protective dispositional factors, in order to inform the development of preventative work and interventions.

Other adaptive strategies identified in the review included being involved in a variety of activities, with both physical and creative activities acting as an outlet for self-expression which served in the development of positive psychological growth and well-being. Other strategies included an internal motivation to push themselves to excel in many areas of life and focusing on strengths rather than ‘defects’. This contributed to adjustment and increased wellbeing (Loewenstein et al., 2008; Searle et al., 2017). Some studies reported that being motivated resulted in participants setting high standards or goals to try to minimise the differences from their peers, challenging stereotypes of ‘disability’, highlighting their competencies and developing a successful life pathway (Beaune et al., 2004). This strategy of ‘compensation’; a defensive-adaptive coping mechanism that individuals with physical differences engage in, to symbolically protect their ‘different self’ may however, have several disadvantages (Timberlake, 1985). These include unrealistic expectations, fear of failure, burn out and excessive pressure and may result in additional stressors.

As suggested by Bronfenbrenner’s (1977) ecological systems theory, developmental stages and life transitions play an important role in acceptance of visible differences. The results of the review suggest that a person’s age and life stages influence adjustment, with individuals becoming more accepting of their difference and developing a more positive self-concept as they aged (Beaune et al., 2004; Stock et al., 2016). This is potentially a result of a combination of factors, including peers being more accepting of difference as they mature (Kappen et al., 2019), finding a romantic partner or secure friendship group (Chetpakdeechit et al., 2009) and individuals placing less emphasis on appearance (Teismens et al., 2011). Reviewing papers with first-hand accounts from individuals across the lifespan enabled insights into these different transitional stages. Further research is needed with children and young people to hear these unique accounts. In addition, to reduce the potential for dilation of developmentally specific findings, studies recruiting from paediatric and adult populations perhaps would benefit from analysing and reporting data in distinct age-related categories.

The review also highlighted the role of systemic influences on individuals and indicated that perceived social support and positive social experiences may act as a buffer against negative experiences. Those with supportive parents, peers and health care professionals reported this support aided development of a more positive self-concept and acceptance of their difference (Havstam et al., 2011; Kappen et al., 2019; Riklin et al., 2019; Stavropoulous et al., 2010). As outlined in Bronfenbrenner's ecological systems theory the microsystem around individuals appears to be the most influential system, having a huge impact on an individual's view of the self and wellbeing.

In one study of adolescents with Treacher Collins syndrome (Beaune et al., 2004) participants identified dating, marriage and having a family as part of their social expectations, which is inconsistent with previous reports about dating and marriage in the physical disability literature (Steven et al., 1996). Other participants recalled how being in a relationship and receiving recognition from significant others had enabled them to focus less on their difference and this contributed to feeling more self- confident and socially accepted (Kappen et al., 2019; Chetpakdeechit et al., 2019). Nonetheless, the lack of research on the romantic experiences among individuals with CFA stands in sharp contrast to the importance of this variable in many people's lives (Furman & Shaffer, 2003). Further exploration of the impact of living with a CFA on romantic relationships is needed.

Another key finding in the review was the perception of difference. Adolescents and adults with various craniofacial conditions described how they saw themselves as no different from others, whilst some commented that they felt unique and liked to stand out (Eiseman, 2000; Loewenstien et al., 2008). These perceptions appeared to positively contribute to individual's identity and acceptance of their identity (Stock et al., 2016). Interactions with others and wider perceptions of difference appears to impact upon an individual's view of themselves and sense of difference or sameness and inclusion (Loewenstein et al., 2008 &

Stock et al., 2016). As a result, the notion of difference can be seen as extrinsic in origin, a consequence of social context.

Prior theories on the development of stigmatisation and more recently the social model of disability suggest that the development of attitudes toward difference is multifactorial and highly complex and that attitudinal barriers are the greatest limitation experienced by individuals with visible differences (Oliver, 1996; Sigelman and Singleton, 1996). These ideas have been demonstrated within previous research on negative attitudes within the general public towards individuals with a visible difference (Grandfield, Thompson, & Turpin, 2005; Stock, Whale, Jenkinson, Rumsey, & Fox, 2013; Partridge & Julian, 2008), where the challenges of adjusting to a visible difference in a society with increasingly high appearance standards and values have been illustrated (Grogan, 2008). Concerns about wider societal views towards difference, reported by parents have been illustrated in previous research (Klein, Pope, Gatahun & Thompson , 2006). Research analysing the content of television programming found people with a visible difference were seldom shown on television and when they where they would often be cast in roles with a negative stereotype (Wardle & Boyce, 2009). This highlights the paradox of visible difference often being ‘invisible’ in mainstream media. Campaigns such as Face Equality (Changing Faces, 2017), are paramount in continuing to challenge prejudice and discrimination related to visible differences.

Clinical implications

The findings from the review can have several important clinical implications to help support individuals living with a CFA.

As highlighted in this review, many individuals living with a CFA live fulfilling lives, feeling confident in themselves and establishing a place in the world. If we increase our awareness about the factors that help individuals manage challenges and thrive, we can use this knowledge in how best to support and build skills in those who are struggling. The identification of patients who may be at risk of experiencing ongoing psychological difficulties related to their condition is key. By increasing psychological thinking within medical teams, this may allow for earlier identification of individuals who are struggling. Predicting which patients will struggle and which will thrive is however, a well-documented challenge. Individual's personality traits and psychological characteristics, as explored above influence wellbeing and adjustment to living with a visible difference.

In recent years there has been a growing interest in the influence of Positive Phenomena (PP) on physical and mental health, a field of study referred to as Positive Psychology (Seligman, Steen, Park, & Peterson, 2005). There are multiple pathways through which these PP are thought to influence health outcomes. Individual's cognitive and behavioural processes are believed to intervene between PP and their physical and emotional health. Although some personality traits and psychological characteristics are considered trait-like in nature, prior research suggests that these factors are modifiable, and interventions targeting specific PP to improve health and wellbeing have been successful (Evans, 2011; Seligman et al., 2005). Such interventions may be particularly applicable to those with a CFA: a context in which patients may accept that some challenges are likely to be permanent and the focus of treatment shifts to "building what's strong rather than fixing what's wrong" (Duckworth, Steen, & Seligman, 2005; Evans, 2011). When considering interventions to assist this population, positive psychology may offer something of benefit, offering the chance to help individuals find their inner strengths, focusing more on proactive steps to use their own abilities to flourish. On reflection more traditional interventions around difference may have unintentionally played a part in sustaining stigma by basing approaches around the view that difference is the problem.

The review also highlighted the role of systemic influences on individuals, particularly the role of parents' responses to difference and reactions to unwanted social interactions. Family systems theories can offer a helpful framework to explore systemic influences on the whole family (Bowen, 1978; Minuchin, 1974). In the context of the present review, it may be helpful for parents to explore their own relationship with difference and bullying, which may unconsciously influence how they interpret and respond to negative social interactions and model responses to their child.

The review also suggests a sense of people feeling appropriately supported and involved with healthcare systems was an important aspect of individuals' experiences. The relationship between healthcare provider and individual has received a lot of attention in the research literature, with positive patient -physician relationships associated with satisfaction of care with health settings (Swedlund, Schumacher, Young & Cox, 2012).

Healthcare professionals have a duty of care to ensure all patients are heard and enable collaborative informed decision making. Clinical psychologists have a role to play here in ensuring the patient is understood, fully- informed and supported in making decisions around potential treatments. This will be dependent on age and cognitive ability and care is needed that children's views or wishes are not overlooked. Within this it is important that individuals with CFAs are given the opportunity to explore positive outcomes from living with a CFA, with personal strengths and capabilities highlighted.

With surgical interventions often recommend for aesthetic reasons (Marsh, 2006), these findings call for clinicians and researchers to move away from the paradigm that an observable visible difference automatically leads to dissatisfaction with appearance and towards an exploration of the factors which may impact upon an individual's self-perceptions. Given the centrality of extensive and long-term treatment for many CFAs, motivations behind appearance alerting surgery and the emotional and psychological impact of treatment on individuals and their families is a key subject for future research.

Limitations

The studies reviewed had several limitations and highlighted a number of methodological challenges within the field of psychological adjustment to CFAs. Firstly, and as anticipated, relatively few studies drew upon large samples, impacting generalisability. Sample size varied from 6 to 52. Qualitative research typically only involves small sample sizes and so perhaps this limitation is in relation to only considering qualitative research. Participants presenting with a variety of CFAs were often mixed within one sample. This is understandable when certain conditions are rare, however this rendered analysis of relevant subgroups and potentially confounding factors very difficult within this review. Future research may benefit from using multicentre collaborations to recruit larger sample sizes (Munafo, 2017). This would allow for additional analysis of data to investigate the effects of variables, such as characteristics of the condition and increase robustness of findings. Further research should be focused on understanding what it is like to live with under researched CFA conditions, such as craniosynostosis or Treacher Collins syndrome. Further research in these areas can potentially have huge clinical implications, guiding the care these populations receive. Whilst the review, along with previous reviews have demonstrated that awareness of positive adjustment and resilience are beginning to be included in research within this population (e.g. Chimruang et al., 2011; Pisula et al., 2014; Stock et al., 2015; Stock, et al., 2016). This progress should be built upon to provide a more balanced view of adjustment and to allow for the assessment of points of opportunity and growth.

Due to the lack of research in the area a broad range of ages were included in the review, although grouping all ages may be viewed as a limitation of the review, it highlighted important themes that may have otherwise been missed. Nevertheless, further research is needed to help us understand how these conditions impact individuals at different stages of their of lives. It is important we broaden our knowledge base of these conditions to enable appropriate support across the life span.

Conclusion

The present review explored factors that influence positive adjustment to living with a craniofacial anomaly. The review synthesised studies from a variety of contexts and highlighted six over-arching themes that encapsulate adjustment to living with a CFA. The findings of this review have important implications clinically and stressed the need for further research in this area. This review offers a contributional perspective and does not address the whole picture of living with a CFA. It does not aim to dismiss or deny the challenges those with a CFA face but rather it aims to highlight the factors that influence positive adjustment, that are often overlooked in research and indeed in practice. Further research is needed into both factors that help or hinder adjustment, but it is important future research offers a balanced view and does not dismiss those individuals who do thrive. It is important we question the ideas that difference automatically equates to negative experiences or disability and give voice to those who flourish.

Disclosure statement

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UNIVERSITY OF LIVERPOOL

Chapter Two: An exploration of child and parent experiences of childhood Midface Advancement surgery and life following surgery: Empirical paper

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Abstract

Objectives: Craniofacial surgery is often recommended to individuals with syndromic craniosynostosis to improve physical function and aesthetic outcomes (Marsh, 2006). However, there is much debate in this area as to whether surgery always improves psychological wellbeing in these children. This was the first study, which aimed to explore experiences of midface advancement surgery, from the perspective of the child and their parents.

Design: Mother and child dyads, engaged in semi-structured interviews to capture their experience of midface advancement surgery. Their interviews were analysed using Interpretative Phenomenological Analysis.

Results: Four master themes encapsulated participants' experiences; 'Life before', 'Transitional stages', 'The intrinsic role of support' and 'Managing and making sense of the new me'.

Conclusions: The findings highlighted the ongoing challenges presented by social responses towards difference. With societal pressures around appearance contributing to the decision-making process to undergo this appearance altering surgery. The study identified whilst for some surgery acted as a catalyst for growth, whilst for others unexpected outcomes occurred. The value of peer support networks and connecting with others was demonstrated both for children and parents. Finally, there is a need for more research in this area to truly capture the experiences of appearance altering surgery and the meanings families ascribe to these interventions.

Keywords: craniosynostosis, midface advancement surgery, interpretative phenomenological analysis, child, young person, parent.

Introduction

Congenital craniofacial anomaly (CFA) and other conditions that affect prominent areas of the body can affect mental health and social functioning (Roberts & Mathias, 2013). CFA is a broad term used to describe a wide range of diagnoses, which can be isolated to the face and head or form part of a wider genetic syndrome (Holmbeck & Aspinall, 2015). Craniosynostosis, a rare CFA results when one or more cranial structures fuse prematurely and can cause numerous difficulties for the individual (McCarthy et al., 2012). Syndromic craniosynostosis is associated with various dysmorphisms involving the face, skeleton, nervous system and can be accompanied by developmental delay (Vlad Ciurea & Toader, 2009). While each condition will affect every child to differing degrees, craniosynostosis often impacts upon several domains of psychological and emotional functioning, such as levels of distress and anxiety (Feragen & Stock, 2016).

Complex syndromic conditions often require regular treatment and multiple surgeries to regularise the shape of the head (Feragen, Stock, Sharrat & Kvalem, 2016). Surgical intervention is often recommended to parents of children with CFAs to improve physical function, aesthetic outcomes and psychological wellbeing (Marsh, 2006). There are three primary motivations for undergoing craniofacial surgery: functional improvement, physical health, and aesthetics. Research exploring children and young people's views on motivations for surgery found that whilst for some individuals with CFAs the motivations for surgery were self-derived, others felt external pressure from parents and outside influences to undergo surgery (Bemmels, Biesecker, Schmidt, Krokosky, Guidotti, Sutton, 2013). Parents are often heavily involved in the decision-making progress of surgery and dependent upon cognitive ability and age, children may have little involvement in this progress. Previous research has found that although some children were comfortable with their parent's decision around surgery, others resented the intimation that there was something about them needed ‘fixing’ (Bemmels et al.,2013).

Midface advancement surgery is typically performed during early adolescence, a time when individuals become more aware of their appearance (Harter, 2012; McAdams, 2013; Levine & Smolak, 2002; Rumsey & Harcourt, 2007) and are trying to establish who they are and where they fit within the world. Adolescence has been characterized by Erikson (1950) as the period in the human life cycle during which individuals try to establish a sense of personal identity and avoid the dangers of identity confusion. Adolescents find their personal identity by establishing a meaningful self-concept. To do this, experiences and perceptions of the past, present, and future are brought together to form a unified whole (Harter, 2012; Klimstra, Hale, Raaijmakers, Branje & Meeus, 2010; Meeus, Van De Schoot, Keijsers, Schwartz & Branje, 2010). Consequently, this task could be more difficult if an individual has experienced conflicts in earlier life stages and when their future is unpredictable. Since surgery involves uncertainty with regard to the ultimate outcome for appearance, finding a stable self-concept is difficult.

Adolescents often seek social feedback from others, which shapes their perception and evaluation of themselves (Feragen, Kvalem, Rumsey & Borge, 2010; Backes & Bonnie, 2019); their appearance is a large part of identity for many. Adolescents' preoccupation with the perception of others is the basis of Elkind's (1967) theory of egocentrism. It suggests that identity is formed through interactions with significant others, a process Erikson refers to as psychosocial reciprocity. Adolescents often seek peer group recognition and involvement and conforming to the expectations of peers helps adolescents find out how certain roles fit with them and how they fit within society (Feragen, Kvalem, Rumsey & Borge, 2010; Cash 2012; Crerand, Sarwer, Kazak, Clarke & Rumsey, 2017). If an individual is segregated or bullied for being ‘different’ and not given recognition from others, it can impact how they view themselves and their sense of identity. It has been suggested that this has led some individuals with a visible difference to seek reconstructive surgery to ‘normalise’ their appearance (Hamlet & Harcourt, 2015), in the hope they would ‘fit in’ with society (Cash, 2012).

Surgery

Whilst many individuals seek reconstructive surgery for various reasons the potential physical risks of craniofacial procedures are well established. However, there is little information about the potential social and psychological implications of surgery (Asch, 2006; Mouradian, Edwards, Topolski, Rumsey & Patrick, 2006). Some studies support the theory that reconstructive surgery for CFAs can increase self-esteem (Arndt, Travis, Lefebvre & Munro,1987; Kay, Tubbergen, Warschausky & Buchman , 2005), perceived improvement in appearance (Hansen, Kreiter, Rosenbaum, Whitaker, & Arpey, 2003), and enhanced social engagement (Arndt et al., 1987). Other research has challenged this and found that although individuals might be initially satisfied with the outcomes of surgery, the satisfaction often dissipates over time, leaving the children susceptible to future psychosocial difficulties (Arndt et al., 1987; Sarwer, Bartlett, Whitaker, Paige, Pertschuk, & Wadden, 1999; Bruce, 2009). The lack of long-term studies measuring long-term satisfaction and psychological benefits following surgery (Marsh, 2006), has encouraged some health professionals to reflect critically on whether surgical intervention to prevent predicted social and psychological challenges is the most effective route to take when weighed against the potential risks of surgery (Mouradian et al., 2006).

Satisfaction with surgery

Although prior research has examined satisfaction with surgical treatment, it has mainly focussed on parental interpretation of success and not the child's. Indeed, most parents say they are pleased with their child's altered appearance after surgery (Kunz, Lehner, Heger, Armbruster, Weigand, Mast & Peraud, 2014; Wong-Gibbons, Kancherla, Romitti, Tyler, Damiano, Druschel & Burnett, 2009). Bemmels et al., (2013) found that although many adolescents and adults with various CFAs were satisfied with the outcomes of the reconstructive surgery, reporting pleasing changes in appearance and improved self-esteem; others felt that the outcome of surgery did not always meet expectations or justify the challenges associated with surgery. Individuals' unrealistic expectations about what surgery will offer them can be a source of dissatisfaction following surgery (Bemmels et al., 2013). Difficulties following surgery have included unexpected emotions arising from adjusting to one's altered facial appearance. In one study with young adults with craniosynostosis some participants reported an 'identity crisis', which resulted in difficulties in social interactions (Stavropoulos, Hallberg, Mohlin, & Hagberg, 2011).

Whilst the area of syndromic craniosynostosis conditions and midface advancement surgery remains relatively under researched, other CFAs such as cleft lip and palate have been more widely researched in relation to the impact on psychological functioning and surgery. Studies on the psychological impact of cleft lip and palate have produced conflicting results. Although some studies suggest those born with a cleft lip or palate experience more psychosocial difficulties than their peers without a cleft lip or palate, other research has found few differences between these two groups (Rumsey & Stock, 2013). Such findings illustrate the complexity of adjustment to a condition that causes visible differences (Rumsey & Stock, 2013). A recent study investigating factors and processes that contribute to psychological adjustment to cleft lip and palate found that a lack of autonomy in regard to treatment choices had a negative impact on adults' level of adjustment, in addition to unrealistic expectations of these treatments (Stock, Feragen & Rumsey, 2016). The study suggests care is needed to avoid unnecessary uptake of surgical interventions and to prevent distress and disappointment arising from unrealistic expectations of surgery. Psychologists have an important role in helping patients and their families clarify their knowledge, motivations, and confidence, to support decision making, adjustment and quality of life (Rumsey & Harcourt, 2005).

Professional guidance and psychological input have been seen to be valued, assisting families to implement helpful coping strategies and improve adjustment after surgery (Beaune, Forrest & Keith 2004; Taylor & Stanton, 2007). In addition, involvement with other individuals who have the same diagnosis and have undergone similar surgeries has been shown to aid positive adjustment and a sense of empowerment as a result (Ussher, Kirsten, Butow & Sandoval, 2006). However, parents and children are encouraged to be cautious and to think about whether surgery is best for them. This is especially important for considering the impact on long term mental health (Bemmels, Biesecker, Schmidt, Krokosky, Guidotti, & Sutton 2013).

As the area of craniosynostosis and surgical interventions remains relatively under researched and given the centrality of extensive and long-term treatment for syndromic craniosynostosis conditions, the psychological impact of this treatment on individuals and their families is an important topic (Feragen et al., 2016). In addition, whilst facial surgery alone does not appear to be enough to achieve a better quality of life, it is crucial to provide individuals and their families, with precise information, explore expectations and provide psychological support to aid adjustment to facial changes. This also warrants further consideration given the clinical implications on practice.

Aims

This qualitative study aims to explore people's experiences of midface advancement surgery who have a diagnosis of syndromic craniosynostosis in childhood from the perspective of the child and their parents. This will include their experiences of life following surgery.

The study was concerned with understanding children's and parents' lived experiences, an under researched area, the utilisation of a qualitative research methodology was appropriate. Semi-structured interviews were used to elicit detailed narratives of the participants' experiences. The transcripts were individually analysed using interpretative phenomenological analysis (IPA), to explore and interpret participants experiences. The approach was chosen as it was congruent with the research question, which was looking to explore the experiences of children who had undergone midface advancement surgery and their parents, and their ascribed meaning to these experiences. Given that the research findings may affect the way children are treated medically and therapeutically, we were guided by the UN Committee on the Rights of the Child. Article 12 states "… every child capable of forming his or her own views has the right to express those views freely in all matters affecting the child…" Thus, parents and children were interviewed together.

Study findings will be used to provide information to families who are considering midface advancement surgery.

Method

Research and Ethical approval

This study was granted approval from the Doctorate in Clinical Psychology Research Committee (see Appendix F), and awarded University Sponsorship (see Appendix G). Ethical approval was then granted by the Health Research Authority (see Appendix H). Considerations were taken when involving children in the research project. Children were asked if they would like to bring an object that represents their experiences having midface advancement surgery or after surgery. This was to provide all children with a voice in the room and engage them in the interview. All children were given a participant information sheet that was deemed suitable to their age and cognitive ability. The lead researcher asked two teachers and three children of various ages to review all participant information sheets and incorporated any suggested changes.

Sampling and participants

In keeping with IPA methodology, the study aimed to recruit between four and ten participants. Studies using IPA are typically conducted with small samples of participants that share or are bound by a particular experience. Smaller sample sizes allow for detailed analysis of individual accounts as well as the development of meaningful cross-case analysis for patterns of similarity and difference (Smith, Flowers & Larkin, 2009). The criteria for inclusion in the study were children with a diagnosis of craniosynostosis, who had undergone midface advancement surgery, under the care of two regional craniofacial services and their parents. The child's age range for inclusion was between 8 and 16 years but were not excluded if they had an intellectual disability. This age range was chosen as typically children undergo this procedure around 9-11 years of age. In discussions with the experienced craniofacial team, the criteria of undergoing the surgery no less than 6 months ago and no more than 5 years ago was also added. It was important for children to have fully recovered from surgery, but to be able to reflect on their experiences, without hindering recall. Parents were required to speak English, since no translator could be used. Parents were excluded if they had known difficulties that would prevent them from giving informed consent or successfully engaging in the interview, such as intellectual disabilities, as this may have reduced their ability to accurately recall historical experiences or meaningfully engage in the interview (King et al., 2003).

In total, five families consented to be contacted by the research team, following recruitment from the medical team. All five families were eligible and recruited to the study. All parents were mothers, with three male children and two female children. One child had a diagnosis of moderate intellectual disability and did not participate in the interview. Another child had social communication difficulties and fully participated. Four families were British, one parent was Columbian with their child being of Columbian and English heritage. Children's ages varied from 10 to 14 years. All had undergone surgery between 6 months and 4 years prior.

Procedure

Participants were recruited through a regional craniofacial service using purposive sampling (Smith et al., 2009). Clinical staff identified potential participants prior to their routine clinical appointments and offered them information about the research when they attended their appointments. Parent participants were given an information sheet and a detailed consent form and parental consent form (see Appendix I-L) covering all aspects of the research including informed consent, confidentiality and anonymity, and interview recording process. Child participants were also provided with an age-appropriate information sheet and consent form (see Appendices L and M). Participants were provided with the researcher's contact details should they wish to receive further information or alternatively interested potential participants could consent to their information being passed to the researcher, who contacted them in a timely manner.

The lead researcher telephoned all families who expressed an interest in taking part. This enabled the researcher to check that individuals met the inclusion criteria and provided an opportunity to answer any questions. Those who wished to proceed arranged a date to meet with the researcher for a face to face interview. At interview, the researcher refreshed participants on the contents of the Participation Information Sheet and gave them the opportunity to ask any questions and sign consent forms. Children were asked if they wish to participate; assent was not assumed from parents.

Four families were interviewed at their local children's hospital, on dates that coincided with an outpatient appointment. One interview was booked for March 2020 and in light of the COVID-19 pandemic, and precautions put in place by local Trusts, this interview took place using video conference software. To aid a deeper understanding of this shared experience one dyadic interview took place with children and their main care-giver. In addition, as children with intellectual disabilities were included in the study, an adult/ parent would have been needed to be present with the child and so for consistency it was decided that families would be interviewed together.

Interviews followed a semi-structured interview schedule (see Appendix N), which was developed in collaboration with a research supervisor who has extensive experience in the field. The schedule was flexibly followed to support the natural flow of conversation (Smith & Osborn, 2008). Following the interview, each participant was thanked for taking part. All interviews were audio-recorded on a digital voice recorder. Two interviews were transcribed by the researcher and the remaining interviews by a University approved transcriber. Once transcribed and data analysis complete the recordings were erased, and any identifiable details deleted or anonymised, with each participant given a pseudonym to protect anonymity.

Data analysis

Interview transcripts were analysed using IPA, as it enabled a focus on the rich and individual experiences of participants (Hugh-Jones, Madill, Gibson, Keane & Beestin, 2012). As children and families were interviewed together data from each interview was analysed together. IPA enables interpretations about how participants make sense of their experiences and the association between their narratives and their actual experience (Shaw, 2010). The primary aim of the analysis can be characterised by moving from the particular to the shared, from the descriptive to the interpretative (Smith et al., 2009).

The researcher analysed the transcripts in line with the framework outlined by Smith et al. (2009). First the researcher immersed themselves in the data by listening to the audio recordings of each interview and reading and re-reading each transcript. Initial codes of descriptive themes for each transcript were generated, while linguistic and conceptual comments also made. Based on shared aspects of experience and meaning, these exploratory comments were developed into emergent themes and then grouped into super-ordinate themes. This was completed for each transcript. Super-ordinate themes across all participants were combined and grouped into higher order master themes. Through multiple discussions with a research supervisor, the researcher renamed and reorganised themes to develop more concise accounts of participants' experiences (Smith et al., 2009).

Quality and validity

The researcher worked closely with their supervisors by sharing transcripts, initial coding and emergent and super-ordinate themes, as they were developed and refined. Final master themes were discussed with supervisors and feedback was sought concerning the level of the interpretation of the analysis. As IPA is a subjective methodology, sharing of this information ensured transparency and aimed to maintain quality and validity.

Reflexivity

To help develop a sense of self-awareness in the research process, the lead researcher made brief notes before and after each interview, acknowledging any feelings, thoughts, or actions that may have influenced the data generated. The researcher's role in the research development is acknowledged through the through a process of double hermeneutics; the acknowledgement that participants are attempting to make sense of their own experiences, which in turn are interpreted by the researcher. Thus, it is important that the researcher attempts to bracket personal beliefs and assumptions and adopts a position of reflexivity (Dahlberg, 2006; Starks & Trinidad, 2007). Since the lead researcher was completing a Doctorate in Clinical Psychology during the data collection period, her place in the process became part of the IPA analysis.

The researcher was aware of how their interest in clinical health psychology and visible difference, influenced their decision to research this field. They reflected on how their interest and expertise in this area potentially influenced their interpretation of participants' accounts. Before the commencement of the research, the researcher did not have any prior experience of working with individuals with craniofacial conditions. However, the research sparked an interest and passion working with this population and the researcher went on to complete a 12-month placement with the craniofacial team as part of their clinical psychology doctoral training and has since started a qualified job within the area. The researcher reflected on how when making interpretations of the participants' experiences, their prior experiences arose in their memory, which may have taken the researcher away from the unique voice of each account. Through the use of supervision and self-reflection

Name	**Age**	**Time since surgery**	**Who participated**
'Anna'	10	9 months	Child and mother
'Amelia'	14	2 years	Child and mother
'Oscar'	14	3 years	Child and mother
'Gregory'	12	4 years	Mother
'Mason'	13	3 years	Child and mother

throughout the process, the researcher was able to identify any unexpected preconceptions as they arose (Smith et al., 2008) and reflect upon this with research supervisors.

Results

Demographic information is included in Table 1, for ease of reference.

Table 1: *Participant demographics*

Four master themes were identified through the analysis and are presented with their corresponding subthemes in Table 2

Table 2: *Master themes and sub-themes*

Master theme	Sub-themes
Life before	• Cruel world • Why are we doing this?
Transitional stages	
The intrinsic role of support	• Developing a collective connection & enabling shared knowledge • True friendship • Healthcare support
Managing and making sense of the new me	• Surgery as a catalyst for growth • Struggles with a new identity • Unexpected consequences

Master Theme one: Life before

This master theme illustrates participants' experiences before surgery, their interactions with others and the sense they made of those interactions. The decision-making process leading up to surgery was also discussed.

Cruel world

"You love your children the way they are but the world is a cruel place" (Mason's mother).

The notion of a cruel world and the intolerance of difference was mentioned by children and parents. All families commented how children received stares, comments or remarks from others about their appearance before the surgery. All parents discussed the challenge of managing and making sense of social reactions toward their child. For some, this challenge provided a direct conflict and tension to a parental instinct to protect their child.

"*As parents we're always checking, no one is being mean to your kid"* (Mason's mother). Children spoke about how peers would tease them about their appearance *"they would call me pop eye"* (Mason). This resulted in feelings of distress, impacting their confidence and how they viewed themselves, "*It made me feel sad and not like how I look"* (Anna). Some parents commented that their child would avoid certain situations and believed this to be due to fear of interacting with others.

Social responses created a considerable challenge to both children and parents. The experience of negative social responses was difficult for all and seemed to agitate parents' own beliefs that their child should not be treated differently from others. Parents commented on how both adults and children would react in a negative way to their child, "*parents moving kids away, which is not nice. It's not contagious"* (Mason's mother).

*"I would often nearly lose my top with people….people can be so ignorant" (*Gregory's mother).

Amelia stated that although others stared or commented on her appearance, she interpreted these interactions as curiosity, which was experienced as a less harmful interaction. Although Amelia appeared to have adopted a positive coping strategy of cognitively reframing events, she appeared somewhat conflicted, as later when asked what did she hope would be different after the procedure she stated *"hmm that I wouldn't get hmm questions about the way I* *looked and hmm… wouldn't get them looks off people",* suggesting that perhaps this did cause her some upset or a desire to be less different.

There was also an aspect of experiencing social reactions that seemed to lead to wider reflections around societal attitudes toward difference with participants suggesting that we live in a world where looks matter and they felt the surgery would be beneficial so they would 'fit in more'.

"because of his appearance we thought well that's not going to help him, living in a society where looks do matter. So, I guess we tried to make it a little bit easier for him" (Mason's mother).

"In society nowadays, I know it shouldn't but it does matter the way you look" (Gregory's mother).

Why are we doing this?

For two participants surgery was suggested for functional improvement, whilst for others aesthetics was the main motivation for surgery. Families' narratives suggested that despite the motivation behind the surgery, all hoped it would improve their/ their child's quality of life.

Two families described how the surgery was recommended to improve breathing and sleeping difficulties. When asked if they would have gone through the procedure if these medical reasons were not present, both parents commented that they likely would have still gone through with the procedure for aesthetic reasons:

"It's very hard because again…I guess we would have…It's hard because you think are we doing this for him or are we doing it for us so we don't have to deal with people looking. It's not good for him either to be the subject of staring, comments, people moving away. So, in a

way you do it to protect them and help them, so I guess we would have, to be perfectly honest" (Mason's mother).

Interestingly, one child commented that he felt the operation was done for functional improvement and although he stated that comments from others upset him, he would not have had the operation only for aesthetic reasons. His mother disagreed and stated she felt aesthetics was more of a concern for him than he was saying.

"*This bit's really hard to answer, especially while he's in the room, but big things were obviously the aesthetics for him. I think it was more of an issue than probably what he's saying. I suppose it's more from my experience and I know what people see and what people and how people are. You can see how people are…reacting, so it was about the aesthetics"* (Oscar's mother).

When discussing aesthetic motivations for surgery, children reflected on their experiences of the world and their perception of difference as negative or shameful. Language used such as "*change me*" (Anna), "*less different*" (Mason) and "*more like others*" (Amelia) conveyed a sense that children believed if they changed something about themselves to fit in more perhaps, they would be more accepting of themselves and accepted by others. This was also echoed in parents' accounts of wanting to "*normalise appearance*" (Amelia's mother) due to outside influences and believing their child would do better in the world if they fitted in more. "*In a way it's almost like…if it wasn't for outside pressures, it wouldn't be needed"* (Oscar's mother).

Amelia spoke about wanting to "*normalise*" her appearance for her own sake and reflected that before the surgery she stood out and had no choice over this. Now however, she can choose to be different:

"I didn't have a choice before… Before you naturally stood out but now I can choose to stand out, if I want to stand out… I have my own style" (Amelia).

Amelia and her mother commented that Amelia dressed differently to her peers and liked to stand out, suggesting that she valued difference, but the important factor was the choice to stand out or not, and perhaps the ability to do something about the difference.

Parents were more vocal than in other parts of the interview, when discussing motivations for surgery. This perhaps mirrored their involvement in the decision-making process. Whilst they spoke about encouraging their child to be involved, consent for the procedure fell to them because the child was not old enough. Children stated that the procedure was explained to them, but their understanding about their change in appearance was dependent upon age and cognitive ability.

Master Theme two: Transitional stages

The second master theme illustrates how participants viewed transitional stages in adolescents as an important stage in children's lives. No distinct subthemes were identified within this master theme.

Three parents reflected on their child's experiences of teasing and staring from others and believed it would be beneficial for the procedure to be done before high school. It appeared these parents believed high school would be more challenging if their child looked different, with these parents stating the timing of surgery was very important and played a role in the decision-making process for them.

Oscar's mother commented *"We talked about it a lot, about that (timing of surgery)… I think it helped his transition into high school as well. It would have been difficult for X to go into high school looking the way he did."*

Parents were more vocal when discussing the timing of surgery, perhaps again mirroring their involvement in the decision- making process.

Amelia was due to have midface advancement surgery before high school, but due to medical reasons, it was postponed until she was 12 years old, end of year 7 of high school. Amelia reported she was glad she waited for the operation; she believed she had a better understanding and had built a support network in school. Thus, she felt it was her choice to have the operation:

"At that age really you can make your own decisions. So I think that was like the best time I think"

Although Amelia's mother commented that having the surgery cancelled before high school was "*bad news*", she reflected that because of Amelia's age she felt Amelia coped better with the procedure.

Mothers reflected on the fact that their children had naturally started to become more aware of their appearance. They recognised that they looked different to others. Parents appeared to interpret this recognition of difference as a negative experience and as reinforcing child's views of themselves as 'different to others' and the need for corrective surgery.

"it wasn't until he was about 8 that he started to be like why are people looking at me?...so we had to explain" (Mason's mother).

Master Theme three: The Intrinsic Role of Support

The master theme of the Role of Support captures participants narratives of the importance of feeling supported and connected to others.

Developing a collective connection and enabling shared knowledge

Three families discussed how connecting with other children, who had undergone midface advancement surgery and their parents, was particularly helpful. This support provided parents with a shared connection and a shared understanding of their parenting role. There was also a benefit of developing these connections that related to a pragmatic benefit of increasing their knowledge in relation to the procedure and after care. Amelia's mother captured the value of parent-to-parent support, which related to a sense of true understanding and relatability:

"I know you're told that (what happens) here (hospital) but in reality, when get home what's it like, really what is it like…(and) all practical things. The mum was really helpful. I think every family should be buddied up with someone…that should be offered or encouraged anyway, definitely".

The importance of connection seemed to also relate to participant's initial feelings of uncertainly and the unknowing aspect of the procedure:

"helpful to alleviate some fears and concerns, from people who truly know" (Anna's mother).

"it was good because could see, like how he changed over the time and hmm he's like transformation from the beginning and afterwards was really good so. It made me more confident about it" (Amelia).

Two families did not speak with other families. However, one mother felt she could offer valuable support and share knowledge with others after her child had the procedure. This mother spoke with other families who were considering this procedure, so she valued this role of support-worker.

Developing these support networks was highly valued and offered both emotional and practical support.

True friendship

Children and mothers also spoke about the importance of supportive friends, who accepted them for who they were and helped them overcome the barriers of returning to school.

Amelia commented *"your friends, got people who really, really know you…look out for you."*

"he had a really good group of friends …that accepted X the way he was. They were going to be there for him. When we went back to school, his friends were always looking after him, making sure" (Mason's mother).

Children spoke about friends visiting them in hospital and at home, with three participants discussing how seeing friends before returning to school boosted their confidence to return to school. These close connections created an underlying sense that their friends cared and would support them through any experience of stigma. This enabled them to feel more secure in themselves.

The role of support offered by healthcare services and professionals was also a prominent narrative in children's and mothers' accounts.

Healthcare support

Child and mother's narratives reflected the sense of trust and gratefulness they had with the multidisciplinary team

"feel know them (team) so well, so they are able to support you and you have a high level of trust for them" (Gregory's mother).

The dyads spoke about how they were grateful for the nursing staff going into schools to explain the procedure, such as having a frame on and other changes in appearances. They felt this helped them prepare for going back to school and *"it kind of like took weight off me when I went in"* (Amelia), it "*takes the pressure off"* (Oscar's mother) parents and children having to explain themselves.

The sharing of knowledge and discussions with the team also helped families feel prepared. Children and parents spoke about the value of psychological support around the decision-making process and preparation for the procedure. Whilst one family spoke of being offered psychological support after the operation due to adjustment concerns, another felt they did not know where to turn to for support after the operation. The family without the psychological support appeared to feel this was inadequate care given the emotional challenge faced during this period.

Master Theme four: Managing and making sense of the "new me"

The fourth master theme illustrates how participants made sense of their life after midface advancement surgery. Their narratives suggest that, looking back on their lives, they believed the surgery to be a success. All mothers emphasised the positive impact it had upon their child's quality of life; children also expressed similar positive thoughts. However, this journey was not always easy since some participants encountered difficulties along the way.

Surgery as a catalyst for growth

Two mothers discussed how functional improvements such as clearer speech, improved breathing, and sleep, had a huge impact on their child's quality of life.

"Amazing results, the CPAP machine has gone, breathing has improved, he can sleep now so he feels better, behaviour has improved. He looks great. Fantastic" (Gregory's mother).

All mothers commented positively on the way their child's appearance had changed. Children also commented similarly. Amelia commented that she now looks the *'way (I am) supposed to look',* similarly Mason's mother stated *"looking normal has helped him".* Statements such as these imply that before the operation these children did not look 'normal' and something about their appearance needed to be corrected. One parent reflected on this process of thinking and reasoning:

"In a way it's almost like…if it wasn't for outside pressures, it wouldn't be needed…I suppose there's a balance between looking back and thinking well what are you saying about how he used to look. It's a really difficult place to fit yourself in to. It's a difficult one." (Oscar's mother).

Children reflected on how they felt changes in their appearance were positive and how this impacted on how they viewed themselves. Mason commented *"I feel better than before…it's changed my life".*

Some made comments about appreciating the fact that they looked more like family members; they also spent more time looking in the mirror admiring their appearance. Gregory's mother commented that Gregory, who does not communicate verbally now kisses himself in the mirror, displaying approval for his new appearance.

The reactions of others appeared to be important for families, with children and parents recalling times when others commented on the changes in their appearance, which strengthened their own approval of these changes.

"*yeah I think it made me feel better about myself because, also after you get quite a lot of compliments, like oh you look really well and hmm it such a big change in a good way …I think also make you feel like better about yourself because you get so many compliments*" (Amelia).

There was also a sense that positive reactions from others offered reassurance to parents that they had made the right decision.

Children spoke about how after the operation, the teasing and staring from others reduced, Oscar recalled how "*People stopped calling me pop-eyed and stuff"* and Anna stated "*people weren't mean anymore.*" Children also commented how they were not longer "*singled out" (*Oscar) for being different.

Having their 'difference' removed and experiencing positive responses from others appeared to leave children feeling as though they now fitted in and were accepted more by peers.

"*I don't get like, yeah I don't get looks, the looks I used to get, I guess people would just, if I'm out and about, like stranger….yeah, I think it makes me feel like I fit in more… I feel more like, accepted. Like people" (*Amelia).

Oscar, also felt that being accepted enabled more positive interactions with peers *"People have like got to know that, to know me a little bit better."*

Parents also reflected about how the reduction in teasing and stares from others had a positive impact on the wider family, with parents and siblings' role as 'protector and defender' no longer needed as frequently.

These factors combined; more positive view of self, positive reactions from others coupled with less negative interactions led to an increase in the children's self-confidence. This increase in confidence was noted by children and parents alike: "*it makes you feel better about yourself and you know more confident*" (Amelia). Anna's mother commented that Anna is now more confident in her own abilities and now holds a positive view of herself. Similarly, Oscar's mother reflected "*I talk about the aesthetics, but for me what goes with that is the emotional impact and your confidence. Your self-esteem".*

This increase in confidence had a further impact on children, such that they were more vocal and increasingly enjoyed socialising and making new friends. Gregory's mother reflected that before the operation he *"used to not like going into places with a lot people, I used to put it down to the noise… He could ask to leave. But now he has new confidence. He loves going to a party, socialising and gets more involved now".* Gregory's mother felt this new confidence was due to a change in his appearance that others would no longer comment on.

Struggles with a new identity

Children and parents recalled the initial shock in seeing the visible changes in appearance after surgery. However, for some people, this change resulted in an unexpected struggle to adjust to their new appearance.

Oscar recalled how after the operation "*I wasn't too happy about pictures being taken, but I was sort of like grown in to having pictures taken*". His narrative suggested that it took time for him to get used to a new appearance. Similarly, Anna's parents reported how although the change in appearance was positive overall, it took some time for the whole family, including Anna to adjust to a different appearance.

Mothers said they often pictured the child before surgery, *"In your mind still think of how looked like befor*e" (Amelia's mother).

Amelia recalled how she also struggled for some time to adjust to a new appearance and sought support from the team:

"obviously my face had changed. It was like getting used to seeing myself looking like the new, new version. I think it was just that, I think that takes time to get used to… But I obviously had a different face, for so much longer, its take you a while…You have got used to it being you."

Mason’s mother also recalled that his behaviour changed following the operation. He became angrier, impulsive and did not want to talk to others about how he was feeling. She stated they never ‘got to the bottom’ of this but there was some suggestion this could be linked to difficulties following a change in appearance and experiences.

Suggestions that a ‘normalised’ appearance would automatically alleviate distress for these children was not founded. Although children described fitting in and feeling more accepted, the changes in appearance had a destabilising effect on their identity. Intriguingly, two children who struggled with adjustments to changes in their appearance reported that they are not satisfied with another facial feature unrelated to surgery. Thus, their insecurities may have shifted to another facial feature.

Mothers stated that people’s comments about their children looking like different people was distressing and neglected the fact that identity was based on more than looks. Some mothers became angry at this questioning of their child’s sense of self. Gregory’s mother recalled how people would often say he is *“different boy”* or it *“doesn’t look like him”*. She recalled how although people were not purposely cruel with these comments, she felt it was important that others recognised that he was still Gregory inside. Similarly, Amelia expressed a dislike for people commenting she was a different person, leading her to question her identity. She reflected on a few occasions her annoyance “*oh everyone said not look different but different person. Because I’m not a different person. Because my personality is still the same, it’s just my bone structure, that’s changed.”, “I’m not a different person, I just have a different face saying I’ve changed completely didn’t like that. Can still tell it’s me”.*

Unexpected consequences

Whilst some struggled with adjustment and self-identity, other unexpected consequences were also discussed. Two children recalled feeling anxiety following surgery. They felt fearful around hospitals and about future operations; they become unwell as a result. This increase in anxiety may have also been related to having limited understanding and less control over the decision to have surgery, due to age.

Mason's mother recalled how when he needed other procedures after midface advancement surgery, he became anxious:

"*start shaking a bit saying I don't want this, don't want that. That he's sick. So, we're working on that a bit…But that has been one of the major things after the operation, he goes into a panic"*

Similarly, Anna and her mother explained how since the procedure, Anna had been fearful of future operations, fearing a further hospital admission. Anna recalled the pain of the operation and the long recovery process, which fed her fears about the future. These narratives show that participants developed an unhealthy association between hospital environments and the pain and distress of surgical operations.

Discussion

This is the first study, to the researcher's knowledge to explore and make sense of children's and parents' experiences of undergoing midface advancement surgery. Using semi-structured interviews with five families, the results produced four main themes. The themes will be discussed in relation to existing research, highlighting significant issues and clinical implications.

When discussing life before surgery, all families commented on how children received unwanted social reactions about their appearance. This resulted in feelings of distress, impacting children's self-confidence and identity. Previous research has also found that children with a visible difference are often subjected to teasing and bullying (Hamlet & Harcourt, 2015; Dunaway, 2004) impacting a children's self-esteem. Participants reflected on wider societal attitudes towards difference, suggesting that we live in a world where appearance matters. Thus, they felt that if they changed something about their appearance, they would ‘fit in’ and be more accepted by others. On reflection, the problem with intolerance of difference is not difference itself but societies’ negative view of difference. It has been suggested that societal attitudes to visible differences have improved in recent years (Pausch et al., 2016), often with difference being more accepted in early adulthood and beyond (Livneh, 2012). Nevertheless, social discrimination of difference in appearance still occurs, with previous research demonstrating an association between perceived social discrimination and reduced satisfaction with appearance (Roberts, 2014). Those with a visible difference have described the ongoing challenge of living in a modern society that simultaneously tries to accept difference, whilst also placing more emphasis on appearance than ever before (Hamlet & Harcourt, 2015). Social media perhaps playing a role here, portraying unrealistic expectations of body image. In addition, visible differences are often portrayed in negative stereotypes or are often ‘invisible’ in mainstream media (Wardle & Boyce, 2009).

Participants’ narratives suggested the surgery had been successful, with all participants emphasising the positive impact it had on their own or their child’s quality of life. Some spoke of the functional improvements following surgery; all spoke of the pleasing changes to appearance. Participants spoke of a more ‘normal’ appearance that enabled them to fit in more with peers, reduced unwanted social reactions and increased self-confidence. Similar findings have been noted in previous research with individuals with visible differences (Arndt et al., 1987; Hansen, Kreiter, Rosenbaum, Whitaker, & Arpey, 2003; Tubbergen, Warschausky & Buchman, 2005). Despite successful outcomes, some described struggles adjusting to an altered facial appearance. This change in appearance led some children to question their sense of self, with unexpected emotions arising from a change in appearance. These individuals are not alone in this feeling. Bemmels et al. (2013) suggested that the outcomes of surgery do not always meet people's expectations and sometimes the difficulties associated with surgery outweigh the positive outcomes. Even young adults experience an 'identity crisis' following surgery, resulting in other unexpected outcomes from surgery (Stavropoulos et al., 2011).

Despite some unexpected outcomes, such as difficulties with adjustment and heightened anxiety around future operations and hospital environments, all parents reported that they felt the surgery was a success and on reflection were happy with the decision for their child to have the procedure. As a way of potentially managing the anxiety around making this decision and going through with the surgery, there may be a bias amongst parents to not recognise at least some potential consequences of the surgery.

Participants discussed the importance of timing of midface advancement surgery. Whilst for some the functional need for the procedure dictated timings, for others they spoke about children becoming more aware of their appearance and a hope for it to completed before high school. Children naturally start to become more aware of their appearance around 8 to 9 years of age (Backes & Bonnie, 2019) and begin to notice differences or similarities between themselves and others. If this difference is interpreted by a child with a visible difference or their parents as negative or shameful in some way, it can lead to distress and families to seek appearance altering interventions.

Timing was important to mothers and children, such that people valued earlier treatment. Parents spoke about the importance of their child having the procedure before high school. During discussions around motivation and the decision-making process prior to surgery, mothers more vocal than in other parts of the interview, perhaps mirroring their involvement in the decision-making process. Mothers reported societal pressures and made predictions that high school would be more challenging for a child with a visible difference. They spoke about hoping the surgery would reduce unwanted social reactions and enable their child to fit in more with peers and be accepted by others. This was supported by previous research where decisions about the timing of appearance altering surgery was often influenced by psychological issues, such as a desire to avoid teasing at high school (Dunaway, 2004).

Participants in the present study underwent the procedure during pre or early adolescence. According to Eriskon's model of development (1950) adolescence is a time when individuals try to establish a sense of personal identity and figure out where they fit within the world. During the adolescence period of life, the self is not fully developed, leading to an increased vulnerability in changes of the appearance or the self (Stavropoulos et al., 2011).

Children's natural curiosity about their appearance and the development of identity in adolescence raises the question of when the best time is, if at all, to intervene with appearance altering surgery. Should interventions be recommended in early adolescence before children fully adjust to their appearance and as a way to protect them from potential future unwanted social reactions or predicted difficulties in high school. Alternatively, one could argue children with a visible difference should be supported in developing a positive sense of self and allow natural adjustment to appearance. With maturity these individuals perhaps could then decide for themselves whether to proceed with surgery, with the ability to better comprehend potential outcomes. With this said there is no one size fits all approach. Decisions will be down to individual choice, depend upon cognitive ability, experiences, available support networks, personality, and medical considerations.

The role of social support was important for mothers and for children, highlighting the need for multiple support systems; friendships and parent-to-parent support was valued by both children and parents. The positive influence of social support is well documented within paediatric models of adjustment (Thompson, Gil, Burbach, Keith, & Kinney 1993; Wallander & Varni, 1992) and cited as an important feature in the resiliency model of family stress, adjustment and adaptation (McCubbin & McCubbin, 1993). Participants valued connecting with other individuals who had the same diagnosis as themselves or their child and had undergone the same surgery. The importance of this support was also highlighted by Ussher et al (2006), where connecting with similar individuals was shown to aid positive adjustment and a sense of empowerment as a result. Parents in this study valued the pragmatic aspects of information sharing in addition to emotional support, helping them feel more prepared in supporting their child during this time.

Participants wanted to feel supported by healthcare professions. The relationship between patients, parents and healthcare professions is prominent in the research literature, with positive child-physician and parent-physician relationships associated with satisfaction of care within paediatric settings (Swedlund, Schumacher, Young & Cox, 2012; Taylor et al., 2007). Within these relationships however, healthcare professionals have a duty of care to be aware of and reduce the inherent power imbalances that can occur. This may offer a role for psychologists, in developing team formulations, to highlight the role of professional power more explicitly within parent-provider relationships. Trust within these relationships was also reported to be an important factor by parents. Research has however suggested that trust, can at times be fatalistic, and appeal to a surgical heroism, whereby possible complications or risks of surgery could be downplayed by parents to protect the trust they place in healthcare professionals (Nelson et al., 2012). With this in mind and coupled with a parent’s moral desire to ‘do the right thing’ and enhance social inclusion for their child (Nelson, Caress, Glenny & Kirk, 2012), great care is needed when discussing potential surgeries with families for aesthetic reasons. There is a potential that feeling grateful that a child can be offered an intervention to ‘normalise’ their appearance and having full trust in a health professional, that parents would not question their own motivations for surgery. Nor question reasons why perhaps they would decline this opportunity for their child. Furthermore, ‘Regret theory’ proposes that seeking to avoid feelings of regret arising from not taking action, maybe a strong influence on individual’s drive to do something rather than nothing (Loomes & Sugden, 1987).

Clinical implications

The results have several clinical implications that can help inform effective patient-centred care for both children and parents of children considering or undergoing this surgery.

Although shared decision making is not always easy to navigate, Healthcare services aim to adopt a model of patient-centred care which includes shared decision making between patient and provider (Dwamena et al., 2012). Research suggests decision making around treatment options in healthcare settings can be influenced by parents’ moral desire to ‘do the right thing’ (Nelson, Caress, Glenny & Kirk, 2012). Supporting parents to consider their personal meanings and feelings attached to reconstructive surgery for their child, may help parents to feel more confident in their decision making and ability to support their child. It is also of the upmost importance to support children and parents in setting realistic expectations of surgery and provide psychological support not only in the preparation phase, but as routine follow up post-surgery. This may capture any difficulties and enable the implementation of timely interventions, if necessary.

Support exploring children’s experiences of social interactions and the meanings they construct from this should also be considered. This could be done through a Cognitive

Behavioural model, for example. Research suggests that reappraisal of events is a key component in individual's adjustment to living with a visible difference and sense of self-worth. Where social stares or comments are reappraised as curiosity, rather than a threat, this appears to reduce the emotional impact of the event. It can also lead to a reconstructed meaning of difference and increase in self-acceptance (Beaune et al., 2014). This sits in line with Roger's (1959) theory of personality development that suggests that the Self is influenced by the experiences one has and their interpretation of those experiences. Parents' experiences of perceived negative social reactions toward their child should also be considered. Previous research suggests parents' reactions and management of social interactions, along with their feelings about their child's appearance can be assimilated and internalised by their child (Kearney-Cooke, 2002). This can in turn influence a child's own perceptions of body image and feelings of self-worth (Kearney-Cooke, 2002). Exploring this with families, through a systematic model or Cognitive Analytical model with individuals in a non-stigmatizing and non-judgemental way may be of value.

Another key clinical implication involves the co-ordination of healthcare services with relevant charities and agencies to ensure support networks can be developed with those with similar conditions or considering comparable surgeries. In addition to the emotional support participants gained from a network of peer support, there was also a practical aspect that related to information sharing and sharing of resources. Within services 'buddy systems' could also be set up to help families make these connections. In addition, it hoped the information from this study will be used to produce information documents to provide to families who are starting out on this journey.

Limitations

The limitations of the current study should be considered. Firstly, this research was exploratory in nature, employing IPA methodology, using a small but detailed account of children's and parents' experiences. Therefore, it is important to acknowledge that the results are not attempting to generalise to the experiences of all children and parents who undergo midface advancement surgery. However, it gives a rich understanding that readers can place into the context of their own knowledge (Smith et al., 2009).

It is important to acknowledge the impact of completing interviews with children and parent together. Whilst this was done to generate a deep understanding of the area of interest and gain children's perspectives, it does pose a limitation. Some parents stated that it was difficult to speak freely in front of their children and it may have hindered some children's accounts, impacting results.

In consideration of the sample, the absence of any fathers in the present research is another limitation. It is plausible that the results do not portray salient aspects of fathers' experiences. For instance, Mason's mother indicated her husband responds differently to comments or stares from others towards Mason, which may have produced different narratives around how parents manage and make sense of social interactions. The absence of fathers within paediatric health research remains an ongoing issue (Kazak, Rourke & Navsaria, 2009). This study utilised purposive sampling which may have led to an unintentional bias.

Another limitation concerns a broader methodological issue: the influence of the researcher in the study. Viewing this as a limitation may well be fallacious as IPA is interpretative in nature. As Yardley (2008) comments, qualitative researchers accept the inevitable influence they have on the research process, with reflexivity considered throughout. The generation of an audit trail (Appendices O to Q) offers assurances to the reader regarding the credibility and validity of the analysis.

Research implications

The present study has captured the rich experiences of life before and after midface advancement surgery and has provided insight into both the triumphs and emotional challenges of this surgery. Future qualitative research is recommended and should consider longitudinal designs using multiple interviews to better capture experiences over time. Future research should prioritise both children and parent's personal and social meanings attached to reconstructive surgery. Children's experiences are often overlooked in research (Hadfield-Hill & Horton, 2014). However, the present study has demonstrated the value and importance of hearing children's narratives. The experiences of struggling with an altered appearance should also be further explored in more detail. This would offer valuable insights, impacting on clinical practice and the care individuals receive. It would also be of interest to explore the long-term emotional wellbeing and social outcomes of operated individuals compared to unoperated individuals, with a diagnosis of craniosynostosis.

Conclusion

This study utilised IPA to explore children's and parents' experiences of midface advancement surgery. The findings highlighted the ongoing challenges presented by social responses towards difference. With societal pressures around appearance contributing to the decision-making process to undergo this appearance altering surgery. The study identified that whilst for some, surgery acted as a catalyst, for others unexpected outcomes occurred. The value of peer support networks and connecting with others was demonstrated both for children and parents. The findings also highlight the importance of including both children's and parents' experiences, with children's experiences often overlooked in research. Finally, there is a great need for more research in this area to truly capture the experiences of appearance altering surgery and the meanings families ascribe to these interventions.

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Appendices

Appendix A. *The Journal of Psychology and Health Instructions for authors*

About the journal

Psychology & Health is an international, peer reviewed journal, publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

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Manuscripts should be compiled in the following order: title page; abstract; keywords; main text; acknowledgements; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figure caption(s) (as a list). **Word limits**

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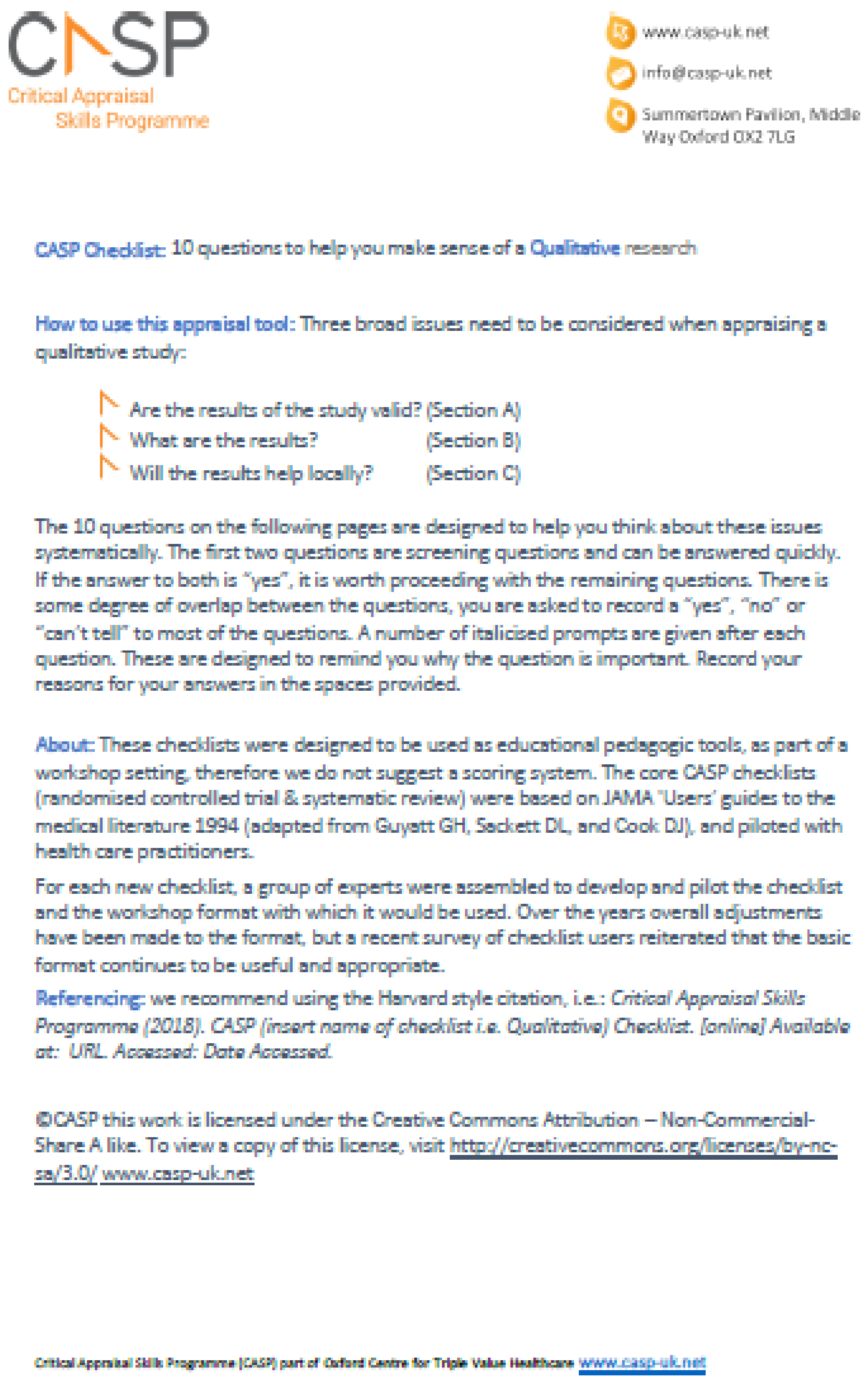
Appendix B. *SPIDER tool for the identification of search terms.*

Sample	Children Adults Individuals
Phenomenon of **I**nterest	Craniofacial conditions The databases will be searched using the key words. Population terms will include: "visible difference", "visible disfigurement", “visibly different", "visible deformity", “disfigurement”, "altered appearance", "Cleft Palate", "Cleft lip", craniofacial, "cranio-facial" ,"craniofacial anomaly”, Craniosynostosis, synostosis, "apert syndrome", "crouzon syndrome", “muenke syndrome", “pfeiffer syndrome", "saethre-chotzen syndrome", "craniofrontonasal syndrome", "Antley-Bixter Syndrome", "Baller-Gerold Syndrome", “Carpenter Syndrome", "Cloverleaf Syndrome", "Jackson-Weiss Syndrome", “Multiple Suture Craniosynostosis", “Opitz C Syndrome", "Osteoglophonic Dysplasia", "Sagittal synostosis", "Coronal synostosis", “Metopic synostosis", “Lambdoid synostosis", "Syndromic synostosis","treacher collins syndrome".
Design	Interviews Surveys Open ended questionnaires Focus groups Case study In depth
Evaluation	Adjustment Psychological factors will include: Adjustment, "emotional adjustment", “psychological adaptation", " psychological adjustment", "psychosocial adjustment", “adjustment disorder*", "positive adjustment", "adaptive behavio*r*", coping, "emotional wellbeing", "emotional well-being" The search strategy will be amended appropriately for each database
Research type	Qualitative studies/research Mixed methods Grounded theory Narrative Thematic analysis Phenomenology

	Discourse IPA

Search terms (combined using Boolean operator “OR” within terms and “AND” across terms)

Appendix C. *CASP blank form*



CASP
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CASP Checklist: 10 questions to help you make sense of a **Qualitative** research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

- Are the results of the study valid? (Section A)
- What are the results? (Section B)
- Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is "yes", it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a "yes", "no" or "can't tell" to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

CASP
Critical Appraisal
Skills Programme

Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- what was the goal of the research
- why it was thought important
- its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
- Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

4. Was the recruitment strategy appropriate to the aims of the research?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If the researcher has explained how the participants were selected
- If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
- If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If the setting for the data collection was justified
- If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
- If the researcher has justified the methods chosen
- If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
- If methods were modified during the study. If so, has the researcher explained how and why
- If the form of data is clear (e.g. tape recordings, video material, notes etc.)
- If the researcher has discussed saturation of data

Comments:

6. Has the relationship between researcher and participants been adequately considered?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider

- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

8. Was the data analysis sufficiently rigorous?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider
- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
- To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes ☐
Can't Tell ☐
No ☐

HINT: Consider whether
- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

CASP
Critical Appraisal
Skills Programme

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix D. *CASP scores for included studies*

Study	Aims	Methodology	Research Design	Sampling/ recruitment	Data collection	Reflexivity	Ethical issues	Data analysis	Findings	Value of Research	Overall
Lowenstein et al. (2008)	1	3	2	3	3	1	1	2	3	3	22
Beaune et al. (2004)	3	3	3	3	3	1	2	3	2	3	26
Chetpakdeechit et al. (2009)	3	3	3	3	3	1	1	3	3	3	26
Eiserman (2001)	1	3	3	3	3	1	1	2	3	3	23
Havstam et al. (2011)	3	3	3	3	3	2	3	3	3	3	29
Riklin et al. (2019)	3	3	3	3	3	1	1	2	3	3	25
Roberts & Shute (2010)	3	3	3	3	3	1	2	2	3	3	26
Searle et al. (2017)	3	3	2	3	3	1	2	3	3	3	26
Stavropoulous et al. (2010)	3	3	2	3	3	2	2	3	3	3	27
Stock et al. (2015)	3	3	3	3	3	1	3	3	3	3	28
Stock et al. (2016)	3	3	3	3	3	1	3	3	3	3	28
Tiemens et al. (2013)	3	3	3	3	3	1	3	3	3	2	27
Kappen et al. (2019)	3	3	3	3	3	1	2	2	3	3	26

1=little or no justification or explanation for a particular area
2=moderate justification or explanation but not fully elaborated
3=strong justification or explanation and explained relevant issue at hand

Appendix E. *Table of quotes illustrating analytical and descriptive themes*

Analytical and descriptive themes	*Participants' quotations* and/or authors' explanations
Sources of support	
Family	*"So you know, right now I owe everything to my parents, you know, bringing me up to be confident"* *"… throughout all of it my parents were supportive. They certainly never talked down or you know, made me feel stupid, not at all."* Support and encouragement from parents and siblings…were credited for nurturing positive adjustment. Individuals said that their family background was an overwhelmingly positive experience where having CLP didn't single them out for differential treatment. As a result, the family context emerged as a 'needs supportive' environment, one that nurtured the whole child, rather than dwelling on their CLP.
Friends	*"Friends have made a difference for me...they have helped me meet more people."* *"I'm lucky because I have a good group of friends that are there for me all the time even if I have one of those bad moments."* *"I have such good friends and I think friends play a big huge role in growing up and finding who you are and feeling comfortable with yourself."* All participants spoke about the support they received from friends. They described an underlying sense that their friends cared about them and would support them through emergent difficulties of stigma and adjustment.
Romantic relationships	*"She (girlfriend) really supported me and helped me to focus on my positive qualities"* Participants reported that having a relationship had contributed to them developing more self-confidence and social acceptance. Having encouraging supportive ….friends, and influential adults were factors that adolescents described as contributing to positive adaptation. Participants who received recognition from significant others, such as partners, felt that they were just as valuable as everyone else. This made them focus less on their condition, thus increasing their self-esteem.
Psychological services	*"Seeing the psychologist as part of the cleft team has made a massive, massive impact and it's completely turned round the way I see myself and where I go from here".* Several participants discussed using religion and therapy as additional coping methods. Others believed that receiving professional psychological support had been very beneficial.

Analytical and descriptive themes	***Participants’ quotations*** **and/or authors’ explanations**
Dispositional Style	
Optimism	*“I’m happy. I know I still have a road ahead of me, but I’m happy about it. I’m happy that everyday I’m closer to becoming the person I want to be and learning more about myself and my condition and helping other people.”* *“You have to find the positives . . . and have a sense of humour about it I suppose . . . I could get bogged down in thinking “oh poor me,” but where would that get me?”* In general, participants who identified as having adjusted positively to their cleft stated having a positive look upon life and a certain resilience against the challenges associated with CLP.
Openness	Openness was described as an important way of dealing with the cleft experience overall and crucial for dealing with other people’s reactions. Another strategy was being open about the condition and prepared to talk to others which served to increase relatedness.
Reappraisal	*“When someone makes a nasty remark, it tells me more about that person than anything else”* Adolescents who described less concern over others’ perspectives and opinions of them appeared to have engaged in a substantial extent of personal cognitive ‘‘inner work’’ in grappling with negative images regarding differences and their constructive personal meanings. Participants often stated that remarks about appearances were made by people who were not happy themselves. Many participants mentioned various coping skills that helped with stressors and bullying. Participants mentioned skills such as restructuring negative thoughts or mindfulness.
Resilience	*“As I look back, I can say that it’s been really good for me to have had this experience, because it has made me strong and has made me who I am today.”* “*.....But that kind of spurred me on to, basically be the person that I am today, you know, I am very determined, and if someone says, ‘You can’t do this because of this, that and the other,’ I try and set out to prove that I can. So it’s made me the person I am.”* *‘‘A lot of people, I think, wonder if people who look like me are confident. I guess sometimes I may not be as confident walking into a social situation where beauty and appearance seems to be one of the most important points of the gathering. But what is much more important is a far deeper sense of self-confidence I have. I feel confident that I have the strength to get me through whatever life brings my way—those real and inescapable challenges of life that include disappointments, loss, fear, etc. Those I do feel very capable of confronting and I attribute that, at least partly, to all of the experiences I had living with a facial difference.’’*

Analytical and descriptive themes	***Participants’ quotations* and/or authors’ explanations**
Difference in self and others	
Perceptions of difference	*“…I wasn’t made to feel any different than anyone else. So that has kind of been one of my values, you know, to get me through school and everything else, that you know, I don’t, it doesn’t make me a different person; it’s just I look different.”* *“I like to stand out slightly I think, it’s nice to have some individuality and be unique”* *“To be honest I don't see myself as being any different from anyone else”.* Some did not perceive themselves to be different.
Family appraisal of difference	*“… my parents were very secure in the fact that I was no different to my siblings or you know, I was still a, a person inside, and that was more important to them than the way that I looked. So, I think because they were comfortable with it, I was comfortable with it.”* *“I’ve always felt special, because my mother, she has always told me that the fact that you’re born with a cleft makes you who you are. … and I’ve felt special somehow, instead of feeling weird or strange.”*
Difference in others	*“ I truly have a diverse set of people (in my life) and I value the richness of this diversity greatly. I have friends who are disabled, from diverse cultural backgrounds, one who is a burn survivor, gay and lesbian friends. . . . And I think that my own experience of difference helped me open to people I most likely would not have been open to. . . . I feel very good about that part of who I am.”* Becoming a more tolerant person *“It’s taught me to accept other people … you don’t judge them by how they look”.* Participants spoke about experiences resulting in an increased understanding of others with disabilities or differences.
Positive self-concept	
Identity; ‘part of me’	Many participants were accepting of their cleft and viewed it as being a part of who they are. *‘‘While I would give up the hard part in a second, I have a feeling that I would give up a central part of who I am that makes me the person I am if I sacrificed the experience of having a facial difference. I just could not do this in good conscience.”* *‘‘I imagine that if I hadn’t been born with a facial difference, my life would very likely have been easier. But honestly, it may have been more mundane. I doubt that I would have the passions I do to do good in the world or the*

Analytical and descriptive themes	***Participants' quotations*** **and/or authors' explanations**
	same lust I have for experiencing all the things life has to offer, and I know I wouldn't have the same sense of purpose and meaning to my life…" *"I would have viewed the chance to get rid of my cleft like 'shedding a skin.' Today I think it would feel more like losing a limb since it's been such an integral part of my life. I don't think I even know how much it works for me.''*
Capabilities and potential for success	
	"I played quite a lot of sport when I was younger . . . being part of a team really helped because everyone is equal, and I think you get quite a lot of respect… I got involved in a wide variety of activities and focused on what I was good at . . . I think that stood me in good stead". After being involved in such a special interest, they felt less focused on their facial appearance during the performance of the activity and they felt an increased wellbeing. "*For me to have high standards is to make a difference, to go further in life, do well in life, and that is just what I want to do. In some cases in this world, it is pretty much based on looks, and I personally believe that with Treacher Collins syndrome that I can make a difference but showing that you do not have to have good looks to do well in this world."* Participants also believed that focusing on their strengths, had contributed to their adjustment.
Transitional stages	Most participants identified the ways in which their self-concept changed over time, feeling better about themselves as they got older. *"College for me was a great experience because I think when I got to college I was very self-conscious and not very confident at all, and then the course was very confidence boosting"* *"I used to value appearance a great deal, but as one grows older, over 30, one starts to look at it differently and it becomes less of an issue. When I finished high school and went to higher education it suddenly seemed all OK. People acted normal, nobody teased. It was a turning point for me."* Social confidence was often lowest at primary school or high school, due to teasing, and gradually increased as participants got older. Factors such as having an enjoyable education, a satisfying job, or a romantic relationship often helped improving their overall and social confidence.
Surgical intervention	
Decision making	"… yeah don't kind of just discuss things with their parents: make sure they're involved in those conversations as well. I was always there for the conversations and kind of knew everything as they knew it, which was good. But if I hadn't known those things I'd be a lot more uncertain about it."

Analytical and descriptive themes	*Participants' quotations* and/or authors' explanations
	Some younger individuals born with CLP reported feeling involved in decision-making prior to surgical intervention. These individuals reiterated the value of talking to both parents and the individual concerned to ensure the rationale and expected outcomes of treatment are well understood by all parties.
Realistic expectations	Participants generally believed that this surgery would provide them with a desired change to feel more ''normal.'' Perhaps as another coping strategy, they were hopeful for small changes that would be enough to make a difference, but not so much that it would change the essence of who they were. During these facial and identity changes from major surgical interventions, the participants in the study thought that it was crucial to receive appropriate psychological support from both their families and friends, as well as from professionals Expectations of treatment; participants discussed the importance of psychological input in relation to managing patients' expectations of surgical outcomes.

Appendix F. *Approval from the Doctorate in Clinical Psychology Research Committee*

D.Clin.Psychology Programme
Division of Clinical Psychology
Whelan Building, Quadrangle
Brownlow Hill
LIVERPOOL
L69 3GB

Tel: 0151 794 5530/5534/5877
Fax: 0151 794 5537
www.liv.ac.uk/dclinpsychol

Meghan Owens
Clinical Psychology Trainee
Doctorate of Clinical Psychology Doctorate Programme
University of Liverpool
L69 3GB

6 December 2018

RE: An exploration of child and parent experiences of childhood midfacial surgery and adjustment to life following surgery

Trainee: Meghan Owens Supervisors: Luna Centifanti & Anna Kearney

Dear Meghan,

Thank you for your response to the reviewers' comments of your research proposal submitted to the D.Clin.Psychol. Research Review Committee.

I can now confirm that your amended proposal (version number 4.1, dated 28/11/18) meets the requirements of the committee and has been approved by the Committee Chair.

Please take this Chairs Action decision as ***final*** approval from the committee.

You may now progress to the next stages of your research.

I wish you well with your research project.

Dr Catrin Eames
Vice-Chair D.Clin.Psychol. Research Review Committee.

A member of the

Russell Group

Dr Laura Golding	Dr Gundi Kiemle	Dr Jim Williams	Dr Beth Greenhill	Dr Ross White	Mrs Sue Knight
Programme Director	Academic Director	Joint Clinical Director	Joint Clinical Director	Research Director	Programme Co-ordinator
l.golding@liv.ac.uk	gkiemle@liv.ac.uk	j.r.williams@liv.ac.uk	bethg@liv.ac.uk	rgwhite@liv.ac.uk	sknight@liv.ac.uk

Appendix G. *University Sponsorship Letter of Approval*

Miss Lara Lavelle-Langham
Research Integrity and Governance Manager

University of Liverpool
Research Support Office
2nd Floor Block D Waterhouse Building
3 Brownlow Street
Liverpool
L69 3GL

Tel: 0151 794 8373
Email: sponsor@liverpool.ac.uk

18/04/2019

RE: 4723 - An exploration of child and parent experiences of childhood midfacial surgery and adjustment to life following surgery

Dear Dr Weatherhead,

After consideration at the JRO Non-Interventional Sponsorship Sub Committee on 15th March 2019.

I am pleased to confirm that the University of Liverpool is prepared to act as Sponsor under the UK Policy Framework for Health and Social Care Research (v3.2 10th October 2017) for the above study.

The following documents have been received by the Research Support Office:

Document Type	File Name	Date	Version
Evidence Of Peer Review	Owens%2c Meghan_Approval_06.12.18	06/12/2018	1
Chief Investigator CV	Ste CV	01/03/2019	1
Project Protocol/Clinical Investigation Plan	Final research protocol 4.1	08/03/2019	4.1

Please note this letter does NOT allow you to commence recruitment to your study.

A notification of Sponsor Permission to Proceed will be issued when governance and regulatory requirements have been met. Please see Appendix 1 to this letter for a list of the documents required.

If you have not already applied for regulatory approvals through IRAS you may now do so at https://www.myresearchproject.org.uk/Home.aspx (see SOP013).

In order to meet the requirements of the UK Policy Framework for Health and Social Care Research (v3.2 10th October 2017), the University requires you to agree to the following Chief Investigator responsibilities. Please see SOP006 for further details of delegated responsibilities;

- Comply with the UK Policy Framework for Health and Social Care Research (v3.2 10th October 2017) and all relevant legislation, including but not limited to the Data Protection Act 1998 and subsequently the EU General Data Protection Regulation (2016), the Mental Capacity Act 2005 and the Human Tissue Act 2004;
- Inform the Research Support Office as soon as possible of any adverse events especially SUSARs and SAE's, Serious Breaches to protocol or relevant legislation or any concerns regarding research conduct;
- Approval must be gained from the Research Support Office for any amendments to, or changes of status in the study prior to submission to REC and any other regulatory authorities (as per SOP018);
- It is a requirement that Annual Progress Reports are sent to the NHS Research Ethics Committee (REC) annually following the date of Favourable Ethical Approval. You must provide copies of any reports submitted to REC and other regulatory authorities to the Research Support Office;
- Maintain the study/trial master file (as per SOP005);
- Make available for review any study documentation when requested by the sponsors and regulatory authorities;
- Upon the completion of the study it is a requirement to submit an End of Study Declaration (within 90 days of the end of the study) and End of Study Report to REC (within 12 months of the end of the study). You must provide copies of this to the Research Support Office;
- Ensure you and your study team are up to date with the current RSO SOPs throughout the duration of the study.

The University also requires you to comply with the following:

University professional indemnity and clinical trials insurances will apply to the study as appropriate. This is on the assumption that no part of the clinical trial will take place outside of the UK. If you wish to conduct any part of the study in a site outside the UK or you wish to subcontract any part of the study to a third party specific approvals and consideration of appropriate indemnity would be required;

If you have any queries regarding the sponsorship of the study, please do not hesitate to contact the Clinical Research Governance Team on 0151 794 8373 (email sponsor@liverpool.ac.uk).

Yours sincerely,

Miss Lara Lavelle-Langham

Research Integrity and Governance Manager

Research Support Office

Chief Investigator Sponsor Declaration

I, the Chief Investigator, agree to the terms and conditions of the University of Liverpool Sponsorship approval for **Experiences of childhood midfacial surgery and adjustment to life following surgery**, **4723** and I am aware of my responsibilities under the UK Policy Framework for Health and Social Care Research. I also agree to provide the Research Support Office with the documents listed overleaf when available.

By signing this agreement you also declare you have read and understood all applicable Sponsor SOPs, including but not limited to those referenced in this letter.

All Sponsor Policies, SOPs and related templates policies are available via the Research Support Office internal webpage. By signing this you also agree that you have made these available to all

Please return a scanned, signed copy of this declaration to the Research Support Office (sponsor@liverpool.ac.uk) within 30 days of the date of this letter.

The original version should be retained for inclusion in the Study Master File.

Failure to return this declaration may result in Sponsorship being withdrawn.

Appendix H. *Health Research Authority Approval*

Ymchwil Iechyd
a Gofal Cymru
Health and Care
Research Wales

NHS
Health Research
Authority

Dr Stephen Weatherhead

University of Liverpool, Whelan Building

Email: hra.approval@nhs.net

HCRW.approvals@wales.nhs.uk

Brownlow hill

Liverpool

L69 3GB

29 October 2019

Dear Dr Weatherhead

HRA and Health and Care

Study title:	**An exploration of child and parent experiences of childhood midface surgery and adjustment to life following surgery**
IRAS project ID:	**261595**
Protocol number:	**4723**
REC reference:	**19/NW/0396**
Sponsor	**University of Liverpool**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **261595**. Please quote this on all correspondence.

Yours sincerely,

Helen Penistone

Approvals Specialist

Email: hra.approval@nhs.net

Telephone: 0207 104 8010

Copy to: *Mr Alex Astor*

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

Document	*Version*	*Date*
Copies of advertisement materials for research participants	1	04 June 2019
Costing template (commercial projects) [Research budget]		30 May 2017
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only)		31 July 2019
HRA Schedule of Events	1	04 June 2019
Interview schedules or topic guides for participants	1	30 May 2019
IRAS Application Form [IRAS_Form_27082019]		27 August 2019
Letter from sponsor [Sponsorship]	1	18 April 2019
Other [Organisational Information document]	1	06 June 2019
Other [Response to Assessment Queries Only]		
Other [Response to assessment queries]		
Participant consent form [Adult Consent Form]	2	04 July 2019
Participant consent form [Consent form parents]	3	20 May 2019
Participant information sheet (PIS) [Children's information sheet and assent form]	2	20 May 2019
Participant information sheet (PIS) [Adult]	4	10 October 2019
Participant information sheet (PIS) [Young Persons]	4	10 October 2019
Research protocol or project proposal [Research proposal]	2	10 February 2019
Summary CV for Chief Investigator (CI) [Ste Weatherhead CV document one]		
Summary CV for student [Summary Research CV]	1	05 February 2019
Summary CV for supervisor (student research) [CV supervisor]		08 May 2019

IRAS project ID	261595

Information to support study set up

The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	Expectations related to confirmation of capacity and capability	Agreement to be used	Funding arrangements	Oversight expectations	HR Good Practice Resource Pack expectations
There will be one study site type.	Research activities should not commence at participating NHS organisations in England or Wales prior to their formal confirmation of capacity and capability to deliver the study.	An Organisation Information Document has been submitted and the sponsor is not requesting and does not expect any other site agreement to be used.	No external funding has been sought. No funding will be available to site.	There will be a local collaborator at site to facilitate access of the researcher to carry out interviews.	Where arrangements are not already in place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtain a Letter of Access based on enhanced DBS checks (no barred list check) and occupational health clearance.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.
The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.

Partner Organisations: Health Research Authority, England NIHR Clinical Research Network, England NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales HSC Research & Development, Public Health Agency, Northern Ireland

Notification of non-substantial / minor amendments; version 1.0; November 2014 **Page 1 of 4**

Notification of Non-Substantial/Minor Amendments(s) for NHS Studies

This template **must only** be used to notify NHS/HSC R&D office(s) of amendments, which are **NOT** categorised as Substantial Amendments.

If you need to notify a Substantial Amendment to your study then you MUST use the appropriate Substantial Amendment form in IRAS.

Instructions for using this template

- For guidance on amendments refer to http://www.hra.nhs.uk/research-community/during-your-research-project/amendments/
- This template should be completed by the CI and optionally authorised by Sponsor, if required by sponsor guidelines.
- This form should be submitted according to the instructions provided for NHS/HSC R&D at http://www.hra.nhs.uk/research-community/during-your-research-project/amendments/which-review-bodies-need-to-approve-or-be-notified-of-which-types-of-amendments/ . If you do not submit your notification in accordance with these instructions then processing of your submission may be significantly delayed.

1. Study Information

Full title of study:	An exploration of child and parent experiences of childhood midface surgery and adjustment to life following surgery
IRAS Project ID:	261595
Sponsor Amendment Notification number:	Amendment 2
Sponsor Amendment Notification date:	23rd April 20
Details of Chief Investigator:	
Name [first name and surname]	Ste Weatherhead
Address:	University of Liverpool Whelan Building Brownlow Hill Liverpool
Postcode:	L69 3GB
Contact telephone number:	0151 794 5025
Email address:	Stephen.weatherhead@liverpool.ac.uk

Partner Organisations: Health Research Authority, England NIHR Clinical Research Network, England NHS Research Scotland NISCHR Permissions Co-ordinating Unit, Wales HSC Research & Development, Public Health Agency, Northern Ireland

Notification of non-substantial / minor amendments; version 1.0; November 2014 **Page 2 of 4**

Details of Lead Sponsor:	
Name:	Alex Astor
Contact email address:	sponsor@liverpool.ac.uk
Details of Lead Nation:	
Name of lead nation *delete as appropriate*	England
If England led is the study going through CSP? *delete as appropriate*	No
Name of lead R&D office:	University of Liverpool

Partner Organisations:

Health Research Authority, England	NIHR Clinical Research Network, England
NHS Research Scotland	NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland	

2. Summary of amendment(s)

This template **must only** be used to notify NHS/HSC R&D office(s) of amendments, which are **NOT** categorised as Substantial Amendments.
If you need to notify a Substantial Amendment to your study then you MUST use the appropriate Substantial Amendment form in IRAS.

No.	**Brief description of amendment** ***(please enter each separate amendment in a new row)***	**Amendment applies to** ***(delete/ list as appropriate)***		**List relevant supporting document(s), including version numbers** ***(please ensure all referenced supporting documents are submitted with this form)***		**R&D category of amendment** ***(category A, B, C)*** ***For office use only***
		Nation	**Sites**	**Document**	**Version**	
1	In light of the current COVID-19 situation, following advice from local NHS trust, two remaining interviews will need to be carried out over video calls. Two families had agreed to take part in the study prior to the COVID-19 situation. No one more families will be recruited. Interviews will be carried over secure platforms; Zoom or Microsoft teams. They will be recorded using the secure Dictaphone, as previously approved. Consent forms and participant information sheets will password protected and emailed to parents from a NHS email address. Consent forms will be printed out, deleted from email and stored in line with previous documents.	England	Alder Hey Children's Hospital	Adult participant information sheet Young person's information sheet Research protocol	4.1 4.1 4.3	C
2						
3						
4						
5						

[Add further rows as required]

Partner Organisations:

Health Research Authority, England	NIHR Clinical Research Network, England
NHS Research Scotland	NISCHR Permissions Co-ordinating Unit, Wales
HSC Research & Development, Public Health Agency, Northern Ireland	

3. Declaration(s)

Declaration by Chief Investigator

- I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.
- I consider that it would be reasonable for the proposed amendment(s) to be implemented.

Signature of Chief Investigator: ....

Print name: ...Dr Stephen Weatherhead..

Date: ...18.5.20..

Optional Declaration by the Sponsor's Representative (as per Sponsor Guidelines)

The sponsor of an approved study is responsible for all amendments made during its conduct.

The person authorising the declaration should be authorised to do so. There is no requirement for a particular level of seniority; the sponsor's rules on delegated authority should be adhered to.

- I confirm the sponsor's support for the amendment(s) in this notification.

Signature of sponsor's representative: .... A. Ast ....

Print name:.............Alex Astor...............................

Post: ......Research Support Office.......................................

Organisation:......University of Liverpool.......................................

Date:......12th June 2020.......................................

Appendix I. *Parent information sheet*

Version 3 Date 20th May 2019

Title of study: An exploration of child and parent experiences of childhood midface distraction surgery and adjustment to life following surgery

You are being invited to participate in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and feel free to ask us if you would like more information or if there is anything that you do not understand. Please also feel free to discuss this with your friends, relatives and GP if you wish. We would like to stress that you do not have to accept this invitation and should only agree to take part if you want to.

Thank you for reading this.

1. What is the purpose of the study?

The study aims to explore the experiences of midface distraction surgery for the treatment of syndromic craniosynostosis in childhood from the perspective of the child and their parents/ guardians. This will include their experiences of adjusting to life following surgery; exploring factors which influence perceived success and difficulties.

We hope to gain a better understanding of how surgery has impacted on your families' life and will use this knowledge to inform other families considering this journey and help guide what the craniofacial service offer.

Research has tended to focus on parent's experiences and children's views have often been overlooked. We feel it is very important to include children in this study and their views are very valuable to us.

2. Why have I been chosen to take part?

You have been invited to take part in this research as you been identified as a parent/ guardian of a child between the ages of six and twelve years old who has undergone midface distraction surgery. We are seeking participants from two nationally designated craniofacial centres; Alder Hey Children's Hospital and Great Ormond Street Hospital. We hope speaking to children and parents/ guardians will give us in-depth overview of the journey through and after surgery.

3. Do I have to take part?

No. Participation is voluntary and you are free to withdraw your participation at any time, without explanation, and without incurring any disadvantages.

4. What will happen if I take part?

If you would like to take part in the study a member of your child's care team will ask your permission to pass on your details to the research team. They will also ask for your consent for the research team to access your child's medical records. This will only be used to help the researcher understand what diagnosis and treatment your child has received and if any adjustments are needed for the study.

You and your child will then be asked to take part in an interview with a trainee Clinical Psychologist. They will ask you both some questions about the surgery your child has had and about life following the surgery. To ensure the environment is as child-friendly as possible, children will be provided with paper, colouring pens and toys.

The interviews will be audio-recorded but when transcribed all identifiable information will be anonymised. Only people involved in the research and transcribing process will have access to the recordings.

The whole process should take no longer than 60 minutes.

5. How will my data be used?

The University processes personal data as part of its research and teaching activities in accordance with the lawful basis of 'public task', and in accordance with the University's purpose of "advancing education, learning and research for the public benefit". Under UK data protection legislation, the University acts as the Data Controller for personal data collected as part of the University's research. The Principal Investigator acts as the Data Processor for this study, and any queries relating to the handling of your personal data can be sent to Dr. Stephen Weatherhead ste@liverpool.ac.uk

Further information on how your data will be used can be found in the table below.

How will my data be collected?	During an interview
How will my data be stored?	On password-protected computers/ locked cabinets within the hospital or university.
How long will my data be stored for?	10 years

What measures are in place to protect the security and confidentiality of my data?	Computers will be password-protected. Information will also be password-protected.
Will my data be anonymised?	Yes.
How will my data be used?	Student research project, journal article, and conference presentations.
Who will have access to my data?	The investigators who part of the research team and transcribers.
Will my data be archived for use in other research projects in the future?	No
How will my data be destroyed?	All data will be deleted from computers after 10 years or paper copies destroyed in confidential waste.

6. Expenses and / or payments

There are no payments for taking part in this study, however, each family who completes the interview will receive a £20 Amazon voucher, as reimbursement for taking part in the study.

Travel costs up to the value of £30 will be covered for each family.

7. Are there any risks in taking part?

There are no anticipated risks to you if you take part. If you experience any discomfort or disadvantage as part of the research, please let the researches know immediately. You will not have to answer any questions that you do not feel comfortable with.

8. Are there any benefits in taking part?

There are no direct benefits from taking part, apart from the opportunity to contribute to psychology research that may guide future interventions and clinical practice. We then hope we can help other children and families.

9. What will happen to the results of the study?

All the information collected about you and your child during the course of the research will be kept strictly confidential. Your data will be stored in an anonymized format (identified by a random number only). We plan to publish the results from this study in a scientific journal and in an information leaflets for families considering this surgery. It is important that you understand that your results will be completely anonymous, so there is no way that you can be identified.

10. What will happen if I want to stop taking part?

Your participation is voluntary and you are free to stop taking part and can withdraw from the study at any time without giving any reason and without your rights being affected. Just let a member of the research know you wish to withdraw. Results may only be withdrawn prior to anonymization.

11. What if I am unhappy or if there is a problem?

If you are unhappy, or if there is a problem, please feel free to let us know by contacting Meghan Owens on 0151 794 5877 or Stephen Weatherhead on 0151 794 5025 and we will try to help. If you remain unhappy or have a complaint which you feel you cannot come to us with then you should contact the Research Ethics and Integrity Office at ethics@liv.ac.uk. When contacting the Research Ethics and Integrity Office, please provide details of the name or description of the study (so that it can be identified), the researcher(s) involved, and the details of the complaint you wish to make.

The University strives to maintain the highest standards of rigour in the processing of your data. However, if you have any concerns about the way in which the University processes your personal data, it is important that you are aware of your right to lodge a complaint with the Information Commissioner's Office by calling 0303 123 1113.

12. Who can I contact if I have further questions?

Meghan Owens, University of Liverpool, Clinical Psychology programme

Whelan Building, Brownlow Hill, Liverpool, L69 3GB.

Email: m.owens@liv.ac.uk Tel: 0151 794 5530

Thank you for reading.

Appendix J. *Parent consent form*

Participant consent form

Title of the research project: An exploration of child and parent experiences of childhood midface surgery and adjustment to life following surgery

Name of researcher(s): Meghan Owens, Dr Stephen Weatherhead, Dr Anna Kearney and Dr Luna Centifanti

Please initial box

1. I confirm that I have read and have understood the information sheet dated [14/01/19] for the above study, or it has been read to me. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that taking part in the study involves participating in an interview with a researcher. ☐
3. I understand and agree that my participation will be audio recorded and I am aware of and consent to your use of these recordings for the following purposes: transcription and analysis. ☐
4. I understand that my participation is voluntary and that I am free to stop taking part and can withdraw from the study at any time without giving any reason and without my rights being affected. In addition, I understand that I am free to decline to answer any particular question or questions. ☐
5. I understand that I can ask for access to the information I provide and I can request the destruction of that information if I wish at any time prior to publication. I understand that following publication I will no longer be able to request access to or withdrawal of the information I provide. ☐
6. I agree that my anonymised information can be quoted in the publication of this research. ☐
7. I understand that the information I provide will be held securely and in line with data protection requirements at the University of Liverpool until it is fully anonymised and then deposited in the archive for sharing and use by other authorised researchers to support other research in the future. ☐

8. I understand that my responses will be kept strictly confidential. I give permission for members of the research team to have access to my fully anonymised responses. I ☐

understand that my name will not be linked with the research materials, and I will not be identified or identifiable in the report or reports that result from the research.

9. I understand that confidentiality and anonymity will be maintained and it will not be possible to identify me in any publications ☐

10. I understand that signed consent forms, audio recording and transcripts of the interview will be retained in a locked cabinet within the psychological services departments of the hospital sites or stored in a locked cabinet within the clinical psychology department at the University of Liverpool. At the end of the study, the data custodian (primary supervisor- Dr Stephen Weatherhead) will store any hard copies securely in locked cabinets at the University of Liverpool and will be responsible for the data for ten years after which it will be destroyed. ☐

11. For children only, I agree that my children's current and historical medical records may be accessed for the purpose of this study by members of the NHS Trust or regulatory authorities. ☐

12. I agree to take part in the above study. ☐

_______________________ __________ ____________________

Participant name Date Signature

_______________________ __________ ____________________

Name of person taking consent Date Signature

Principal Investigator	**Student Investigator**
Dr Stephen Weatherhead	Meghan Owens
University of Liverpool	University of Liverpool
Department of Clinical Psychology	Department of Clinical Psychology
Whelan Building	Whelan Building
Brownlow Hill	Brownlow Hill
Liverpool	Liverpool
L69 3GB	L69 3GB
Tel: 0151 051 7945025	0151 794 5877
ste@liverpool.ac.uk	m.owens@liv.ac.uk

Appendix K. *Parental consent form*

Parental consent form

Researchers' names: Meghan Owens, Dr Stephen Weatherhead, Dr Anna Kearney and Dr Luna Centifanti

Research institution: University of Liverpool

To be completed by the PARENT/GUARDIAN

Please circle the relevant answer

- Have you been fully informed about the research study? **YES/ NO**
- Are you aware that your child's medical records will be accessed? **YES/ NO**
- Do you know that your child can withdraw from this study at any time, with no need to give a reason and without any negative consequences? **YES/ NO**
- Do you know that your child's answers are confidential?

(The only exception for this would be if during the research it becomes clear that a child is at risk from someone or a risk to themselves then this would be reported to appropriate services. Where possible this would be discussed with you first.]. **YES/ NO**

- Do you agree that your child can take part in this study, whose results could be published (e.g., in a report or book)? Neither you nor your child will be identified in any way in these publications. **YES/ NO**

If **YES to all**, please fill in the details below.

Child's name: ____

Child's date of birth: _________________________________

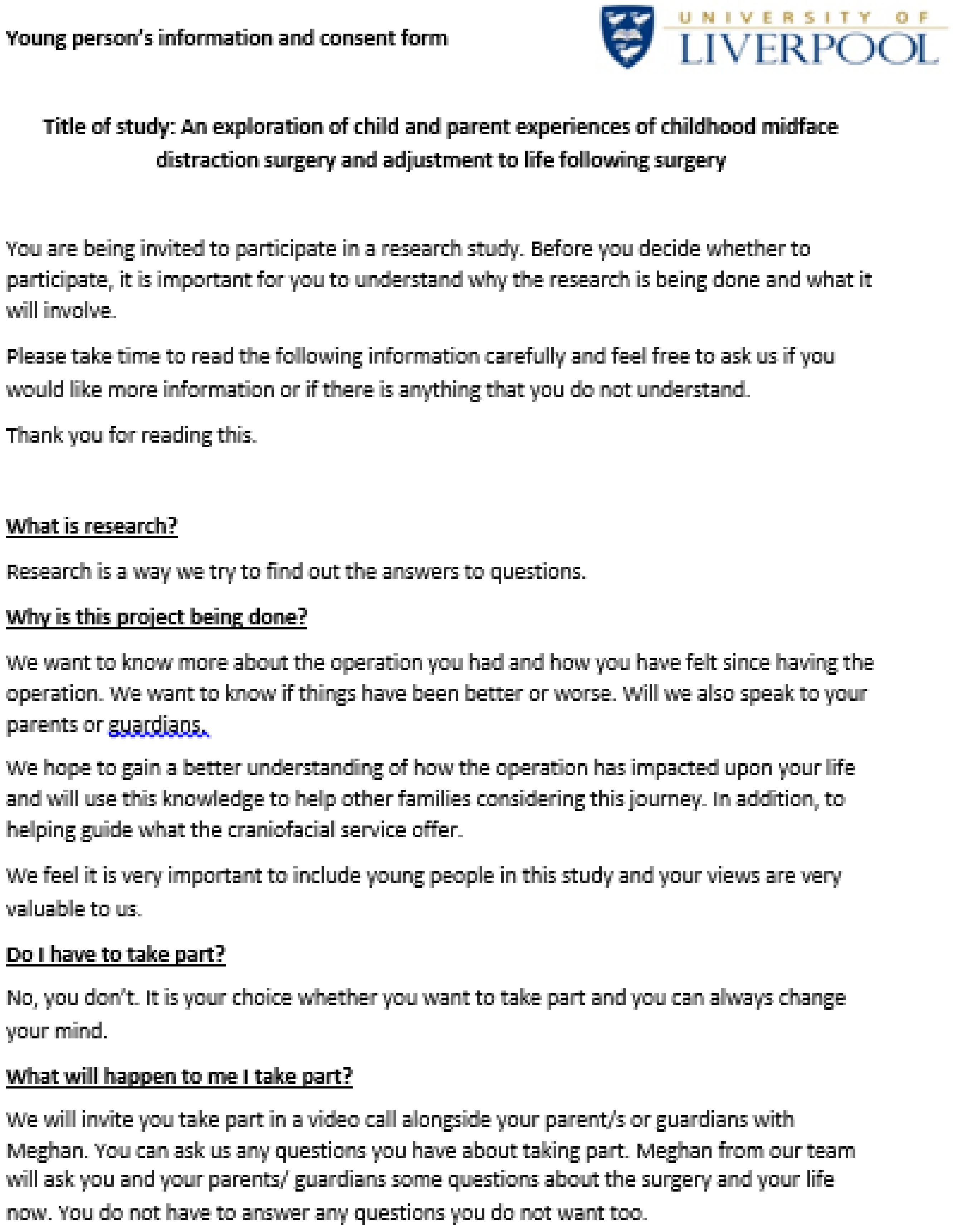
Signed by parent/guardian: ____________________________________

Date: ____

Name in block letters: ____

For any questions about the study, please contact Meghan Owens at m.owens@liv.ac.uk

Appendix L. *Young persons’ information sheet and consent form*



Young person’s information and consent form

UNIVERSITY OF LIVERPOOL

Title of study: An exploration of child and parent experiences of childhood midface distraction surgery and adjustment to life following surgery

You are being invited to participate in a research study. Before you decide whether to participate, it is important for you to understand why the research is being done and what it will involve.

Please take time to read the following information carefully and feel free to ask us if you would like more information or if there is anything that you do not understand.

Thank you for reading this.

What is research?

Research is a way we try to find out the answers to questions.

Why is this project being done?

We want to know more about the operation you had and how you have felt since having the operation. We want to know if things have been better or worse. Will we also speak to your parents or guardians.

We hope to gain a better understanding of how the operation has impacted upon your life and will use this knowledge to help other families considering this journey. In addition, to helping guide what the craniofacial service offer.

We feel it is very important to include young people in this study and your views are very valuable to us.

Do I have to take part?

No, you don’t. It is your choice whether you want to take part and you can always change your mind.

What will happen to me I take part?

We will invite you take part in a video call alongside your parent/s or guardians with Meghan. You can ask us any questions you have about taking part. Meghan from our team will ask you and your parents/ guardians some questions about the surgery and your life now. You do not have to answer any questions you do not want too.

You will be asked to sign your name on a consent sheet sent to your parent/s or guardians via email.

Our conversation will be audio-recorded, so nothing is missed but your name will be removed so all your information is kept anonymous.

The whole process should take no longer than 60 minutes.

Will anything about the research upset me?

We will not ask you to talk about anything you do not want to and someone will be there to help you if needed.

Will taking part help me?

It might not help you right now but it will help us to know more about how you found life after the operation. We then hope we can help other children and young people. The study will also be used to guide the services the craniofacial team offer.

Will my information be kept private?

Your information will be kept private. We will only speak with your team at the hospital.

Your name won't be used, we will give all the young people a number instead.

How will my information be used?

The University of Liverpool will protect your personal data and act as something called a Data Controller, this means they will keep your information safe.

A member of the research team Ste Weather will act as something called a Data Processor, who will too help keep your information safe. If you are concerned about how your personal data is used you can ask to speak to Ste.

Further information on how your data will be used can be found in the table below.

How will my data be protected by law?	All personal data will be collected and stored in line with the General Data Protection Regulation (GDPR).
How will my data be collected?	During an interview

How will my data be stored?	On password-protected computers/ locked cabinets within the hospital or university.
How long will my data be stored for?	10 years
What measures are in place to protect the security and confidentiality of my data?	Computers will be password-protected. Information will also be password-protected.
Will my data be anonymised?	Yes.
How will my data be used?	Student research project, journal article, and conference presentations.
Who will have access to my data?	The investigators who part of the research team and transcribers.
Will my data be archived for use in other research projects in the future?	No
How will my data be destroyed?	All data will be deleted from computers after 10 years or paper copies destroyed in confidential waste.
What if I am concerned about how my data is handled?	You can speak to the research team at the University of Liverpool or you have the right to lodge a complaint with the Information Commissioners Office

Can I change my mind about taking part or answering?

Yes. Just tell your parents or the researchers that you don't want to take part anymore. You don't have to give any reason, but we will keep information about you that we already have. We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

What happens afterwards?

All the information collected about you and your parents/ guardian will be kept strictly confidential.
We plan to publish the results from this study in a scientific journal and in information leaflets for families considering this surgery.
It is important that you understand that your results will be completely anonymous, so there is no way that you can be identified.

Expenses / payments

There are no payments for taking part in this study, however, each family who completes the interview will receive a £15 Amazon voucher, as reimbursement for taking part in the study.

Travel costs up to the value of £30 will be covered for each family.

Can I get more information or ask a question?

Of course. You can phone Meghan or ask your parent/s or guardians to phone Meghan. You can also ask Meghan when you meet her or ask the doctor or psychologist at the hospital.

Thank you for reading.

Young person's consent

I agree that I have read the information above and understand what will happen if I take part. ☐

I agree I have been able to ask any questions I have and these have been answered ☐

I agree that members of the research team can access my medical notes ☐

I understand that our conversation will be audio-recorded and only used for research purposes ☐

I understand that I do not have to take part in the study if I do not wish to and can stop taking part at any point. ☐

I understand all my information will be kept confidential and stored safely ☐

I agree to take part in the above study. ☐

My name ____

Today's date ____

A bit for the researcher / your parent or guardian/ health professional to fill in:

I have witnessed ________________________ give their consent to participate in this study.

Name and occupation (researcher/parent or guardian/ health professional) of witness:

____

Signature: _________________________Date: ____________________

Appendix M. *Child information sheet and consent form*

Children's information sheet

What is research? Research is a way we try to find out the answers to questions.

Why is this project being done?

We want to know more about the operation you had and how you have felt since having the operation. We want to know if things have been better or worse. Will we also speak to your parents or guardians. We can then help other children who are thinking about having the same operation.

Do I have to take part?

No, you don't. It is your choice whether you want to take part and you can always change your mind.

What will happen to me I take part?

The research team will look at your medical notes to help them understand more about your condition and see what operation or other treatments you might have had.

We will invite you to come to the hospital with your parent/s or guardians. You can ask us any questions you have. One person from our team will ask you and your parents/ guardians some questions about the surgery and your life now. We can draw or play games while we talk and you can take some breaks.

We will voice record our chat so that we do not forget anything important.

Will anything about the research upset me?

We will not ask you to talk about anything you do not want to and someone will be there to help you if needed.

Will taking part help me?

It might not help you right now but it will help us to know more about how you found life after the operation. We then hope we can help other children and families.

What happens afterwards?

The research will be talked about and written down to help other children.

Will my information be kept private?

Your information will be kept private. We will only speak with your team at the hospital.

Your name won't be used, we will give all the children a number instead. It's top secret!

Can I change my mind about taking part or answering?

Yes. Just tell your parents or the researchers that you don't want to take part anymore. You don't have to give any reason.

Can I get more information or ask a question?

Of course. You can phone Meghan or ask your parent/s or guardians to phone Meghan. You can also ask Meghan when you meet her or ask the doctor or psychologist at the hospital.

Children's assent

I agree to take part in an interview for the research project

My name ______________________________

Today's date ______________________________

A bit for the researcher / your parent or guardian/ health professional to fill in:

I have witnessed ____________________ give their verbal assent to participate in this study.

Name and occupation (researcher/parent or guardian/ health professional) of witness:

____

Signature:____________________Date: ____________________

Appendix N. *Semi-structured interview topic guide*

Interview topic guide

An exploration of child and parent experiences of childhood midface distraction surgery and adjustment to life following surgery

Topics/questions: Notes for the Interviewer it is not necessary to stick to the exact line of questioning or order. The purpose of these prompts is to be open and explorative. It is important, however, that the subject matters in the topic guides are explored and the style of questioning kept open and explorative. Not leading.

Before surgery

Can you tell me about your decision to have midface surgery?

Exploring reasons behind/ Who's decision/ Perceptions of self before/Hopes for surgery/ Sense made of decision/ Understanding of procedure

How where you prepared for the surgery?

Was any support was offered to you in preparation?

During

How did you find having the operation to fit the frame?

Views of frame/ Being in hospital/ Recovery/

Interested to know, how did you find going home with the frame on?

How did you feel about the frame once home/ Other people's responds/ School/ peers/ going out

How long did you have the frame on for?

Did your feeling changes towards the frame change over the …months?

How did you find having the frame removed?

After frame off

Functional results

Changes to appearance

Interested to know, what it was like for you seeing yourself with the frame off for first time?

What did you think? Feel?/ Did you have any preparation or conversations with anyone about looking at yourself for first time?

How did you feel about your appearance?

What was it like first seeing your daughter/son with the frame off?

What did you think? Feel? / What did you think about child's appearance once the swelling and bruising had gone?

If not answered-

What did you think of the results of the procedure?

After frame- going home

How did you find going back home?

How did other people respond to seeing you after the frame was removed?

Going back to school/ activities/ daily life

Was there anything else particularly challenging

Was there anything that helped settling back into life? *(after coming home)*

Did you have any support during this time?

Do you feel having this procedure has made any difference to your life? Both child and parent

Did surgery meet expectations?

Is there anything you wish would have been different about the procedure? Or the care/ support you received?

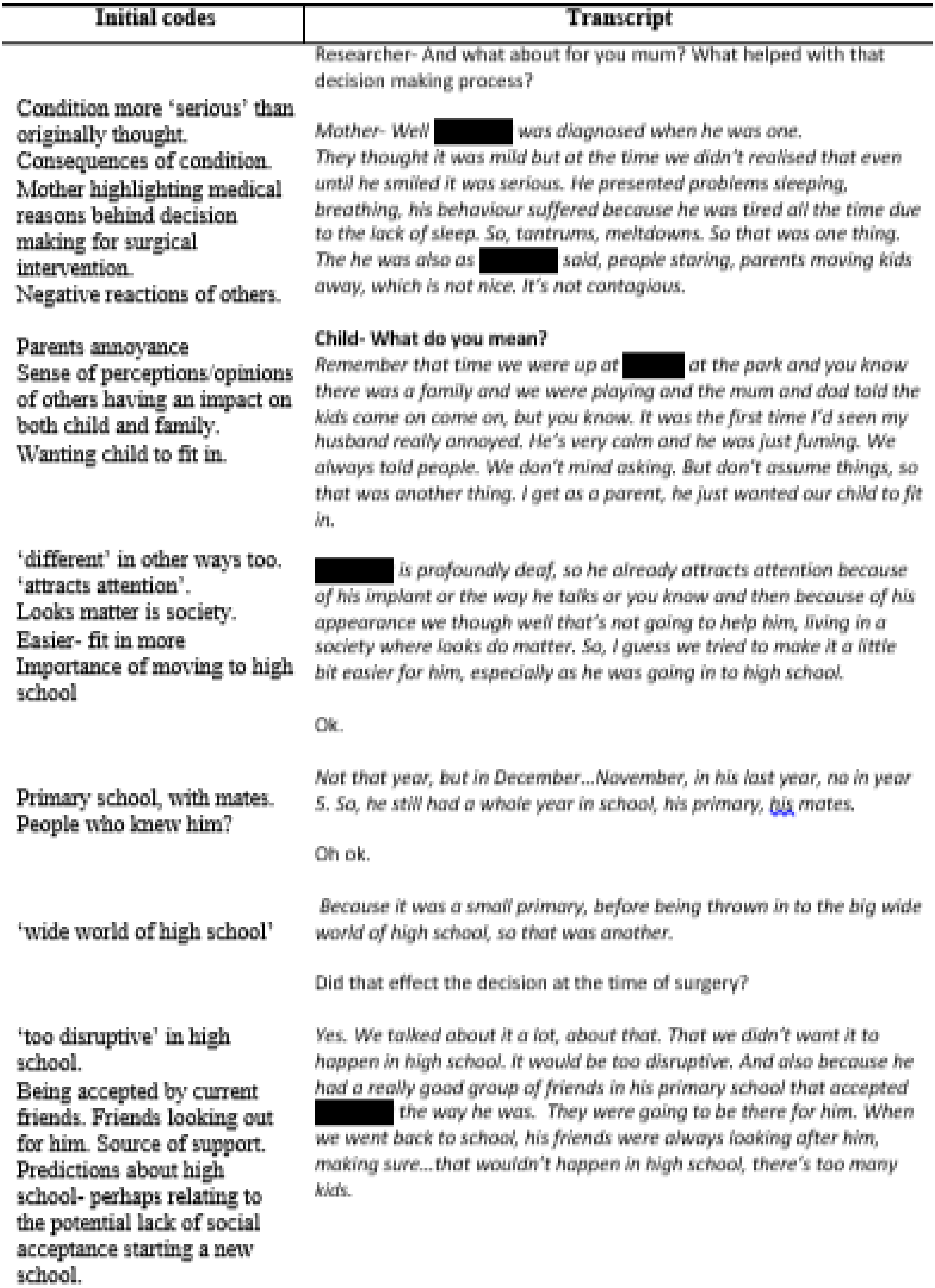
What would you tell others considering this surgery?

Was there anything else you would like to mention that you think is important that we have not discussed? Child and Mum

Thank you for taking part, do you have any final thoughts or reflections a

Provide details of dissemination and signposting if necessary.

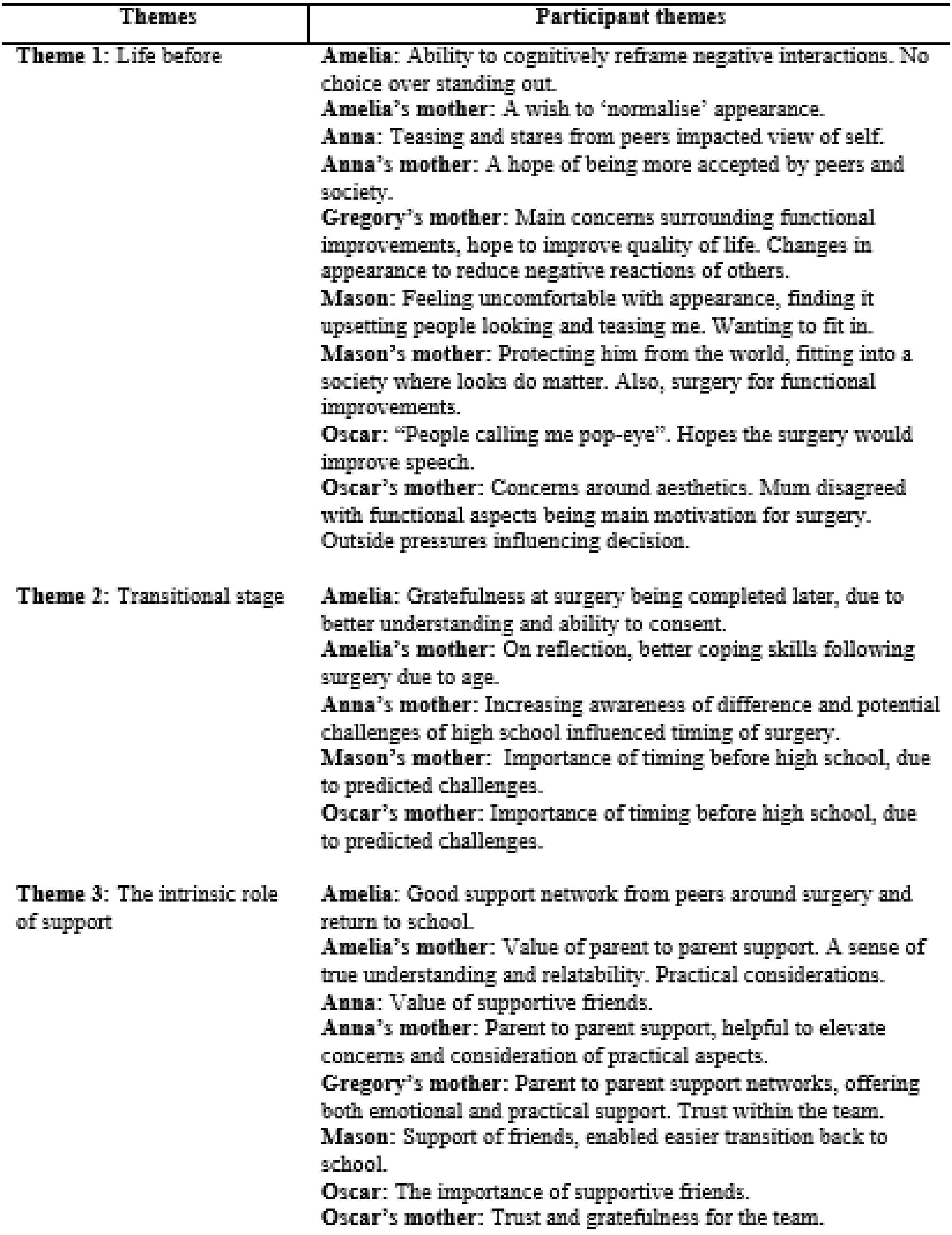
Appendix O. *Initial coding exert*



Initial codes	Transcript
	Researcher- And what about for you mum? What helped with that decision making process?
Condition more 'serious' than originally thought. Consequences of condition. Mother highlighting medical reasons behind decision making for surgical intervention. Negative reactions of others.	*Mother- Well [redacted] was diagnosed when he was one. They thought it was mild but at the time we didn't realised that even until he smiled it was serious. He presented problems sleeping, breathing, his behaviour suffered because he was tired all the time due to the lack of sleep. So, tantrums, meltdowns. So that was one thing. The he was also as [redacted] said, people staring, parents moving kids away, which is not nice. It's not contagious.*
Parents annoyance Sense of perceptions/opinions of others having an impact on both child and family. Wanting child to fit in.	**Child- What do you mean?** *Remember that time we were up at [redacted] at the park and you know there was a family and we were playing and the mum and dad told the kids come on come on, but you know. It was the first time I'd seen my husband really annoyed. He's very calm and he was just fuming. We always told people. We don't mind asking. But don't assume things, so that was another thing. I get as a parent, he just wanted our child to fit in.*
'different' in other ways too. 'attracts attention'. Looks matter is society. Easier- fit in more Importance of moving to high school	*[redacted] is profoundly deaf, so he already attracts attention because of his implant or the way he talks or you know and then because of his appearance we though well that's not going to help him, living in a society where looks do matter. So, I guess we tried to make it a little bit easier for him, especially as he was going in to high school.*
	Ok.
Primary school, with mates. People who knew him?	*Not that year, but in December...November, in his last year, no in year 5. So, he still had a whole year in school, his primary, his mates.*
	Oh ok.
'wide world of high school'	*Because it was a small primary, before being thrown in to the big wide world of high school, so that was another.*
	Did that effect the decision at the time of surgery?
'too disruptive' in high school. Being accepted by current friends. Friends looking out for him. Source of support. Predictions about high school- perhaps relating to the potential lack of social acceptance starting a new school.	*Yes. We talked about it a lot, about that. That we didn't want it to happen in high school. It would be too disruptive. And also because he had a really good group of friends in his primary school that accepted [redacted] the way he was. They were going to be there for him. When we went back to school, his friends were always looking after him, making sure...that wouldn't happen in high school, there's too many kids.*

Timing of surgery important. Want for your child to fit in 'just another kid'.	*You know. So, the timing was really good as well. He was mainly, apart from the health issues, they just wanted* [redacted] *to be just another kid.* Ok.
Vain- carrying about appearance. 'one less thing to worry about'. Removing what can change.	*It may sound a bit vain. But he spent a lot of time in hospitals, we just wanted one less thing to worry about.*

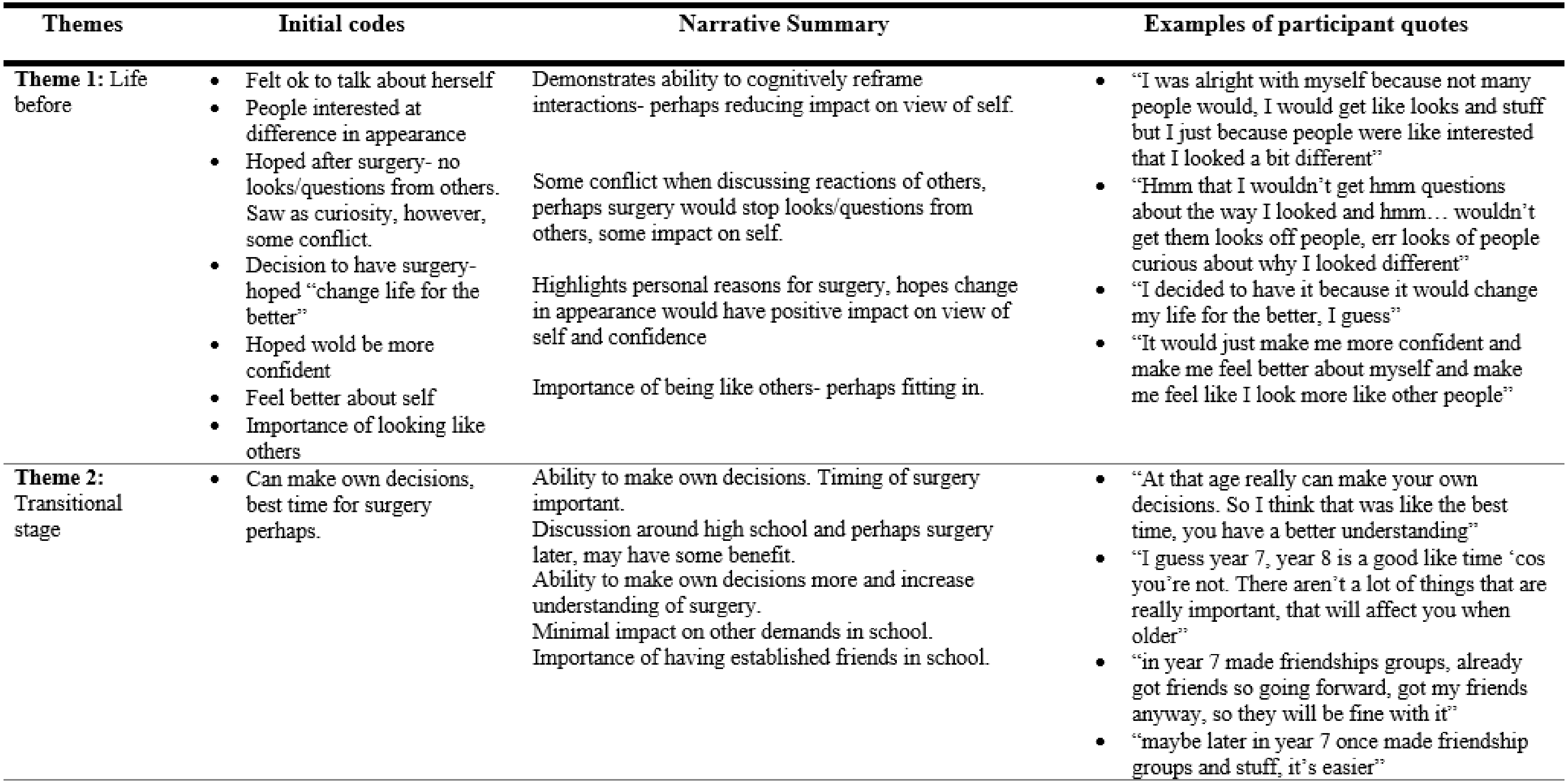
Appendix P. *Examples of participant themes*



Themes	Participant themes
Theme 1: Life before	**Amelia**: Ability to cognitively reframe negative interactions. No choice over standing out. **Amelia's mother**: A wish to 'normalise' appearance. **Anna**: Teasing and stares from peers impacted view of self. **Anna's mother**: A hope of being more accepted by peers and society. **Gregory's mother**: Main concerns surrounding functional improvements, hope to improve quality of life. Changes in appearance to reduce negative reactions of others. **Mason**: Feeling uncomfortable with appearance, finding it upsetting people looking and teasing me. Wanting to fit in. **Mason's mother**: Protecting him from the world, fitting into a society where looks do matter. Also, surgery for functional improvements. **Oscar**: "People calling me pop-eye". Hopes the surgery would improve speech. **Oscar's mother**: Concerns around aesthetics. Mum disagreed with functional aspects being main motivation for surgery. Outside pressures influencing decision.
Theme 2: Transitional stage	**Amelia**: Gratefulness at surgery being completed later, due to better understanding and ability to consent. **Amelia's mother**: On reflection, better coping skills following surgery due to age. **Anna's mother**: Increasing awareness of difference and potential challenges of high school influenced timing of surgery. **Mason's mother**: Importance of timing before high school, due to predicted challenges. **Oscar's mother**: Importance of timing before high school, due to predicted challenges.
Theme 3: The intrinsic role of support	**Amelia**: Good support network from peers around surgery and return to school. **Amelia's mother**: Value of parent to parent support. A sense of true understanding and relatability. Practical considerations. **Anna**: Value of supportive friends. **Anna's mother**: Parent to parent support, helpful to elevate concerns and consideration of practical aspects. **Gregory's mother**: Parent to parent support networks, offering both emotional and practical support. Trust within the team. **Mason**: Support of friends, enabled easier transition back to school. **Oscar**: The importance of supportive friends. **Oscar's mother**: Trust and gratefulness for the team.

Theme 4: Managing and making sense of new me	**Amelia:** Struggling to adjust to altered appearance. Compliments from others both a help and a hindrance. **Amelia's mother:** Notion of choice to be different. **Anna:** Reduced teasing and feeling more accepted, increased confidence. Some time to adjust. **Anna's mother:** Unexpected consequences: heightened anxiety. **Gregory's mother:** Great improvements in quality of life. More confidence. Positive impact on whole family. Dislike for perception that he was a difference person. **Mason:** "It's changed my life". More time admiring appearance and more vocal. **Mason's mother:** Unexpected outcomes: behavioural and emotional changes. Reduced role as protector and defender. Increased social confidence. **Oscar:** No longer singled out or teased. Changes enabled more positive interactions with peers. **Oscar's mother:** Positive emotional impact and increase in confidence.

Appendix Q. *Audit Trail Example – Sample extract from ‘Amelia’*



Themes	Initial codes	Narrative Summary	Examples of participant quotes
Theme 1: Life before	• Felt ok to talk about herself • People interested at difference in appearance • Hoped after surgery- no looks/questions from others. Saw as curiosity, however, some conflict. • Decision to have surgery- hoped “change life for the better” • Hoped wold be more confident • Feel better about self • Importance of looking like others	Demonstrates ability to cognitively reframe interactions- perhaps reducing impact on view of self. Some conflict when discussing reactions of others, perhaps surgery would stop looks/questions from others, some impact on self. Highlights personal reasons for surgery, hopes change in appearance would have positive impact on view of self and confidence Importance of being like others- perhaps fitting in.	• “I was alright with myself because not many people would, I would get like looks and stuff but I just because people were like interested that I looked a bit different” • “Hmm that I wouldn’t get hmm questions about the way I looked and hmm… wouldn’t get them looks off people, err looks of people curious about why I looked different” • “I decided to have it because it would change my life for the better, I guess” • “It would just make me more confident and make me feel better about myself and make me feel like I look more like other people”
Theme 2: Transitional stage	• Can make own decisions, best time for surgery perhaps.	Ability to make own decisions. Timing of surgery important. Discussion around high school and perhaps surgery later, may have some benefit. Ability to make own decisions more and increase understanding of surgery. Minimal impact on other demands in school. Importance of having established friends in school.	• “At that age really can make your own decisions. So I think that was like the best time, you have a better understanding” • “I guess year 7, year 8 is a good like time ‘cos you’re not. There aren’t a lot of things that are really important, that will affect you when older” • “in year 7 made friendships groups, already got friends so going forward, got my friends anyway, so they will be fine with it” • “maybe later in year 7 once made friendship groups and stuff, it’s easier”

Theme 3: The intrinsic role of support	• Seeing transformations of other child who had midface surgery. Good outcome increasing own confidence • Importance of having friends, who know you well, will help, helped transition back to school. • Medical team offering support, speaking to school about procedure, making return easier.	Importance of shared connection with others in similar position. Seeing outcomes, increased confidence in procedure. Relatability support network, connections with peers who know you. Feeling they will support and care for you. Increasing confidence to see others, to transition back to day to day life. Gratefulness of nurses taking a burden off her- helping her transition back to high school.	• "It was good because could see, like how he changed over the time and hmm he's like transformation from the beginning and afterwards was really good so it made me more confident about it" • "Your friends, got people who really know you…look out for you" • "they (friends) visited and made me feel better, confident about going to school" • "the nurses, going into school to explain, It kind of like took weight off me when I went in"
Theme 4: Managing and making sense of new me	• Recalling the initial shock of seeing self after surgery. • Huge changes in appearance.	Shock at altered appearance.	• "It was a shock because can see yourself, can see the difference as like it goes along, as each day with it on, but you can't see everything like at the same time, the whole thing. But when I got it off I was like wow it's a big difference"
	• Changes in appearance took time to get used to 'new version'. • Recalling previous appearance that had for so long, struggling with change. • Grown to get used to new appearance, please with outcomes. • Concerns about other aspect of appearance, others disagree.	Feeling as though 'a new version of self' Changes in appearance resulted in an unexpected struggle to adjust to overtime, gradually adjusted and pleased with outcomes. Times to adjust needed. Focus on insecurities shifted to another feature. Conflict between others perception of this and her own.	• "obviously my face had changed. It was like getting used to seeing myself looking like the new, new version. I think it was just that, I think that takes time to get used to… But I obviously had a different face, for so much longer, its take you a while…You have got used to it being you, I had a wobble, it's a huge change" • "I have kind of grow to get used to it, you grow to like see yourself looking like I do now. And you do. I like the way I look now" • "It was quite different because obviously moved your face forward which meant that my nose looked different but apparently it doesn't look different. To me it looks different".

	• Outcome- more confident, socially too. • Feel 'fit in more' and more accepted 'like people'	Surgery as a catalyst for increased confidence/social confidence. With 'difference' removed, now feel more accepted by others/fit in more, seen as an important outcome.	• "I think it's probably made me bit more confident, like talking to people and like things like that" • "I think it makes me feel like I fit in more" • "I feel more like, accepted. Like people" • "Yeah I think it made me feel better about myself because, also after you get quite a lot of compliments like oh you look really well and hmm it such a big change in a good way" • "My own style. Before you naturally stood out but now I can choose to stand out, if I want to stand out".
	• Receiving compliments from others	Comments from others strengthening own approval of changes, offering reassurance.	• "oh everyone said not look different but different person. Because I'm not a different person. Because my personality is still the same, it's just my bone structure, that's changed."
	• Differences making her naturally stand out, can choice to stand out • 'Different person' not difference appearances. Expressing annoyance at this, feels still the same person, just different appearance.	Valuing difference- but important factor being the choice to stand out or not on own terms. Comments from others that different person, after this surgery, distressed and neglected the fact that identity was based on more than looks, a want for others to acknowledge this.	• "I do look different but I'm not a different person, I just have a different face saying I've changed completely didn't like that"