Parents' experiences of stepping back in the decision-making process during transition in the cleft pathway

Jana Safarikova

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University of East Anglia

Faculty of Medicine and Health Sciences

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Thesis Portfolio Abstract

Background

Cleft lip and/or palate (CL/P) is the most common congenital craniofacial condition that can cause physical health complications, psychological difficulties and challenges in social interactions. Parents of children with CL/P are also affected as they face challenges of demanding care needs and long-term treatment. It is important to understand parents' experiences and their needs to support them when caring for their children.

Method

First, a systematic review was conducted to appraise and synthesise evidence of parental attachment in the early years of lives of children with CL/P and subsequent parental. Second, an empirical study explored parental experiences of a decision-making process about an elective, orthognathic surgery when the responsibility of the decision moves from parents to the young person affected.

Results

The systematic review found that after some changes during the first year, there are no differences in attachment between mothers of children with and without cleft at five years. Findings on parenting were mixed but the majority highlighted either no differences in comparison to parents of healthy children or fostering autonomy despite worries about children's health and physical safety. The empirical study identified three main themes, '"Our" journey', 'Stepping back' and 'Helping with the bigger picture', reflecting the changing role in responsibility for decision-making.

Conclusions

The thesis focuses on two key time periods identified in literature, the start and the end of the cleft journey, to better understand parental needs. Parents develop attachment to their child while adjusting to their needs and undergoing initial surgeries during the first year.

Another crucial period is the transition of decision-making regarding the orthognathic surgery when parents are expected to hand over responsibility to their adolescent child. The thesis highlights complexities and potential key factors impacting on parental experience, i.e., the age of the young people and parenting styles.

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Acknowledgements

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Chapter 1.

Introduction Chapter

Cleft lip and/or palate malformations are the most common congenital abnormality affecting the craniofacial region (Goodacre & Swan, 2008, 2012; NHS England, 2013). Approximately one in 700 babies in the United Kingdom are born with this condition. It occurs when embryological parts forming the lip, nose and palate do not join correctly during early pregnancy and create a gap. Although some potential risk factors have been found, the causes are thought to be complex, an interplay of genetic and environmental factors that cannot be predicted. The most frequent type is cleft palate only (45%), cleft lip only (23%), followed by unilateral cleft lip and palate (22%) and bilateral cleft lip and palate (10%) (NHS England, 2013).

This condition can cause orthodontic complications, negatively impact facial growth, speech and hearing (NHS England, 2013), making people with cleft stand out. It can therefore cause psychological difficulties and challenges in social interactions for the individuals. Young people reported being bullied because of their appearance and wanting to fit in better (Liddle et al., 2018). Parents considered their children to have more internalising and externalising behavioural problems such as anxiety, lower self-esteem, being less happy and teased more due to their condition (Hunt et al., 2007).

Parents are greatly affected as well (Breuning et al., 2020; Hlongwa & Rispel, 2018). Their psychological burden starts when their child's condition is diagnosed, which can be during pregnancy or at birth, and continues throughout their child's life. These challenges can centre around feeding, worries their child will choke as milk comes out through the nose because of the hole in the mouth, paying extra attention to their breathing and numerous visits to the hospital and clinics during the long-term treatment. Up to 25% of cleft lip and palate conditions are diagnosed prenatally during a 20-week scan (NHS England, 2013) with cleft lip only in around 70% of cases (Goodacre & Swan, 2008, 2012). Although this represents an increased worry in expecting parents for the second half

of the pregnancy, it also gives them a chance to adapt to the diagnosis. Mothers of newborns with cleft lip and/or palate who received diagnosis postpartum reported higher anxiety, more frequently feeling scared and depressed than mothers who were given diagnosis prenatally (Johns et al., 2018).

Clinical care comprises advice on feeding and care, hearing assessments of babies, counselling and psychological support for parents, and continues into early adulthood (NHS England, 2013; Colbert et al., 2015). Children undergo several surgeries during their childhood (Cleft Lip and Palate Association, 2019; Colbert et al., 2015; Goodacre & Swan, 2008, 2012) starting with a lip repair surgery at three to six months of age. This is followed by a palate repair surgery at six to 12 months, speech surgery and lip revision around three to five years of age, and Alveolar Bone Graft surgery alongside extensive orthodontal work to help with teeth growth at six to 12 years old. Numerous eye, nose, throat (ENT), ear and speech assessments are conducted alongside surgical interventions to ensure clear hearing, breathing and speech production. Given the above raising and caring for a child with cleft leads to an additional burden of care and responsibility for decision-making for parents due to their involvement in the treatment of their children (Hlongwa & Rispel, 2018; Maurien et al., 2019; Stock & Feragen, 2016).

Around the child's age of 15 years, an elective orthognathic pathway is offered to some young people as a prospective treatment for aesthetic and functional reasons. This pathway takes several years during which extensive orthodontic work is done in preparation for the main orthognathic surgery (Royal College of Surgeons of England, 2013). The surgery itself takes place when the young person is around 20 years old, at a time when there are no more changes to their bone structure through growth. The purpose of this complex surgery is to realign the upper and lower jaws to improve biting and create a more typical facial profile, and correct other structures to help with speech production or

breathing. This surgery is understood to represent the final stage of the lengthy treatment process. With it also comes a change in decision-making as the young person themselves is expected to make the decision about the pathway, not their parents who will have held the responsibility for their child's treatment decisions for at least the previous 15 years (Nelson et al., 2012).

It is crucial to understand parents' experiences of caring for their child and their needs throughout the different stages of the cleft pathway (Nelson et al., 2011). This thesis therefore aims to develop and report a greater understanding of the impact and experiences of caring for a child with craniofacial condition. Chapter 2 contains a systematic review of literature addressing parent-to-child attachment and parenting styles. It explores whether cleft diagnosis and the child's additional needs affect bonding of the parent to their child and subsequent parenting style. Chapter 3 briefly connects the systematic review and empirical study, drawing on the link between parenting styles and their impact on decision-making in children. Chapter 4 presents an empirical study that explores parents' experiences of and readiness for handing over responsibility for decision-making to their children after many years of making decisions about their treatment.

The thesis portfolio finishes with Chapter 5 that brings together findings of both papers and offers a reflection on the process of conducting and reporting the systematic review and empirical paper.

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Chapter 2.

Systematic Review

Prepared for submission to The Cleft Palate-Craniofacial Journal (Appendix A).

Parental attachment and parenting when having a child born with cleft lip and/or palate: A systematic review

Jana Safarikova and Dr Paul Fisher

Department of Clinical Psychology and Psychological Therapies, University of East Anglia, UK

Corresponding author:

Jana Safarikova, Department of Clinical Psychology and Psychological Therapies, Norwich Medical School, University of East Anglia, Norwich NR4 7TJ. E-mail. j.safarikova@uea.ac.uk

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Abstract

Objective: This systematic review aims to identify and evaluate available research related to parent-child attachments and parenting in parents of children born with craniofacial anomalies (CFA).

Design: A systematic search following Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidance was conducted using three databases (Medline EBSCO Complete, PsycINFO and Scopus) from their inception to March 2021.

Results: The search identified 1,188 articles of which 12 were eligible to be included in the review. Six studies addressed parent-child attachment and seven studies parenting style/approach. Only one of the studies was longitudinal and focused on both concepts. Considering mothers of children with cleft no evidence for bonding disorders was found. Maternal representations seemed to improve by 12 months, and no attachment differences were found by 5 years of child's age between the groups of mothers. In terms of parenting style, while one study found mothers of children with CFA more authoritarian, other studies found no differences between the groups in parenting, or even fostering more independence.

Conclusions: Bonding and attachment to a child with CFA seem to undergo development during the first year, showing no differences in maternal attachment compared with mothers of healthy children at 5 years. Due to varied methodologies findings on parenting seem mixed. The review emphasised factors influencing both attachment and parenting. It highlighted underrepresentation of fathers in research, with no paternal attachment investigated, and the need to assess parental attachment and parenting in longitudinal studies.

Keywords

Attachment, maternal representations, parenting, cleft, review

Introduction

Cleft lip and/or palate, the most common congenital condition of the face and oral cavity, affects approximately one in 700 babies in the United Kingdom (UK) (Goodacre and Swan, 2008, 2012). Parents' reactions to the news of having a baby with cleft were addressed internationally, describing shock, shame, worry, self-blame, sadness or disappointment (eg, Dapaah et al., 2020; Hlongwa and Rispel, 2018; McCorkell et al., 2012; Nelson et al., 2011; Nelson et al., 2012). Having a child born with cleft represents a long treatment journey (NHS England, 2013) that is intensive from the beginning and can be found overwhelming in terms of information (Costa et al., 2019) and increased burden of care (Razera et al., 2017). However, more positively, Stock et al. (2020) suggested that parents adjust to the diagnosis, and factors such as satisfaction with health care support and familial relationship serve as protective.

Adjusting to the diagnosis and demands on care such as feeding takes place during bonding stages. Forming an emotional bond and attachment to a child is generally considered the most important relationship for the child's social, emotional and cognitive development (Clark and Ladd, 2000). A secure attachment reflects the child's feelings of safety, closeness and trust. However, some factors have been found to impact on the bonding process (eg, Martins and Gaffan, 2000). A meta-analytic investigation of early maternal depression and attachment of healthy infants suggested that infants of depressed mothers were less likely to form a secure attachment. Depression was also highlighted as a predictor for insecure attachment in infants with cleft, alongside younger maternal age and this being the woman's first baby, rather than child's facial appearance (Speltz et al., 1997). In contrast, although mothers of children with cleft were found to be more likely depressed than mothers of healthy children, there was no evidence that maternal depression affected interactions with their children (Murray et al., 2008). Similarly, maternal

depression was not found to significantly affect behaviours towards children with cleft but factors such as marital stress and socioeconomic circumstances did (Pelchat et al., 2003).

Research focused on early attachment of infants with medical conditions indicated more insecure attachment in comparison to healthy children (Ward et al., 1993). However, when specifically assessing attachment of infants with and without cleft no differences were found (Habersaat et al., 2013; Hoeksma et al., 1996; Speltz et al., 1997). Furthermore, there is evidence that attachment can change and by 24 months children's attachment patterns are no different (Maris et al., 2000). The greatest change was found in children with cleft palate only. If they were insecurely attached at 12 months, they were more likely to have secure attachment at 24 months. Because of these changes Maris et al. (2000) highlighted the need to address child attachment longitudinally as assessment is usually conducted once, at 12 months of infants' age, using the Strange Situation (Ainsworth et al., 1978).

Nevertheless, a recent study explored the adult attachment style of people born with cleft (Ardouin et al., 2021). They reported less secure attachment in adult relationships than the general population while having good relationships with family members who can provide emotional support.

The research above focused on evaluating attachment quality in infants or adults who were born with cleft. The attachment of parents was not addressed although some parental attributes such as protectiveness and perceived vulnerability of children were considered (Collet and Speltz, 2007; Hlongwa and Rispel, 2018; Speltz et al., 2000). These factors were also considered in parents of children with other health conditions. Increased protectiveness and/or perceived vulnerability was found in parents of children with chronic illness such as cancer (Colletti et al., 2008), diabetes (Mullins et al., 2004) or rheumatoid arthritis (Power et al., 2003).

The attachment theory (Ainsworth and Bowlby, 1991) suggests that primary caregivers should be responsive and available for the child to thrive and develop a secure attachment. However, times of increased stress and threat could impact on the caring behaviours in parents and establishing bonds with their children. Upon receiving their child's diagnosis of cleft lip and/or palate followed by experiences of shock, grief or shame (Nelson et al., 2011), parents might be mourning their 'perfect child', which negatively affects their developing attachment (Bowlby, 1982). In contrast, research also shows that parents expressed elation at having a newborn and considered the diagnosis of cleft unremarkable (Nelson et al., 2011), able to form a loving attachment.

Pinquart (2013) conducted a meta-analysis exploring parent-child relationships and parenting behaviours relating to various chronic illnesses. There was evidence, despite small effect sizes, that the parent-child relationships tended to be less positive in families with children with specific chronic illness such as epilepsy, asthma and hearing impairment, most likely due to more stressors associated with these conditions. Families with children with cleft, despite the additional burden of care and psychological strain, seemed to adjust well.

Parental characteristics such as protectiveness, control or perceived vulnerability are linked with the parenting dimensions (Baumrind, 1996) of responsiveness and demandingness. Responsiveness is conceptualised as fostering individuality in the child, being supportive, and showing warmth. Demandingness is displayed in actions to regulate child's behaviour or expectations of child's own control over their behaviour. These attributes are over time reflected in parenting styles (Baumrind, 1971), which in turn impact on the development of decisional capacity in children (Partridge, 2010). Authoritative parenting style was found most supportive of developing autonomy in children. Furthermore, there is evidence of a link between specific parenting styles and

parents' adult attachment, namely, secure adult attachment and authoritative parenting style (Nanu and Nijloveanu, 2015).

Considering the parental involvement during the whole cleft treatment, parents play a crucial, demanding role. Their attachment and parenting style will be reflected in decision-making and affect their readiness to hand over responsibility for decisions to their children (Partridge, 2010). Within this context the current systematic review aimed to present a synthesis of literature on parental attachment and parenting of a child with cleft lip and/or palate and examine whether there are any changes in them over time.

Methods

Search Methods

This systematic review followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Statement (PRISMA; Moher et al., 2009) to report on the process and results. The protocol was registered on PROSPERO, the International prospective register of systematic reviews (PROSPERO 2021 CRD42021240453). Three electronic databases (MEDLINE EBSCO Complete, PsycINFO and Scopus) were searched for research articles from inception to 6 March 2021. Guidance was sought from a dedicated academic librarian. Search keywords are available in Table 1. Reference lists of included studies and wider cleft literature were also searched for additional potentially eligible research papers.

Table 1. Systematic Review Search Strategy Keywords.

	Concept 1 (AB)	AND	Concept 2 (AB)	AND	Concept 3 (AB)
Search terms	Cleft		parent or parental or mother or maternal or father or paternal or stepparent or stepparental or		Attachment style or relationship or interaction

	stepmother or stepfather or caregiver

Selection Criteria

All retrieved records were gathered into an Excel spreadsheet and duplicates removed. Two independent researchers screened the titles and abstracts using set eligibility criteria. Remaining studies underwent a full-text screening. Studies which focused on parent-child bonding, parental attachment style with children born with cleft lip and/or palate (but not child's attachment), parenting style and parenting/caring behaviours were selected for the review. No restrictions were applied to interventions or control groups specifications due to the expected varied methodology. Both qualitative and quantitative studies were included to maximise the findings in the area of interest (Harden and Thomas, 2005). Records were excluded if they were not published in English, were editorials, review articles or not related to cleft lip and/or palate. Figure 1 represents the PRISMA flow diagram detailing the number of studies and reasons for exclusion.

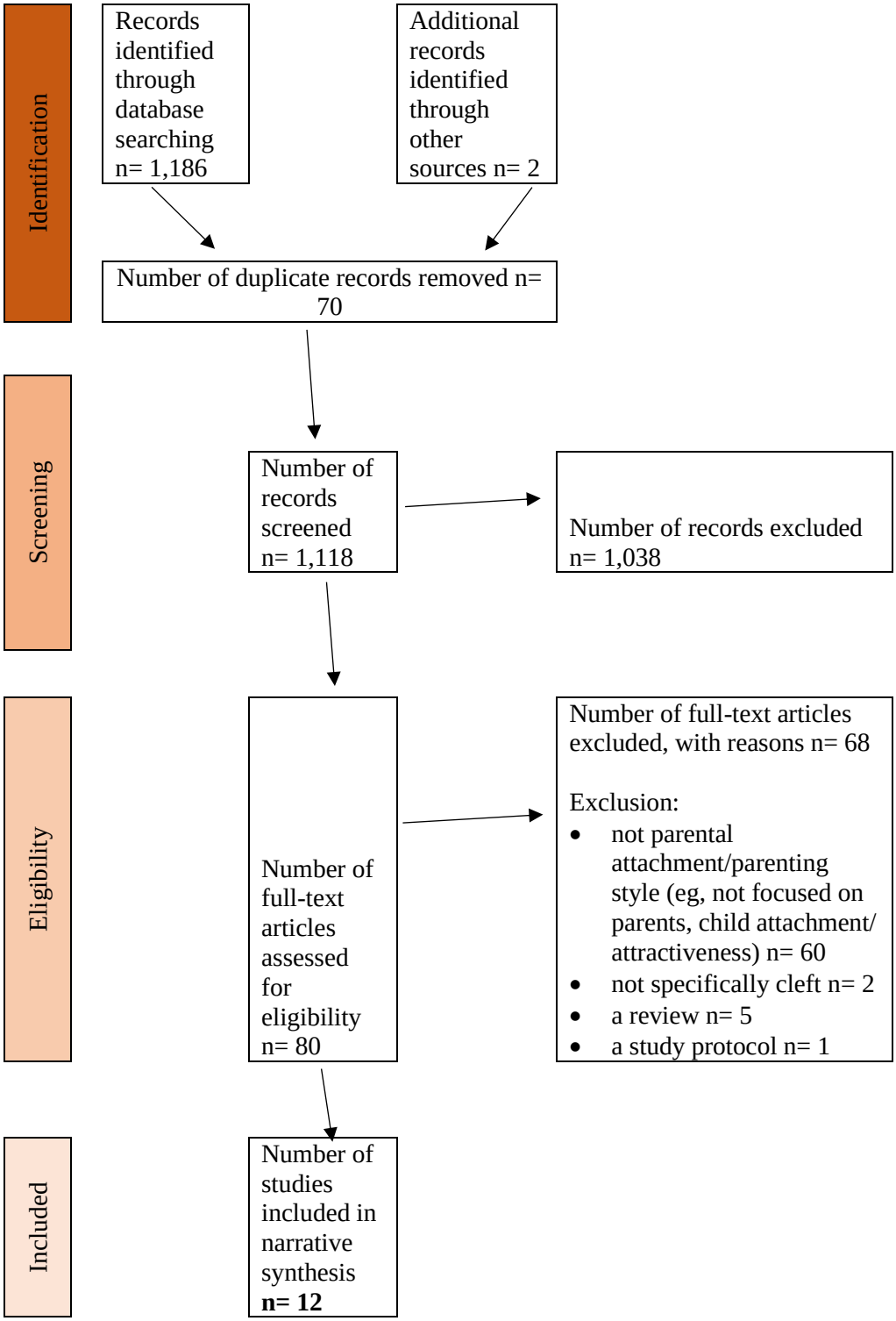

Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Flow Diagram of Search and Eligibility Process.

Data Collection

Remaining studies were reviewed in full text by the lead reviewer who also conducted the data extraction. A list of key details was developed into an excel tool for data extraction containing the following fields: authors, year, country/setting, study title, methodology (qualitative/quantitative), study focus/aim, methods, intervention, target population including gender, sample size, age of children whose parents took part, results, strengths and limitations, findings and notes. All study articles were used in a narrative synthesis.

Methodological quality and risk of bias assessment was conducted by the lead reviewer and separately 25% of studies were assessed by a second reviewer to ensure a robust process. Any disagreements were resolved between the reviewers. Studies were assessed using the Critical Appraisal Skills Programme (CASP, 2018). The CASP Cohort Study Checklist was used for quantitative studies and the CASP Qualitative Checklist for qualitative studies. Following other research that applied scoring criteria alongside CASP or similar checklists (Hendry et al., 2018; Kmet et al., 2004; Rushbrooke et al., 2014) a scoring system was used in this review. A score of 2 was assigned to 'yes', a score of 1 to 'can't tell', a score of 0 to 'no'. Where items on the quantitative studies checklist were not applicable, eg, no follow-up, those were assigned a score of 2 and deducted from the possible total score (Kmet et al., 2004). Studies that achieved a percentage of 85% and above were considered of high quality, a percentage of 51%-84% was considered moderate quality and below 50% a low quality.

Data Analysis

Due to varied methodology across the studies a narrative synthesis was used (eg, Harden and Thomas, 2005; Popay et al., 2006). Narrative synthesis is a widely used approach for systematic reviews, especially if meta-analysis is not possible, and allows the researcher to

synthesise a range of questions and summarise findings. In undertaking the synthesis studies were initially categorised into two groups based on the concepts they explored, ie, attachment and parenting, and the findings of papers were reviewed. Connections within and between the groups were considered, leading to the synthesis of findings that are discussed.

Results

Search Outcome

As Figure 1 shows, the search identified 1,188 articles. Following the removal of duplicates, 1,118 records remained for title and abstract screening during which 1,038 records were excluded with 80 records left for more detailed screening. Forty-nine articles were excluded, leaving 31 records for full text review. Of those, 12 articles published between 1993 and 2020 were included for data extraction. Nine studies used quantitative methodology (Table 2) and three studies qualitative (Table 3). One study was longitudinal (Habersaat et al., 2018), the remaining studies were cross-sectional. Five studies focused on the first year after giving birth, four studies addressed time period between 3 to 9 years, two studies explored parents' experiences of children aged 9 to 14 and one study captured a wider age range 8 to 18.

Participants

In total, the 12 studies represented 79,966 participants, of which 753 were participants in the groups of interest, eg, cleft, congenital anomaly (634 parents, 119 children), and 79,213 in control groups (79,151 parents, 62 children). Participants across the 12 studies were predominantly mothers. Whereas parenting was explored with both mothers and fathers, parental attachment was investigated only in mothers. The overall high number of

participants is due to one nation-wide study accounting for 79,140 participants (Tsuchiya et al., 2019).

Table 2. Summary of Quantitative Studies Included in the Systematic Review.

Authors (year)	Country/ setting	Study design	Measure	Target population	Sample size	Results
Boztepe et al. (2016)	Turkey	Cross-sectional; questionnaires, face-to-face interviews	Maternal Attachment Inventory (MAI), Structured Questionnaire Form	Mothers of children with CL/P, congenital heart disease and healthy controls (infants 1-12 months old)	50 CL/P, 50 congenital heart disease, 100 controls	Mothers of infants with CL/P who did not experience psychological problems during pregnancy had higher MAI scores than did mothers who experienced psychological problems ($z = –2.060$, $P < .05$). Mothers of infants with CL/P were more likely to have future concerns (CHD: 62%, CL/P: 66%) than the mothers of infants with CHD. A statistically significant difference was found in maternal attachment scores between the mothers of infants with CL/P and those of healthy infants ($P = .001$).
Cinar et al. (2020)	Turkey	Cross-sectional; quasi-experimental study with a pre-test and post-test control group design (questionnaires)	Maternal Attachment Inventory (MAI), Parental Self-Efficacy (PSE)	Mothers of children with CLP who had undergone CL repair surgery (age 3-7 months)	32 mothers of infants with CLP (16 intervention group, 16 control)	The difference in the mean post-test maternal attachment scores between the intervention and control groups ($t = 6.670$, $P = .001$). The maternal attachment score and the parental self-efficacy score ($r = 0.555$, $P < .001$).

Table 2. (Continued)

Authors (year)	Country/ setting	Study design	Measure	Target population	Sample size	Results
Despars et al. (2011)	Switzerland	Cross-sectional, semi-structured interview	The Working Model of the Child Interview, The Impact of Event Scale	Mothers of children with cleft lip and/or palate (but not cleft palate only) and a control group	22 mothers of children with CL/P, 36 healthy controls	Mothers of infants with CL/P presented significantly fewer balanced ($x^2 = 3.84$, $P = .05$) and more disengaged attachment representations ($x^2 = 3.97$, $P = .05$) than did mothers of healthy infants. The cleft complexity did not significantly influence the quality of mother's representations. Mothers of infants with a cleft showed significantly more posttraumatic stress symptoms than the control group ($F_5 = 5.032$, $P = .03$). The complexity of the cleft did not significantly influence maternal stress ($F_5 = 2.54$, $P = .09$). Mothers of CL/P children who showed a relatively low level of PTSD symptoms, were more frequently categorized as disengaged, as compared with mothers of the control group and with mothers of CL/P children with a relatively high level of PTSD symptoms ($x^2 = 11.29$, $P = .01$).
Gassling et al. (2014)	Germany	Cross-sectional; experimental design (observation)	Observation and coding of parent-child interactions	Families of children with non-syndromic CLP (both parents), no history of migraine, learning difficulties or psychological problems in children	15 families with CLP child (7-9 years old), 20 families with a healthy child, 20 families with a child with migraines	Interactions with mother: significant differences between the groups for the purpose-related control (specific instructions, $F_{2,54} = 7.185$, $P = 0.002$), positive reinforcement ($F_{2,54} = 10.514$, $P < .001$) from the mother, through questions ($F_{2,54} = 13.144$, $P < .001$), interruptions ($F_{2,54} = 4.553$, $P = .015$), and autonomous problem solving, ie, control (autonomous problem solving, $F_{2,54} = 4.743$, $P = .0133$) by the child. Interactions with father: significant differences between the groups for help ($F_{2,52} = 9.370$, $P < .001$) from the father, questions ($F_{2,52} = 8.378$, $P < .001$), interruptions ($F_{2,52} = 9.404$, $P < .001$), and autonomous problem solving, ie, control ($F_{2,52} = 4.693$, $P = .014$) by the children. Compared with healthy children, there was a significant tendency towards more autonomous behaviour among CLP-affected children ($P = .12$).

Table 2. (Continued)

Authors (year)	Country/ setting	Study design	Measure	Target population	Sample size	Results
Habersaat et al. (2018)	Switzerland	Longitudinal; semi-structured interviews, questionnaire	The Working Model of the Child Interview [WMCI]) at 2 and 12 months of children's age; Parent Development Interview [PDI], Parenting Style and Dimensions Questionnaire (PSDQ) at 5 years of children's age	Mothers of children with an orofacial cleft	30 mothers of children with an orofacial cleft and 14 mothers of children without a cleft	No statistically significant difference across groups (cleft/non-cleft) in maternal representations at the 2-month, 12-month, and 5-year assessments. In the cleft group, significant differences were shown between 2 and 12 months in caregiving sensitivity (t = -2.12, P < .05), perceived infant difficulty (t = -2.51, P < .05), fear for the infant's safety (t = -3.43, P < .005), and parental pride (t = -2.20, P < .05), all factors being higher at 12 months. Such differences in parental representations over time were not found in the non-cleft group. Mothers of the cleft group were significantly more authoritarian than mothers of children without a cleft (t[32.04] = 2.07, P = .046).
Krueckeberg et al. (1993)	USA	Cross-sectional; questionnaires, interview, facial encoding and decoding	Parenting Stress Index, Modification of the Block Child Rearing Practices Report (nurturance and restrictiveness), Social Relationship Scale; Four Factor Index of Social Status, Social Skills Questionnaire, Facial Encoding and Decoding tasks, Enactive social knowledge interview	Preschool age children (3-6) and their families, children with craniofacial anomaly (CFA) and without	30 families with a child with CFA, 22 control families (parental gender not specified)	Parenting Stress Inventory: no significant main effects or interactions found on any of the parent measures between groups. Impact of visibility of cleft in the cleft group (out of 30 children 8 had invisible cleft) showed three statistically significant results. Parents of children with visible defects found their social network more helpful (t = 2.92, P ≤ .01), and favoured parenting styles that were more nurturant (t = 3.20, P < .01) and less restrictive (t = -2.54, P < .05) than parents of children with invisible defects. Child Rearing Practices Report: no significant differences between the cleft and non-cleft groups, both had similar nurturant and restrictive average scores.

Table 2. (Continued)

Authors (year)	Country/ setting	Study design	Measure	Target population	Sample size	Results
Shapiro et al. (2018)	USA	Cross-sectional; questionnaires	The activities subscale of the Occupations, Activities, and Traits–Attitudes Measure (C/OAT-AM), selected items from the Parent Perception Inventory (PPI), the nurturance subscale of the Parenting Dimensions Inventory–Short Form (PDI-S) and the Parenting Stress Scale (PSS)	Children with craniofacial difference (CFD) and their parents	74 dyads of children (8-18 years old) with CFD (n = 36 female), including cleft lip/palate, and a parent (n = 56 female).	Parents' flexible views about gender were associated with marginally lower parenting stress ($F_{1, 67}$ = 3.12, P = .08, η^2 = .04), and higher parent-reported nurturance ($F_{1,67}$ = 10.40, P = .002, η^2 = .13). There was also a main effect suggesting that more flexible parents were perceived by their children to be warmer and more positive ($F_{1,67}$ = 5.36, P = .02, η^2 = .07). However, this main effect was qualified by an interaction between child gender and parents' gender views ($F_{1,67}$ = 6.08, P = .02, η^2 = .07). Specifically, females with parents with more flexible views rated their relationship with their parent more favourably than did females with parents with stereotypical views ($F_{1,33}$ = 11.41, P = .002, η^2 = .26), for the simple main effect.
Tsuchiya et al. (2019)	Japan	Cross-sectional; questionnaires	Mother-to-infant bonding scale 12 months after childbirth; covariates [age at birth, parity, infant sex (from medical records), smoking, drinking, feeding, Psychological distress scale (PDS) (questionnaires)]	Mothers of children with CL/P or CP at 12 months post birth	211 pairs of mother-infant with cleft, 78,929 healthy controls	Risk of bonding disorders among all the mothers of infants with CL/P or CP (0.97 [0.63-1.48], P = .880). A significant association of CL/P or CP with bonding disorders was found only among advanced-age multiparae (odds ratio [95% confidence interval] = 2.51 [1.17–5.37], P = .018), but it was weakened after additional adjustment for maternal depression.

Table 2. (Continued)

Authors (year)	Country/ setting	Study design	Measure	Target population	Sample size	Results
Yilmaz et al. (2011)	Turkey	Cross-sectional; questionnaires	Maternal Attachment Inventory (MAI), demographic information	Mothers of infants (1-8 months) born with congenital anomalies (curable, incurable, Down syndrome)	70 mothers of infants (42 curable condition inc. CL/P, 17 incurable condition, 11 Down syndrome)	MAI averages for mothers with infants that could not be cured surgically were lower (M = 91.47, SD = 13.99) than MAI scores of the other mothers (curable: M = 98.57, SD = 4.72; Down syndrome: M = 97.72, SD = 7.41). There was a relationship between a mother's economic situation and the MAI point average, with mothers with higher income having a closer attachment (t = 2.100, P = .039). Mothers with problems during their pregnancies had lower MAI averages than mothers with a normal pregnancy (t = 2.286, P = .028). Mothers with poor relationships with their own mothers had lower MAI scores compared with mothers who had good relationships (t = 2623, P = .011).

Abbreviations: CL, cleft lip; CLP, cleft lip and palate; CL/P, cleft lip and/or palate; CP, cleft palate.

Table 3. Summary of Qualitative Studies Included in the Systematic Review.

Authors (year)	Country /setting	Study design	Method	Target population	Sample size	Results
Breuning et al. (2020)	Canada	Cross-sectional; interview	Semi-structured interviews (inductive content analysis)	Parents of children younger than 7 years of age with CL/P	Parents of 14 children (14 mothers, 3 fathers)	Four themes identified, one relevant: Psychosocial (school/daycare, family, coping strategies, child's pain).
Klein et al. (2006)	USA	Cross-sectional; interview	The maternal interview (narrative inductive approach)	Mothers of children with congenital craniofacial anomalies	9 mothers	Three themes identified, two of them relevant: Maternal worries and concern (physical/medical/safety, social exclusion/teasing/reaction of others, forming social relationships, emotional well-being), Maternal proactive behaviours (school related, social/peers, encouraging independence/letting go).
Klein et al. (2014)	USA	Cross-sectional; interview	The maternal interview (thematic narrative approach), a Social coaching task	Mothers of children with congenital craniofacial anomalies	9 mothers	Maternal Interview: Mothers' reports of child's social experiences (positive and negative experiences), Mother's views of other people, Mother's responses to other people (eg, actions taken to support her child's social interactions). Social coaching task: Maternal Framing (situational factors, factors in the other children, factors in her own child), Maternal Advice (prosocial behaviour, self-reliance/withdrawal, seeking help).

Grouping Based on Study Focus

Overall, studies were divided into two groups based on the concepts they addressed. Attachment was the focus of six studies and parenting was assessed and explored in seven. One study (Habersaat et al., 2018) is included in both groups as it addressed both attachment and parenting (Table 4).

Table 4. Grouping of Included Studies.

Authors (year)	Attachment	Parenting
Quantitative studies		
Boztepe et al. (2016)	X	
Cinar et al. (2020)	X	
Despars et al. (2011)	X	
Gassling et al. (2014)		X
Habersaat et al. (2018)	X	X
Krueckeberg et al. (1993)		X
Shapiro et al. (2018)		X
Tsuchiya et al. (2019)	X	
Yilmaz et al. (2011)	X	
Qualitative studies		
Breuning et al. (2020)		X
Klein et al. (2006)		X
Klein et al. (2014)		X

Quality Assessment

Methodological quality and risk of bias of studies was assessed using the CASP checklists (Critical Appraisal Skills Programme, 2018) and scores applied (eg, Kmet et al., 2004). A second reviewer assessed three out of the 12 studies, ie, 25%. Table 5 offers an overview of quality ratings with nine studies being rated as high quality and three as moderate quality. The scoring for individual studies including inter-rater checks is available in Appendix B.

Table 5. An Overview of Quality Assessment in Individual Studies.

Quality assessment	Definition	Studies
High	A study achieved 85% or above in terms of the possible maximum score based on answers to CASP criteria.	Boztepe et al. (2016) Breuning et al. (2020) Cinar et al. (2020) Despars et al. (2011) Klein et al. (2006) Klein et al. (2014) Krueckeberg et al. (1993) Tsuchiya et al. (2019) Yilmaz et al. (2011)
Moderate	A study achieved 51%-84% in terms of the possible maximum score based on the answer to CASP criteria.	Gassling et al. (2014) Habersaat et al. (2018) Shapiro et al. (2018)
Low	A study achieved 50% or below in terms of the possible maximum score based on answers to CASP criteria.	N/A

All studies had clearly stated aims of their research and all quantitative studies used appropriate measures in an attempt to minimise bias. Seven of the quantitative studies used questionnaires to gather data (Boztepe et al., 2016; Cinar and Koc, 2020; Habersaat et al., 2018; Krueckeberg and Kapp-Simon, 1993; Shapiro et al., 2018; Tsuchiya et al., 2019; Yilmaz et al., 2011). Although there is the potential of participant and/or social desirability bias, all questionnaires were validated. Interview methods and analysis of qualitative studies (Breuning et al., 2020; Klein et al., 2006, 2014) were considered appropriate and rigorous. However, consideration of the relationship between the researchers and participants was not clear.

Apart from one longitudinal study (Habersaat et al., 2018) all of the studies in the review were cross-sectional giving evidence for one point in time. Some studies highlighted the need for a longitudinal approach especially when exploring attachment (eg, Boztepe et al., 2016; Yilmaz et al., 2011).

Half of the studies (Boztepe et al., 2016; Habersaat et al., 2018; Klein et al., 2006, 2014; Krueckeberg and Kapp-Simon, 1993; Shapiro et al., 2018) were considered at risk of bias in terms of recruitment or their recruitment methods were not clear. Some authors themselves commented on the implications of their recruitment strategy (Habersaat et al., 2018; Klein et al., 2006, 2014; Tsuchiya et al., 2019). Although overall scoring 'high' on quality assessment for their studies, Klein et al. (2006, 2014) highlighted that their participants were volunteers in a support group, from a majority ethnic background, middle to upper-middle class, and therefore the authors were wary of generalising outside of these characteristics. Habersaat et al. (2018), scoring 'moderate' on quality assessment, reported on a small group of mothers of healthy children in their study leading to using non-parametric tests in their analysis due to a lack of power. They also commented that highly motivated parents were likely to participate in their longitudinal study over the 5 years.

In contrast, Gassling et al. (2014) stated that there was no recruitment bias in their study as all eligible participants that attended clinical appointments in the recruiting hospital within the time frame of the study were approached to participate and all of them consented and took part. No one declined or withdrew. However, this would indicate that only the first 15 eligible families out of the caseload were approached before reaching the required number of participants. It is questionable whether the 15 families were representative of the whole caseload. The study only included 'complete families' with both mothers and fathers present, which might not be representative of families that are separated although both parents care for the child.

Three studies were considered not fully taking into account potential confounding variables such as level of support from family or clinical team, impact of visibility of cleft (Habersaat et al., 2018; Shapiro et al., 2018), satisfaction with repair surgeries (Habersaat et al., 2018), and parenting style more broadly (Yilmaz et al., 2011).

Taken both attachment and parenting together, findings are limited to the age groups of children whose parents participated and cannot be generalised beyond that. Attachment was addressed mainly in the first year after giving birth (five out of six studies; Boztepe et al., 2016; Cinar and Koc, 2020; Despars et al., 2011; Tsuchiya et al., 2019; Yilmaz et al., 2011) and then at 5 years (Habersaat et al., 2018). Parenting was explored during a time period between 3 to 9 years in four studies (Breuning et al., 2020; Gassling et al., 2014; Habersaat et al., 2018; Krueckeberg and Kapp-Simon, 1993), ages 9 to 14 in two studies (Klein et al., 2006, 2014), and 8 to 18 in one study (Shapiro et al., 2018). Each study therefore focused on a specific period during the cleft journey and parenting within that context, with varied methodologies, which limits the scope for comparison of the studies.

Attachment

Having a child with cleft was not associated with increased risk of bonding disorders such as decreased maternal affection to their child in the first year (Tsuchiya et al., 2019). Although mothers of children with CL/P seem to present with fewer balanced (secured) and more disengaged (insecure) attachment representations than mothers of healthy children at 2 months (Despars et al., 2011), these changed during the first year and by 5 years no differences between mothers of children with and without CL/P were found (Habersaat et al., 2018). However, several factors were found to influence mothers' attachment. Lower economic situation, poor quality of relationship with their own mother (Yilmaz et al., 2011) and physical or psychological problems during pregnancy (Boztepe et al., 2016; Yilmaz et al., 2011) all had negative impact on attachment. Advanced maternal age and having more than one child were also highlighted as antecedents for difficulties bonding in the first year if mothers of children with CL/P had depression (Tsuchiya et al.,

2019). Nevertheless, attachment improved when mothers' level of perceived self-efficacy increased (Cinar and Koc, 2020) following additional nursing care provided to them. More broadly, attachment was also found to be influenced by the type of congenital anomaly of the child and whether it can be treated surgically (Yilmaz et al.). Mothers of children with CL/P had higher level of attachment than mothers of children with incurable conditions, eg, osteogenesis, hematologic disease.

Parenting

Studies addressing parenting used diverse methods, focused on different aspects of parenting and participants within the studies presented a more heterogenous group as their children quite noticeably differed in age. The children's age range across these seven studies was 3 to 18 years. Findings are therefore presented within the contexts of how parenting was explored. Due to varied methodologies across the studies the results seem mixed.

In a study specifically using Baumrind's (1971) parenting style classifications mothers of children with cleft reported more authoritarian parenting style when their child was 5 years old, especially if they had more negative affective experiences with them (Habersaat et al., 2018). This would imply lower responsiveness and higher demandingness in terms of parenting dimensions (Baumrind, 1996). However, another study with parents of older children, 7 to 9 years old (Gassling et al., 2014), assessing parent-child interactions during a task found parents of children with CL/P more encouraging and less directive. These parents promoted greater independence and autonomy in their offspring than parents of healthy children or children suffering from migraines.

No difference was found in parenting styles in terms of nurturance and restrictiveness between families with children aged 3 to 6 years old with CL/P and without (Krueckeberg and Kapp-Simon, 1993). However, within the group of parents with children with CL/P or other craniofacial differences some factors were found to impact on parenting. More visible clefts seemed to promote a more nurturant and less restrictive parenting, indicating a more authoritative style, than parents whose children did not have a visible cleft (Krueckeberg and Kapp-Simon, 1993). More nurturing parenting behaviours were also found associated with parents of children 8 to 18 years old with craniofacial differences who held more flexible gender views (Shapiro et al., 2018). Furthermore, levels of parenting stress were associated with the complexity of children's diagnoses, with a more severe diagnosis leading to higher parental stress. However, when compared to parents of children without cleft, stress levels were not statistically different (Krueckeberg and Kapp-Simon, 1993). This was explained as possibly due to the age of the children in the study, 3 to 6 years old, during which time there are usually no craniofacial surgeries being undertaken.

In-depth qualitative studies showed that despite worries about their children's health and physical safety parents actively promoted autonomy (Klein et al., 2006) and self-reliance (Klein et al., 2014), even more than in their healthy children. Participants in these studies were volunteers in a support group, which might be an acceptance-based coping strategy similar to joining online communities as highlighted by Breuning et al. (2020). They also identified protective coping strategies such as assistance at school which is consistent with proactively establishing links with school to ensure child's success (Klein et al., 2006).

Discussion

This systematic review presented evidence of maternal attachment and parenting styles of parents of children born with craniofacial anomalies. No evidence for increased risk of bonding disorders in mothers of children with CL/P was found (Tsuchiya et al., 2019). Furthermore, there seems to be a change in maternal attachment during the first year. Mothers experienced a more insecure attachment to their baby 2 months after delivery (Despars et al., 2011) but at 5 years there was no difference in attachment between mothers of children with CL/P and without (Habersaat et al., 2018). Several factors were highlighted as impacting on attachment in mothers of children with CL/P such as economic situation, the quality of relationship with own mother, whether child's condition can be treated surgically (Yilmaz et al., 2011), problems during pregnancy (Tsuchiya et al., 2019; Yilmaz et al., 2011) and perceived self-efficacy (Cinar and Koc, 2020). Although there is evidence for more authoritarian parenting style when children are 5 years old (Habersaat et al., 2018), nurturance and restrictiveness were found not to differ in another study (Krueckerberg and Kapp-Simon, 1993). Furthermore, parents of older children fostered greater independence and autonomy in their offspring (Gassling et al., 2014; Klein et al., 2006, 2014).

Findings in Context

Mothers of children with CL/P were not found at increased risk of bonding disorders in nation-wide research (Tsuchiya et al., 2019), however, the study highlighted potential antecedents such as maternal age (35 years or older) and having more children as impacting negatively if maternal depression was also present. Maternal depression itself was found in some studies more likely linked with insecure attachment in infants (Martins and Gaffan, 2000; van Ijzendoorn et al., 1992). Speltz et al. (1997) also found certain

maternal characteristics in combination with greater depressed mood associated with insecurely attached infants, however, the trend was opposite to Tsuchiya et al., ie, young age and being a first-time mother. Nevertheless, child's cleft and appearance were not found to negatively affect attachment in children (Speltz et al.), which is consistent with Tsuchiya et al.'s findings.

Maternal attachment to children with cleft seems to develop and improve during the first year (Habersaat et al., 2018), which is consistent with research into attachment of children with cleft showing changes in attachment during the first 24 months (Maris et al., 2000). It seems that as maternal attachment changes, the infant attachment changes as well or vice versa. When assessing maternal attachment at 2 months after delivery (eg, Despars et al., 2011), the infants await their first corrective surgery and mothers are likely to experience feeding problems and other complications, which might impact on their perception of their child at the time. Consistent with this are findings that the level of perceived self-efficacy in mothers affects their attachment to their child (Cinar and Koc, 2020). Feeling more able to care for their infants, mothers' attachment improved. This is supported by another study in which offering early intervention support to adapt to children's additional needs led to lower stress levels and improved perceptions of their children (Pelchat et al., 1999). Further research shows that parents of children with cleft adapt to the diagnosis and children's needs well (Stock et al., 2020), which might be reflected in the evolving attachment.

Considering the findings that maternal attachment was no different between mothers of children with and without cleft at 5 years (Habersaat et al., 2018), this seems to be mirrored in research into attachment of infants with and without cleft. No differences were found (Hoeksma et al., 1996; Speltz et al., 1997) and some researchers even found that infants with cleft were more likely to show secure attachment (Coy et al., 2002).

Several risk factors were identified to impact on maternal attachment (Boztepe et al., 2016; Yilmaz et al., 2011). These are consistent with research in attachment in general, eg, income, family size, stressful events, marital relationships (Moss et al., 2004; Nair and Murray, 2005). However, none of the studies investigated father-infant attachment, highlighting a lack of research in the field. As Stock and Rumsey (2015) reported fathers are generally underrepresented in cleft research.

Findings related to parenting were more mixed, in part reflecting the diversity of the included studies. There is some evidence that mothers of children with cleft were more authoritarian (Habersaat et al., 2018), which could be linked with protectiveness and perceived vulnerability of children (Collet and Speltz, 2007; Hlongwa and Rispel, 2018; Speltz et al., 2000). However, the remaining studies found either no differences (Krueckeberg and Kapp-Simon, 1993), or parents of children with cleft were less directive, more encouraging and fostering autonomy in their children notwithstanding concerns about their health (Gassling et al., 2014; Klein et al., 2006, 2014). Studies comparing CL/P and chronic illness (Pinquart, 2013) indicate that despite the demands and parental stress, coping with CL/P is different to chronic illnesses and parents adjust well. Parents of children with cleft reported less distress than parents of children with chronic illness such as congenital heart disease and Down syndrome (Pelchat et al., 1999). Parental overprotection associated with lower levels of autonomy in children seen in spina bifida (Holmbeck et al., 2002), type 1 diabetes and asthma (Mullins et al., 2007) might be reflective of the ongoing level of perceived vulnerability of their children.

Although parents of children with craniofacial differences seem to adjust well to the needs of their children and their diagnosis (Pinquart, 2013; Stock et al., 2020), this review highlighted changes in attachment during the first year as well as factors impacting on the development of attachment and parenting style. As secure attachment in parents is

associated with authoritative parenting style (Nanu and Nijloveanu, 2015), which is linked to greater fostering of decisional capacity in children (Partridge, 2010), it is important for clinical teams to continue to support parents throughout the whole treatment journey and be aware of potential risk factors impacting on attachment and parenting. These will play a key role when children and young people are expected to make decisions about surgeries (NHS England, 2013).

Strengths and Limitations of the Review

To our knowledge this is the first review of literature addressing parental attachment and parenting style in parents of children with CL/P. It provides a rigorous account of the research in this area to clinicians and researchers. Reflecting on parenting and its impact on fostering decisional capacity in children should inform clinicians when decisions about surgeries are being made. Reflecting on their attachment and parenting should allow clinicians to better support parents and their children accordingly. The eligibility of included studies and inter-rater checks of methodological quality were conducted by two reviewers to enhance the quality of the process. The review identified possible future research directions by highlighting a lack of studies in parent-child attachment in fathers of children with CL/P and the need for longitudinal studies within cleft in both attachment and parenting.

By addressing a broad question to capture both parental attachment and parenting, the review included several related but different concepts including bonding disorders, attachment, parenting approach and parenting style. Whilst links between these concepts are evident they are also distinct which made aspects of results challenging. Similarly, the included papers used varied methodologies which presented some challenges. While participants in some studies were specifically parents of children with CL/P compared to

healthy controls (eg, Despars et al., 2011), in other studies participants represented a broader category, eg, craniofacial (Klein et al., 2006, 2014) or congenital anomalies (Yilmaz et al., 2011), within which they were assessed. Ages of participants' children also varied and comparing parenting approach in terms of fostering autonomy in a 7-year-old would be different to an 18-year-old. These differences in children's ages or specific conditions made the participants a less homogenous sample overall and therefore conclusions across different age groups cannot be made.

The review included only research published in peer-reviewed journals and in English and therefore some studies published in different languages might have been missed. Similarly, the grey literature was not included in the search and therefore the review might reflect a publication bias.

Conclusions

This review presented that despite additional stress and burden of care that could negatively affect attachment to a child following the attachment theory (Ainsworth and Bowlby, 1991), mothers of children with cleft are not at increased risk of experiencing bonding disorders. The review found no evidence for decreased affection in mothers or behaviour towards their child. Their attachment is no different in comparison to mothers of healthy children at 5 years. However, it highlighted that maternal attachment can change during the first year of having a child with cleft and presented several factors that might impact on bonding with a child. Parenting was assessed using varied methodology and as such findings were mixed. The majority of studies reported either no difference to parenting children without cleft or fostering autonomy and self-reliance while negotiating concerns about children's health and safety. However, the review also highlighted the lack of studies exploring paternal attachment and general underrepresentation of fathers in

research as well as the need for longitudinal studies in that area. It is important for clinical teams to continue to support parents throughout the whole treatment journey and be aware of potential risk factors impacting on attachment and parenting.

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Declaration of Interest

The authors report no conflict of interest.

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Chapter 3.

Bridging Chapter

The systematic review identified and synthesised literature addressing parental attachment to children born with craniofacial anomalies and their subsequent parenting. Exploring these is important as they affect parents' care for their children and their development (Clark & Ladd, 2000). Parents form bonds to their children while dealing with initial reactions to having a child with cleft lip and/or palate such as shock, disappointment or worry (e.g., McCorkell et al., 2012; Nelson et al., 2011) as well as experiencing an increased burden of care and parental stress around issues such as difficulties feeding and preparation for surgery (Grollemund et al., 2020; Razera et al., 2017). The qualitative studies in the review (Breuning et al., 2020; Klein et al., 2006, 2014) highlighted the ongoing concerns parents carry regarding their children's health and safety. While in other long-term medical conditions parents might become overprotective (e.g., Mullins et al., 2007) some research suggests that parents of children with craniofacial anomalies seem to be aware of the need to negotiate between their worries and their children's autonomy (Klein et al., 2006, 2014). Although the parents worry about their child's safety and it might be hard for them, they encourage and foster independence in their children.

Within this context parents are required to make decisions about the surgical treatments that their children undertake. They must consider the risks of surgeries involved, their benefits as well as pain and discomfort their children will go through. For these reasons parents shared being conflicted about making decisions about surgeries (Nelson et al., 2012b) as they want their children to reach their full potential but also balance this with the risks of any procedure and pain their child is likely to experience (Nelson et al., 2012a). Research would indicate that when making treatment choices (especially for elective surgeries) for their offspring the attachment relationship between parent and child and parenting style will influence the parents' decision-making (Partridge,

2010). Authoritative parenting style was found to support the development of adolescents' capacities to become competent adult decision-makers.

A significant elective surgery, an orthognathic surgery, is offered to some young people with cleft lip and/or palate around the age of 15 (NHS England, 2013). Cleft lip and palate teams are quite unique in terms of providing care across all ages so service users do not change from paediatric services to adult services unlike in other long-term conditions (Ludvigsen et al., 2021). Moving from paediatric to adult services illustrates a tangible change and expectation that the young person will be responsible for their healthcare decisions. The orthognathic pathway is therefore considered to represent that transition from paediatrics in cleft lip and/or palate. The young person, rather than their parents, is expected to take on the responsibility for the decision about their forthcoming treatment (NHS England, 2013). However, after many years of holding the responsibility for treatment decisions, parents might find it difficult to let go, as seen in other health conditions (Betz et al., 2015). The next chapter presents an empirical piece of research that explores parents' subjective accounts of the transition in cleft lip and palate and the changing decision-making process.

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Chapter 4.

Empirical Paper

Prepared for submission to The Cleft Palate-Craniofacial Journal (Appendix A).

Stepping back: Parents' experiences of the decision-making process for elective orthognathic surgery in cleft lip and palate (An Interpretative Phenomenological Analysis)

Jana Safarikova[1], Dr Eliane Young[2], Kiki Mastroyannopoulou[1] and Dr Paul Fisher[1]

[1]*Department of Clinical Psychology and Psychological Therapies, University of East Anglia, UK*

[2]*Cambridge University Hospitals and Cambridgeshire and Peterborough NHS Foundation Trust, Cambridge, UK*

Corresponding author:

Jana Safarikova, Department of Clinical Psychology and Psychological Therapies, Norwich Medical School, University of East Anglia, Norwich NR4 7TJ. E-mail. j.safarikova@uea.ac.uk

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Abstract

Objective: The current study explored parents' experiences of decision-making about an elective orthognathic surgery in cleft lip and/or palate (CL/P).

Design: A qualitative design was used. Semi-structured interviews were conducted with 11 participants using online videoconferencing platforms. The data were transcribed and analysed using interpretative phenomenological analysis (IPA).

Settings: Participants were recruited via Cleft Lip and Palate Association (CLAPA) and from a Cleft lip and palate team in the National Health Service (NHS).

Participants: Participants were parents of young people who made a decision whether to undergo the surgery and were either in the preparatory phase awaiting it, or not if they decided against it. Participants were 6 females and 5 males, aged 41 to 60 years.

Results: Three main themes were identified. '"Our" journey', relating to participants' involvement in the cleft process, 'Stepping back' as the responsibility for the decision-making shifted to their children and 'Helping with the bigger picture' while adjusting to their new role.

Conclusions: Participants were invested in the cleft journey and stepping back comprised a spectrum of experiences from a relief to a shock and upset. Participants went through sometimes difficult negotiating of their ongoing involvement while supporting their child. The study highlighted the need to explore parents' understanding of the process and forthcoming changes prior to offering the orthognathic pathway. Clinicians can support parents to recognise the complexities involved, especially if young people are making decisions before reaching adulthood.

Keywords

Cleft, parent, decision-making, orthognathic surgery, qualitative, interpretative phenomenological analysis

Introduction

Approximately 1 in 700 babies in the United Kingdom (UK) is born with a cleft lip and/or palate (CL/P), the most common congenital conditions of the face and oral cavity (Goodacre and Swan, 2008, 2012). It develops in early pregnancy and the causes are considered complex, a combination of environmental and genetic factors that cannot be predicted (NHS England, 2013). Despite this, parents often feel guilty and blame themselves for having done something wrong during pregnancy and causing their child's condition (Nelson et al., 2011; Zeytinoglu et al., 2016). Receiving a diagnosis was reported by parents as a shock (McCorkell et al., 2012), an emotional and traumatic experience and parents can be left feeling overwhelmed with information (Costa et al., 2019). While parents can be elated at birth, they also experience a sense of loss, grief and mourning (Beaumont, 2006; Nelson et al., 2011) and perceive their children to be more vulnerable than children without cleft (Hlongwa and Rispel, 2018).

Cleft lip and/or palate impacts on the child's feeding, breathing, hearing as well as speech, facial development and appearance. The National Health Service (NHS) offers specialist cleft lip and palate services across the UK with an approximately 20-year care pathway involving several surgeries starting at 3 months of child's age (Cleft Lip and Palate Association, 2019; Colbert et al., 2015; Goodacre and Swan, 2008, 2012; NHS England, 2013). The long-term cleft treatment with numerous appointments and surgeries are experienced by parents as a burden of care with ongoing emotional impacts (Breuning et al., 2020; Hlongwa and Rispel, 2018; Maurien et al., 2019; Stock and Feragen, 2016).

From around the age of 15 years old, some young people are offered an elective, orthognathic pathway. It is a combination of orthodontic and surgical treatment offered for functional and/or aesthetic reasons with the orthodontic part taking a few years before the actual surgery (Royal College of Surgeons of England, 2013). During the orthognathic

surgery, also known as corrective jaw surgery, the jaws and chin can be realigned to help with biting, and nose structure and nasal pathways can be corrected to improve breathing or speech. Further surgeries such as lip or nose revision can also be considered afterwards. This orthognathic surgery can dramatically alter the appearance of the young person and therefore can have a significant impact on their self-image (Alansari et al., 2014; Tevik and Feragen, 2014) and identity (Cadogan and Bennum, 2011; Liddle et al., 2018). However, when making a decision about this surgery, young people do not know what their new appearance might be (Liddle et al., 2018), making it perhaps more difficult to decide.

This complex elective surgery represents the first time that young people themselves are expected to be the main decision-makers. This is often also the first time in a young person's pathway where the treatment is optional, given that this surgery aims to improve their condition and functioning rather than being critical to their survival. Up until this point parents mainly hold responsibility for making decisions (Nelson et al., 2012a). Parents usually consider all previous surgeries as necessary, whether for functional or aesthetic reasons, and explain their pro-treatment stance as not wanting to fail their children by declining offered cleft treatments.

Cleft and palate services do not divide into paediatric and adult care unlike other long-term health conditions such as diabetes or cardiology (Ludvigsen et al., 2021). As there is no obvious change into adult services, this surgery and associated decision-making represents such a shift and therefore is a significant transition time for the young people as well as their parents. Young people need to consider the treatment, seriousness of the surgery, timing, recovery and outcome with its impacts on their appearance and identity (Liddle et al., 2018; Nelson et al., 2012a).

Similarly, their parents are at a transition point when their children are developing independence and are expected to take on responsibility for their care (Heath et al., 2017;

Miller, 2009). The parent-child relationship undergoes a complex realignment as their roles change and parents can find it difficult to 'let go' (Betz et al., 2015). Letting go can be problematic for some parents of healthy children in emerging adulthood, ie, 18-25 years, as well (Kloep and Hendry, 2010). However, the difficulties for parents of children with cleft might be intensified by the fact that the decision is often made or required while the young person is still in their teens, not legally an adult. The shift of responsibility for the decision might therefore come earlier than expected. Furthermore, some parents might also feel very protective of their children whom they consider more vulnerable due to their condition (Coy et al., 2002).

Given the importance of this transition the decision-making process experienced by young people was recently explored (Acum, 2018), which highlighted the influence of parents' and professionals' values and motivations when supporting young people to make decisions. Another qualitative study exploring young people's experiences prior and following orthognathic surgery (Liddle et al., 2018) identified the influence of significant others such as parents. Young people felt that parents' views were particularly important, with parents being usually encouraging towards the surgery.

Research found that throughout the treatment journey, parents seem to be conflicted about surgeries (Nelson et al., 2012b). They saw operations as a means for their child to get closer to 'normality' while they also considered the risks, discomfort and stress associated with them. They understood operations as a 'necessary evil' and felt a 'moral obligation to be "good" parents', wanting their children to achieve their full potential (Goodacre and Swan, 2008, 2012; Nelson et al., 2012a). It transpired that more support for parents themselves was needed during the treatment journey to address their emotional and social well-being (Nelson et al., 2012b). Communication skills and use of language by

clinical teams were also found to be important factors (Alansari et al., 2014; Beaune et al., 2004; Myhre et al., 2019).

The above literature addressed parents' experiences of caring for their child throughout the whole cleft journey. The significance of the orthognathic surgery and the associated transition in decision-making has been recognised and researched with young people. However, parents' experiences and needs are missing (Nelson et al., 2011). The aim of the current study was to therefore address this gap in research and explore parents' experiences of this particular stage of the cleft journey.

Methods

A qualitative methodology was chosen to explore parents' experiences of the decision-making process regarding orthognathic surgery. Semi-structured interviews were transcribed and analysed using interpretative phenomenological analysis (IPA), which is a prominent method in health psychology (Smith et al., 2009). IPA was chosen as the most appropriate approach to explore in depth how each participant made sense of their experience (Larkin and Thompson, 2012). IPA is interested in how individual people relate to the world through the meaning-making process and exploring their 'lifeworld' (Smith and Osborn, 2008), which is in line with person-centred care promoted in the NHS (Reid et al., 2005).

This study received the required approvals from the University of East Anglia, NHS research ethics committee and Health Research Authority (HRA, Appendix C).

Participants and Recruitment

Participants represented a purposive sample and were parents of young people born with cleft lip/palate who have been through the decision-making process regarding the

orthognathic surgery in the last 5 years. If the decision was in favour of the operation, their child had not undergone the surgery at the time that they participated in the study. This was to avoid the outcome of the surgery impacting on the memory or experience of the decision-making process. Participants were required to have a good understanding and use of English language.

Six females and 5 males took part. Eight of them represented 4 heterosexual married couples. As each participant in these married dyads was considered to have a different experience of the phenomenon in question based on IPA (Smith et al., 2009), all participants were interviewed individually to gain insight into their personal experience. All participants were White British with the exception of 1 participant who was Asian/Asian British. Their age range was 41 to 60 years. In terms of surgeries they have been through with their child prior to the orthognathic surgery, the number varied from 2 (cleft related only) to 8 (6 cleft related). Their children's age when the decision was made was between 14 to 21. Table 1, using pseudonyms, presents a more detailed overview.

Table 1. Participants' Characteristics.

Pseudonym	Gender	Age	Number of child's prior surgeries	Age of child when decision made
Emma	Female	46-50	2	16-17
Peter	Male	51-55	2	16-17
Joanne	Female	45-49	5	16-17
Pat	Female	46-50	4	14-15
Sophie	Female	41-45	4	16-17
Adam	Male	56-60	3	14-15
Eve	Female	51-55	3	14-15
Andrew	Male	51-55	5-6	16-17
Daniel	Male	56-60	4-5	20-21
Mary	Female	46-50	7-8	16-17
Richard	Male	46-50	7-8	16-17

Participants were recruited via advertisement through the Cleft Lip and Palate Association (CLAPA) and from a Cleft lip and palate team in the National Health Service (NHS). Participants who engaged via CLAPA contacted the lead researcher directly. The NHS clinical team identified potential participants, shared the study details with them and obtained consent to be contacted by the lead researcher. Two initially identified participants were subsequently not interested in the study.

Procedure

The lead researcher initially contacted all prospective participants by phone, explained what the study involved, this included discussions about confidentiality and anonymity in

accordance with the General Data Protection Regulation (UK Government, 2018). Prospective participants were then sent the Participant Information Sheet (Appendix D), consent form (Appendix E) and demographic information form (Appendix F) by email and arrangements for the interviews were made.

Due to Covid-19 restrictions all interviews were conducted via online video platforms and ranged in duration from 45 to 90 minutes. All participants confirmed their consent to take part in the study. Interviews were audio and video recorded to ensure the best quality sound. Participants were interviewed during a single appointment in a semi-structured manner using a Topic guide (Appendix G). All participants were sent an electronic shopping voucher worth £10 after the interview as a 'thank you' for their time.

Analysis

Each interview audio recording was transcribed by the researcher into a Word document and analysed before moving to the next one (Smith et al., 2009). To ensure familiarity with each transcript, the process started with reading and re-reading of the individual accounts followed by making initial notes on the descriptions, use of language and concepts.

Initial exploratory comments and interpretations developed into emergent themes and further into superordinate themes within each transcript (Smith et at., 2009). Table 2 offers an example of the analytic process leading to 1 main theme.

Table 2. Example of the Analytic Process from a Transcript to a Main Theme.

Quote	Coding	Emergent theme	Superordinate theme within transcript	Main theme
"I just think however old your kids get, as a parent... Um, I don't know. It's hard to take that step back. It's just hard, because you- from day one, you know, you have made that decision, it's really, really hard to take that step back, really hard."	Her role as a mother is changing. She is expected to hand over the responsibility to her child but it doesn't come naturally, it's not a welcome change, it's hard for her to "take that step back". Stepping back is as if her 'mother' identity is lessened?	Stepping back is really hard, not a natural process.	Natural process (*not*).	Stepping back

Once all transcripts were analysed, patterns across them were sought. Themes present in at least 5 transcripts were selected to form superordinate themes (Smith et al., 2009). To ensure the quality and validity of the themes, the transcripts and quotes for each superordinate theme were checked back for evidence. As Smith and colleagues (2009) further suggest, themes were consulted with the supervisor who also conducted a mini audit from the first transcripts, annotations, codes and themes as well as the structure and evidence of themes across.

Reflexivity

IPA requires researchers to have a reflective stance, to bracket themselves from the participants' experiences (Larkin and Thompson, 2012). The lead researcher was a female, not a parent herself. As a Trainee Clinical Psychologist, the researcher had 6 years' experience in health research and/or clinical practice at the time of the interviews. The researcher was not part of the Cleft lip and palate team but engaged closely with the clinical staff to research and gain understanding of the cleft pathway and the orthognathic surgery. A reflective journal was kept allowing the researcher to note thoughts and reflections, and supervision took place throughout this piece of research.

Results

Three main themes arose from participants' accounts with 6 subthemes as presented in Table 3. The first main theme '"Our" journey' relates to the participants being part of the cleft journey along their child from the start. It was this context that framed how they experienced the decision-making related to the orthognathic surgery. The second theme 'Stepping back' captures the experience of handing over the responsibility for the decision to their child. The third theme 'Helping with the bigger picture' reflects participants' new role and their negotiation of it.

Table 3. Main Themes and Subthemes.

Main theme	Subtheme
"Our" journey	Through it together
	Nearly at the end
Stepping back	A natural process (*or not*)
	I'm not the one going through it
Helping with the bigger picture	There to give advice and guidance
	Aware of my influence and motives

"Our" Journey

This theme encapsulates participants' involvement in the cleft journey that often started before their child was born, attending appointments with clinicians and making decisions about surgeries and care. Seven participant accounts were drawn on in developing this theme.

> You go through initially where you're responsible for everything and you are doing all the appointments in the diary and it's such a big part of your life. (Joanne)

Joanne highlights the intensity of going through the cleft journey, especially in the beginning. In "it's such a big part of your life" she conveys the meaning of being a key figure in quite an overwhelming process. Furthermore, she uses the present tense which might also reflect how intense and lasting the engulfing feeling is for her, yet distancing herself from it by talking in the second person.

Subtheme: Through it together

The transition of the decision-making process moves the responsibility for the decision to the young people, yet participants still felt that this was their journey. They talked about the cleft journey and the surgery as much as theirs as their children's, with a united voice and a sense of togetherness.

> ...after she was 14 years old, this particular operation suddenly became a possibility for us. (Adam)

Adam talks about "a possibility for us", not for his daughter. This reflects the togetherness, the participation of the whole family in the cleft journey.

> They're gonna have to obviously wire her jaw together for a little bit so, yeah, it's gonna be, it's gonna be fun. We'll get through it together. (Emma)

Emma considers her role in the ongoing journey thinking about the surgery. She uses dark humour in recognition of what awaits: her daughter having her jaw wired, being in pain and discomfort.

> I think there is a part of you as an adult that kind of oh, your stomach almost flips a bit as a kind of like oh, gosh, you know that means that we've got to go through this surgery altogether. Because it is just such a big, I feel it's a big thing. (Eve)

Eve reflects on being part of the surgery. Her use of a metaphor that closely mirrors her physical reaction upon hearing her daughter's confirmation that she wants to undergo

the surgery, portrays the complex position of her as a mother, thinking about the scope of the surgery.

Subtheme: Nearly at the end

As the surgery usually takes place around the young person's age of 20, the treatment pathway is perceived as a long one. The orthognathic surgery represents the end goal of this pathway following years of orthodontic treatment. However, the orthognathic surgery does not always mean the very end as other corrections might be suggested and required afterwards. This was found helpful by some participants but surprising by others.

> It's a long journey … It seems like yesterday he was born and then you think you're nearly at the end of that journey. Um, but, after maybe two, possibly three more [surgeries], then the journey is complete. (Sophie)

Sophie talks about the journey as a long one, yet going quickly, being near the end. She refers to 2 more possible surgeries after the orthognathic one as an expected process.

Joanne saw the orthognathic surgery as the end of the cleft pathway that could not arrive early enough. This was connected to her feeling of being "on a conveyor belt of treatment" that she wanted to finish. However, the 'end' seemed to be pushed further and further away, which Joanne found difficult.

> You feel you're kind of going down this conveyor belt … I just wanted it all done out of the way … What is this magical goal where everything is complete? 'cause, 'cause then they started talking about and you might need this doing or you might

> need a further revision or this or that. There's not like a fixed line in the sand, that this will be finished because it won't be. (Joanne)

Stepping Back

This theme captures the experience of realising that after years of being responsible for all decisions on behalf of their children participants are expected to hand the responsibility for making a decision about the orthognathic surgery over to their children although they might not be 18 years old yet. All participants shared their experience of stepping back, the impact it made, highlighting a spectrum of experiences and different levels of being comfortable with the change.

Subtheme: A natural process (*or not*)

Four participants understood the shift of responsibility for the decision to their child as a natural process reflecting the developmental stage of their child as well as what the surgery entails and therefore the need for their child to decide themselves. Participants felt that their children reached an age when they were able to consider implications, pros and cons of having or not having the surgery and make a decision that would be right for them.

> As a parent, you feel proud because your child is now taking the ownership for his or her life ahead and deciding this is what I want to do and I'm taking ownership of that decision … To me it was the right thing to do, let go of the reins, because she has to live with her choice and she has to be happy with the choice. (Daniel)

Daniel's metaphor "let go of the reins" represented a proud moment for him, seeing his daughter as a mature young woman.

However, 3 participants described the process of handing over responsibility as difficult or surprising. Joanne talks about a need to adapt.

> It's quite difficult, that period of adjustment, to be in charge of everything and to be making all the decisions and then suddenly at this one it's like, oh no, now it's over to your son. It would help to have a bit more of a sliding scale. (Joanne)

Similarly, Peter describes a stunned surprise.

> They suddenly hit us with it in a roundabout way. To say, this has to be decision that your daughter has to make. And it's up to your daughter to decide if she wants the op. And I mean, it's her decision only. It's nothing to do with what would anybody else- … It's just a fact of life. She's growing up. She has to make the decision. There's got to be a time when you let go of the of the purse strings, isn't it? (Peter)

Peter describes the moment using a metaphor "they hit us" signifying an unexpected surprise. Yet, on reflection he comes to a new understanding and accepting the shift as "a fact of life", and further normalises the timing of it by another metaphor "let go of the purse strings".

Sophie found it incredibly hard and upsetting that her son was to make the decision before he reached the age of 18. In her understanding of being a mother, she felt she should be responsible for all decisions, more notably voice them until her son reached adulthood.

This seems greatly embedded in her identity as a mother, that is very important to her, so the news came as a shock.

> …the shock of that being announced … It was like somebody had taken my voice away from me … It sounds selfish, I just felt that I was no longer needed as mum. Yeah, the only way I can explain it, really. (Sophie)

In contrast, 2 participants felt a welcome relief when it was suggested to step back. The previous ownership of decisions for the participants was laden with worries whether they were making the right decision for their children. They appreciated that the young people had a voice and could decide for themselves.

> It sounds really awful, but in a way it was a little bit of a relief. Because it's really hard- Yeah, in a way it was a little bit of a relief because as a parent with a child that needs treatment it's all on you. I've got to make this decision. What if I make the wrong one? … It was no longer my right or privilege to do that for her … It was always going to be her decision. It wasn't something that we actively had to make ourselves think: Oh, she's going to make this decision. We knew that she would make it for herself and I don't think it's anything to do with the cleft. I think she's just quite a determined personality. (Pat)

Pat describes her worry associated with potentially making the wrong decision for her daughter yet talks about her daughter being "a determined personality" who always made her decisions. This seems to make stepping back easier for Pat.

> In some ways it's a bit of a relief to know that you don't have to make that decision completely on your own, and that your daughter is now old enough to make the decision with you. (Mary)

Mary considers the orthognathic surgery a major, complex procedure with significant impact on her daughter. She is therefore relieved that she no longer has the responsibility for the decision.

Adam understood the shift as natural but something that was to happen after the operation, not at the time of his daughter making a decision.

> Once that big operation is out of the way we can start taking a step back and because she's now an adult and she can take all those decisions for herself, the shift from us to her is slowly happening. I'm sure that after her major- after her jaw surgery the shift is gonna be 75% her and 25% us. We shall see. (Adam)

Subtheme: I'm not the one going through it

Eight participants shared a strong feeling that the decision had to be down to their child due to the procedure itself, the pain and discomfort involved, the recovery time and the unknown impact on their appearance as they were "going into it blind".

Six participants explained that it was not their place to make the decision as they were not the ones going through the surgery.

> She's ultimately the one who has to have the surgery. It's not me that's going through recovery. It's not me who is not gonna be able to eat for two weeks. It's not. It's her. (Emma)

Andrew felt that if his daughter was aware of the procedure and what it involved, despite perhaps his preference, it was not his place to try and dissuade her. He respected her decision.

> When they're describing shaving bits of bone off her hip to insert into her gum, there's always that horrible cold stomach feeling … So if she can go through that and not worry about it and decide to go a step further, who are we to argue with her? (Andrew)

Helping with the Bigger Picture

Stepping back did not represent withdrawing from the decision-making process altogether. Participants were adapting to their new position and a role they could and/or should play going forward. All 11 participants were involved in supporting the young person whilst making their decision.

Subtheme: There to give advice and guidance

Participants reflected on the developmental stage and age of their child and ability to decide themselves, how significant the surgery was and/or what a big decision it represented. Participants talked about having discussions with their children, exploring advantages and disadvantages of the surgery, impact on their future plans, some shared their preference with their child and for others it was important not to, to avoid influencing

their decision. They offered advice, support as well as space for the young people to establish what they wanted.

> It's something that you know as far as we're concerned, this decision that *she*'s gotta make and she's gotta be 100% comfortable with. We would review the facts, go over everything, and then just yeah, and respect, respect the decision she makes. We were just talking to her, helping, supporting, advising. (Richard)

The age of participants' children differed and Eve in particular highlighted the influences young people are under.

> They're making this decision during a time when they're very influenced by lots of external factors. I'm beginning to wonder whether they make those decisions based on the right reasons. (Eve)

Eve acknowledged the influence of media and portrayal of perfect-looking faces and questioned whether making this decision when her daughter was perhaps 25 would bring a different outcome. Although Eve trusted her daughter's judgement and knew she made the right decision for herself at that time, she wondered about the "right reasons" for the surgery at any particular time.

Two participants also highlighted that using humour was important in the family dynamics when talking about the surgery with their child. Joanne in particular.

> There were lots of conversations going on and you know lots of, lots of discussions around it. Would he wanna wear braces if he went to university and he didn't think that would do much for his attractiveness to women (laugh) and all these conversations … And a lot of it was like good humoured so there was a lot of laughter going on. (Joanne)

Subtheme: Aware of my influence and motives

This subtheme was linked with participants' awareness of their influence on their children. Participants reflected on their position and whether they supported the young people objectively, or consciously tried to influence them. Some participants talked about how their worries of pain and potential disadvantages of the surgery might translate in conversations and influence their children. Two participants made a conscious effort not to share their worries with their children.

> If I thought about it just as a mum and that's my child, yeah, if you don't have to have it done, don't do it. But that's, that's my view and I was determined that I wasn't going to let her know that that's perhaps how I felt, because that's not fair … I can't put my worries onto her. (Pat)

> You don't want to try and influence her decision by putting your worries too far to the front. (Mary)

Both Pat and Mary felt it unfair sharing their worries and influencing their daughters. They did not consider it a supportive and helpful role.

Pain seems to be the overarching theme in Andrew's account. Based on previous operations and seeing his daughter in pain that he could not take away, the idea of the orthognathic surgery seems rather difficult. He uses generalisation to other parents to normalise his concerns about his daughter being in pain, and so did 3 other participants.

> [The surgery] sounds like some sort of medieval torture really, doesn't it? … At the back of my mind is how much it will be hurting. So no, I myself, I wouldn't wish the pain (pressured outbreath) on anybody … I'm sure most people don't want to see their children in pain, do they? (Andrew)

Whereas for some participants it was crucial not to influence their child's decision, 2 participants, a married couple, felt that as parents who loved their daughter, knew her and her needs, they also knew what was best for her. However, they were also aware that it was their daughter's decision and she needed time and space to feel comfortable making it.

> If she was completely against it because she was too scared, you have to respect it, but my initial things were I'm going to do what I can to put my point across to her because I know it would be for the best … As parents we see a bigger picture… You have to convince her to try and see, see this bigger picture which as 16-, 17-year-olds or now 18 doesn't always see. (Peter)

Peter acknowledges the developmental stage of his daughter and the need to support her around the complexity of the decision, to see "the bigger picture". Nevertheless, he is open about his determination to influence her decision-making and "convince her" if he can.

Discussion

This study aimed to gain a greater understanding of parents' experiences of their involvement in decision-making about an orthognathic surgery for their child. A greater insight into their experiences would help better understand the support needs parents have during this transitional period.

Three main themes captured parents' involvement in the cleft journey and changes associated with the orthognathic surgery: '"Our" journey', 'Stepping back' and 'Helping with the bigger picture'.

It became clear that participants were invested in the treatment pathway after many years of holding responsibility for their child's healthcare needs, considering it also their own journey. This journey reflected a significant burden for both the young people and the parents (Breuning et al., 2020; Hlongwa and Rispel, 2018; Nelson et al., 2011; Stock and Feragen, 2016). However, as the end stage of the treatment was approaching, they found out that it was no longer their place to decide about it.

Being told about the transition in decision-making moving to the young people generated a spectrum of experiences in participants from a relief (eg, Pat) to a shock and upset (Sophie). Even if it was a welcome change for the participants, they were facing a complex situation. They still had their worries and preferences, wanted their child to make the right decision for themselves, while being supportive and respectful of the young person's choice. If the shift came unexpectedly, it took time to adjust to the idea of transition, to consider their new position and come to terms with it. This adjustment was evident in Peter's reflection on the transition. Coy and colleagues (2002) found extraordinary protectiveness in some mothers of children with CL/P, which might have been at play for some female participants in the current study. Sophie talked about the

impact of the transition on her role as a mother. The experience of stepping back and knowing they had to, was influenced by the degree to which participants felt this shift being a natural process (or not). As others (Betz et al., 2015) addressing transition in long-term conditions found, some parents struggled with not being responsible and accountable for their child's care and had difficulty letting go.

Control and protectiveness are characteristics of the parenting dimensions (Baumrind, 1996) of demandingness and responsiveness. Demandingness is portrayed by the extent of regulating a child's behaviour and/or expecting a child to control their own behaviour, ie, having a varying level of control over the child's actions. Responsiveness represents the extent of fostering individuality in the child, showing warmth, supporting the child's autonomy and being responsive to their communication. This is particularly evident in the second and third themes where the participants let the young person decide about the surgery while providing guidance and support. Whether participants initially felt they wanted to be the ones making the decision or not, they accepted that it had to be a decision made by their child considering what the surgery involved. The extent of demandingness and responsiveness adjusted through the process of transition. However, this highlighted a conflicting experience some participants had, yet not necessarily shared with clinicians and others at the time. It is therefore important that clinicians are aware of the potential internal conflict parents might go through around this surgery and explore it with them. Checking parents' understanding of the process and exploring their experience of the journey as well as preparing them for the forthcoming changes is important as it will shape their expectations and give them time to adjust to the transition. This corresponds with the delivery of person-centred care in the NHS (Department of Health and Social Care, 2021).

A key factor in terms of stepping back might have been the age at which the young people decided about the orthognathic pathway. Daniel talked about feeling proud of his daughter making the decision, however, she was in the oldest group. In contrast, Eve highlighted influences on young people in the lowest age group and the reasons for their decision. The developing brain undergoes changes linked with decision-making between 15 and 20 years (eg, Partridge, 2010), especially in the capacity to consider long-term implications or risk. Research with young people undergoing the orthognathic surgery identified support with decision-making as a key need (Acum, 2018; Liddle et al., 2018). This is worth taking into account not only from the young person's perceptive but how their age might impact on parents' readiness to step back as in the current study.

Participants often used metaphors to convey their message and illustrate their experience, several of them depicted in the quotes. Talking particularly about the orthognathic surgery, they used metaphors such as a medieval torture (Andrew), a feeling that their stomach flips (Eve). Such metaphors were very emotive, striking, helping the researchers to understand the participants' meaning-making (Smith et al., 2009) and the impact on them. While these intense thoughts and feelings were happening within the participants, they did not express them, or their internal conflict, in front of their children or clinicians. This means that clinicians are not aware of what is going on within parents.

Methodological Considerations

Due to COVID-19 restrictions, the study was conducted online using videoconferencing platforms, which is considered an effective alternative to face-to-face research with many advantages (Archibald et al., 2019; Bolderston, 2012; Irani, 2019). Participants were in their own homes, there was no need to travel, which decreased the burden on participants. However, ensuring privacy was crucial as other family members were sometimes present.

For example, a participant had to change rooms to continue to speak freely without being overheard when their child arrived home during our interview.

A strength of this study was addressing experiences of both mothers and fathers (6 and 5 respectively), the latter being underrepresented in cleft research (Nelson et al., 2011). Parents of children who decided for as well as against the surgery were encouraged to take part. In this study only 1 participant's child decided against the surgery in comparison to 6 young people who were awaiting the procedure. The experiences of this 1 parent contributed to all resulting themes, adding to their breadth.

This was a cross-sectional study capturing one point in time. Longitudinal research starting when the orthognathic pathway is offered until the end of the surgery with both the young people and their parents would offer greater insight into their experiences over time.

Clinical Implications

The study found that the cleft journey represented a significant part of parents' lives. Their understanding and experience of the changing role surrounding the orthognathic pathway varied. The experience of the transition seemed to be especially difficult for participants whose child was not an adult yet. They were used to making decisions about care throughout the cleft journey and expected to do so until their child reached adulthood. Some felt it was part of their parental role. This sudden change to parental expectations can therefore challenge their sense of themselves and their parental role and make it more difficult to let go.

To support parents with this complex transition it is suggested to communicate with them about the end stages, including the decision-making processes around the orthognathic pathway in the years before this point is reached. It is important to highlight that their children are likely to still be underage at the time this decision point is reached.

Parents also need to be aware that their involvement will still be welcomed and required to support their child with the decision. Explaining this to parents earlier on, rather than when presenting the opportunity of the orthognathic pathway, might help shape expectations and prevent feelings of stunned surprise.

Furthermore, clinicians can support parents in recognising the complexity and importance of the transition in the decision-making, their expectations around it and being sensitive to the parents' potential internal conflicts around these changes. These approaches could serve to better prepare parents for their final part of the orthognathic journey.

Future Research

Future research can build on the current findings by using a longitudinal approach to explore parents' and their children's experiences over time, starting when the orthognathic pathway is offered until after the surgery. Research can also focus specifically on experiences of parents whose children decided against the surgery, considering that only 1 participant in this study represented that population. Furthermore, research could explore clinicians' experiences of the orthognathic pathway as their views are currently missing from the literature.

Conclusions

Exploring parents' experience of the transition in cleft pathway when they are expected to hand over responsibility for decision-making about an orthognathic surgery to their child brought to light a complex picture. Participants were invested in the cleft journey over many years and stepping back was represented by a spectrum of experiences, from a welcome change and a relief to a shock and upset. Participants shared about sometimes

difficult negotiation of their ongoing involvement when supporting their child to make the right decision for themselves. The study highlighted the need for parents' understanding of the process and forthcoming changes to be explored prior to the appointment during which the orthognathic pathway is offered. Exploring their experiences of the journey and helping them to recognise the complexities involved in stepping back, especially that their children might be in middle teenage years, should better prepare parents for the transition. Future research could address clinicians' perspective on decision-making about the orthognathic pathway as it is missing.

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Declaration of Interest

The authors report no conflict of interest.

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Chapter 5.

Discussion and Critical Evaluation

This chapter presents an overall discussion of the findings from both the systematic review and empirical paper, addresses their strengths and limitations, and suggests directions for future research. It also offers a reflection on the thesis process.

Overview of results

Systematic review

The systematic review synthesised literature on parent-child attachment and parenting style. Twelve studies were included in the review, nine quantitative and three qualitative, and assessed for methodological quality and risk of bias.

Only maternal attachment, not paternal, was addressed in the studies included in the review. Attachment seems to undergo changes during the first year of having a baby with cleft lip and/or palate while parents adjust to the additional needs of their children (Habersaat et al., 2018). However, no differences in attachment were found at five years in comparison to healthy children. This is consistent with research in attachment of infants with the condition (Maris et al., 2000) where changes in attachment were found during the first two years. Other studies also support that there are no differences in infant attachment in comparison to children without cleft (Hoeksma et al., 1996; Speltz et al., 1997). Although parents were protective of their children with craniofacial anomalies, overprotectiveness seen in other medical conditions or chronic diseases such as type 1 diabetes, asthma (Mullins et al., 2007) or cancer (Colletti et al., 2008) was not found in this review. Despite worries and concerns about children's physical health and safety parents fostered autonomy (Gassling et al., 2014; Klein et al., 2006) and self-reliance (Klein et al., 2014) in children seven to 14 years old. This review identified several factors that can influence maternal attachment such as perceived level of self-efficacy (Cinar & Koc, 2020)

which is supported by other research (Pelchat et al., 1999), family size, income and stressful events, also found in other studies (Moss et al., 2004; Nair & Murray, 2005).

Empirical paper

The empirical study focused on parents' experiences of a decision-making process about an elective surgery during a transition when the responsibility for the decision moves from the parents to their children. A qualitative method of semi-structured interviews was used to allow for in-depth exploration of participants' experiences and data were analysed employing an interpretative phenomenological analysis.

The empirical paper identified three main themes. '"Our" journey', 'Stepping back' and 'Helping with the bigger picture' reflected parents' involvement over many years of being in charge of their children's healthcare decisions. In the theme '"Our" journey' participants talked about the burden of the cleft treatment in terms of the previous surgeries their child had experienced and the many varied appointments, worries and concerns they had experienced over the years as are documented in other literature (Breuning et al., 2020; Hlongwa & Rispel, 2018; Nelson et al., 2011; Stock & Feragen, 2016). The second theme highlighted a whole spectrum of experiences when asked to step back, from a relief of not making the decision to a shock. Difficulties in letting go of the responsibility for decisions is consistent with evidence in other long-term conditions (Betz et al., 2015). The last theme captured parents' new role during the changing process and their ongoing support. Participants considered the orthognathic surgery to be a big decision requiring a thorough deliberation and assisted their children in thinking about the impact of the surgery, timing and implications for the future, a breadth of aspects that the young people might not necessarily consider in middle adolescence themselves (Partridge, 2010). The need for parents to support their children during the decision-making process was also acknowledged and appreciated by young people in other studies (e.g., Liddle et al., 2018).

Critical appraisal of the research

Systematic review

The completed systematic review had a number of strengths. One strength was related to consultations with an experienced academic librarian to ensure the most appropriate search strategy in terms of keywords and databases. Another strength was registering the review protocol on PROSPERO, an international prospective register of systematic reviews, which is considered a good research practice as it supports research transparency and avoids potential duplication (Stewart et al., 2012). A further strength was the involvement of a second reviewer to ensure a robust process (Charrois, 2015; Moher et al., 2009). The second reviewer assisted with the selection of papers included in the review and conducted inter-rater checks of methodological quality of 25% of studies in the review. Another strength was synthesising available evidence on the topic which has not been done before, offering a helpful summary for clinicians.

There are a number of potential limitations and areas for future development with the current review. The main limitation is that addressing attachment and especially parenting presented the lead researcher with varied concepts linked to the phenomena such as bonding disorders (decreased maternal affection and behaviour to their child), attachment, parenting style, parenting dimensions, etc. Different studies based around particular concepts naturally used varied methodology and population sampling, it was therefore not possible to compare studies at the level of measures used and this impacts on the ability of any review to generate generalisable conclusions. Due to the lack of homogeneity across the included papers it was more difficult to synthesise measures and findings which presented a challenge. This was dealt with by focusing on the findings within each phenomenon and population groups and exploring similarities and differences across them (Popay et al., 2006). Findings should be understood only within the

populations they were studied in and not generalised beyond the age groups of children whose parents were involved or to other sociodemographic groups. For example, generalisations relating to maternal attachment beyond child's age of five years should be avoided as no research addressed maternal attachment to older children in the review.

Empirical paper

The empirical paper explored parents' experiences of decision-making about a specific elective surgery that represents a significant time during the treatment pathway of their children. It adds a new perspective to the knowledge in the area that has been explored with the young people themselves (e.g., Liddle et al., 2018), however, parents' experiences of this were missing. Another strength was the number of participants in the study. With eleven participants, which is at the higher end of the recommended number, it offered a more multifaceted account of experiences (Reid et al., 2005). Additionally, both mothers and fathers took part, with the latter being underrepresented in research (Nelson et al., 2011).

A further strength was the level of engagement of the lead researcher from the initial stages of the study when preparing a protocol throughout. The researcher closely liaised with the clinical team in the NHS to learn about the care provision as well as in preparation of study materials. Drawing on Patient and Public Involvement in research (National Institute for Health Research, 2014) study documents were sent to parents of service users who provided valuable feedback that was used in finalising the documents prior to the study approval process. A pilot interview was also performed with a fellow Trainee Clinical Psychologist to ensure a smooth process when conducting research with participants. This allowed testing of online technologies and highlighted a need for study documents to be amended to reflect NHS and non-NHS participants, which was submitted

as a non-substantial amendment to the research authorities. Conducting the pilot therefore proved invaluable.

Another strength is related to the researchers' commitment to the quality of the study. Sensitivity to context was demonstrated in several ways (Yardley, 2017). For example, choosing a phenomenological approach to studying the experiences of participants represents the most appropriate approach to achieving an in-depth understanding of their experiences. An important part of interviews was building rapport with participants and making them feel comfortable to share their experiences. Furthermore, the resulting themes were generated from the data, based on the participants' accounts, not being imposed prior to analysis.

Commitment and rigour were evident in thorough and complete data collection, in-depth interviews and analysis as well as consultations between the researchers. The study reported on variation and complexity of accounts, providing illustrations in carefully selected quotes. This is demonstrated, for example, in the subtheme 'Natural process (*or not*)' where participants' experiences differed dramatically. Sophie's account portrayed the most difficult experience when she described feeling as if her voice was taken away from her, no longer needed as a mum.

Transparency and coherence were demonstrated in the reported recruitment process. Moreover, the interpretation of research data was documented in transcripts during analysis and development of themes within and across accounts and checked back for evidence. A reflective stance of the lead researcher was embraced, reflections noted in a reflective journal and supervision used throughout the research process.

The empirical study also had a number of limitations. Firstly, in terms of the participants, although parents of young people who decided either for or against the surgery were invited to take part, only one parent whose child decided not to have surgery

was represented, which might be considered a limitation. However, their account contributed to all themes, adding to their scope. Another limitation might be linked to the recruitment from two different organisations, i.e., the NHS and the Cleft Lip and Palate Association (CLAPA). Although all participants received treatment in the NHS, they were from different teams across the UK and therefore potentially having different experiences of information delivery and the process of the transition in decision-making, making for a less homogeneous sample. However, all were suitable to participate because they had the experience of the process that was being explored. Another limitation might be seen in context of COVID-19 restrictions and conducting interviews online. However, research suggests that online interviews are a suitable alternative to interviews in person (Archibald et al., 2019; Bolderston, 2012; Irani, 2019).

Reflections on the Thesis Portfolio Process

Systematic review

This was the first systematic review the lead researcher conducted and it represented a range of valuable learning opportunities. The search strategy was consulted with a dedicated academic librarian to ensure that no studies were missed. Broad search terms were used to identify all eligible papers. Despite this one of the papers included in the review was identified through hand searching as it did not appear in database searches, which highlighted the need for reference lists checking and searching in related literature as database searches might not be exclusive.

Assessing methodological quality of studies in the review also generated a key learning. Using the Critical Appraisal Skills Programme (CASP, 2018) Checklists allowed for the appraisal, however, they did not offer a method of scoring to establish the level of quality. The lead researcher therefore drew on studies that had used CASP and added

scoring system to determine the level of quality (Hendry et al., 2017; Kmet et al., 2004; Rushbrooke et al., 2014).

Trying to synthesise heterogenous concepts and methodologies introduced some challenges. The process of bringing the results together appeared rather disjointed at first due to various aspects such as different population characteristics, methods and measures used. This was dealt with by keeping two overarching concepts, maternal attachment and parenting, that served as umbrella terms for research in these areas.

A substantial learning point was related to the process of conducting the systematic review. Published systematic reviews are presented as a linear, streamlined process yet conducting it revealed how iterative it in reality is and how much of the revision processes might not be acknowledged in the paper itself.

Empirical paper

The lead researcher chose the area of cleft lip and palate due to her interest in clinical health psychology with no previous clinical experience of the condition. That offered valuable learning of a new field of expertise, opportunity to expand knowledge and understanding of the cleft journey for the individuals as well as for their parents. However, at the beginning of learning about the condition the researcher had to reflect on some emotional reactions to photographs of babies affected by cleft.

Conducting the qualitative study was met with several challenges due to COVID-19. At first, all recruitment in the National Health Service (NHS) stopped for several months, which threatened the feasibility of the study that almost had to be abandoned. After being invested in the study for over a year and being just days away from starting recruitment at the time COVID-19 restrictions struck, this was unsettling and stressful for the lead researcher. However, the doctoral programme staff at the University of East

Anglia were supportive and acknowledged the impact of COVID-19 by offering a later thesis portfolio submission to accommodate delays caused by restrictions.

For the study to continue and to gain some level of control during the times of uncertainty and delay, the lead researcher proactively identified another, non-NHS recruitment channel and adapted methodology to comply with restrictions by conducting the study solely remotely. Online research interviews presented a range of new learning in terms of technology and experiences of adapted rapport building. Interviewing in person seemed much more natural to the researcher, yet participants seemed to be almost more relaxed and causal in their own environment online than in usual clinical settings in person.

Participants shared lots of experiences and offered various reflections, some of which unfortunately could not be included in the empirical paper due to methodological limitations of theme frequency or word count. It highlighted how much of research data can be lost in the process between analysis and presentation of findings. Thinking about the end stages of the cleft journey, participants talked about its beginning, receiving their child's diagnosis of cleft lip and/or palate, reflecting on the very first surgery their child underwent, their love for their child pouring through. Several participants also reflected on the impact of COVID-19 on the timing of their child's operation. Although parents were encouraged by clinical teams to get in touch with any questions, this opportunity to find out about delays or impact of COVID-19 was not taken up by many participants, highlighting parents' potentially ambivalent position of wanting to know but not wanting to put further strain on the team during the pandemic. Yet, some parents shared frustrations with delays and uncertainty about timing that subsequently impacted on plans for their children's education or employment. Some parents considered their child to be 'in limbo', their 'life on hold' while waiting for the surgery. That highlighted the significance of the long-awaited surgery for the whole family.

Conducting 11 interviews meant spending many hours on transcription and working through masses of data during analysis. It was a long process that the lead researcher broke down into smaller parts and carefully planned to meet deadlines. A lot of effort was put into organising the data, identifying, structuring the themes and checking back for evidence, which at times felt overwhelming. Supervision was invaluable during this process when ideas and reflections were discussed leading to the themes structure being amended to best capture parents' experiences. It helped the lead researcher in crystalising her understanding and interpretation of the data.

Clinical and Theoretical Implications

Systematic review

Although the systematic review of included studies found no differences in attachment in mothers of children with craniofacial anomalies in comparison to mothers of healthy children at five years, it highlighted changes in maternal attachment during the first year. It is therefore crucial to bear in mind that attachment evolves during that time, potentially in a different way to typically developing children. Parents adjust to the additional needs of their children, feel more confident in taking care of their children over time (Cinar & Koc, 2020) and go through the first repair surgeries with their children (NHS England, 2013) leading to yet again adjusting to their child's new needs and appearance.

As attachment feeds into the developing parenting styles (Nanu & Nijloveanu, 2015), it is crucial to support parents in establishing effective and positive relationships with their child. Psychological support for parents is part of the cleft lip and palate service provision (NHS England, 2013) where parents can address their initial reactions to the diagnosis, stress related to surgeries or whether they perceive their child to be more

vulnerable and they feel they need to be perhaps more protective of them by holding more control.

Being aware of potential risk factors for developing attachment such as stressful events, psychological or physical problems during pregnancy (Boztepe et al., 2016; Yilmaz et al., 2011) as well as the evolving attachment process during the first year will allow clinicians to better support parents throughout the cleft journey.

Empirical paper

Research suggests that parents can find it difficult to 'let go' and support independence in their children whether this is within healthy population of young adults (Kloep & Hendry, 2010) or young people with health conditions (Betz et al., 2015). The more challenging fact in decision-making about the orthognathic pathway might be that young people are not yet adults, usually in their middle teenage years, and there is no tangible transition between paediatric and adult services. This might be making it more difficult for parents to hand over responsibility for treatment choices.

Clinicians are aware of the intensity of the cleft treatment pathway and the commitment of parents in the process over many years (e.g., Nelson et al., 2011). However, they might not be aware of the potentially conflicting processes that take place within parents during the transition of responsibility in decision-making around the orthognathic surgery. The empirical study found a wide spectrum of parents' experiences that participants did not readily share with clinical teams. It is therefore recommended to prepare parents for this change in decision-making gradually so they are aware of the shift happening while their child is still an adolescent, which for some parents might feel too early. Clinicians can introduce the idea of the shift ahead of actually offering the orthognathic pathway to the young people, this should enable parents to have more space to share some of their experiences and concerns about these changes. Clinicians can

explore parents' potential internal conflict of not wanting to let go and feeling that it is their responsibility to make decisions about their child's healthcare, a role they took on when their child was born.

Direction for Research

Systematic review

The systematic review gathered and synthesised available evidence on parental attachment and parenting style in the population of interest. It highlighted a lack of research into father-child attachment and the need for longitudinal studies in attachment as well as parenting to gain a clearer understanding of parenting and to see any changes over time. For example, fostering autonomy in pre-school children during play is different to encouraging autonomy during decision-making about surgeries when the children are adolescents.

Empirical paper

The empirical paper explored parents' experiences of a changing decision-making process in a cross-sectional study. A longitudinal approach capturing several crucial time points, e.g., when parents are told about the change in who decides about the surgery, while they are stepping back and after the surgery if the young people opt in, would be helpful to enhance understanding of the evolving process during the transition. The empirical study featured parental reflections on the age of their children and their motives for making a decision about the surgery. The young person often needs to wait several years for their bones to mature while undergoing orthodontic treatment, during which time their level of self-acceptance and motivation for the surgery can change. What might have seemed the right decision at the age of 15 might be different when the young person reaches adulthood, has new opportunities, interests and responsibilities, e.g., university or

employment. Considering the cognitive development that occurs during the late teenage years, making the decision later and consequently undergoing the surgery later or checking with the young person that their decision stands might be helpful. Longitudinal research could address the development and potential decisional changes.

Research could specifically explore experiences of parents of children who decided against the surgery. Literature suggests that parents are usually in favour of 'normalising' surgeries (Nelson et al., 2012) and thus parents of children who do not want to undergo the surgery might have different needs and require different support from the clinical team, e.g., accepting that what they might have wanted for their child to reach their 'full potential' is not the young person's preference. Another area to address is how clinicians themselves view the orthognathic surgeries as their accounts are missing in literature. Research could explore clinicians' experiences of the orthognathic pathway, their role in the decision-making, their drives and narratives used when talking about it within the team and with families.

Thesis Portfolio Conclusion

Taken together, the systematic review is a helpful summary of research and what is known on the topic of maternal attachment and parenting style in parents of children born with craniofacial anomalies. The review will inform clinicians when supporting parents during the cleft journey and highlights to researchers important areas for future studies.

The empirical paper focused on parents' experiences of the end stages of the cleft journey, reflecting their parenting styles and attitudes. After many years of making decisions about their child's treatments parents are expected to hand over the responsibility for a decision about a complex surgery to their child who might still be in their middle teenage years. Parents in the study shared their readiness to step back and let go of the

responsibility for decisions about the orthognathic surgery. The study highlighted a whole spectrum of experiences, a shock, feeling as if their voice was taken away, as well as a relief, that are not necessarily shared with the clinical team. Parents talked about trying to present in front of clinicians as if the change did not make any impact on them, they reflected on the need to adjust to that change over time and talked about their ongoing involvement in the long treatment process as they considered it their journey as well. Clinicians thus need to be aware that this change in decision-making may have a profound impact on parents in the moment whilst they try to keep their emotional reactions hidden. Clinicians can prepare parents for the transition gradually and support them in exploring the complexities involved. Further research in the field is also suggested.

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A List of Appendices

Appendix A. The Cleft Palate-Craniofacial Journal Manuscript Submission Guidelines

https://journals.sagepub.com/author-instructions/CPC

Manuscript Submission Guidelines:

Due to the worldwide impact of the COVID-19 pandemic, we are very aware that many researchers and reviewers will have difficulty meeting the typical timelines associated with our journal's peer review process. Our editorial office will continue to send reminders, but we intend to be very flexible during this time. Please do let us know if you will need additional time. Furthermore, journal submissions are currently substantially higher for *CPCJ* and the availability of reviewers in some cases is limited. This may cause delays, but please be rest assured that our journal team is working to ensure the timely management of your submission.

This Journal is a member of the Committee on Publication Ethics.

This Journal recommends that authors follow the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals formulated by the International Committee of Medical Journal Editors (ICMJE).

Please read the guidelines below then visit the Journal's submission site https://mc.manuscriptcentral.com/cpcj to upload your manuscript. Please note that manuscripts not conforming to these guidelines may be returned.

SAGE Publishing disseminates high-quality research and engaged scholarship globally, and we are committed to diversity and inclusion in publishing. We encourage submissions from a diverse range of authors from across all countries and backgrounds.

Only manuscripts of sufficient quality that meet the aims and scope of *The Cleft Palate-Craniofacial Journal (CPCJ)* will be reviewed. *CPCJ* is directed to a multidisciplinary readership of clinicians and scientists interested in craniofacial anomalies, including cleft lip and cleft palate. The *CPCJ* publishes original research articles, clinical reports, brief

communications, articles related to new ideas or innovations, letters to the editor, editorials, invited book reviews, and meeting announcements.

There are no fees payable to submit or publish in this journal.

As part of the submission process you will be required to warrant that you are submitting your original work, that you have the rights in the work, that you have obtained and can supply all necessary permissions for the reproduction of any copyright works not owned by you, that you are submitting the work for first publication in the Journal, and that it is not being considered for publication elsewhere and has not already been published elsewhere. Note that the Journal may accept submissions of papers that have been posted on pre-print servers; include the DOI for the preprint in the designated field during the submission process. Authors should not post an updated version of their paper on the preprint server while it is being peer reviewed for possible publication in the journal. If the article is accepted for publication, the author may re-use their work according to the Journal's author archiving policy. If your paper is accepted, you must include a link on your preprint to the final version of your paper.

If you have any questions about publishing with SAGE, please visit the SAGE Journal Solutions Portal

1. What do we publish?

1.1 Aims & Scope

Before submitting your manuscript to *CPCJ*, please ensure you have read the Aims & Scope. *CPCJ* publishes manuscripts of the highest scientific quality on all topics related to orofacial clefts and other craniofacial anomalies in order to advance the global education of scientists and clinicians

1.2 Article Types

Original Articles: 7 typeset pages as they appear in the journal (about 7,000 words, with up to 6 figures or tables combined)

What I (We) Do: 2 typeset pages as they appear in the journal (about 1,000 words, with up to 3 figures or tables combined and up to 5 references)

Case Reports: 4 typeset pages as they appear in the journal (about 4,000 words, with up to 6 figures or tables combined)

Ethics / Health Policy / Ideas and Innovations / Brief Communications: 3 typeset pages as they appear in the journal (about 3,000 words, with up to 3 figures or tables combined)

Perspectives / Letters to the Editor / Editorials: Should provide thoughtful, scientific, constructive commentary pertaining to articles or research published in The Cleft Palate-Craniofacial Journal. 1.5 typeset pages as they appear in the journal (about 1,500 words, with up to 1 figure or table).

A single figure may include multiple images (a, b, c, etc.) but all must appear on the same page.

Supporting material that is not essential to an understanding of the article may be posted with the article as supplemental online-only material.

CPCJ allows as many citations and references as the authors feel necessary for the manuscript.

1.3 Writing your paper

The SAGE Author Gateway has some general advice and on how to get published, plus links to further resources.

1.3.1 Make your article discoverable

When writing up your paper, think about how you can make it discoverable. The title, keywords and abstract are key to ensuring readers find your article through search engines such as Google. For information and guidance on how best to title your article, write your abstract and select your keywords, have a look at this page on the Gateway: How to Help Readers Find Your Article Online

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2. Editorial policies

2.1 Peer review policy

Two independent peer reviews are typically solicited. At the discretion of the Section Editor, a third review by a biostatistician may also be solicited. The Editor is responsible for all final decisions regarding acceptance or rejection, recommendations for revision, and final editing. Manuscripts will be evaluated according to various criteria, including scientific methodology, level of evidence, novelty, clarity, and conciseness. Accepted articles describing novel findings or methods with high levels of evidence may be advanced in the publication queue at the discretion of the Editor.

All submitted articles are "double-blinded" to ensure an unbiased review. Reviewers will not have access to author names or affiliations. Authors will not have access to reviewer names or affiliations.

The Editor or members of the Editorial Board may occasionally submit their own manuscripts for possible publication in the journal. In these cases, the peer review process will be managed by alternative members of the Board and the submitting Editor/Board member will have no involvement in the decision-making process.

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>
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Medical research involving human subjects must be conducted according to the World Medical Association Declaration of Helsinki. Compliance with these guidelines should be indicated in the Methods section of the manuscript, along with Institutional Review Board approval if appropriate.

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3. Publishing Policies

3.1 Publication ethics

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Page 1: Title The first page of the manuscript text file should include only the title used on the Title Page (above).

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Structured Abstract:

Objective: State the main question or objective of the study and the major hypothesis tested, if any.

Design: Describe the design of the study indicating, as appropriate, use of randomization, blinding, criterion standards for diagnostic tests, temporal direction (retrospective or prospective), etc.

Setting: Indicate the study setting, including the level of clinical care (for example, primary or tertiary; private practice or institutional).

Patients, Participants: State selection procedures, entry criteria, and numbers of participants entering and finishing the study.

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The metric system is preferred for expressing units of measure. Abbreviations may be used for terms. The full term for each abbreviation should appear at its first use in the text, unless the abbreviation is a standard unit of measure. Abbreviations used in a table must be explained in a footnote below the table. For a list of standard abbreviations, consult the Council of Biology Editors Style Guide (available from the Council of Science Editors,

9650 Rockville Pike, Bethesda, MD 20814; http://www.councilscienceeditors.org) or other standard sources.

The table below lists standard accepted abbreviations for typical cleft-type classifications and study groups. Other abbreviations may be proposed for classifications and groups not listed.

ABBREVIATION USED TO DESCRIBE A SUBJECT GROUP THAT INCLUDES:	
	cleft lip (excludes (1) cleft lip and alveolus, (2) cleft lip and palate, and (3) cleft palate)
CL	cleft palate only (excludes (1) cleft lip and (2) cleft lip and palate)
CP	cleft lip and palate (excludes (1) cleft lip and (2) cleft palate)
CLP	cleft lip with or without cleft palate = cleft lip + cleft lip and palate (excludes cleft palate)
CL±P	cleft palate with or without cleft lip = cleft lip and palate + cleft palate (excludes cleft lip)
CP±L	cleft lip and/or cleft palate = cleft lip + cleft lip and palate + cleft palate (no exclusions)
CL/P	cleft lip with or without cleft alveolus = cleft lip + cleft lip and alveolus (excludes (1) cleft lip, (2) cleft lip and palate, and (3) cleft palate)
CL±A	

TERMS THAT MAY BE ADDED TO THE ABBREVIATIONS ABOVE (IF APPROPRIATE):

i	isolated
I	incomplete

U unilateral

B bilateral

SM submucous

Phonetic Symbols

Authors who use phonetic symbols are required to use Unicode-compliant fonts in their manuscripts. This will ensure the symbols display properly both during peer review and in the final published article. Examples of acceptable fonts include Charis SIL, Doulos SIL, and Gentium Unicode. Times New Roman is also acceptable, as it includes most IPA symbols and is Unicode compliant.

Citations/References

Single Author Article
Citation: Mantel (1963) or (Mantel, 1963)
Reference: Mantel N. Chi-square tests with one degree of freedom; extensions of the Mantel-Haenszel procedure. *J Am Stat Assoc*. 1963;58:690–700.

Two Author Article
Citation: Rasheed and Munshi (1996) or (Rasheed and Munshi, 1996)
Reference: Rasheed SA, Munshi AK. Electromyographic and ultrasonographic evaluation of the circum-oral musculature in children. *J Clin Pediatr Dent*. 1996;20:305-311.

Three Or More Author Article
Citation: Lilja et al. (2000) or (Lilja et al., 2000)
Reference: Lilja J, Elander A, Lohmander A, Persson C. Isolated cleft palate and submucous cleft palate. *Oral Maxillofac Surg Clin N Am*. 2000;12:455–468.

Two or more works by the same first author in the same year
Citation: Smith (1975a), Smith (1975b) or (Smith, 1975a) etc
Reference: Smith RC. Long term effects of smoking on fetal development. *Teratology* 1975a;42:75-84.

Monograph

Citation: Bardach (1967) or (Bardach, 1967)

Reference: Bardach J. *Cleft Lip and Palate* (Monograph). Warsaw: Polish Institute of Medical Publications; 1967.

Thesis

Citation: Dowden (1992)

Reference: Dowden PA. The Effects of Listener Training on the Speech Intelligibility of Severely Dysarthric Individuals. Seattle, WA: University of Washington; 1992. Dissertation.

Book

Citation: McWilliams et al. (1990) or (McWilliams et al., 1990)

Reference: McWilliams BJ, Morris HL, Shelton RL. *Cleft Palate Speech*. Philadelphia: BC Decker; 1990: 40-49. (only list pages if specific pages are cited).

Chapter in Book

Citation: Eliason (1990) or (Eliason, 1990)

Reference: Eliason MJ. Neuropsychological perspectives of cleft lip and palate. In: Bardach J, Morris HL, eds. *Multidisciplinary Management of Cleft Lip and Palate*. Philadelphia: WB Saunders; 1990:825–831.

Conference Presentation

Citation: Parke and Sawin (1975) or (Parke and Sawin, 1975)

Reference: Parke RD, Sawin DB. Infant characteristics and behavior as elicitors of maternal and paternal responsivity in the newborn period. Presented at the Meeting of the Society for Research in Child Development; April 1975; Denver, Colorado.

Website

Citation: World Health Organization (2005)

Reference: World Health Organization. International database on craniofacial anomalies. Available at: www.who.int/genomics/anomalies/. Accessed June 27, 2005.

When multiple references are cited simultaneously in the text, they should be arranged in chronological order, for example: (Smith, 1975; Jones et al., 1981; Brown, 1986). References should be double-spaced, and listed in alphabetical order (unnumbered)

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Tables should be numbered consecutively using Arabic numerals. Each table should have an appropriate title and explanation at its head. Abbreviations used in a table must be explained in a footnote below the table. Submit tables as separate files, with one table per file, in either .doc (text) or .xls (spreadsheet) format.

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All figures and illustrations must be original photographs or artwork. For figures or illustrations reprinted from published work, the author must obtain written permission from the copyright holder and upload that permission as an "Additional Information" file at submission. Figures should be numbered consecutively in the order in which they appear in the manuscript, using Arabic numerals. A "List of Figure" Legends must be included on a separate page following the body of the manuscript. The legend should explain each figure in detail. Authors will be responsible for the following charges for each color figure submitted: $75.00 for online only; $400.00 for both online and print for ACPA members or $500.00 for non-members. A single figure may include multiple images (a, b, c, etc.) but all must appear on the same page.

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6. On acceptance and publication

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details are correct, and that Funding and Conflict of Interest statements, if any, are accurate. Please note that if there are any changes to the author list at this stage all authors will be required to complete and sign a form authorising the change.

6.2 Online First publication

Online First allows final articles (completed and approved articles awaiting assignment to a future issue) to be published online prior to their inclusion in a journal issue, which significantly reduces the lead time between submission and publication. Visit the SAGE Journals help page for more details, including how to cite Online First articles.

6.3 Access to your published article

SAGE provides authors with online access to their final article.

6.4 Promoting your article

Publication is not the end of the process! You can help disseminate your paper and ensure it is as widely read and cited as possible. The SAGE Author Gateway has numerous resources to help you promote your work. Visit the Promote Your Article page on the Gateway for tips and advice.

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7. Further information

Any correspondence, queries or additional requests for information on the manuscript submission process should be sent to the CPCJ editorial office as follows:

Editor: Jamie Perry, PhD
Editorial Office: The Cleft Palate-Craniofacial Journal
Email: perryja@ecu.edu

7.1 Appealing the publication decision

Editors have very broad discretion in determining whether an article is an appropriate fit for their journal. Many manuscripts are declined with a very general statement of the rejection decision. These decisions are not eligible for formal appeal unless the author believes the decision to reject the manuscript was based on an error in the review of the

article, in which case the author may appeal the decision by providing the Editor with a detailed written description of the error they believe occurred.

If an author believes the decision regarding their manuscript was affected by a publication ethics breach, the author may contact the publisher with a detailed written description of their concern, and information supporting the concern, at publication_ethics@sagepub.com

Appendix B. Quality rating tables of studies with inter-rater checks

Table 1. CASP quality rating for quantitative studies with inter-rater checks

CASP criteria (inter-rater score)	Boztepe et al. (2016)	Cinar et al. (2020)	Despars et al. (2011)	Gassling et al. (2014)	Habersaat et al. (2018)	Krueckeberg et al. (1993)	Shapiro et al. (2018)	Tsuchiya et al. (2019)	Yilmaz et al. (2011)
1.Did the study addressed a clearly formulated issue?	2	2	2	2	2 (2)	2	2	2 (2)	2
2.Was the cohort recruited in an acceptable way?	1	2	2	2	1 (1)	1	1	2 (2)	2
3.Was the exposure accurately measured to minimise bias?	N/A	2	N/A	N/A	N/A	N/A	N/A	N/A	N/A
4.Was the outcome accurately measured to minimise bias?	2	2	2	2	2 (2)	2	2	2 (2)	2
5a.Have the authors identified all important confounding factors?	2	2	2	2	0 (0)	2	0	2 (2)	2
5b.Have they taken account of the confounding factors in the design and/or analysis?	2	2	2	2	1 (1)	2	1	2 (2)	1
6a.Was the follow-up of subjects complete enough?	N/A	2	2	N/A	2 (2)	N/A	N/A	N/A	N/A
6b.Was the follow-up of subjects long enough?	N/A	2	2	N/A	2 (2)	N/A	N/A	N/A	N/A
9.Do you believe the results?	2	2	2	2	2 (2)	2	2	2 (2)	2
10.Can the results be applied to the local population?	2	2	2	1	2 (2)	2	2	2 (2)	2
11.Do the results of this study fit with other available evidence?	2	2	2	0	1 (1)	N/A	2	2 (2)	2
12.What are the implications of this study to practice?	2	2	2	1	2 (2)	2	2	2 (2)	2
Possible total score per study	18	24	22	18	22	16	18	18	18
Total score achieved (percentage)	17 (94%)	24 (100%)	22 (100%)	14 (78%)	17 (17) (77%)	15 (94%)	14 (78%)	18 (18) (100%)	17 (94%)

Note (Table 1): Questions 7 and 8 do not use answers 'yes', 'can't tell', 'no' and therefore could not be scored in the same way.

Table 2. CASP quality rating for qualitative studies with inter-rater checks

CASP criteria (inter-rater checks)	Breuning et al. (2020)	Klein et al. (2006)	Klein et al. (2014)
1.Was there a clear statement of the aims of the research?	2	2	2 (2)
2.Is the qualitative methodology appropriate?	2	2	2 (2)
3.Was the research design appropriate to address the aims of the research?	2	2	2 (2)
4.Was the recruitment strategy appropriate to the aims of the research?	2	1	1 (1)
5.Was the data collected in a way that addressed the research issue?	2	2	2 (2)
6.Has the relationship between researcher and participants been adequately considered?	1	1	1 (1)
7.Have ethical issues been taken into consideration?	2	2	2 (2)
8.Was the data analysis sufficiently rigorous?	2	2	2 (2)
9.Is there a clear statement of findings?	2	2	2 (2)
10.How valuable is the research?	2	2	2 (2)
Possible total score per study	20	20	20
Total score achieved (percentage)	19 (95%)	18 (90%)	18 (90%)

Appendix C. HRA approval

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Miss Jana Safarikova
Trainee Clinical Psychologist
Cambridgeshire and Peterborough NHS Foundation Trust
Elizabeth House, Fulbourn Hospital
Fulbourn
Cambridge
CB21 5EF

Email: hra.approval@nhs.net
HCRW.approvals@wales.nhs.uk

25 February 2020
Reissued 02 March 2020

Dear Miss Safarikova

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Parents' experiences of the decision-making process for elective surgery at transition in cleft lip and palate: An Interpretative Phenomenological Analysis**
IRAS project ID:	**266133**
REC reference:	**20/WS/0034**
Sponsor	**University of East Anglia**

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 **266133**. Please quote this on all correspondence.

Yours sincerely,
Catherine Adams

Approvals Manager

Email: hra.approval@nhs.net

Copy to: *Ms Polly Harrison*

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 [Poster_266133_v1_2020-01-17]	v1	17 January 2020
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Public_Liability_and_Product_Liability_(to_May_2020)]		01 May 2019
Interview schedules or topic guides for participants [Topic_guide_266133_v1_2020-01-10]	v1	10 January 2020
IRAS Application Form [IRAS_Form_24012020]		24 January 2020
Letter from sponsor [Insurance_and_Indemnity_confirmation_2020-10-17]		17 January 2020
Letters of invitation to participant [Participant_invitation_letter_parents_266133_v1_2020-01-17]	v1	17 January 2020
Letters of invitation to participant [Participant_invitation_letter_YP_266133_v1_2020-01-17]	v1	17 January 2020
Organisation Information Document		03 February 2020
Other [Professional_Indemnity_(to_May_2020)]		01 May 2019
Other [Research_CV_Eliane_Young_2019-09-20]		20 September 2019
Other [Consent_to_contact_266133_v1_2020-01-17]	v1	17 January 2020
Other [Demographic_information_form_266133_v1_2020-01-17]	v1	17 January 2020
Other [Voucher_receipt_form_266133_v1_2020-01-17]	v1	17 January 2020
Other [Signature_And_Delegation_Log_Template_v1-2_IRAS266133_2020-01-17]	v1-2	17 January 2020
Other [Confirmation Email re CI/Student role]		26 January 2020
Participant consent form [Consent_form_266133_v1	V2	18 February 2020
Participant information sheet (PIS) [PIS_266133_v1	V2	18 February 2020
Research protocol or project proposal [Thesis_Protocol_266133_v1_2020-01-10]	v1	10 January 2020
Schedule of Events or SoECAT [IRAS schedule-events-excel-template-1_0_266133_2020-01-17]	v1_0	17 January 2020
Summary CV for Chief Investigator (CI) [Research_CV_Jana_Safarikova_2019-09-23]		23 September 2019
Summary CV for supervisor (student research) [Research_CV_Paul_Fisher_2019-11-04]		04 November 2019
Summary CV for supervisor (student research) [Research_CV_Kiki_Mastroyannopoulou_2019-09-20]		20 September 2019

Appendix D. Participant Information Sheet (NHS and non-NHS)

Participant Information Sheet (NHS)

[Trust logo]

PARTICIPANT INFORMATION SHEET

'Parents' experiences of decision-making about orthognathic surgery'

My name is Jana Safarikova, Trainee Clinical Psychologist at the University of East Anglia. I am carrying out a research study about parents' experiences of decision-making about orthognathic surgery which is conducted as part of my Doctorate in Clinical Psychology course.

Please take time to read through this information sheet. You can ask me for clarification if anything is not clear or if you would like further details.

Why me?
You are being invited to take part in this study because you are a parent of a young person who was born with cleft lip and/or palate and who has been cared for by the Cleft Lip and Palate Team at [redacted]. By taking part in the study, you could help us improve and shape our services to deliver the best support for children, young adults and their families.

What is the study about?
We understand that orthognathic surgery is offered to young people during a transition time when they are expected to be the main decision-makers. It is therefore a significant time for them and consequently for many parents. A recent study addressed the experiences of young people making a decision about this elective surgery but there is limited knowledge about the experiences of parents during the decision-making process. We would like to interview 8-10 parents to share their experiences with us.

Who can take part?

- Parents of young people who have made a decision about whether or not to undergo orthognathic surgery.
- If the decision is 'yes', the young person is on the treatment pathway to undergo the surgery. It is expected that this decision has been made in the last five years.
- If the decision is 'no', then this decision was made in the last five years as well.

Do I have to take part?
No, you do not have to take part. Taking part in NHS research is voluntary, it is up to you whether you want to take part or not. If you decide against taking part, this will in no way affect the care provided to the young person in your family. It is an opportunity for you to share your experiences.

What would taking part in this study involve?
If you are interested in taking part, you should contact either me (Jana) directly or the Cleft Lip and Palate Team using the contact details provided at the end of this information sheet Once you make contact, we will arrange a meeting for our interview. Considering the current COVID-19 restrictions, the interview will be done via Skype or similar online platform.

We will start our meeting by going through the study details to ensure that you are well informed and agree to take part. We will then read through a consent form to confirm your agreeing to taking part and I will audio record your consent. This might take up to 15 minutes.

During our interview I will start by asking you some basic information about yourself, what type of cleft the young person in your family was born with and how many surgeries they have had (approx. 10 minutes). I will not need to know the gender or name of your child but you can share it with me if you wish. We will then focus on your experience of the decision-making process, how involved you were, what it was like for you and how the decision was reached. The interview is expected to last approximately 45-60 minutes and it will be video and audio recorded. The video recording will be deleted once the audio is confirmed as of satisfactory quality.

What will happen to the information I provide?
Your interview will be transcribed and anonymised so you cannot be identified. Your interview and other parents' interviews will be considered in detail in order to try and understand what the experience of the decision-making process has been like. With your consent I would like to share my findings from the study with other professionals and researchers by publishing the findings, so they and future patients can benefit from your expertise. The Cleft Lip and Palate Service will use the information to develop their service and this part of the pathway in particular. If you are interested, you will be able to receive a summary of the anonymised findings.

Following the Data Protection Policy in accordance with GDPR the information you provide (including your non-identifiable data) will be stored securely for 10 years in an encrypted file on a password-protected computer at the University of East Anglia.

The University has a dedicated Data Protection Officer: Ellen Paterson, The Library, University of East Anglia, Norwich Research Park, Norwich NR4 7TJ, dataprotection@uea.ac.uk, 01603 592431.

What are the benefits of taking part?
By taking part you will be helping us to understand more about your experiences and you may be able to help other people going through the same process in the future.

What are the possible disadvantages and risks of taking part?
As mentioned above, you will need to offer your time to conduct the study, which might be around 60-80 minutes.

Although I hope you will find the interview straightforward, there is a small chance that it might cause you upset as it will address your personal experiences. Ethics committees have to review all planned studies before they can start. The purpose is to protect people taking part by ensuring they do not come to any harm and the studies conducted have value to science and society. This research

study has received the required approvals: Health Research Authority (HRA) approval and favourable opinion from West of Scotland REC 5. [REC reference: 20/WS/0034; IRAS project ID: 266133]

Can I change my mind?
Yes, you can change your mind whether you would like to take part in the study. This can be prior to our meeting, during our interview or up to a week later. However, after this time the analysis will start and it will *not* be possible to withdraw your data. Hopefully, this approach will offer you enough opportunities to consider your participation and allow you to withdraw in time if you change your mind.

Reimbursement
As a 'thank you' for giving up your time to take part you will receive a £10 voucher.

Confidentiality and anonymity
All information you provide during the interview will be for this research purposes only. However, if you share something in your interview about harming yourself or others, this will need to be shared with the Cleft Lip and Palate Team to ensure that you and others are safe. This would be discussed with you first.

As I will be using your data in the analysis and might use some quotes, full confidentiality of what you share with me cannot be guaranteed. However, all information that identifies you will be changed and therefore your anonymity won't be broken. A pseudonym of your choice will be used to protect your and your child's identity in any data or quotes used in publications.

The University of East Anglia (UEA) is the sponsor for this study based in the United Kingdom. It will be using information from you in order to undertake this study and will act as the data controller for this study. This means that UEA is responsible for looking after your information and using it properly. UEA will keep identifiable information about you for no longer than it is required after the study has finished, e.g. to share a summary of findings with you if you are interested in seeing them.

You can find out more about how we use your information at https://portal.uea.ac.uk/information-services/strategy-planning-and-compliance/regulations-and-policies/information-regulations-and-policies/data-protection.

General Data Protection Regulation (GDPR) transparency
In this research study we will use information from you. We will only use information that we need for the research study. We will let very few people know your name or contact details, and only if they really need it for this study.

Everyone involved in this study will keep your data safe and secure. We will also follow all privacy rules.

We will make sure no-one can work out who you are from the reports we write.

Interested?

If you are interested and would like to be interviewed to share your experiences, please contact me and we can arrange a date and time for your interview. Please ask me any questions you may have about participating in this research.

We appreciate that life is busy and therefore if neither the team or I hear from you, someone from the Cleft Lip and Palate Team will contact you in approximately a week's time to see if you have had a chance to look through study information.

Complaints
If you wish to make a complaint about any aspect of this research, please contact Professor Niall Broomfield, Programme Director, Doctorate in Clinical Psychology (n.broomfield@uea.ac.uk).

How to contact us
If you have any queries or would like further information, here are the best contact details to get in touch:

Jana Safarikova, Trainee Clinical Psychologist
j.safarikova@uea.ac.uk
[redacted]

Dr Paul Fisher
Registered Clinical Psychologist, Senior Clinical Lecturer in Clinical Psychology at UEA, Supervisor of this research study
p.fisher@uea.ac.uk

Alternatively, you can contact the Cleft Lip and Palate Team:
Dr [redacted]
Lead Clinical Psychologist
[redacted] option [redacted] (Secretary [redacted])

If you are interested in taking part and would like to arrange the interview, please contact Jana or the Cleft Lip and Palate Team.

Thank you for taking time to read through the information and for considering taking part in the research study.

Participant Information Sheet (non-NHS)

PARTICIPANT INFORMATION SHEET

'Parents' experiences of decision-making about orthognathic surgery'

My name is Jana Safarikova, Trainee Clinical Psychologist at the University of East Anglia. I am carrying out a research study about parents' experiences of decision-making about orthognathic surgery which is conducted as part of my Doctorate in Clinical Psychology course.

Please take time to read through this information sheet. You can ask me for clarification if anything is not clear or if you would like further details.

Why me?
You are being invited to take part in this study because you are a parent of a young person who was born with cleft lip and/or palate. By taking part in the study, you could help us improve and shape NHS services to deliver the best support for children, young adults and their families.

What is the study about?
We understand that orthognathic surgery is offered to young people during a transition time when they are expected to be the main decision-makers. It is therefore a significant time for them and consequently for many parents. A recent study addressed the experiences of young people making a decision about this elective surgery but there is limited knowledge about the experiences of parents during the decision-making process. We would like to interview 8-10 parents to share their experiences with us.

Who can take part?

- Parents of young people who have made a decision about whether or not to undergo orthognathic surgery.
- If the decision is 'yes', the young person is on the treatment pathway to undergo the surgery. It is expected that this decision has been made in the last five years.
- If the decision is 'no', then this decision was made in the last five years as well.

Do I have to take part?
No, you do not have to take part. Taking part in this research is voluntary, it is up to you whether you want to take part or not. If you decide against taking part, this will in no way affect the care provided to the young person in your family. It is an opportunity for you to share your experiences.

What would taking part in this study involve?
If you are interested in taking part, you should contact me (Jana) using the contact details provided at the end of this information sheet. Once you make contact, I will answer any questions you might have about the study, check that you are eligible to take part and we will arrange our interview via Skype or a similar online platform.

We will start our interview by going through the study details to ensure that you are well informed and agree to take part. We will then read through a consent form to confirm your agreeing to taking part and I will audio record your consent. This might take up to 15 minutes.

During our interview I will start by asking you some basic information about yourself, what type of cleft the young person in your family was born with and how many surgeries they have had (approx. 10 minutes). I will not need to know the gender or name of your child but you can share it with me if you wish. We will then focus on your experience of the decision-making process, how involved you were, what it was like for you and how the decision was reached. The interview is expected to last approximately 45-60 minutes and it will be video and audio recorded. The video recording will be deleted once the audio is confirmed as of satisfactory quality.

What will happen to the information I provide?
Your interview will be transcribed and anonymised so you cannot be identified. Your interview and other parents' interviews will be considered in detail in order to try and understand what the experience of the decision-making process has been like. With your consent I would like to share my findings from the study with other professionals and researchers by publishing the findings, so they and future patients can benefit from your expertise. The Cleft Lip and Palate Service in the NHS will use the information to develop their service and this part of the pathway in particular. If you are interested, you will be able to receive a summary of the anonymised findings.

Following the Data Protection Policy in accordance with GDPR the information you provide (including your non-identifiable data) will be stored securely for 10 years in an encrypted file on a password-protected computer at the University of East Anglia.

The University has a dedicated Data Protection Officer: Ellen Paterson, The Library, University of East Anglia, Norwich Research Park, Norwich NR4 7TJ, dataprotection@uea.ac.uk, 01603 592431.

What are the benefits of taking part?
By taking part you will be helping us to understand more about your experiences and you may be able to help other people going through the same process in the future.

What are the possible disadvantages and risks of taking part?
As mentioned above, you will need to offer your time to conduct the study, which might be around 60-80 minutes in total.

Although I hope you will find the interview straightforward, there is a small chance that it might cause you upset as it will address your personal experiences. Ethics committees have to review all planned studies before they can start. The purpose is to protect people taking part by ensuring they do not come to any harm and the studies conducted have value to science and society. This research study has received the required approvals: Health Research Authority (HRA) approval and favourable opinion from West of Scotland REC 5. [REC reference: 20/WS/0034; IRAS project ID: 266133]

Can I change my mind?
Yes, you can change your mind whether you would like to take part in the study. This can be prior to our interview, during our interview or up to a week later. However, after this time the analysis will start and it will *not* be possible to withdraw your data. Hopefully, this approach will offer you enough opportunities to consider your participation and allow you to withdraw in time if you change your mind.

Reimbursement
As a 'thank you' for giving up your time to take part you will receive a £10 voucher.

Confidentiality and anonymity
All information you provide during the interview will be for this research purposes only. However, if you share something in your interview about harming yourself or others, this will need to be shared with CLAPA (or the Police) to ensure that you and others are safe. This would be discussed with you first.

As I will be using your data in the analysis and might use some quotes, full confidentiality of what you share with me cannot be guaranteed. However, all information that identifies you will be changed and therefore your anonymity won't be broken. A pseudonym of your choice will be used to protect your and your child's identity in any data or quotes used in publications.

The University of East Anglia (UEA) is the sponsor for this study based in the United Kingdom. It will be using information from you in order to undertake this study and will act as the data controller for this study. This means that UEA is responsible for looking after your information and using it properly. UEA will keep identifiable information about you for no longer than it is required after the study has finished, e.g. to share a summary of findings with you if you are interested in seeing them.

You can find out more about how we use your information at https://portal.uea.ac.uk/information-services/strategy-planning-and-compliance/regulations-and-policies/information-regulations-and-policies/data-protection.

General Data Protection Regulation (GDPR) transparency
In this research study we will use information from you. We will only use information that we need for the research study. We will let very few people know your name or contact details, and only if they really need it for this study.

Everyone involved in this study will keep your data safe and secure. We will also follow all privacy rules.

We will make sure no-one can work out who you are from the reports we write.

Interested?
If you are interested and would like to be interviewed to share your experiences, please contact me and we can arrange a date and time for your interview. Please ask me any questions you may have about participating in this research.

Complaints
If you wish to make a complaint about any aspect of this research, please contact Professor Niall Broomfield, Programme Director, Doctorate in Clinical Psychology (n.broomfield@uea.ac.uk).

How to contact us
If you have any queries or would like further information, here are the best contact details to get in touch:

Jana Safarikova, Trainee Clinical Psychologist
j.safarikova@uea.ac.uk
[redacted]

Dr Paul Fisher
Registered Clinical Psychologist, Senior Clinical Lecturer in Clinical Psychology at UEA, Supervisor of this research study
p.fisher@uea.ac.uk

Thank you for taking time to read through the information and for considering taking part in the research study.

Appendix E. Consent form (NHS and non-NHS)

Consent form (NHS)

[Trust logo]

IRAS ID: 266133

Study Number:

Participant Identification Number for this trial:

CONSENT FORM

Title of Project: **Parents' experiences of decision-making about orthognathic surgery**

Name of Researcher: **Jana Safarikova**

Please initial box

1. I confirm that I have read the information sheet dated 19/06/2020 (version 5) for the above study. ☐
2. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
3. I understand that my participation in this research is voluntary. ☐
4. I understand that my interview will be audio recorded. ☐
5. I agree for my video interview to be video recorded. ☐
6. I understand that I can withdraw from the study without giving any reason and this will not affect my and my child's (young person's) care. ☐
7. I understand that once my interview data have been anonymised and entered into analysis they can no longer be withdrawn. Therefore, I have one week after my interview to request withdrawal of my interview data. After this time anonymised data will be used in the study. ☐
8. I understand that my data collected during the study may be looked at by individuals from University of East Anglia, regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to these records for the purpose of audit and monitoring of the research. ☐

When completed: 1 for participant; 1 for researcher site file
IRAS 266133, REC 20/WS/0034
Consent_form_NHS_v5_19062020

9. I understand and agree for my anonymised interview data to be used in publications. ☐

10. I understand that if the researcher is concerned about my or someone else's safety, then information may be shared with the clinical team. However, this will be discussed with me first. ☐

11. I would like to receive a summary of findings and agree for my contact details being kept by the researcher to be able to do so. ☐

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

Name of Participant	Date	Signature
Name of Person taking consent	Date	Signature

Consent form (non-NHS)

IRAS ID: 266133

Study Number:

Participant Identification Number for this trial:

CONSENT FORM

Title of Project: **Parents' experiences of decision-making about orthognathic surgery**

Name of Researcher: **Jana Safarikova**

Please initial box

1. I confirm that I have read the information sheet dated 19/06/2020 (version 5, non NHS) for the above study. ☐
2. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
3. I understand that my participation in this research is voluntary. ☐
4. I understand that my interview will be audio recorded. ☐
5. I agree for my video interview to be video recorded. ☐
6. I understand that I can withdraw from the study without giving any reason and this will not affect my and my child's (young person's) care. ☐
7. I understand that once my interview data have been anonymised and entered into analysis they can no longer be withdrawn. Therefore, I have one week after my interview to request withdrawal of my interview data. After this time anonymised data will be used in the study. ☐
8. I understand that my data collected during the study may be looked at by individuals from University of East Anglia where it is relevant to my taking part in this research. I give permission for these individuals to have access to these records for the purpose of audit and monitoring of the research. ☐

9. I understand and agree for my anonymised interview data to be used in publications. ☐

10. I understand that if the researcher is concerned about my or someone else's safety, then information may be shared with CLAPA (if applicable) or the Police. However, this will be discussed with me first. ☐

11. I would like to receive a summary of findings and agree for my contact details being kept by the researcher to be able to do so. ☐

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

_____	_____	_____
Name of Participant	Date	Signature
_____	_____	_____
Name of Person taking consent	Date	Signature

Appendix F. Demographic information form

University of East Anglia

Demographic information form

Participant ID: _____

Date: _____

DEMOGRAPHIC INFORMATION FORM

Study name: Parents' experiences of decision-making about orthognathic surgery

In order to learn about the range of people taking part in this study, we would be grateful if you could answer the following questions. All information provided is anonymous, it will be linked only to your Participant Identification Number above. Please write your answer in the space provided, or circle the answer that best applies to you.

ABOUT YOUR CHILD

What type of cleft does your child have?	Unilateral cleft lip and palate (right/left) Bilateral cleft lip and palate Cleft palate only Other (please specify) .....
How many surgeries has your child had prior to the decision regarding orthognathic surgery?	.....
How old was your child when the decision about orthognathic surgery was made?	.....
How long ago was that decision made?	.....

Have you ever had a session with the Clinical Psychologist within the Cleft Lip and Palate Service?	Yes No Not sure
If yes, how many times have you met with the Clinical Psychologist?	Once 2-3 times 3-6 times 6 times or more (please specify) ..... Not sure
Does your child have any other diagnosis?	Please specify ....

ABOUT YOU

How old are you?	
Please indicate your gender	Female Male Other (please specify) .....
How would you describe your racial/ethnic background?	White British/White other (please specify) ..... Asian/Asian British Black/Black British Mixed ethnicity (please specify) ..... Other ethnic group (please specify) ..... Prefer not to say

Thank you for your time completing this form.

Appendix G. Topic guide

Parents' experiences of the decision-making process for elective surgery at transition in cleft lip and palate: An Interpretative Phenomenological Analysis

Topic guide

Research question:

What are the experiences of parents of young people with cleft lip/palate, of the decision-making process in elective orthognathic surgery?

Topic guide:

1. In particular, what are parents' experiences of their involvement and role in this process?

What was it like for you? Can you describe/give me an example of your role?

2. How do parents experience the transition in decision-making from themselves to their child (young person)?

Was there a moment when you realised that your child started to have responsibility for their decision regarding the surgery? Can you describe the moment? Where were you, who was with you…?

3. How do parents experience the milestone in their child's (young person's) care?

What was it like for you after years of making decisions about your child's care to reach a time when your child was expected to decide themselves? Do you recall a particular moment when you felt it?

4. Was the process of decision-making what parents expected?

Were there any surprises along the way of making the decision? Can you give me an example?