The experience of families receiving a diagnosis of craniosynostosis for their child

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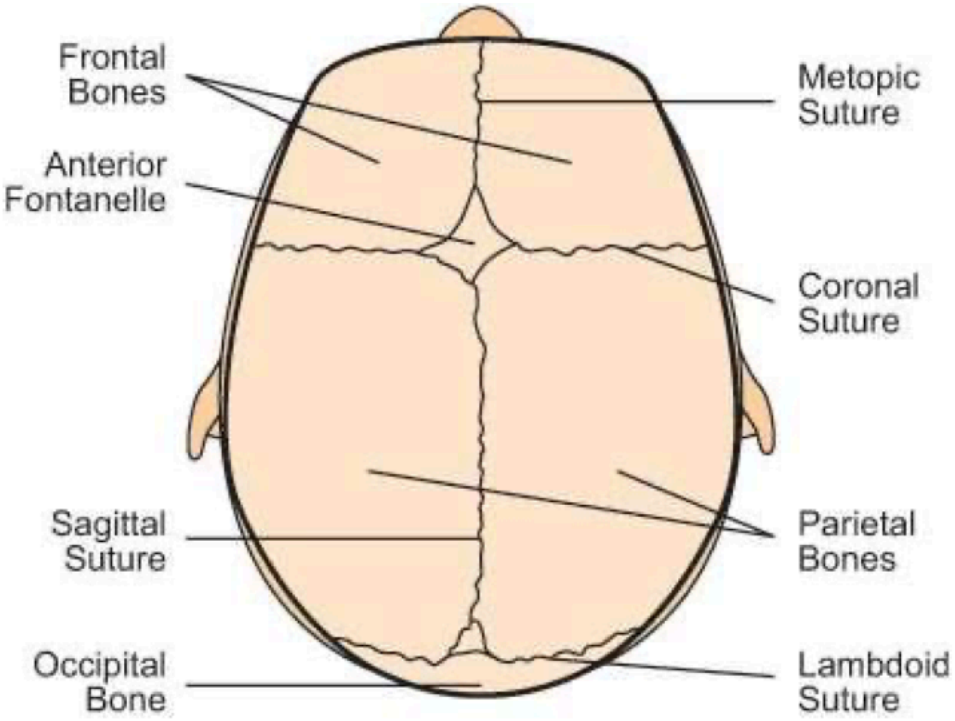
Thesis Overview

A congenital craniofacial anomaly (CFA) is a broad term used to describe a wide range of diagnoses, which affect the head and facial bones or form part of a wider genetic syndrome.[1] Prevalence rates and characteristics vary across the different craniofacial conditions, with some diagnoses considered more common (such as cleft lip and/or palate) than others (such as craniosynostosis).[2] The following thesis is specifically interested in one of the rarer CFAs - craniosynostosis.

Figure 1. Skull of the Newborn.[3]



If we consider Figure 1, the skull is comprised of several ‘plates’ of bone which are not tightly joined together at birth to allow for brain growth. The lines at which the plates meet are termed ‘sutures’.[3] As an individual develops, these sutures fuse together. However, craniosynostosis occurs if one or more of the sutures have fused

prematurely before birth.[4] This can affect the area and direction at which growth can occur, resulting in an abnormal head shape. The rate of which the condition is detected can differ but is primarily soon after a child is born.[5]

Craniosynostosis is classified as either syndromic (involving multiple sutures shown in Figure 1; often an underlying genetic cause; accompanied by physical features affecting other parts of the skull, face or body) or non-syndromic (typically involving only one suture from Figure 1; minimal impact on other parts of the body).[6]

Whilst the diagnosis will impact each child to varying degrees, craniosynostosis often requires multidisciplinary treatment throughout childhood and into adulthood.[7] Parents can therefore find themselves immersed in an unexpected healthcare journey, which can be emotionally demanding and stressful.[8] Indeed, there is literature within the paediatric population to suggest adjusting to a child's health needs may affect the psychological wellbeing of parents, or the family as a whole.[9,10]

Currently, there are four specialist craniofacial centres in England and one in Scotland. The evolution of this thesis project stemmed from discussions within these centres about developing greater support for families that access craniofacial services. In order to do this, an understanding and awareness of families' experiences was warranted and prompted the following chapters:

1. A mixed-methods systematic review that aims to synthesise the existing literature about the experiences of families caring for a child with craniosynostosis.

2. An empirical project exploring parental experiences of a particular type of craniosynostosis – non-syndromic. Grounded theory methodology was utilised to analyse the qualitative accounts of fifteen parents. The project aimed to extend the previous literature synthesised in the systematic review by (i) identifying risk and protective factors that contribute to adjustment and psychological wellbeing in parents whilst (ii) specifically focusing on the time of diagnosis. In this chapter, a theoretical model is presented to summarise these experiences.

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Chapter 1

Families' experiences of caring for a child with craniosynostosis: A mixed-methods systematic review*

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Abstract

Objective: Receiving a diagnosis of a congenital craniofacial anomaly (CFA) not only affects the psychological health of the individual themselves, but also the wider family unit. Previous reviews have either focused on the most common CFA, cleft lip and/or palate, or grouped all CFAs together. This review summarises the evidence base for a specific CFA diagnosis – craniosynostosis – to answer the question: What are the experiences of families caring for a child with craniosynostosis?

Design: Five electronic databases were searched. Extracted data from eligible studies were synthesised using an integrated framework for mixed-methods systematic reviews. The quality of included studies was critically evaluated using the Mixed Methods Appraisal Tool.

Results: Twenty papers pertaining to families' experiences of craniosynostosis were identified. Findings are presented according to emotional, social and service-related experiences. Caregivers experienced a range of difficult emotions including frustration, stress and anxiety. Support from family, friends, peers and staff were crucial in navigating the healthcare journey. This included the prospect of surgery, accessing information and attending regular appointments.

Conclusions: Craniosynostosis has a significant and varied psychological impact on families, spanning from detection of the condition through to long-term treatment, and is accompanied by a range of emotional and practical support needs. The review identifies clinical implications, methodological challenges and areas for further research.

Keywords: craniosynostosis, family, parent, craniofacial, psychosocial, treatment, systematic review

Introduction

Craniosynostosis is a diagnosis that falls under the umbrella term of 'congenital craniofacial anomalies' (CFA), which refers to a wide range of conditions that are present from birth and affect the form and function of the head and face.[1] Craniosynostosis is the third most common CFA, after cleft lip and/or palate and craniofacial microsomia.[11] It is characterised by the premature fusion of one or more sutures of the skull and can be divided into two classifications: syndromic and non-syndromic. Most syndromic craniosynostoses have a genetic origin, are associated with other difficulties, and frequently involve multiple sutures. The most common include Crouzon, Apert, Pfeiffer, Muenke and Saethre-Chotzen syndromes.[12] In comparison, non-syndromic cases typically involve a single suture; the most common types being sagittal, unicoronal, bicoronal, metopic, and lambdoidal.[6] Non-syndromic diagnoses are more prevalent, accounting for 45-60% of detected craniosynostosis cases.[13]

The current management of the condition tends to be surgical correction within the first year of life. Typically, there are two methods of surgical intervention: minimally invasive surgery (e.g., endoscopic) and open surgery (cranial vault remodelling). This is often followed by multidisciplinary treatment throughout childhood and into early adulthood.[14] As with other rare conditions,[15] individuals diagnosed with craniosynostosis can be exposed to challenges with social interaction, well-being, cognitive development, appearance, treatment and overall quality of life.[16-18] The role of the caregiver has been shown to help navigate and potentially mediate the impact of these challenges in paediatric health settings.[19,20] It follows that an understanding of families' experiences is important for both child and familial adjustment.

So far, the impact of a craniosynostosis diagnosis has been qualitatively explored in parents and the literature summarised in two prior reviews.[10,21] These indicate an adaptation process in which parents experience a range of difficult emotions, such as guilt, disappointment and anxiety.[21] Quantitative studies, on the other hand, are less conclusive. Whilst a number of studies report increased levels of stress in parents of children with CFAs, others find no differences.[10,21] The inclusion of multiple types of CFAs in these reviews means various conditions were mixed within samples. This is understandable given the limited literature available in any one area, but makes it difficult to compare between diagnoses.[14]

A seminal paper by Nelson et al[22] instead chose to focus on one particular population and completed a review on parental experiences of cleft lip and/or palate. This showed how research has tended to focus on negative emotional reactions in parents, such as grief, shock and worry, as opposed to positive aspects. Parents were seen to report challenges in coping with others' reactions to their child's appearance. In addition, service-related experiences encompassed parental wishes for greater information and involvement in decision making about treatment.

Relative to research into cleft lip and/or palate, a focus on other CFAs remains scarce. Of the literature that is available, previous reviews have grouped rarer CFAs together. This prompted the present review. To the author's knowledge, no prior reviews have exclusively focused on craniosynostosis in the context of the family perspective. The present review therefore summarised all available literature on the

topic, irrespective of methodology. The aim of the present review was therefore as follows:

Aim

To provide a comprehensive synthesis of the qualitative and quantitative literature available to help answer the question: What are the experiences of families caring for a child with craniosynostosis?

Method

A protocol was registered with the PROSPERO database (CRD42022349537) and the paper written in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.[23] Prior to conducting this review, provisional literature searches were undertaken and researchers in the field were contacted to ensure this review was not a duplication.

Search Strategy

Search terms were generated using the PiCo tool[24] (Table 1) and used in the following databases: MEDLINE, Scopus, Web of Science, CINAHL and PsychINFO. Literature searches were carried out in September 2022 and later replicated in July 2023 to identify any further eligible papers that had been published more recently. At this time, no new studies were identified. Hand-searching took place in key journals (The Cleft-Palate Craniofacial Journal, Psychology & Health), cited references in eligible papers and key review articles. One well-established craniofacial researcher (NS) was also contacted to advise on any key research studies in the topic area.

Table 1. PiCo tool and Search Terms.

Domain		Criteria	Search Terms
P	Population	Family members/caregivers	Craniosynostosis OR craniosynostos* OR craniofac* OR cranio-facial AND Child* OR infant OR pediatric* OR paediatric* OR "young people" OR "young person*" OR "young adult" OR teen* OR adolescen* OR juvenile* AND Parent* OR mother* OR father* OR famil* OR caregiver*
I	Interest	Any experiences relating to craniosynostosis	
Co	Context	Child with diagnosis of craniosynostosis	

Eligibility Criteria

The inclusion criteria for this review were: (a) studies which focused on the experiences of caregivers or family members, (b) related to a child with any type of craniosynostosis diagnosis, (c) papers relating to any stage of a families' craniofacial journey, (d) peer-reviewed, primary research, (e) papers available in English and (f) reporting qualitative or quantitative data, or mixed-method studies.

Date parameters were not applied to the search strategy as no previous systematic review had previously summarised families' experiences of craniosynostosis.

Papers were excluded if they were unpublished dissertations/theses, posters, commentaries, opinion pieces, posters or studies not subject to peer-review. Any

animal studies were also excluded. Additionally, exclusion criteria included papers solely focusing on the child's experience, investigating other craniofacial conditions (such as cleft lip and/or palate) or material only available in languages other than English.

Study Selection

The search yielded a total of 2,871 articles. A further 6 articles were identified by a combination of citation searching and suggestions from a researcher in the field (NS). Reference management software EndNote X9 assisted in the removal of 1516 duplicates. The remaining articles were screened by title and abstract applying eligibility criteria using Rayyan, an online review software platform. This left 31 papers for full-text screening, of which 20 were deemed suitable for the review. Figure 2 captures the screening process including reasons for exclusion. Two external reviewers (EG, BC) were provided with the review eligibility criteria and independently screened 10% of articles at both title/abstract and full-text stages. There were no discrepancies in the decisions made.

Quality Assessment

Articles were quality assessed using the Mixed Methods Appraisal Tool (MMAT; Appendix 2).[25] The MMAT was chosen because it was designed to critically appraise various research designs across qualitative, quantitative and mixed method studies. An overall quality score was given using stars (ranging from 1* = 20% quality criteria met, to 5* = 100% quality criteria met; see Appendix 3 for scoring guidance). An independent reviewer (EG) appraised 10% of eligible articles chosen at random. There

were two discrepancies in scoring, in relation to which research design had been employed. Any disagreements were resolved through discussion.

Data Extraction

Information was extracted from the included studies using a data extraction tool developed by the first author (Appendix 4). The independent reviewer (EG) completed extraction for 10% of the studies to establish validity. Table 2 details the key characteristics of the final 20 eligible papers.

Data Synthesis

The use of mixed systematic reviews has been gaining traction in recent years.[26] This approach to reviews is considered advantageous for gaining a broader and comprehensive knowledge of a topic area due to the inclusion of various methodologies. Moreover, this was deemed suitable as capturing all existing research about families' experiences of craniosynostosis has not previously been summarised in a systematic review.

Several frameworks have been proposed through which to conduct mixed systematic reviews.[27,28] The present review was undertaken in accordance with the convergent integrated framework proposed by JBI, formerly known as the Joanna Briggs Institute, who have issued methodological guidance on this review approach.[29] An integrated framework assumes that both quantitative and qualitative data can address the same review question and be assimilated into a single synthesis. It allows research findings from all eligible studies to be synthesised together, as opposed to a more segregated approach.[28,29] This was beneficial to the present review as one type

of data could then contextualise and explain the findings of the other type of data, rather than presenting each methodology in isolation. Using this framework, papers are grouped for synthesis using findings that address the same phenomenon and not by study methods.[28]

For this to occur, all data must be transformed into the same format. JBI recommends converting quantitative data into qualitative data (i.e., qualitising).[26] This is because coding quantitative data into 'textual descriptions', such as themes or categories, is considered less error-prone than giving numerical values to qualitative data.[30] As per JBI[26,29] guidance, this involved data and study outcomes from quantitative studies being transformed into categories and narrative interpretations. These data were then integrated and pooled with the findings extracted directly from the qualitative papers. The first author examined the integrated dataset to identify themes, based on similarity in meaning, that helped answer the review question. These themes were identified as emotional, social and service-related experiences (Appendix 5). The transformation of data was checked and refined through discussions with the independent reviewer (EG). Finally, a narrative description summarising the themes was produced which forms the results section of this paper.

Figure 2. PRISMA Diagram.

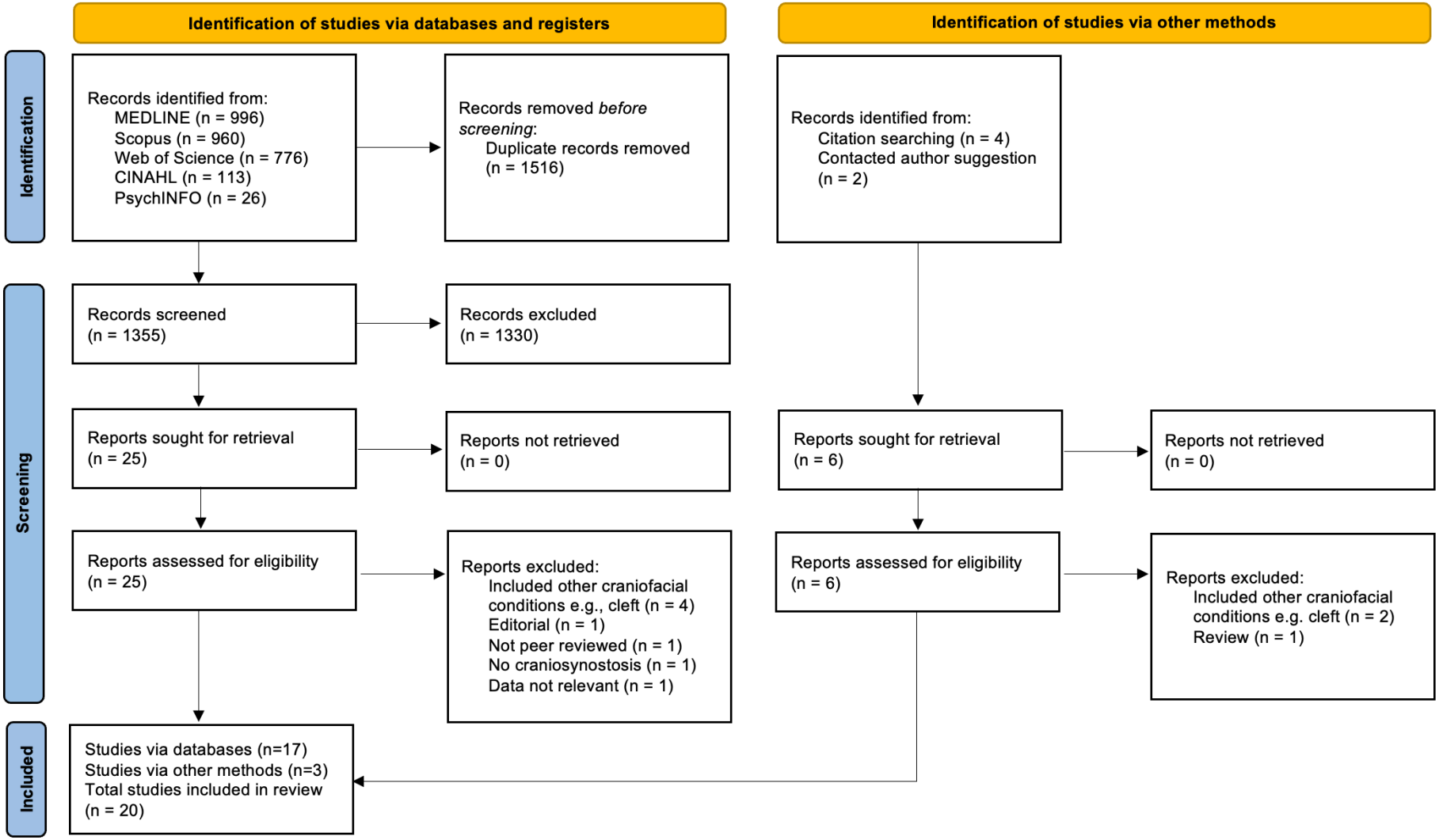

Table 2. Key Characteristics of Included Studies.

	Author(s) and year	Participant characteristics	Country	Method	Focus and aim of the study	Key findings	Study setting	Measures	Quality
Mixed methods									
1	Costa et al., (2022)	Mothers (n = 109) and fathers (n = 9) of children aged 3 months to 49 years with non-syndromic (63%) or syndromic (33%) craniosynostosis. Children and parents had a mean age of 10 and 40.9 years respectively.	UK	Mixed-methods survey	Psychosocial adjustment Aim: to conduct an initial investigation of psychosocial adjustment among parents of individuals with craniosynostosis	In comparison to the general population, parents of individuals with craniosynostosis reported higher levels of stress, anxiety and depression. The same parents reported lower levels of resilience and optimism. The majority of parents (92.8%) reported feeling upset at the time of diagnosis, in addition to shock (73.9%), worry (99.1%), guilt (68.4%) and blame (10.8%). Parents' experiences included missed/delayed diagnosis and dismissal of concerns. Parents	Online	- Conor-Davidson Resilience Scale (CD-RISC10) - Perceived Stress Scale (PSS) - Hospital Anxiety and Depression Scale (HADS) - Strengths and Difficulties Questionnaire (SDQ) - Revised Life Orientation Scale (LOT-R) - Healthcare experiences (open-ended) - Parent well-being (open-ended) - Child Wellbeing (open-ended) - Relationships (rated on Likert scale)	*****

						discussed the impact on their own wellbeing. Some positive outcomes were noted, such as satisfaction with craniofacial specialists, but there was also recognition of the need for more information/support than what is currently available for families.			
Quantitative									
2	Coulter et al., (1991)	Parents of 27 children with "pure craniosynostosis", which was defined as non-syndromic. Age ranges for children were 2 months – 15 years, with a mean age of 3 years. Ethnicity data included: 24 White, 2 Black and 1 Asian.	USA	Survey	Stress Aim: to assess psychological and emotional stress in parents of children with craniosynostosis	There were certain factors associated with higher levels of parental stress: number of older children in the home, poor current health of participant, using transportation other than one's own on a regular basis, not having prior experience caring for a person with additional needs, having someone to help care for child, dissatisfaction with	Private craniofacial centre	- Questionnaire on Resources and Stress (QRS) - Family Characteristics Form, adapted from Maternal Social Support Index	***

						the amount of time clinic staff spend with child.			
3	Gray et al., (2015)	Parents of children with unoperated single-suture craniosynostosis (cases) and those without the diagnosis (controls). For cases, 247 mothers and 211 fathers participated at first visit. Corresponding numbers for controls were 254 mothers and 220 fathers. The majority of children were male (63.9% cases and 63.7% controls) and White (74.7% cases, 70.7% controls).	USA	Prospecti ve	Stress Aim(s): To examine longitudinal differences in reported stress between parents of children with and without single-suture craniosynostosis and to compare the stress reports of mothers and fathers	No significant differences in caregiver stress between children with or without craniosynostosis. In general, mothers reported greater stress than fathers.	Clinical visits at three time points	Parenting Stress Index (PSI)	*****
4	Kilipiris et al., (2022)	Parents of 256 children with operated single	Slovak Republic	Survey	Experiences of service	High satisfaction from parents about the use of virtual follow-up of	Online	9 question survey about satisfaction level for	**

		suture (n=246) and syndromic (n=10) craniosynostosis. Just over half of children were male (56.3%).			Aim: to evaluate the satisfaction level of parents from telemedicine use in the long-term follow-up of children operated for craniosynostosis during the COVID-19 pandemic	children operated for craniosynostosis. Vast majority answered that they would prefer in-person visits in the future.		telemedicine, devised for this study	
5	Kim et al., (2008)	47 parents of children with operated non-syndromic craniosynostosis. Sample included 36 mothers and 10 fathers, with one participant classed as 'unlisted'. Children were mostly male (n=29), with the remaining female (n=18). A total of 32 participants	USA	Survey	Stress Aim: to determine if there is a difference in parenting stress status-post open versus minimally invasive craniosynostosis procedures	There was a decrease in total stress from the perspective of parents with children who had minimally invasive surgical procedures, when compared to open procedures.	Hospital	Parenting Stress Index-Short Form (PSI-SF)	****

		identified as White, 7 as Hispanic, 1 African American, 1 Iraqi and 6 identifying as 'Other'.							
6	Kluba et al., (2016)	Parents of 46 children with operated craniosynostosis. This encompassed both single suture and syndromic. Children were mostly male (n=26), with the remaining female (n=20).	Germany	Survey	Experiences of service Aim(s): to examine parental perceptions of the quality of medical service, communication and aesthetic outcomes	Parents were satisfied with the craniofacial service and all indicated that they would opt for surgery in retrospect. Parents perceived the communication pre- and postoperatively, as well as at discharge, to be 'perfect' or 'largely appropriate'.	Medical setting	Questionnaire devised for this study looking at quality of service, communication and surgery outcomes	**
7	Nieroba & Larysz (2020)	54 parents of children with craniosynostosis. Diagnoses were predominantly non-syndromic. Sample consisted of mainly mothers (95%), with an average age of 32.25 years.	Poland	Survey	Support Aim(s): to expand knowledge of how social media support groups function, and what impact they have on members	The main reason for joining support group was to seek information on their child's diagnosis. One of the benefits of the online group was the feeling of being better informed. Thanks to the group, some parents felt	Online	Survey about use of support group, devised for this study	****

		Children were mostly male (59.3%), with an average age of 25.8 months.				more confident for meeting with doctors; felt less alone; and had increased optimism for the future.			
8	Rosenberg et al., (2011)	Parent dyads (n=246) of children with unoperated single suture craniosynostosis and matched controls (n=253). The majority of both cases and controls were male (64%), with an average age of 7.3 months.	USA	Case control	Stress Aim: To compare relative levels of stress reported by mothers and fathers of children with and without cranio-synostosis	Parents reported similar levels of stress to controls. Parents, regardless of case/control status, showed a consistent pattern in which mothers reported higher parent-related stress than fathers.	Medical setting	PSI	*****
9	Rotimi et al., (2021)	Parents (n=59) of children with operated craniosynostosis. Mean age of children was 7.8 years (range 3 months to 22 years), with 36 non-	UK	Survey	Parental anxiety Aim: to investigate parents of children with craniosynostosis regarding anxiety in under-	Vast majority of children were involved in sporting activities. Parental anxiety increased with increasing intensity type of sport.	Medical setting	Questionnaire on sporting activity and parental anxiety, devised for this study	****

		syndromic and 23 syndromic cases.			taking sporting activity				
10	Sarimski (1998)	41 parent dyads of children (22 female, 19 male) with Apert syndrome, who had undergone surgery.	German y	Survey	Stress Aim(s): to assess the emotional and behavioural status of children with Apert syndrome and examine parental stress factors	Difficulty with acceptance of child's appearance, behavioural problems, and low parental self-esteem were associated with higher parental stress levels.	Postal questionnai re	- Society for the Study of Behavioural Phenotypes Postal Questionnaire (SSBP) - PSI - Family Functioning Style Scale - Vcrhaltensbeuncilun gsbogen fix Vorschulkinder (VBV) - Child Behaviour Checklist (CBCL)	***
11	Shaw et al., (2022)	Parents of 462 children (325 male, 136 female) with operated single suture craniosynostosis.	UK	Survey	Surgery Aim: to assess the opinion of children and their families regarding coronal access incisions and their perceived psychosocial impact	Majority of parents were not 'bothered' by appearance of surgical scar. Parent perceptions improved with age and therefore time post-surgery.	Medical setting	Questionnaire relating to perceptions of scar, devised for this study	***

12	Tang et al., (2022)	Caregivers (n=106) of children with craniosynostosis. This consisted of 62 mothers, 40 fathers, 1 grandparent and 3 listed as 'other'. Caregivers were predominantly White (84%). Other ethnicity data reported: Black or African American (10%), Hispanic or Latino (4%), Native American/America n Indian (1%) and Other (1%). Only 3 caregivers noted their child to have syndromic craniosynostosis.	USA	Prospecti ve	Stress Aim: to compare caregiver stress in children with craniosynostosis at diagnosis and postoperatively	Stress decreased at 3 months following surgical intervention, but this was not sustained at 6 month follow-up visits. Female sex, uninsured status and open surgery predicted higher stress levels in caregivers. Syndromic craniosynostosis was associated with higher stress levels across all time points.	Clinical visits at three time points: pre-operatively, 3 and 6 months post-operatively	- PSI-SF - Pediatric Inventory for Parents (PIP)	***
13	Wong-Gibbons et al., (2009)	82 mothers of children with craniosynostosis. Children were predominantly male (n=52).	USA	Case control	Experiences of service Aim: to examine maternal perceptions of satisfaction with	Majority of mothers reported to be satisfied with care received from professionals.	Medical centres	Authors state structured telephone interview	***

		The mean ages of children and their mothers were 4.4 and 34.8 years, respectively.			medical and surgical care, treatment outcomes, perceived overall health of their child, and access to team care and support	About one half of mothers received "excellent answers" to questions regarding diagnosis and treatment options.			
Qualitative									
14	Dangsomboon & Jirapaet (2017)	Mothers (n=14) of children with operated syndromic craniosynostosis. Mothers were aged 25-39 years.	Thailand	Interviews	Post-operative experiences Aim: to examine the lived experience of caregivers having children with craniosynostosis using distractor devices	Mothers described caring for their child after surgery as scary and challenging. For example, looking after the wound or keeping up with follow-up appointments. Mothers reported a variety of emotions, ranging from hope to fear. To help with this, support from family, friends or healthcare teams was fundamental. Mothers also mentioned using a positive mindset.	Hospital		*****

15	Kuta et al., (2020)	Mothers (n=12) of children newly diagnosed with single suture craniosynostosis. The mean age of mothers was 32.4 years, with a range of 19-42 years. Children were predominantly male (n=10).	Canada	Telephone interviews	Experiences from diagnosis - postoperative period Aim: to examine the psychosocial experience of families with a child diagnosed with craniosynostosis	During the pre-operative interviews, mothers shared frustration with diagnostic delays. They discussed the importance of knowing what to expect and the struggle to reach a decision about surgery. In the post-operative interviews, mothers shared fear and how professionals helped them cope with this emotion. All mothers expressed a sense of relief following surgery.	Health centre		*****
16	Letourneau et al., (2003)	A total of 18 parents representing eleven families (11 mothers, 7 fathers) of children with operated non-syndromic craniosynostosis.	Canada	Focus groups	Surgery decision making Aim(s): to explore the process of parental decision making regarding surgery and the information/ strategies that	Parents described varying degrees of difficulty in reaching a decision about surgery. They spoke about an active search for information about the diagnosis itself and treatment. Several sources of information were mentioned, although	Craniofacial clinic		**

					help facilitate this	there were mixed feelings about their helpfulness. Parents thought about what life would be like for their child in the future when making decisions about surgery. Parents mentioned certain events that solidified their decision, such as people staring at their child or asking what was 'wrong'.			
17	Netherton et al., (2021)	Mothers of four children (2 males) diagnosed with Apert syndrome, all of which identified as White. The mean age of children was 15 years and they had all received some type of surgery.	UK	Interviews	Adjustment Aim: to explore psychological adjustment to Apert syndrome from the perspectives of young people and their parents	Parents described the early years as being the most difficult period. As time progressed, there was an ongoing journey of learning about and accepting child's diagnosis. One of the biggest barriers to this acceptance was the navigation of healthcare services, both in terms of	Craniofacial centres		*****

						physical access and the content of the appointments themselves. Parents had used various techniques to facilitate adjustment, and particularly the role of social support was emphasised.			
18	Zeytinoglu-Saydam et al., (2021)	Parents (n=21) of 12 children with Apert syndrome, who had undergone surgery. Parents' ages ranged between 29 and 49 years. The age of the children ranged between 1-12 years, with a mean age of 5.6 years. The majority of the children were male (n=14).	Turkey	Interviews	Social experiences Aim: to explore the social experiences of parents raising children with Apert syndrome, including the difficulties they face and the strategies they use to cope	Parents reported social stigmatising experiences, such as stares and questions when out in public, as the most significant challenges. Parents stated that their own acceptance helped them to either educate people, or just ignore these reactions. Social support, religion and actively promoting their child's social inclusion were all identified as factors that help in navigating challenges associated with visible difference.	Hospital		*****

19	Zerpe et al., (2020)	10 parent dyads of children with operated non-syndromic craniosynostosis. The mean age of parents was 35 years, with a range of 27-43 years.	Sweden	Telephone interviews	Pre-operative experiences Aim: to investigate parents' experiences of having a child with craniosynostosis and their perceptions of care	The themes identified represented a timeline of events from the detection of an abnormal head shape to waiting for surgery. The knowledge of parents varied, although many had searched the Internet and/or social media. Most parents had experienced heightened worry and thoughts prior to their appointment with the craniofacial team. The same also applied when waiting for surgery. Various factors seemed to help this, such as positive experiences with the specialist team or social support.	Craniofacial clinic		*****
20	Zerpe et al., (2022)	19 parents (11 mothers, 8 fathers) of 12 children with	Sweden	Interviews	Post-operative experiences	There was relief following surgery, especially after	Craniofacial clinic		*****

		operated non-syndromic craniosynostosis. The mean age of parents was 35 years, with a range of 26-40 years.			Aim: to explore parents' experiences of hospital care after their child's craniosynostosis surgery and their perception of support during the year after discharge	seeing their child sad, afraid or in pain. Parents were at risk of feeling alone during their child's hospital stay but did note the kindness of staff. The importance of information and support was highlighted, although sometimes parents found it difficult to ask for help. Some parents remained worried about risks during recovery and long-term consequences on child development.			

Results

Study Characteristics

Twenty papers were included for analysis. Of these, one was a mixed-method study,[31] twelve were quantitative[32-43] and the remaining seven used a qualitative design.[44-50] All qualitative papers used semi-structured interviewing techniques, with the exception of one which utilised focus groups.[48] Qualitative studies either took place in person,[44,48,50] via the telephone,[45,47] or a combination of the two.[46,49] The approach to qualitative data analysis varied between studies. Three used thematic analysis,[45,47,49] two adopted a phenomenological approach,[44,50] one used thematic content analysis[48] and another used inductive content analysis.[46] Quantitative methodology tended to include surveys using previously validated measures[31-33,35,38,40,43] or questionnaires that had been specifically designed for the purposes of the study.[31,34,36,37,39,41] Two of these quantitative studies adopted a longitudinal design.[33,43]

The populations ranged widely. Six papers originated from the USA,[32,33,35,38,42,43] four reported findings from a UK-based population,[31,39,41,50] two each from Germany,[36,40] Canada[47,48] and Sweden[45,46] and individual papers reported from Slovak Republic,[34] Poland,[37] Thailand[44] and Turkey.[49] Participants were predominantly recruited within clinical settings, with 16 papers mentioning hospitals, health centres and/or craniofacial clinics.[32,33,35,36,38,39,41-50] The remaining samples were recruited via the Internet[34,37,51] or a postal questionnaire.[40]

In terms of diagnosis, six papers included both syndromic and non-syndromic types of craniosynostosis.[31,34,36,37,39,43] An additional nine studies solely focused on

non-syndromic craniosynostosis,[32,33,35,38,41,45-48] four papers recruited caregivers of children with syndromic craniosynostosis[40,44,49,50] and one paper simply stated craniosynostosis in general.[42] Similarly, there was variation in the detail provided about any surgical procedures across the included studies. Twelve papers stated that surgery had already taken place,[34-36,39-41,44-46,48-50] one paper specified no surgical intervention[38] and two papers gathered data at both pre-operative and post-operative time points.[33,43] The remaining studies either did not explicitly specify surgical data[31,32,42] or had a mixed sample whereby only some had undergone surgery.[37,47]

The sample sizes differed according to the methodology used. The smallest sample comprised 4 participants using a qualitative approach,[50] and the largest consisted of 462 participants utilising a survey design.[41] Four studies exclusively recruited mothers,[42,44,47,50] whereas one study recruited more broadly and included mention of grandparents.[43] The other studies aimed to recruit both mothers and fathers, although in these cases samples were still predominantly female. Children were predominantly male, which is reflective of what is known about the incidence of craniosynostosis in the medical field.[52,53] Only five papers reported ethnicity data, with the majority of participants identifying as White.[32,33,35,43,50]

Evaluation of Quality

The findings of the quality assessment are displayed in Table 2 and further detail can be found in Appendix 6. No studies were excluded based on the MMAT, as using overall quality scores to determine inclusion/exclusion can result in important findings being discounted.[54] Instead, consideration has been given to the quality of each study and used to contextualise the subsequent synthesis. Of the twenty papers

appraised using the MMAT, nine studies were rated as 5*,[31,33,38,44-47,49,50] three studies had a score of 4*,[35,37,39] five studies rated as 3*[32,40-43] and three papers received a score of 2*.[34,36,48]

Those with higher overall scores tended to have well-reported sample and methodological information. Their findings were adequately derived from the data and the reader could easily follow the links between data collection, analysis and interpretation. The most common weaknesses for those papers with lower scores were insufficient detail given to recruitment, methodology and demographics. For this reason, attempts were made to contact the authors of one paper[42] and request supplementary material to establish further details about what research design was used. In the absence of a response, the paper has been quality appraised based on the limited information available within the published manuscript. The heterogeneity of the studies reviewed will be referred to and discussed herein.

Emotional Experiences

This first theme covered the various emotional reactions experienced by parents. These were noted across the healthcare journey, ranging from frustrations when seeking a diagnosis to post-surgical worries.

Detection

Three papers qualitatively captured how craniosynostosis was detected in children.[31,45,47] Across each, it was the parents that predominantly noticed the abnormal head shape of their child at birth. Two of the studies referenced how parents became increasingly frustrated as they were offered explanations that the head was

compressed as a result of the birthing process, and it would correct itself over time.[45,47] This resulted in feeling like any concerns they raised were dismissed and often meant diagnoses were delayed. Families were left seeking medical advice from numerous professionals or requesting referrals themselves. This was depicted by the theme 'a need to advocate' in the paper by Costa et al[31] whereby one parent stated they had contacted three general practitioners, two midwives and three health visitors before a specialist referral was made. Interestingly, the three papers that discussed the detection of craniosynostosis were from three different countries – UK, Canada and Sweden – indicating that diagnostic delays and parental frustrations may be shared across healthcare systems.

Difficult Emotions

Approximately half of the included quantitative papers investigated parental stress, albeit with a variety of measures.[32,33,35,38,40,43] Two studies[33,38] found no significant differences in the stress levels among parents of children with craniosynostosis, when compared to controls. In contrast, Costa et al[31] indicated that parents experienced higher levels of stress, anxiety and depression compared to the general population. They also found that parents of individuals with craniosynostosis reported lower levels of resilience and optimism.[31] A great proportion of this sample recruited by Costa et al[31] were mothers which did not allow for consideration of gender differences. Previous studies however seem to suggest higher stress levels in mothers.[33,38,43]

Moreover, three papers[32,40,43] focused on factors associated with higher parental stress levels. These papers were all quality appraised as lacking detail in

regard to samples and differed in terms of the type of craniosynostosis recruited, making it difficult to compare findings across diagnoses. Coulter et al[32] found increased stress was associated with poor current mental health of the parent, a higher number of children in the home, not having their own transportation, no prior experience of caring for additional needs, having someone to help support them in their caring responsibilities, and dissatisfaction with clinic staff. Sarimski[40] identified difficulty with accepting the child's appearance and low parental self-esteem as contributory factors to increased stress. Additionally, Tang et al[43] found that higher stress levels in caregivers were predicted by open surgical procedures. The latter was echoed in another study[35] which found open surgeries were more stressful when compared to minimally invasive procedures.

Qualitative accounts provided information to contextualise this stress. Parents reported worry and fear at not knowing what to expect from the future and the enormity of the surgical procedure,[46] plus the potential for their child to experience developmental delay.[47] They described anxiety at their children engaging in activities in case they were to hurt themselves.[47] This may in part also explain a pattern identified by Tang et al[43] whereby parental stress initially decreased following surgery, but was not sustained at a 6-month follow-up. Emotions that last post-surgery may reflect parental fears about their child's physical health, neurological and/or social development.

Mention of parental wellbeing within the included papers were largely related to distress or negative emotions. There was only a brief mention in two qualitative papers about positive feelings experienced by caregivers, namely hope[44] and gratitude[31].

These focused predominantly on the prospect of surgery, as parents expressed appreciation that treatment options were available and the hope that a procedure would help their child.

Social Experiences

This second theme encapsulated all experiences related to a social setting. This was a positive experience when parents were supported by loved ones and/or felt understood, but more difficult when receiving judgement from others.

Challenges

A smaller body of research has offered insight into the social challenges that families may face. This perhaps marks a move away from exploring social difficulties with the individual, and instead inviting a parental perspective. Both papers in the present review doing this were qualitative, of high quality and focused on Apert syndrome.[49,50] The main challenge for parents were receiving stares, unkind comments and/or questions about their children when out in public. They reported finding these intrusive, judgemental and a reminder of their child's visible difference. In response to this, Zeytinoglu-Saydam et al[49] suggested that parents either try to ignore these reactions or attempt to educate others. Caregivers also spoke about aiming to empower their child, encouraging them to socialise and be proud of their appearance.

Support

Social support was noted as a protective factor for parents. Family and friends were frequently mentioned as not only helping emotionally but also practically, such

as with childcare.[44,50] Zeytinoglu-Saydam et al[49] also described the importance of spousal support. Within this study, parents felt understood and comforted by their spouses as they could relate to everything the other was experiencing. For the same reason, seeking guidance or support from other parents in the same situation was emphasised as valuable in five papers.[37,45-48] In the study undertaken by Kuta et al[47], parents felt it was important to hear the stories of families who had been through the same process previously. This prompted them to reach out to online support groups. Nieroba and Larysz[37] evaluated a craniosynostosis group via social media and highlighted how this made parents feel less alone and better informed. Whilst these studies suggested that parents seek out these groups of their own accord, families in Letourneau et al's[48] research were offered the option by their craniofacial clinic to contact other parents who had experienced surgery before, to help with their own medical decision-making.

Any negative experiences of support were given less research attention. One study made a subtle reference to how parents could feel a disconnect with friends and family if they did not feel understood or did not make an effort to help.[31] Moreover, two papers mentioned that gathering information from others also had the potential to overwhelm and increase feelings of anxiety; especially if hearing about complications.[47,48]

Service Experiences

This third, and final, theme summarised parental satisfaction with their child's medical care. This care comprised of different elements; such as information, treatment experiences and outcomes, plus the perceived burden of care experienced by attending appointments.

Overall Satisfaction

The reviewed studies highlighted how parents perceived their child's medical care. In the paper by Wong-Gibbons et al,[42] the authors stated that many parents were "very satisfied" with the healthcare that was provided at the time of diagnosis. The limited clarification on what other responses could have been available to participants and/or further details about what parents found particularly satisfying was one of the reasons for the lower quality appraisal of this study. In other papers, parents supplied more detail and named having access to psychology[45] or a named contact to which they could ask questions[47] as positive aspects of their service experience.

Furthermore, two groups of researchers wrote about the service experiences of parents post-surgery.[34,46] Again, parents mentioned the ability to call the ward or craniofacial nurse as comforting, which implied families can be left with worries or questions that they appreciate being answered by a specialist. In addition, the survey by Kilipiris et al[34] explored parental satisfaction of virtual follow-up consultations. Whilst the majority of parents reported a high satisfaction rate for telemedicine, only a small proportion said they would opt for a virtual appointment in the future. This indicated that perhaps parents appreciate the more 'human' parent-clinician

relationship, although this conclusion is inferred as Kilpiris et al[34] did not present any qualitative responses to support this.

In contrast, several papers highlighted potential reasons for parental dissatisfaction. This involved both non-specialist health professionals (such as paediatricians and general practitioners) and specialist craniofacial teams. For Costa et al.,[31] dissatisfaction was directed more at non-specialists about service-level delays in treatment. Parents interviewed by Zerpe et al[46] felt specialist health professionals had left them on their own after being discharged from hospital, although did not always find it easy to ask for help. Furthermore, Kuta et al[47] found meeting the craniofacial team was overwhelming for some parents. This was often due to being in a novel medical environment or the number of professionals involved in the consultation, to the point where parents found it difficult to process information or express themselves.

Information

A large component of parents' experiences of services was the degree and quality of information they were provided with. Almost half of the included papers made reference to either gathering or receiving information.[31,36,37,42,45-48,50] Of these, two quantitative papers suggested parents were satisfied with the information and communication from healthcare professionals.[36,42] Quality appraisal highlighted that these findings were gathered using measures devised for the purpose of each study. The use of non-standardised measures could raise questions about reliability, but it is encouraging that both papers drew similar conclusions.

Moreover, data collected by Nieroba and Larysz[37] showed that the main purpose of parents joining an online support group was to seek information. This potentially suggested a parental desire to gather more information, beyond that supplied by the medical setting. This was echoed by Zerpe et al[46] and again by the open-ended responses within Costa et al's[31] survey whereby parents commented on the need for more information than is currently available. Specifically, they mentioned wanting a timeline for treatment, greater detail on support groups and expected development, plus more in-depth leaflets. Similarly, parents interviewed by Kuta et al[47] thought additional written resources would be beneficial, as a lot of the information seems to be communicated via verbal means. The same sample also requested for staff to signpost them to reputable resources. This may be because some of the information encountered by parents can be frightening[45] and/or misleading.[50]

Parents reported mixed feelings about researching craniosynostosis themselves. In the accounts captured by Zerpe et al[45], at least one parent in most families had been searching the Internet for answers about the condition. Some found this reassuring and comforting, but others found it distressing. Parents within the focus groups facilitated by Letourneau et al[48] identified challenges with information overload, misinformation and an inability to find websites relevant to their country and healthcare system.

Treatment experiences and outcomes

Treatment-related experiences across the studies related to surgical procedures. In most cases of craniosynostosis, surgery is indicated to reduce the risk of elevated intracranial pressure and improve head shape.[55] The decision of whether

to operate at all is sometimes available but dependent on many factors, including the presence of a syndrome. Syndromic cases tend to require multiple surgeries, whereas individuals with non-syndromic craniosynostosis often have a singular operation or, in 'mild' cases, may be offered the option.[56] One paper[48] focused on the latter, families making a decision about surgery, and illustrated the complexities involved in this process. Parents in this study reported varying degrees of difficulty in reaching a decision; drawing on factors such as ethical principles, life experiences, appearance concerns and anticipation of their child's future to help with this.

For those children who underwent surgery, parents reported worries and fears prior to hospital admission.[44,45,47] The accounts depicted by Zerpe et al[46] described struggling with seeing their child sad, afraid or in pain. These worries extended somewhat after surgery. For example, quotes in two papers[31,46] indicated how parents were protective following surgery in case their child hit their head. This was echoed by Rotimi et al[39] in relation to sports, indicating that parents would value more guidance about physical activity post-surgery.

Similarly, parents interviewed by Dangsomboon and Jirapaet[44] found themselves always 'keeping an eye' on their child so they did not cause any damage to the surgical site. The same sample talked of the intricacies involved in wound care, and how being responsible for this was a daunting prospect when thinking about discharge from hospital. Ongoing support from a healthcare team was noted as valuable at this time in three papers,[44,46,47] for the likes of questions or being shown ways in which to provide post-surgical care.

In general, parents seemed satisfied with surgical results and expressed a sense of relief post-operatively. Zerpe et al[46] depicted this as 'the worst part had passed'. One study went on to investigate perceptions of surgical scars.[41] This found that parents did not feel their child's scar was that noticeable and that it also improved with time. Parents surveyed by Kluba et al,[36] in the most part, also reported satisfaction with aesthetic outcomes. It was unclear if this survey had been pilot tested as it had been devised for the purposes of the study, although parents interviewed by Kuta et al[47] reinforced findings by praising the successful changes in their child's head shape, face and/or features. Those who indicated dissatisfaction in the survey[36] had more than one undesired result or surgical complication.

Burden of Care

The navigation of health services sometimes led to additional challenges. These were aptly summarised by Costa et al[31] with the theme 'burden of care' whereby parents spoke of the impact of attending a high volume of appointments, which often required a lengthy commute. This impacted on family life, employment and childcare. Quotes within the Costa et al[31] study mentioned having to fly to appointments, reducing to part-time working hours and emotionally struggling to leave their other children at home. The emotional and physical toll of healthcare was also explored by Netherton et al.[50] Parental accounts within this study highlighted how appointments took place in their nearest specialist centre but the ease of access varied, resulting in one family relocating to reduce disruption and travelling time. The financial cost incurred through travel was also raised in two papers.[44,50]

Discussion

This review is the first to synthesise and critically evaluate the existing evidence base about the experiences of families caring for a child with craniosynostosis. It provides a contribution to the craniofacial literature, which has typically focused upon cleft lip and/or palate or CFAs more generally. Overall, it was striking that a high proportion of the reviewed studies had been conducted in recent years. This indicated an increased interest in the topic area, and perhaps marks a shift in research attention towards craniosynostosis. The review findings were summarised into emotional, social and service-related experiences. Each of these themes will now be discussed in relation to the wider craniofacial and healthcare literature, which captures similarities across other conditions. It is thought that presenting these shared experiences is an step toward increasing knowledge about rare diagnoses in healthcare systems.[15,57-59]

Emotional Experiences

Parents reported a range of difficult emotions such as worry, fear and anxiety. Unsurprisingly, this emotional response is common in parents of children with other long-term conditions within the paediatric population[20,60] and was highlighted within Nelson et al's[22] cleft review. This distress is perhaps compounded for craniosynostosis due to the rare nature of the condition, as the review highlighted how parents frequently face a notable delay in diagnosis and treatment due to dismissal of their concerns. Indeed, Feragen et al[8] interviewed parents of children with rare CFA and illustrated the long-lasting nature of these stress reactions, as parents could vividly recall their emotions even 15 years post-diagnosis.

Qualitative studies suggested that parents tended to be worried due to concerns over their child's future, delays in development and surgery. Findings from quantitative papers were less clear; some studies identified increased levels of parental stress[31] whereas others found no differences between parents and controls.[33,38] The same conflicting results were found in a review of caregiver stress in CFAs by Lim et al.[10] This may be explained by small sample sizes that make it difficult to sufficiently power a study and detect any statistical differences, with recruitment made more limited given the rare nature of certain diagnoses.[61] For instance, only four out of seven papers with quantitative data regarding parental wellbeing had more than 100 participants in the present review.[31,33,38,43] Even then, these samples were predominantly non-syndromic and therefore the parental stress levels of syndromic craniosynostosis have not been suitably captured.

Moreover, there is a lack of studies which measure familial stress levels at the beginning of the diagnostic process and throughout the course of post-operative care. Lim et al[10] also proposed that identifying risk factors related to increased distress may aid in explaining why some parents report stress, and others do not. Several studies in the present review suggested some preliminary factors[32,40,43] but there is still not a clear picture. More research has been conducted in this area within the cleft population[62-64] with the likes of social support, particular coping strategies and stressful life events identified as factors influencing psychosocial adjustment.

Social Experiences

Parents of children with syndromic craniosynostosis reported being stared at and receiving comments/questions when out in public.[49,50] Previous studies in the

craniofacial field and the wider visible difference literature have demonstrated how the perception of ‘difference’ can result in children facing these social challenges, as well as teasing, bullying and social isolation.[65] These have the potential to contribute to long-term psychological distress and low self-confidence for the child.[66] The present review highlighted the impact this may also have on the parent, as families reported feeling judged. This echoed findings from a study conducted by Feragen et al[67] who interviewed parents of children with rare craniofacial conditions. They deemed other people’s reactions as insensitive, hurtful, and felt a strong need to protect their child.

In response, one reviewed paper[49] listed how parents opt to ignore, educate others and/or promote their child’s independence. However, this is not always an easy feat as parents elsewhere in the literature have stated reluctance at talking to children about their visible difference in case this generates any further insecurities.[67] If communicated effectively though, promoting their child’s socialisation has been seen to mitigate the harmful effects of negative social experiences.[68,69]

One further social experience illustrated in the review was the role of support. The help of family, friends, spouses and other parents was seen as key to managing the emotional and practical demands of everyday life, alongside caring for a child with craniosynostosis. All included studies that mentioned social support referenced this at different time points – both pre-operatively and post-operatively – suggesting the value of support throughout the process. Previous research into CFAs has established that social support can mediate the relationship between parental quality of life and psychological distress.[70] Similarly, social support was associated with reduced family

impact and better adjustment in parents of children with cleft lip and/or palate,[71] signifying the importance of support across all craniofacial conditions.

Moreover, the wider literature on rare conditions has echoed the value of connecting with peers (i.e. other parents of children with the same diagnosis) for gathering information and feeling less alone.[58] Similar to most of the experiences in the present review, Baumbusch et al[58] suggested that parents seem to seek out peer groups themselves and not be signposted by professionals. A recent study conducted an analysis of discussion boards related to craniosynostosis and found that parents utilise social media to find this sense of community online.[72]

Service Experiences

A key feature of many families' experiences of craniosynostosis was the prospect of surgery. Parents reported pre-operative anxiety and worries about their child hurting themselves following surgery, which is also reflected in the wider paediatric population.[73]

A number of included studies took a focus on satisfaction with the medical care and treatment received for craniosynostosis. The impact of this has been highlighted previously in a cleft population whereby satisfaction with healthcare was found to be a protective factor for parental distress.[64] Overall, the reviewed studies suggested that parents are indeed satisfied with their child's craniofacial care and surgical outcomes. The extent and way in which this was measured determined how much detail was reported about this satisfaction. This may explain, in part, why the views of parents who were less satisfied came out more in qualitative studies. These highlighted

service-related issues and feeling overwhelmed in a multidisciplinary setting as reasons for dissatisfaction. Previous research has therefore suggested that parents need to be prepared for the number of specialists present within craniofacial consultations.[74]

Moreover, a number of cleft researchers aimed to further explore which elements of care parents were dissatisfied with and found issues were raised with the quality of information families received.[51] The same could be applied to the present review as parents wished for more personalised information, signposting to reliable sources and greater written resources. This is important as the wealth of information available on the Internet had the potential to misinform or invoke anxiety. Previous healthcare research has highlighted the need for accurate information and closer monitoring of online support groups for this reason.[75-77]

Clinical Implications

The review highlighted that parents experience a range of emotions throughout their entire clinical journey. The value of healthcare support was noted, although not all parents would ask for help or reach out for support when required. The reasons for this were not explicitly stated but could be hypothesised as not knowing how to access appropriate support, not feeling comfortable or perceiving themselves as a burden. Professionals should consider the reasons for why families may not ask for help, directly explain what support is available to them and ask if they would appreciate accessing any of these options, rather than a reliance on parents to seek out this support themselves. Further, all papers concentrated on parental experiences but the emotions and needs of other family members should not be neglected.

Information was a key theme mentioned across studies. Leaflets have been designed with promising results in the UK[78,79] but there is a need for this to be more widespread. The suggestions by parents within this review for more details on timeline of treatments, developmental delays, post-surgical care and peer support could be a place to start. This information would benefit from being disseminated to non-specialist health professionals in the hope that more information will generate an increased awareness of craniosynostosis, and subsequently reduce delays in diagnoses and treatment.

To also assist with information provision, ensuring that someone additional to the parent is present at clinic appointments with specialist staff could be crucial. This may be a spouse, partner or in the case of single parenting, a friend or relative. The importance of support has already been highlighted herein, but support during appointments specifically could lessen the pressure felt by parents to digest all the information offered during consultations. Nonetheless, parents may inevitably have further queries or questions and the availability of specialist staff to answer these or provide reassurance has been positively noted, and should not be underestimated.

Within the broader service context, parents are expected to attend regular appointments which is most often reliant on parents travelling, taking time out of work, arranging childcare and having the financial means to do so. Even for studies within the UK whereby the National Health Service is free, these costs can accumulate quickly.[50] Given that this review was produced in the context of a socio-political climate

whereby the cost of living is rising, it is likely that systems will increasingly need to bear these factors in mind.

Limitations and Methodological Considerations

The included studies had several limitations and quality appraisal of these highlighted certain methodological challenges. Firstly, quantitative papers used a variety of measures even when investigating the same construct (e.g., stress) making it more difficult to directly compare results. The same limitation has been reflected within the cleft literature.[14,80] The most frequently used standardised measure was the Parenting Stress Index[81] (PSI) which was originally developed as a generic questionnaire, as opposed to being condition-specific. As such, the measure may not capture the context of families with experience of craniosynostosis. Feragen and Stock[14] have previously proposed that a research priority for the craniofacial field should therefore be to reach a consensus about key outcome measures that are clinically relevant to this population. A certain number of recommended measures have been adopted within the field of cleft lip and/or palate but there is still an acknowledgment that there is a way to go in terms of implementation, especially for other CFA diagnoses.[82-84]

Second, there were inconsistencies with the terminology used to describe diagnoses. This ranged from non-syndromic/syndromic, single-suture, specifically naming a syndrome or referring to the suture that was fused. Some papers mixed these within one sample, whereas others recruited distinct diagnoses. This affects the conclusions that can be drawn about any potential differences between conditions, such as non-syndromic versus syndromic. As mentioned before, small sample sizes

also add to the complexities of this. Moreover, some studies relied on self-reported diagnoses, as opposed to clinical records, which is problematic because this further dictates the terminology that is then used.

Similarly, there was variation in the timing of studies which contributes to different levels of understanding about certain stages of the medical journey. Research predominantly took place post-operatively or some even included a combination of operated/unoperated in the same sample. This may be, in part, for methodological purposes, such as increasing the likelihood of recruitment, but also means there is less detail on specific time points in the process. Several key milestones have been identified in a family's journey (pregnancy and birth, diagnosis, accessing treatment, treatment decisions, surgery and post-surgery[79]) which could be a useful framework to guide future research into specific time points. Alternatively, the present review highlighted the paucity of longitudinal research in this population. This has been noted as a limitation of the evidence base previously[80] but seems just as valid today. More longitudinal approaches could help with building up a detailed picture of the different phases.

Further, samples were predominantly female and therefore may not be representative of all caregivers. The review has therefore been unable to draw conclusions regarding any effect of gender on parental experiences. There is preliminary research to suggest that fathers may differ in terms of attitudes toward receiving a CFA diagnosis[85] and have information and support needs of their own[86] but the review reinforces how studies exploring paternal perspectives are currently limited.

Future Research

This review consciously chose families as an area of interest, to allow the experiences of all relatives to be heard. However, it was highlighted that all studies, with the exception of one,[43] solely recruited parents. The experiences of other family members, such as siblings and grandparents, would make an interesting future addition. Impact on the wider family has been considered within the cleft[87,88] and paediatric literature,[89] which suggested the importance of an inclusive approach in healthcare that considers the needs of all family members.

Moreover, the present review highlighted the importance of social support but this was limited to professionals, family/friends, spouses and other parents. The value of other sources of support, such as religion, was only raised in one study[49] and warrants further attention. Within the context of support, no studies investigated in detail the degree to which parents accessed psychology provision within the craniofacial or physical health setting. Those who completed the survey by Costa et al[31] made reference to having access to psychologists, but further research could build on this to explore the proportion of parents that utilise this formal support, the mechanisms by which they access it and their views on its value. Conversely, the same could be explored for those who do not access specialist psychology provision.

Future research would also benefit from looking at and reporting any positive feelings or strengths of families, given that narratives of parental wellbeing were largely focused on distress. Measures of optimism and resilience are beginning to be used in the cleft literature[64] and one qualitative study into CFAs included questions

about personal growth.[8] The same could be extended to craniosynostosis, to generate a more balanced view of parental experiences and help identify protective factors related to familial distress or adjustment. This would work well in combination with Lim et al's[10] recommendation for research into the identification of risk factors for caregiver stress.

Conclusion

This review has synthesised the existing quantitative and qualitative research evidence about the experiences of families caring for a child with craniosynostosis. Experiences spanned from detection of the condition to the use of healthcare systems, and the emotions that accompanied this journey. Following a diagnosis, families were met with social challenges, frequent appointments and treatment options. Parents reported a need for quality information and support. Inconsistences in study methods and gaps within the literature leave scope for further research to advance the evidence base.

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Chapter 2

Exploring the Experience of Parents Whose Children Have Been Diagnosed with Non-Syndromic Craniosynostosis: A Focus on Protective & Risk Factors for Psychological Wellbeing*

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Abstract

Objective: Whilst the psychosocial literature into craniosynostosis is gradually increasing, studies have not tended to concentrate on particular phases of a families' journey. This paper thus aimed to focus specifically on diagnosis, by exploring the experiences of parents with children recently diagnosed with non-syndromic craniosynostosis. The study further aimed to identify risk and protective factors that contributed to adjustment and psychological wellbeing in parents at the time of diagnosis.

Design: Qualitative study, utilising a semi-structured topic guide to conduct virtual interviews. Anonymised transcripts were analysed using Grounded Theory.

Participants: A total of fifteen parents (10 mothers, 5 fathers) were recruited and interviewed. Parents of children diagnosed with non-syndromic craniosynostosis since September 2021 were eligible to take part. Recruitment took place across specialist craniofacial units, social media and via snowball sampling.

Results: A theoretical model was developed to summarise parental experiences of receiving a diagnosis for their child. This encompassed distinct stages involved in the journey to diagnosis and was represented by three themes: starting point, 'the unknown' and accessing the craniofacial service. An additional three themes – information, support and predisposing influences - were identified that influenced parents' trajectories on this journey. Factors found to be protective were access to trustworthy information, positive relationship with the healthcare system, support

networks and ability to employ healthy coping strategies. Risk factors included professionals' lack of awareness and negative life events/stressors.

Conclusions: Findings illustrated that the diagnostic process starts much earlier than any contact with specialist craniofacial teams. The study has implications for training, peer support, psychological screening of parents and fostering therapeutic relationships between staff and families.

Keywords: craniosynostosis, non-syndromic, parental wellbeing, risk and protective factors, qualitative, parents

Introduction

Craniosynostosis is a condition in which one or more of the cranial sutures are prematurely fused. It is estimated to affect one in every 2000 live births[5] and can be divided into two main groups: syndromic and non-syndromic. Receiving such a diagnosis can significantly impact on the psychological well-being of parents and the family unit.[14] Parents must begin to process the implications of their child's condition and embark on an often daunting, long-term multidisciplinary pathway which frequently involves surgical intervention within the first year of life.[90] Complex emotional responses of grief, shock, anger and worry have been reported, in addition to raised levels of stress.[91,92] In parents of children with a craniofacial diagnosis, these emotional responses have been associated with altered psychosocial outcomes for the child and potentially the parents themselves.[10] This highlights the importance of investigating the factors that affect psychological adjustment in this population.

As such, several reviews have recommended for research to clarify the risk and protective factors related to parental distress.[10,93] One sample of professionals working in craniofacial care suggested that factors such as prior traumatic life events, pre-existing mental or physical health concerns and insensitively delivered diagnoses were potential risk factors for poor psychological adjustment in parents.[94] They also labelled protective factors, such as reliable support networks, access to information, confidence in the medical team and having an optimistic worldview.[94] Whilst informative, it is not clear whether these risk/protective factors identified by clinicians are reflective of parents' views. The lack of craniofacial research specifically focusing on parental perspectives has been noted previously.[14,91]

Research focusing on parents in recent years has begun to address this gap in the literature. One such study surveyed parents of children with cleft lip and/or palate

and found optimism, healthcare satisfaction, relationship satisfaction, and several preexisting factors (such as mental health or life events) were associated with psychological outcomes for both mothers and fathers.[92] Cleft lip and/or palate is the most common craniofacial anomaly and as such, the evidence base is largely weighted in this direction. Other diagnoses, including craniosynostosis, have received much less attention.[95]

Of the research conducted into craniosynostosis, studies indicate that parents experience a range of challenges when supporting their child through diagnosis and treatment. For instance, a mixed-methods survey by Costa et al[96] concluded that parents of children with craniosynostosis reported higher levels of stress, anxiety and depression compared to the general population. This was derived from data collected at a single time point and the survey was open to parents at any stage of the healthcare journey. Overall, parents described frustrations with the diagnostic process, including dismissal of their concerns and a need to advocate for their child, worries about their child's wellbeing and a need for more information. This survey recruited via Headlines, a UK based craniofacial charity, which led the authors to acknowledge that findings may not be representative of all parents and therefore proposed that further research could use a multi-centre approach. For this reason, the current study aimed to include NHS centres in its recruitment strategy.

Furthermore, parents eligible for the mixed-methods study[96] could have a child with either syndromic or non-syndromic craniosynostosis. The authors stated that no significant differences were found in parent outcomes depending on the presence/absence of a syndrome. However, one methodological limitation commonly reported in the craniofacial literature is the use of small sample sizes, which can potentially make it difficult to discern differences between diagnoses due to a lack of

power.[93,95] This is important as there are not only differences between craniofacial conditions (such as cleft lip and/or palate) but also within (i.e. syndromic vs non-syndromic cases).[14] The current study thus aimed to focus on one particular diagnosis - non-syndromic craniosynostosis – for an in-depth exploration. Non-syndromic craniosynostosis was chosen as the focus for the current study as it accounts for most detected cases of the condition.[97]

One recent Swedish study also solely focused on non-syndromic craniosynostosis and interviewed 20 parents about their experience of diagnosis and the initial care process. This indicated that parental worries were mostly related to a limited understanding of the diagnosis, which often resulted in searching the Internet for further information.[98] A subsequent paper by the same authors interviewed a sample of parents with experience of non-syndromic craniosynostosis, but this time focusing on those that had received surgery.[99] This highlighted the importance of support from hospital staff, family, friends or peers; especially as parents described feeling alone and worried.

Similar findings were captured in a qualitative Canadian study of mothers interviewed about their journey from diagnosis through to post-surgery.[100] Again, parents expressed frustration at diagnostic delays and suggested ways to increase awareness of the condition to allow for earlier detection. Parents described their first interaction with the medical team as overwhelming but valued their support, as well as the support of connecting with other families. Parents reported relief following surgery but reflected on the fear and difficult emotions they had experienced prior to surgery taking place. This reiterated the findings of other studies which found the time of diagnosis, and before surgery, as the most challenging period.[94,99] An awareness of this early, yet critical, stage of the healthcare journey is therefore a crucial priority;

especially if services wish to identify those parents at risk of psychological distress. Many studies, as referenced here, encompass the whole patient journey which, although extremely valuable, perhaps does not allow for a thoroughly detailed account of the diagnosis time frame. The current study thus aimed to concentrate on the specific period of receiving a diagnosis.

Aims

(1) To explore the experiences of parents with children recently diagnosed with non-syndromic craniosynostosis

(2) To identify risk and protective factors that contributed to adjustment and psychological wellbeing in parents at the time of diagnosis

(3) To construct a model for understanding parental experiences of receiving a child's diagnosis of non-syndromic craniosynostosis, to ultimately help inform the care and support for families facing this journey in the future.

Method

Ethical Considerations

The study was granted ethical approval by North West – Greater Manchester East Research Ethics Committee and the Health and Research Authority (reference number: 22/NW/0190; Appendix 7). The ethical guidelines of the British Psychological Society were followed throughout.[101] The study was also reviewed by the Cleft and Craniofacial Clinical Studies Group which includes clinicians, researchers and patient representatives. Participants were informed of their right to withdraw, the confidential storage of data and how they would not be identifiable during dissemination of the findings. In transcripts, names were anonymised and places/locations were redacted.

All eligible participants provided informed consent electronically prior to participation, via a consent form (Appendix 8) that could be completed electronically. Due to the potentially emotive nature of the interviews, a distress protocol was developed (Appendix 9) to help support participants should they become upset or distressed. There was also a researcher safety procedure in place (Appendix 10) to ensure the lead researcher adhered to lone working policies.

Design

Given the research aim of exploring parental experiences, a qualitative design was used to provide a deeper understanding of the time period of diagnosis. There is a known paucity of qualitative research within the craniofacial evidence base, despite the area being well suited to the methodology due to the limited awareness of the condition.[14,95] Qualitative designs are known to be valuable as they provide an important and detailed insight into aspects of the patient experience that are under-researched and/or not well understood.[102]

Specifically, the qualitative method applied in this study was a constructivist grounded theory approach.[103,104] This differs to earlier positivist grounded theory approaches[105-107] due to its acknowledgement that the social world is complex and contains multiple perspectives. As such, constructivist grounded theory recognises the ongoing interaction between researcher and participant; whereby the researcher's assumptions and prior understanding combine with the participant's experience to form a meaningful theoretical model.[108] As those involved in the authorship of the current study had previous knowledge of craniofacial conditions and the existing literature, the constructivist mode of grounded theory represented by Charmaz[103] was

deemed most appropriate. Additionally, the goal of grounded theory to develop an explanatory theory[109] meant this approach was best suited to meet the research aim of generating a model to understand parental experiences.

Participants and Recruitment

Participants were initially approached at two of the five specialist craniofacial units in the UK. This began with a purposive sampling strategy, before participants were sampled theoretically as the study progressed. Theoretical sampling is a feature of grounded theory research and includes the process of actively seeking out individuals with varied views and experiences to add to the emerging theory.[110]

Potential participants were initially identified by designated Clinical Nurse Specialists and/or Clinical Psychologists at each NHS site who offered parents a leaflet about the study (Appendix 11) and an information sheet (Appendix 12). Interested parents were then invited to contact the researcher to discuss their participation and schedule a time for interview. Alternatively, parents interested in taking part could consent to their information being passed to the lead researcher (Appendix 13) who then made contact in a timely manner. The number of eligible participants approached by staff members was unfortunately not collected so hereby not reported, although a total of nine participants were recruited via this method.

From this, participants were asked if they knew of parents in their social networks who would be interested in taking part (i.e. snowball sampling), resulting in four more participants. Of these, two parents received medical care from a third specialist craniofacial unit.

Additionally, the study leaflet was posted on the social media page for Headlines to specifically invite fathers to participate as the study was originally limited from a paternal perspective. This attracted two further participants. Each parent/caregiver (e.g. mother or father figure) qualified as a single participant. Overall, three dyads participated but were interviewed separately.

Following an expression of interest, the lead researcher screened each potential participant based on study inclusion and exclusion criteria. Participants were eligible for the study if they were parents of a child diagnosed with non-syndromic craniosynostosis since September 2021. This was to ensure a focus on the period of diagnosis and to recruit parents at an early point in their healthcare journey. It was recognised that this early stage is often accompanied by a complexity of emotions and therefore parents whose child received a diagnosis in the last 3 months were not eligible for interview, to allow for a natural window of adjustment. This was decided following clinical discussions and consideration of the healthcare literature regarding optimal recruitment times, namely not approaching parents immediately after learning of their child's condition.[111,112] Participants were also excluded if their child had received a diagnosis of syndromic craniosynostosis, and whom the medical or research team judged to be too unwell or distressed to be able to provide informed consent.

There were an additional eleven parents who expressed interest in participating but were not included in the final sample. Six of these were identified via the NHS, with one person withdrawing due to personal circumstances and the others withdrawing their interest with no explanation. Three parents learnt of the study by word of mouth

but did not meet inclusion criteria. Similarly, one father responded to the Headlines post but did not meet criteria.

Participant demographic information is displayed in Table 3. Pseudonyms chosen by participants are used in place of their names.

Table 3. Participant Demographic Information.

Participant	Age	Gender	Ethnicity	Family set-up	Age of child at time of interview (nearest month)	Gender of child	Age of child at diagnosis (nearest month)	Diagnosis	Surgical intervention	Other health conditions	Family history of condition
Linda	27	Female	White British	Partner, child	11	Male	5	Sagittal	Awaiting	No	Yes
Daisy	30	Female	White British	Partner, 2 children	10	Male	2	Sagittal	Yes	No	No
Kim	22	Female	White British	Child	9	Female	5	Unicoronal	Yes	No	No
Ada*	37	Female	White British	Partner, child	17	Male	11	Metopic	Awaiting	No	No
Steven*	40	Male	White British	Partner, child	18	Male	11	Metopic	Awaiting	No	No
Adella	35	Female	White British	Partner, 3 children	14	Female - twins	6	Unicoronal	Yes	No	No
Lucy	35	Female	White British	4 children	28	Male	19	Sagittal	Yes	Innocent heart murmur	No
Dave	38	Male	White British	Partner, 2 children	14	Male	7	Sagittal	Awaiting	No	No
Katie	37	Female	White - Other	Partner, child	36	Male	26	Sagittal	No	Eczema	No
Daisie	42	Female	White British	2 children	20	Male	16	Metopic	No	No	No

Thom*	31	Male	White British	Partner, child	15	Male	6	Sagittal	Yes	No	No
Helen*	28	Female	White British	Partner, child	15	Male	6	Sagittal	Yes	No	No
Alexander	34	Male	White British	Partner, 2 children	4	Male	1	Sagittal	Yes	No	No
Owl*	34	Female	White British	Partner, 2 children	11	Male	3	Sagittal	Yes	No	No
Johan*	37	Male	White British	Partner, 2 children	11	Male	3	Sagittal	Yes	No	No

*dyad in this study

Note. Further demographic information is available upon request.

Interviews

Semi-structured interviews took place between October 2022 and February 2023. Each participant was interviewed once. These were conducted via remote methods, using an online video platform (Zoom), and audio-recorded to allow for transcription and coding as soon as each interview was completed. Interviews were between 28 and 76 minutes in length, with an average duration of 54 minutes.

Interviews followed a topic guide (Appendix 14) that was devised in consultation with research supervisors, who had extensive clinical and research experience in the field, and a review of the literature. One parent of a child with craniosynostosis was also consulted and provided feedback on the study design and draft research materials. The topic guide included questions to gather information in four broad areas: demographics, the time of diagnosis, adjustment and wellbeing, and offering recommendations for services and/or other families. See Table 4 for an overview of these areas. Its semi-structured nature allowed for flexibility within interviews to follow the direction taken by participants' experiences. In accordance with grounded theory principles, the topic guide evolved across interviews in response to themes arising from the data (Appendix 15).

Table 4. Interview Topic Guide.

Areas of Interest	**Example Questions**
Demographics	• How old are you? • How would you describe your job role or current employment status? • How old is your child under the Craniofacial team?

Time of diagnosis	• Can you tell me about when your child was diagnosed and what it was like for you? • Can you tell me about the formal/informal support you received?
Adjustment and wellbeing	• Can you describe any impact the diagnosis has had on yourself? • How well do you feel you have adjusted? What has helped/hindered this?
Recommendations	• How could your experiences of diagnosis have been improved and/or how could it be improved in the future for other people? • Based on your personal experiences, what advice would you give to someone in a similar position?

Analysis

The first author transcribed three interviews verbatim to become fully immersed in the data. Remaining interviews were transcribed by an external, University approved service and quality-checked. Transcripts were uploaded and analysed using NVivo 12 software. An example transcript is included in Appendix 16. Data analysis followed the process outlined by Charmaz[103]:

i. In-depth reading(s) of the transcripts and narrative summaries were completed after each interview.
ii. Initial coding to name each line or segment of data (Appendix 17).
iii. Significant or frequent initial codes were then integrated into larger, focused codes (Appendix 18).

iv. Relationships between focused codes were identified and formed into theoretical categories (Appendix 19).

v. Throughout the steps above, memos were written to capture thoughts, comparisons, and connections in the data. These ultimately helped in constructing a theoretical structure.

During this process, the constant comparative method of considering the similarities/differences within interviews, between interviews and between coding levels was helpful in identifying directions for theoretical sampling and ongoing development of the interview topic guide.

As analysis progressed, fewer new codes were constructed and the identified theory seemed to make sense and account for participants experiences. This was an indication that theoretical sufficiency had been achieved.[113] Unlike the term 'saturated' used to refer to when fresh data no longer sparks new insights, 'theoretical sufficiency' has been coined to describe reaching a sufficient depth of understanding that allows the researcher to generate a theory.[103] As it developed, the analysis narrowed in focus and the theoretical model was able to reflect new data without further modifications.

Participants were invited to provide feedback on the theoretical model to check if it captured their experiences. All participants were sent an email with a summary of findings (Appendix 20). Eight parents responded (Appendix 21) and this feedback was incorporated into the final results and write-up.

Reflexivity

Reflexivity is the process of attending to the researcher's influence, considering how their interests, experiences, positions and assumptions may impact upon emerging data and knowledge construction.[103] As a method, grounded theory acknowledges that research data and analysis are co-constructed by participant(s) and researchers, as both bring their own views and ideas to research. In this case, the lead author wrote a reflexive statement prior to data collection (Appendix 22) and kept a research diary (Appendix 23) throughout the study. These were reflected on in supervision to think critically about how the background of the researcher or any pre-existing thoughts and expectations might have shaped how the research was conducted, analysed and/or reported.

Trustworthiness

Elliot et al[114] published guidelines for reviewing and assessing the quality of qualitative research within psychology. The guidelines and how these were addressed in the current study are presented in Table 5.

Table 5. Quality Guidelines.

Guideline	In practice
Owning one's perspective: Researchers specify their personal, theoretical and methodological orientations.	• The lead researcher is a 28-year-old White British female, currently training to qualify as a Clinical Psychologist. She has no children. She has prior clinical experience of working with families who have received a diagnosis of craniosynostosis. • As part of clinical training, the lead researcher has had teaching on research methods and

	conducting interviews. She also has prior research experience and conducted qualitative research at both undergraduate and Masters level. • Supervisors have years of clinical experience working in specialist craniofacial units and/or vast methodological knowledge in applying grounded theory. • As per the methodology, the lead researcher took a social constructivist position.[103] • The lead researcher held a reflexive stance and completed a reflective diary throughout the research process. This was to recognise personal and professional experiences and the role that these play in the emerging data. In addition, it captured any initial ideas/beliefs about the study.
Situating the sample: Researchers describe the sample and their life circumstances.	• Demographic characteristics of each participant are shown in Table 3. • Narrative summaries were completed after each interview (Appendix 24).
Grounding in examples: Researchers provide examples of the data to illustrate the analytical process.	• The lead researcher transcribed a proportion of the interviews to become fully immersed in the data. • Line-by-line coding was completed which allowed the researcher to remain grounded in the data. • In vivo participant quotes were used to form codes and the theoretical model.

	• Direct quotes are embedded within the results section. • The theoretical framework is presented in both diagrammatic and narrative form in the results section to allow readers to conceptualise their own meanings and understandings.
Providing credibility checks: Researchers employ methods to check the credibility of categories, themes or accounts.	• Research supervisors reviewed transcripts. • One research supervisor coded two of the interview transcripts independently of the lead researcher and codes were reviewed in supervision (no % agreement available). • Participants were invited to feedback on the theory to check whether it captured their experiences.
Coherence: The researcher's conceptualisation of the data fits together to form a clear framework of the area under investigation.	• Guided by grounded theory principles, the research aimed to develop a theoretical framework for understanding parental experiences. • The analysis and developing theory was discussed and refined in supervision, as were themes and interpretations. • Examples of coding categories are provided in the Appendices to make the analytical process as transparent as possible. • The theoretical framework is presented in both diagrammatic and narrative form in the results section which illustrates the relationships between codes/categories.

Accomplishing general vs. specific research tasks: The researcher is clear on the generalisability and reliability of their findings, and limitations are addressed.	• The manuscript outlines that the theoretical model has evolved from the perspectives of one particular group studied and therefore may not relate to other diagnoses (such as syndromic craniosynostosis and/or cleft). • The research aimed to focus on a particular group (i.e. parents at time of diagnosis for non-syndromic craniosynostosis) as previous research has grouped diagnoses and time points. • The limitations of the study are included within the discussion section and have been reflected on in supervision throughout the research process.
Resonating with readers: The material is presented in such a way that brings the participants' experience to life and readers judge it to have accurately reflected the subject matter.	• The manuscript has been reviewed by the research supervisory team. • A summary of findings was sent to participants (Appendix 20). • A copy of the manuscript was sent to a parent of a child with non-syndromic craniosynostosis who did not participate in the study. • For all above, feedback was received and used to help refine the final write-up. Positive feedback was given about the model capturing the parental experience, but also articulating a period that is often difficult to put into words.

Results

Parents' accounts of their experiences were organised around distinct periods, or stages, of their journey toward diagnosis. As one parent stated:

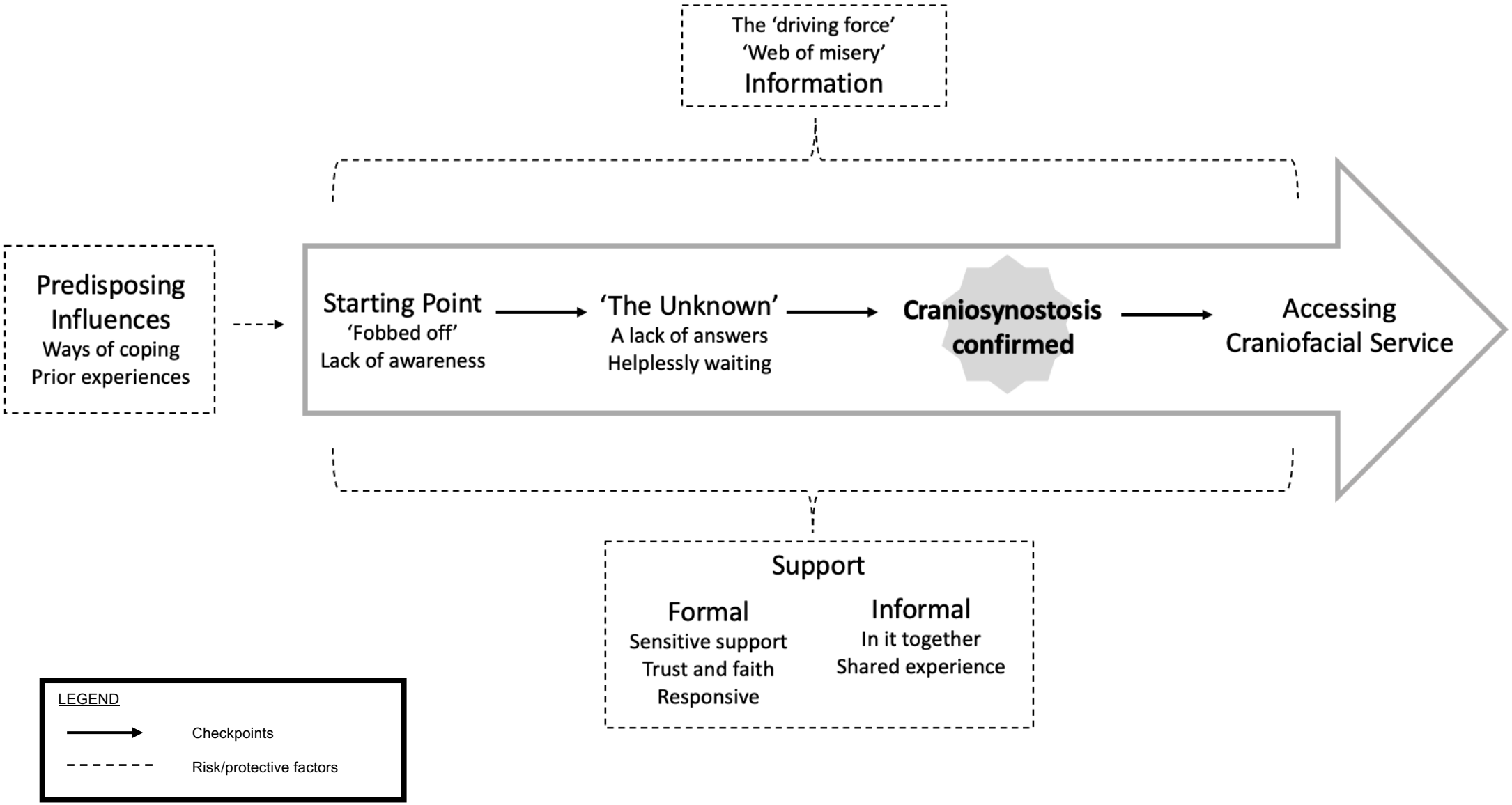
I think there's probably almost like checkpoints of someone's journey (Dave)

Through data analysis, themes were identified that reflected three main 'checkpoints': starting point, 'the unknown' and accessing the craniofacial service. Each one of these was associated with different concerns, questions, levels of understanding and accompanied by a range of varying emotions. Parents moved from detecting an issue with their child and wondering what it could be, through to trying to find answers, before receiving a diagnosis and medical help. The notion of distinct stages was felt to be a meaningful way of structuring parental experiences as it captured how diagnosis was, in fact, a process rather than a single timepoint or event and one that occurred over time. The explanatory model (Figure 3) encapsulates these different checkpoints.

Parents' trajectories across the diagnosis process were influenced by three further themes that represented risk and protective factors: information, support and predisposing influences (Figure 3). These factors affected the length of time parents were in any given stage of the model, and their adjustment and wellbeing.

The results are presented in two sections. First, an overview of the different checkpoints on the healthcare journey are explored from the parental perspective. This is described chronologically, from starting point to diagnosis. Second, the risk and protective factors are outlined. Themes are illustrated with exemplar quotes throughout.

Figure 3. Model.



1. The journey

1.1 Starting Point

This first theme pertained to how symptoms of craniosynostosis were first detected. Receiving a diagnosis, or at the very least a further exploration of these symptoms, required the concerns of parents to be listened to and was dependent on professionals having the knowledge to potentially identify the condition. This was represented in the data by two sub-themes: *'fobbed off'* and a *lack of awareness*.

'Fobbed off'

Thirteen parents spoke of noticing a difference at birth. However, raising concerns or asking questions at this point was not always taken seriously and led to a sense of feeling dismissed. Any noted physical differences with their child, such as an unusual head shape, were often attributed to other factors such as the process of giving birth, and/or parents were led to believe it would get better with time:

> Instead I was told, "It's not a problem. It's a variation of normal. It's just how they look, it's going to get better with time" (Adella)

> I couldn't put my finger on it but I just knew there was something a bit off and he said, "That's absolutely fine, she was just squished, she will even out" (Kim)

All parents pursued their concerns, driven by significant worries about what any physical differences could signify. As Adella stated, this resulted in 'a lot, a lot, a lot, a lot of contact with health care professionals'. This was either within the NHS or three

families accessed support privately; seeking input from the likes of paediatricians, GP, health visitors and consultants to explore what their concerns could be.

Lack of awareness

A professionals' awareness and understanding of craniosynostosis was crucial here. The majority of parents spoke of a lack of awareness of the condition which impacted how quickly a child was diagnosed. This tended to be amongst the professionals that first came into contact with a family locally; such as midwives, health visitors and paediatricians. In a couple of cases, a professional was already aware of craniosynostosis which sped up the process:

> I have seen people say they had to battle to get someone to take them seriously whereas that was never our position because it was so obvious, and our heath visitor had seen it. I think ours was quite straightforward in that case (Helen)

Even if professionals were not familiar with craniosynostosis, there was a consensus that parental concerns should still be listened to and prioritised for referral to further support. In one instance, a family history of the diagnosis meant parents had a clearer idea of what support to request. However, a lack of awareness amongst professionals about the referral processes for the craniofacial specialist centres was felt to be an added barrier when it came to actioning parental concerns:

> GP didn't know about any of the hospitals, or anything like that, which I think was why he was a bit wounded when I'd gone to him and said, "Look, I've got this email, they're happy for a direct referral, please could you go through the

> pathway to do that?" because I don't think he really realised that that was an option (Lucy)

> I would also suggest that this lack of awareness goes beyond just recognition of the condition but the process that should be applied by hospitals, GPs etc when the condition is suspected. For example in our case, the consultant paediatrician who dealt with [child] when he was born actually recognised the condition straight away but was unaware of the process for referral (Alexander)

The extent of this awareness and any subsequent delays in accessing specialist support contributed to increased worry and frustration in parents.

1.2 'The Unknown'

Parents used the terms 'unknown' or 'limbo' to describe the next and most challenging period of the diagnosis process. This tended to be the time between a difference first being noted and receiving a formal diagnosis, which was an emotionally turbulent period for parents:

> I would say, from the thought getting dropped, to the diagnosis, I would call that traumatic (Dave)

> That was the worst [period] we had through the whole process, I think. There was lots of tears, lots of upset, lots of not really being able to compose ourselves very well (Alexander)

Emotions seemed to stem from a state of uncertainty and were compounded by the time taken for professionals to provide clarity and offer any treatment options. This was characterised by the sub-themes: *lack of answers* and *waiting*.

A lack of answers

Parents suspected that there may be an issue worth exploring for their child but, at the same time, had no concrete answers. This left them wondering what the answers could be and contemplating what the future may look like:

> At the time I was very much like, Oh my God, I still don't really know what this means and if it is going to have a big impact on his life (Helen)

Nine parents described these thoughts and feelings as all-consuming and always in the back of their minds. This sometimes led to imagining a plethora of worst-case scenarios. Four parents recalled how a lack of answers contributed to feelings of fear about losing their child:

> The other part is, is he going to be okay? I've never loved anything as much in my life, and I'm petrified of something happening to him (Ada)

> There would be days where I would just cry because I was convinced I was going to lose her (Kim)

From this, it seemed that remaining in 'the unknown' made parents more susceptible to ruminating about what might be in store for their child and the severity

of any potential condition. At times, parents found it difficult to focus and be in the present moment as they were busy pre-empting what may come next.

Helplessly waiting

Parents reported waiting for several things to happen, including the wait for a referral to a specialist centre to be made plus the wait for an initial appointment with specialist staff to then be arranged. Any delays or a longer wait particularly impacted parents who had heard about the prospect of surgical intervention. Some parents were keen to be seen so that they did not miss an opportunity for less invasive surgery:

> And I worry like do we get to a point where surgery might be the best option, but we're now out of the time frame for it to have the best effect or have the least scarring or anything like that (Steven)

> I was kind of really worried then, because from what I'd read from other people, surgery was the only option. So I didn't kind of know what to expect but it was about six months, because I remember thinking, oh, we've passed that cut-off now, and just being really annoyed at everybody that had some involvement with him (Ada)

> Because if it's diagnosed early, parents have the choice of surgery, which I think is important. I'm not saying any one surgery is better than the other, but parents deserve a choice between the two. And, you know, if she had been diagnosed a month later, she would have had to have her whole skull completely taken off,

> reshaped…You know, the difference between treatment options when diagnosed early versus late is absolutely crazy (Kim)

Parents were in a position whereby they were anticipating involvement from specialists but the speed at which this occurred was somewhat out of their control and dictated by service waiting times. This sense of helplessly waiting was likened by Lucy to ‘life being put on hold’. Subsequently, it was difficult to plan for future holidays, occasions, family time or workload capacity as parents did not know what timescale to expect.

1.3 Accessing Craniofacial Service

Following a successful referral to one of the specialist NHS sites, the next checkpoint of a parents’ journey was identified as attending regular appointments with a craniofacial team. There were logistics to consider for attending these appointments; such as childcare and time off work. Ten parents had a long commute to access a craniofacial centre so had additional challenges to navigate such as travel, parking, finances and accommodation:

> It hasn't been cheap going through this whole process (Kim)

> I was panicking, at one point, that I wouldn’t have anywhere for [other children] to go, and I didn’t know how I was going to manage that, which was quite a scary thought. But we figured it out (Lucy)

How a parent was impacted by these was dependent on the resources and support systems they had in place to help manage these challenges. This theme was less detailed in parents' narratives, likely due to the focus during interviews on diagnosis and not on the long-term interaction with services, but felt to be an important stage in parents' experiences nonetheless.

2. Risk and protective factors

2.1 Information

The first factor identified as influencing parents' trajectories related to gathering and seeking out information. For this reason, the theme is positioned in Figure 3 spanning across the checkpoints in the diagnosis journey outlined thus far. All parents, with the exception of one who received a diagnosis promptly, felt a pressure to seek information to some degree. The extent to which parents then accessed quality information characterised this as either a risk or protective factor for psychological adjustment and distress. On the one hand, parents felt information helped prepare them whereas, in other instances, it was challenging and overwhelming. This was represented in the data by two sub-themes: *'the driving force'* and '*web of misery'.*

The 'driving force'

Parents named various avenues to source information including researching online, Googling, reading books or articles, watching videos or social media. Information seeking tended to start early and there was a real sense that parents felt a responsibility to be the driving force and find immediate answers themselves:

> You're putting like a pressure on yourself, or at least I did. It was like, when I'm going to bed, I'm thinking, Have I done everything that I can? You're almost putting too much high an expectation on yourself, because there's only so much I can, but that was a- I wouldn't say it was a source of stress because it got us to where we needed to be, but I would say that was quite a strong emotion of like- You're almost like arguing with yourself, like have you done everything? (Dave)

> I felt like this diagnosis had kind of been- well, half-diagnosis really at that point, had kind of been dumped on us, we'd been left to sort of look at what it could mean and do our own research (Alexander)

The function of this research was two-fold; firstly, to identify what may be happening for their child and secondly, where to access help and support. This meant most parents had an understanding of craniosynostosis from the Internet and therefore receiving a diagnosis came as no surprise. Parents found it helped lessen the emotional impact of the diagnosis as they had time to prepare any questions in advance and process what craniosynostosis was:

> I suppose if you had not known anything, it would have been horrendous, but we kind of knew what was coming (Owl)

For nine parents, researching online is how they found out about the specialist craniofacial centres and even contacted them directly. Subsequently, this prompted a

referral and moved families a step closer to a formal diagnosis. As Johan stated, ‘taking matters into his own hands’ was felt to speed up the process.

‘Web of misery’

One parent coined the term ‘web of misery’ to signify how looking for information could be overwhelming. A couple of parents refrained from the Internet for this reason:

> I think I really tried then to avoid reading anything about what craniosynostosis meant because as I say, like most of the stuff that you find is- you’re sort of diving into you know maybe learning difficulties, and speech problems, and maybe social difficulties later in life, and diving straight into the depths of quite a dark place that I didn’t really want to read about at all (Johan)

Reading content that alluded to developmental delays, as articulated above, seemed to stick with parents. So much so, they found it hard to forget what they had read and held onto this knowledge. If this was to then persist, parents could find themselves at risk of increased worry as their perceptions of craniosynostosis were largely negative.

A mitigating factor for the negative influence of accessing information was how reliable or trustworthy the source was. Three parents were cautious about what information they digested, whereas the remaining parents reflected on how they read everything available:

> It's a broad field, and I don't think in my emotional state at the time I could rationally see what was useful research and what wasn't. So I read a lot of things and took them for gospel, and in some cases that helped me, in other cases it made me feel ten times worse as to what might happen, what the future might look like (Alexander)

This suggests the value of remaining wary of information sources. Placing too much weight on information from unreliable websites or the lack of quality information offered by the healthcare provider seemed to be a risk factor for parental distress, whereas access to quality information had more of a protective role.

2.2 Support

Similar to information, the theme of support is also positioned in Figure 3 spanning across the different checkpoints. This is because the qualities and guidance offered by both formal and informal support networks had an impact on how parents not only explored concerns at the starting point, but also how they adjusted and responded to later stages in the process. The access and quality of such support influenced whether parents were at risk of psychological distress such as feeling dismissed and uncertain, or if it was a protective resource buffering stress.

Formal support

Formal support referred to structures such as local or specialist sites; including facilities, staff and their approach. Despite parents reporting varied levels of satisfaction with teams in their local area(s), they were all mostly satisfied with the

support they received from the specialist craniofacial teams. Particular features were appreciated such as interpersonal skills, trust and responsiveness.

One parent coined the term ‘sensitive support’ to describe how it was not necessarily *what* staff did to help, but *how* they did it. As such, the human connection and relationship with staff was a protective factor for parents. This was characterised by interpersonal skills of clinicians being warm, welcoming and approachable:

> I don’t really like talking about it as a place and as a ward or whatever or as a department. It was the people that were involved that were helpful (Johan)

> That’s the kind of examples of the little sort of interpersonal skills that the team had that were so important in our journey, and it’s those that I felt they did so so well, that kept us in a good place all the way through (Alexander)

For the majority of parents, the first time meeting the craniofacial team was within a multidisciplinary team (MDT) setting. This was most often perceived as daunting and intense as there was typically a lot of professionals together in the same room. Parents described often feeling surprised or taken aback as they had perhaps been focusing on what the outcome of the appointment would be, as opposed to expecting the MDT set-up. ‘Sensitive support’ was less evident inside this setting and, instead, parents reported that information was presented in a factual, clinical way:

> They don’t sugar coat it. When we first met [Consultant Paediatric Neurosurgeon] he was very kind of straight talking, straight to the point but I

> think in situations like this you can't kind of pussy foot around the situation (Linda)

> It was interesting because he wasn't in any way, he wasn't like warm and fuzzy, he didn't say gosh you've had a tough time or what a journey you've been on. He said nothing like that. He just said, "This is the problem, these are the options. You need to decide what you want to do" (Adella)

For four parents, this direct approach was preferable as they wished to know the facts, statistics, risks and/or benefits in order to understand their child's prognosis and to make an informed decision about surgical options if relevant. At the same time, there was a sense that parents also valued a personable approach and to be reminded that their child would be OK.

Parents found comfort in how specialist centres work with craniosynostosis daily and spoke of the importance of feeling able to trust in professionals. There was a sense of relief when a referral was made and accepted to a specialist centre as this signified being in the right place with the 'right people' who could help:

> That's what they do, that's what they do day in, day out. It was very reassuring (Thom)

> Just acknowledging that somebody else who knew more about what was going on was in charge of this show (Johan)

It follows that having confidence in staff and the healthcare system was a protective factor. Conversely, losing faith and confidence impacted parental wellbeing. In the case of Adella, having delays in the diagnostic process resulted in a loss of a trust in a system she actually worked within:

> I'm a health care professional so I tend to trust the system. When I then was told that it was [craniosynostosis] I was shocked but mostly I was really, really angry. I was really angry. I felt like I'd, yes I'd trusted a system that I know well and I'd trusted individuals… I just think oh what more could I have said to have got a diagnosis earlier? (Adella)

This left her ruminating and had long-term negative implications for the relationship she had with healthcare professionals.

Another feature of formal support that was deemed to be valuable was how responsive professionals were to any questions, concerns or comments. Parents spoke of having many questions and valued being able to send these via email or by other means, such as an App. This applied to both pre- and post-diagnosis, as eight parents reported having contact with staff prior to actually visiting a specialist craniofacial team. Often a response would be received promptly which put parents at ease:

> Knowing that their team is so responsive and so helpful. Like obviously they must understand that it is a very stressful or difficult situation for parents to go through so I don't know if that's why they're very quick with their emails (Linda)

Within responses, parents commented on how they appreciated staff being calm and offering their time without feeling rushed. Having a positive experience and feeling satisfied with their healthcare ensured parents knew how to access support and felt comfortable reaching out.

Informal support

Outside of the hospital and medical professionals, parents spoke of how their experiences could be isolating so there was a focus on what helped parents not feel alone during their journey. In most cases, this was the support from the other parent/partner. It helped to have someone to talk about feelings and attend appointments with. Two parents described this as a reciprocal process:

> Being at the right place for each other and at the right times… we've never both been really down in the dumps with this at the same time. I think there's always one of us that's been on a practical road, and one of us that's been on a very emotional road. And then at different points we've switched over, but we've never ended up on the same road together, which actually I think has been the reason we have got through it okay (Alexander)

If this was not possible, feeling alone was a potential risk factor for distress. Three participants identified as single parents and reflected on this:

> Especially when you don't have another adult, like I don't have another adult at home, so not having another person to talk it through with, that is willing to have that conversation with you, that's really hard (Lucy)

In the absence of a partner, the support of friends and family proved integral. Family and friends were also said to provide words of encouragement and/or practical support such as childcare arrangements. Despite their best efforts, parents such as Linda felt this familial support was sometimes 'lost because they'll never have to go through what we will have to'. It was for this reason that parents found connecting with others who shared the same experience (i.e. partner or other parents) of added value.

Connecting to other parents going through the same process was a prevalent theme running throughout the interviews. Eleven parents sought connections with each other by searching for support groups online. Facebook was mentioned on numerous occasions as a place of shared experience and even if parents did not wish to post themselves, reading the stories of other families and seeing photos of children further on in their journeys was felt to be reassuring. Connecting with others in this way sometimes led to meeting up in person, private messaging or group chat conversations. This support was felt to offer something unique:

> It's a completely different dynamic. Doctors are very helpful and they're great if you want reassurance, but they can't connect with you on that 'you know what I'm going through' level. They know what's happening from a 'I've read it in a textbook I've treated this' sort of way, but not in a 'I know the fear you have as a parent' way (Kim)

In several instances, finding these parent forums was how participants found out about craniosynostosis and the specialist centres in the first place. They also provided an opportunity to ask questions that did not require a specialist response.

2.3 Predisposing Influences

The model proposes that the extent to which parents adjusted to the diagnostic process was impacted by how they coped generally, their personal outlook or attitude and past life events. This was represented by the sub-themes *ways of coping* and *prior experiences*. As one parent succinctly described:

> How I feel about that, or how I view this, is in relation to all my other experiences. Like nothing's an island, is it? There's got to be a recognition that people come with their own stuff already (Ada)

Ways of coping

There was a sense that continuing to live everyday life 'as normal as possible' helped alleviate distress. Similarly, keeping busy or having a distraction such as returning to work were protective strategies as they left less time to ruminate:

> There's not really much time to worry, to be honest. But I think that's because I'm busy. I think any parents who have got a lot of free time, then that's when your brain starts turning over. I'm always, you know, very active, really. So I think that's the key (Dave)

Parents talked about their personalities and how their dispositional styles, or the ‘type of person’ they were, impacted how they responded in times of stress. Four parents spoke of finding it easier to adjust when their nature was not to worry until they had the full facts of a situation. These parents were driven to find a solution and would be more inclined to research, rather than jump to conclusions. Three parents mentioned having a positive outlook on life, being ‘level-headed’ or emotionally ‘switching off’. This differed to a couple of parents who described struggling to cope:

> I just sat about and cried. So I don’t think I’m the person to ask about coping strategies. I just cried a bit. I don't cope with things very well (Kim)

Prior experiences

Many parents shared previous life events that had influenced their views about hospitals and their perspective on difficult times. For instance, one mother felt that already being familiar with medical processes had helped her:

> I think it's not been as hard to adjust as it might have been for other people because I'm used to. I've been through this process of what's wrong, new diagnosis, hospital stays, hospital appointments, things going wrong, my whole life (Kim)

Others shared previous negative experiences of hospitals which had led them to expect the worst from medical care, meaning encounters such as delayed diagnosis or dismissal of concerns compounded these feelings. Two parents also shared pre-

existing mental health, such as experiences of anxiety, that they felt left them susceptible to increased worry.

Additionally, several life events were mentioned that seemed to be risk factors for parents' current stress levels and emotional states. For instance, two mothers had recently experienced miscarriages which amplified their fears about craniosynostosis being a life-limiting condition:

> She was born not long after I lost my first baby. So I was like, Oh my God, it's happening again. I'm going to lose her. It was a bit too much all in one space (Kim)

Another example was one family who had suffered several bereavements:

> Obviously we were really sad and I feel like we did deal with it really well, but I don't know how much of it was maybe inflated by the fact [dad] died, you know, it kind of all got merged into one sad period of our lives (Owl)

Naturally, an accumulation of stressors resulted in feeling overwhelmed and it became difficult to disentangle which emotional responses were specific to their child's condition. This highlighted how the diagnosis journey is often not an isolated process, but occurs within the context of busy family lives.

Discussion

This study aimed to explore the experiences of parents with children recently diagnosed with non-syndromic craniosynostosis. Data analysis allowed for a model to

be constructed to encapsulate these experiences. Parents transitioned through different ‘checkpoints’ to obtain a diagnosis; from first detecting signs of craniosynostosis at the starting point, anticipating ‘the unknown’ of what this may mean, to eventually attending appointments and accessing a craniofacial service. Information, support and predisposing influences were highlighted as risk and protective factors that contributed to the adjustment and psychological wellbeing in parents across the journey to diagnosis.

Most parental narratives perceived the journey as long and challenging. This led parents to seek out information, which in most cases served as a protective mechanism to propel parents towards identifying craniosynostosis or enquiring about a specialist referral themselves. The perceived need for information has been highlighted in a recent review (see Chapter 1)[93] which suggested that the parental desire to gather more information signifies that there is a lack of resources readily available, meaning parents are left to ‘hunt’ for material on the Internet. For this reason, parents reflected on how they were felt to be the ‘driving force’ which is similar to how caregivers in past research have viewed themselves as advocates for their child.[96]

At the same time, information had the potential to serve as a risk factor for parental distress. A Swedish study which interviewed parents about their initial care process found some families reported being faced with frightening information when searching craniosynostosis on the Internet. This was also raised in the current study when parents read about the potential of developmental delays. These findings suggest that it is the quality of information and access to reliable sources that determines the impact of such material on parental experiences. Indeed, promising

results have been shown with the introduction of an NHS information leaflet before a family has their first appointment with a craniofacial team.[115] The current study illustrates the value of having these materials accessible online but does suggest that the most trustworthy information is not always the easiest to find in the vast Internet landscape. This was further demonstrated by a group of researchers who objectively scored the quality of websites regarding craniosynostosis and found high-quality information sources did not rank top on search engines.[116]

The role of the Internet was also discussed in relation to parents connecting with others in a similar position. Online support groups were frequently mentioned as the main forum for shared experience, which has previously been cited as a protective factor for parents.[94] In one previous study of a craniosynostosis Facebook group, parents named the main reason for joining as seeking and exchanging information.[37] The current study extends this finding and suggests that parents specifically valued reading other families' stories and seeing pictures. Another study interviewing mothers of children with non-syndromic craniosynostosis proposed that this may be reassuring due to seeing how others have 'made it to the other side'.[100] It is also possible that comfort is best found in those who are felt to understand, or know what it is like to be in the same situation. In the current study, this was experienced with a spouse or partner too. Whilst previous research has made reference to the protective nature of family/friends,[98,117] it follows that the underlying benefit of social support is helping parents to not feel alone.

In addition, the study indicated the role of support offered by specialist craniofacial staff members. Healthcare satisfaction has already been identified as a

protective factor within the cleft population[92] which, given the current findings, also seems applicable to non-syndromic craniosynostosis. Parents spoke of the importance of having confidence and trust in the professionals overseeing the care of their child. Moreover, it is well established within psychological literature that the therapeutic relationship is a key component of care and associated with better outcomes.[118,119] This study suggests traits of warmth, approachability and responsiveness were fundamental to the relationship with craniofacial professionals. In turn, this played a protective role against the negative effects of parental distress (e.g., worry). This is in line with the stress-buffering hypothesis which posits that the more support or resources that a person can draw upon, or perceives to have available, can help them feel in control and alleviate negative emotional outcomes.[120]

This would also help to explain why experiences of formal support were less satisfactory and implicated as more of a risk factor when parents did not feel heard when raising their initial concerns or it was difficult to obtain a referral to a specialist centre. Families in previous research have echoed these experiences, suggesting a need for greater efforts to increase craniosynostosis awareness amongst the professionals likely to first come into contact with families such as midwives and health visitors.[100] Moreover, a study investigating the views of non-specialist health professionals reinforced the need for further education as staff reported that they had received minimal, if any, training on congenital craniofacial conditions.[90] Further education would, subsequently, help reduce parental anxieties given that the rate at which craniosynostosis was detected in the current study was dependent on the awareness and understanding held by professionals.

The theoretical model proposes that the extent to which a parent adjusted to the non-syndromic diagnosis was lastly influenced by predisposing factors. Importantly, there is recognition within the model that a parent did not enter the healthcare pathway or clinic space as a 'blank slate' but rather already possessed their own outlooks, coping styles or ways in which to view the world based on previous life experiences. This aligned with the views of professionals working in craniofacial care who deemed traumatic life events and pre-existing mental or physical health concerns as factors that contribute to parental adjustment.[94] Similarly, a cleft study found that the presence of mental health and stressful life events were risk factors for distress in mothers.[92] Both of these previous studies also mentioned that having an optimistic viewpoint was a protective factor.[92,94] This is reminiscent of how parents commented on their dispositional styles and/or personal outlooks in the current study.

Furthermore, parents spoke about employing different strategies to help them cope with the diagnostic process. Examples included keeping busy, spending quality time together as a family or aiming to find a solution. These varying responses could be distinguished as different coping styles, namely emotion-focused or problem-focused. Emotion-focused coping relates to those strategies that aim to reduce the emotional impact of a diagnosis (e.g., distraction), whereas problem-focused would explain efforts to directly manage or alter the stressful situation (e.g., problem-solving).[121] In most cases, parents engaged in a combination of the two.

Clinical Implications

The theoretical model developed in this study has several practical applications. It was striking that the 'starting point' of a families' journey began long before any contact with a specialist centre. This emphasised the need for professionals to acknowledge the complexity of the process that parents may have experienced before their first appointment, providing compassion and reassurance where possible. Professionals should remain aware that families with more complex journeys may find it difficult to adjust and therefore the timing or way in which information is delivered could be crucial. In practice, this could range from informing parents of the promising outcomes for their child, offering follow-up appointments with fewer professionals present, or even wider communication training for staff.

Parental narratives highlighted how professionals' awareness of craniosynostosis, or lack thereof, was often a barrier to receiving a timely diagnosis. Although training for health professionals who encounter families early on in their journey has been documented previously[94] and forms one of Headlines current research priorities[122], this study reiterated the importance and ongoing need for greater awareness. Parents suggested that training should not only encompass knowledge of craniofacial conditions, but also extend to understanding the referral processes to specialist sites. For this to be achieved, specialist sites should make links with other services - such as perinatal support - and become involved in their training programmes. Ideally, in the long-term, staff training would become part of a core standard for professionals at a national level, as opposed to solely operating within local policies.

The model also illustrated the value of peer support and shared experience throughout the diagnostic process. It would be beneficial for services to promote and support such engagement. Parents reflected how sometimes this is difficult due to sites not wishing to endorse unpoliced social media pages. Instead, parents expressed interest in the idea of support groups which could be facilitated by specialist staff or external charities that would visit and provide a supportive space within hospitals. Whilst it is recognised that the implementation of this may be further complicated due to resources and/or funding issues, the current study certainly showed a demand and need for such support. Alternatively, families' stories could be shared by different mediums (e.g., pre-recorded video, resource pack, audio recording) that could be accessed anytime on relevant NHS webpages. This would also ensure information was from a reliable source, which was noted as an important consideration in this study.

The support of more formal networks, such as the relationship with staff at the specialist sites, was noted as positive and reassuring. A leaflet is currently being developed to summarise this feedback that can be distributed to teams, including examples of good clinical practice that were identified in this study. These examples sometimes differed across NHS sites so the provision of such a leaflet may help toward the standardisation of support across centres.

Lastly, the factors proposed by the model could form the basis of a screening tool to help identify parents at risk of psychological distress. Measures that capture the risk and protective factors such as quality of social support, number of life stressors and coping styles could be compiled to form a tool to then be completed around the time of diagnosis. This is not to say all parents who would score highly on risk factors

would meet clinical levels of distress (e.g., anxiety, depression) but may signify that some families may benefit from additional staff contact or even normalisation of their feelings.

Limitations

In common with many craniofacial studies, more mothers than fathers took part. As such, it is likely that maternal narratives are more dominant in the findings. Moreover, the study did not aim to compare mothers' and fathers' views so any potential gender differences have not been explored.

When considering the sample further, three mothers and three fathers formed dyads. This may have meant findings only captured the experiences of a small subset of families. However, parents were interviewed separately to enable them to speak openly about their own individual experience and hopefully ensure the presence of a partner did not influence any responses. Additionally, 10 of the 15 parents reported a sagittal diagnosis for their child. This is unsurprising given that fusion of the sagittal suture is the most common subtype of non syndromic craniosynostosis.[123]

Another limitation is that the parents recruited predominantly identified as White, UK-born, educated and in employment. This was reflected in how parents spoke of being able to manage the challenges of attending appointments with a craniofacial service, such as navigating healthcare systems and/or long distances to travel. There is more to learn about the experiences and support needs of those not captured in the study and that cannot quite so readily adapt to these challenges. Efforts were made to recruit different groups of parents, such as via snowball sampling, but the voices of minority ethnic groups and those with lower socioeconomic status remain underrepresented. This limitation also applies more widely across the craniofacial

literature which forms a larger call for research to diversify samples, increase participation rates and address health inequalities.

Efforts were also made to recruit parents as close to receiving a formal diagnosis as possible, given the study aims of focusing on the diagnosis time frame. However, it is noted that nine parents reported that their child had already undergone surgery at the time of interview. Taking part post-operatively may have affected the interpretations and reflections made by parents, although it is reassuring that the theoretical model was applicable across families irrespective of surgery status. This does indicate that recruiting a completely unoperated sample has its challenges and highlights that with advances in the medical field, surgical procedures are being undertaken at increasingly earlier ages.

Finally, parents volunteered to participate so there is a possibility that families with particularly negative or positive experiences were more compelled to share. However, this did not appear true of the current study as a wide range of experiences seemed to be reported.

Future Research

Following recommendations for the increased use of qualitative approaches in craniofacial research,[14,102] this study provided a rich and detailed insight into parental experiences of a non-syndromic diagnosis. Future research should build on this and take a focus on receiving a diagnosis of syndromic craniosynostosis, to compare findings and see which components from the theoretical model would generalise more widely. For example, it may be that the checkpoints of the healthcare journey are similar for both syndromic/nonsyndromic craniosynostosis up to the point of surgery.

Given the importance of support in the current study, future research could interview staff about their relationships with families and identify what factors they think help make this a helpful connection. This would aid with understanding both sides of the relationship and provide learning points for the healthcare profession about how to foster good working relationships that are valued by parents.

If the clinical implications of this study were to be implemented and a screening tool developed, further research could evaluate this. This may take the form of a large-scale quantitative design to analyse if items included in the screening tool are in fact associated with parental psychological distress or adjustment, and to what degree. This would help consolidate and reinforce any risk and protective factors that have been suggested in the current study.

Conclusion

This qualitative study used grounded theory methodology to construct a model for understanding parental experiences of receiving a child's diagnosis of non-syndromic craniosynostosis. Parents provided detailed narratives about their journey and the different checkpoints that were involved. The findings suggested that information, support and predisposing influences have a role in the adjustment and psychological wellbeing of parents throughout. Consideration of these factors has the potential to guide further research and inform clinical practice for families facing this process in the future; such as identifying training needs, fostering peer/staff relationships and remaining mindful of parents' previous life experiences.

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Appendix 1. Submission Guidelines.

Please read the guidelines below before visiting the submission site!

Submission site

Manuscript Submission Guidelines:

Important Notice of Changes, effective 01/01/2022: CPCJ recently made changes to these below author guidelines, please carefully review these changes before submitting your paper. One substantial change is moving to strictly follow AMA referencing style. This impacts NEW submissions only (that is, new papers uploaded as of January 1, 2022).

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.

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Article Type	Description	Requirements

Original Research	Reports of original clinical or basic science data pertaining to prevalence, causes, mechanisms, diagnosis, course, treatment, and prevention, including systematic reviews and meta-analysis that represent a new contribution to the field.
	
	Traditional, narrative reviews should also be considered in this category.
		• 7,000 words: Body of the manuscript (excluding abstract, references and figure captions)
		• ≤250-word structured abstract* using the below headings:
		  • Objective
		  • Design
		  • Setting
		  • Patients, Participants
		  • Interventions
		  • Main Outcome Measure(s)
		  • Results
		  • Conclusions
		• Minimum of 3 keywords
		• ≤6 figures and tables, combined
		• For systematic reviews and meta-analyses, please follow the PRISMA checklist and include the checklist in your list of files upon submission
		• For Clinical Trials, please include the CONSORT flow chart as a cited figure and the completed CONSORT checklist should be uploaded with your submission
		
		*Narrative review articles may have an unstructured abstract

Appendix 2. Quality Assessment Tool.

Part I: Mixed Methods Appraisal Tool (MMAT), version 2018

Category of study designs	Methodological quality criteria	Responses			
		Yes	No	Can't tell	Comments
Screening questions (for all types)	S1. Are there clear research questions?				
	S2. Do the collected data allow to address the research questions?				
	Further appraisal may not be feasible or appropriate when the answer is 'No' or 'Can't tell' to one or both screening questions.				
1. Qualitative	1.1. Is the qualitative approach appropriate to answer the research question?				
	1.2. Are the qualitative data collection methods adequate to address the research question?				
	1.3. Are the findings adequately derived from the data?				
	1.4. Is the interpretation of results sufficiently substantiated by data?				
	1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?				
2. Quantitative randomized controlled trials	2.1. Is randomization appropriately performed?				
	2.2. Are the groups comparable at baseline?				
	2.3. Are there complete outcome data?				
	2.4. Are outcome assessors blinded to the intervention provided?				
	2.5 Did the participants adhere to the assigned intervention?				
3. Quantitative non-randomized	3.1. Are the participants representative of the target population?				
	3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?				
	3.3. Are there complete outcome data?				
	3.4. Are the confounders accounted for in the design and analysis?				
	3.5. During the study period, is the intervention administered (or exposure occurred) as intended?				
4. Quantitative descriptive	4.1. Is the sampling strategy relevant to address the research question?				
	4.2. Is the sample representative of the target population?				
	4.3. Are the measurements appropriate?				
	4.4. Is the risk of nonresponse bias low?				
	4.5. Is the statistical analysis appropriate to answer the research question?				
5. Mixed methods	5.1. Is there an adequate rationale for using a mixed methods design to address the research question?				
	5.2. Are the different components of the study effectively integrated to answer the research question?				
	5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?				
	5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?				
	5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?				

Appendix 3. MMAT Scoring Guidance.

Reporting the results of the MMAT (version 2018)

In the version 2018, we advised not to present an overall score. This decision was made from the literature that discouraged to use metrics because it is not informative. By presenting a single number, it is not possible to know what aspects of studies are problematic. We often see people presenting a global score and nothing else in the results or discussion or description of included studies. This often raises the question of why quality appraisal was performed.

This suggestion is, however, problematic for reporting the results of the MMAT. Several MMAT users have contacted us for advice to report their results. If there is a need to report an overall score, here is a suggestion based on the previous version of the MMAT:

For each retained study, an overall quality score may not be informative (in comparison to a descriptive summary using MMAT criteria), but might be calculated using the MMAT. Since there are only a few criteria for each domain, the score can be presented using descriptors such as stars (*) or %:

5***** or 100% quality criteria met
4 **** or 80% quality criteria met
3 *** or 60% quality criteria met
2 ** or 40% quality criteria met
1 * or 20% quality criteria met

For mixed methods studies, since there are 15 criteria to rate (instead of 5), the premise is that the overall quality of a combination cannot exceed the quality of its weakest component. Thus, the overall quality score is the lowest score of the study components. The score is 20% (*) when QUAL=1 or QUAN=1 or MM=1; it is 40% (**) when QUAL=2 or QUAN=2 or MM=2; it is 60% (***) when QUAL=3 or QUAN=3 or MM=3; it is 80% (****) when QUAL=4 and QUAN=4 and MM=4, and it is 100% (*****) when QUAL=5 or QUAN=5 or MM=5; (QUAL being the score of the qualitative component; QUAN the score of the quantitative component; and MM the score of the mixed methods component).

Regarding questions on cut off value, we have not studied values that could characterize low, medium or high quality studies. The categories are arbitrary, but useful for performing qualitative or quantitative sensitivity analysis. We have seen some papers with 2 categories (lower vs higher quality) or 3 categories (e.g., low, medium, and high). What is important is to clearly describe how the results of the appraisal were interpreted and used in the review (transparency).

Appendix 4. Data Extraction Tool.

Author(s) and year	
Participant characteristics	
Country	
Method	
Focus of the study	
Key findings	
Study settings	
Measures (if applicable)	

Appendix 5. Data Transformation.

Extraction of studies into categories:

Costa et al., (2022)
- Stress
- Anxiety
- Depression
- Resilience
- Detection
- Information
- Satisfaction with healthcare
- Impact on wellbeing
- Impact on relationships
- Surgery
- A need to advocate
- Burden of care

Coulter et al., (1991)
- Stress

Gray et al., (2015)
- Stress

Kilipiris et al., (2022)
- Satisfaction with healthcare

Kim et al., (2008)
- Stress

Kluba et al., (2016)
- Surgery
- Satisfaction with healthcare
- Quality of communication

Nieroba & Larysz (2020)
- Peer support
- Gathering information

Rosenberg et al., (2011)
- Stress

Rotimi et al., (2021)
- Post-surgery
- Anxiety

Sarimski (1998)
- Stress

Shaw et al., (2022)
- Surgery

Tang et al., (2022)
- Stress

Wong-Gibbons et al., (2009)
- Satisfaction with healthcare
- Information

Dangsomboon & Jirapaet (2017)
- Surgery
- Burden of care
- Hope
- Worry
- Support

Kuta et al., (2020)
- Detection
- Healthcare support
- Information
- Peer support
- Surgery decision making
- Fear
- Relief after surgery

Letourneau et al., (2003)
- Surgery decision making
- Gathering information
- Social support

Netherton et al., (2021)
- Information
- Burden of care
- Social experiences
- Communication with professionals
- Support

Saydam et al., (2021)
- Social challenges
- Support

Zerpe et al., (2020)
- Detection
- Information
- Craniofacial team
- Waiting for surgery

- Communication

Zerpe et al., (2022)
- Surgery
- Hospital staff
- Alone
- Information
- Worry
- Support

These were then integrated:

Detection + a need to advocate

Stress + anxiety + depression + resilience + hope + worry +fear + impact on wellbeing

Social challenges + social experiences

Social support + peer support + support + impact on relationships + alone

Satisfaction with healthcare + craniofacial team + healthcare support + hospital staff

Information + quality of communication + communication + communication with professionals + gathering information

Burden of care

Surgery decision making + surgery + waiting for surgery + relief after surgery + post-surgery

Themes then identified:

Emotional experiences
- Detection
- Difficult emotions

Social
- Challenges
- Support

Service
- Satisfaction with healthcare
- Information
- Experience of treatment (surgery)
- Burden of care

Appendix 6. Quality Assessment Results.

		1. Qualitative					3. Quantitative non-randomised					4. Quantitative descriptive					5. Mixed methods					Overall quality score
	Author(s) and year	**1.1**	**1.2**	**1.3**	**1.4**	**1.5**	**3.1**	**3.2**	**3.3**	**3.4**	**3.5**	**4.1**	**4.2**	**4.3**	**4.4**	**4.5**	**5.1**	**5.2**	**5.3**	**5.4**	**5.5**	
1	Costa et al., (2022)	Yes	Yes	Yes	Yes	Yes						Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	*****
2	Coulter et al., (1991)											Yes	Can't tell	Yes	Yes	No						***
3	Gray et al., (2015)						Yes	Yes	Yes	Yes	Yes											*****
4	Kilipiris et al., (2022)											Yes	Can't tell	Yes	No	Can't tell						**
5	Kim et al., (2008)						Can't tell	Yes	Yes	Yes	Yes											****
6	Kluba et al., (2016)											Yes	Yes	Can't tell	Can't tell	No						**
7	Nieroba & Larysz (2020)											Yes	Yes	Yes	Can't tell	Yes						****
8	Rosenberg et al., (2011)						Yes	Yes	Yes	Yes	Yes											*****
9	Rotimi et al., (2021)											Yes	Yes	Can't tell	Yes	Yes						****
10	Sarimski (1998)											Yes	Can't tell	Yes	Can't tell	Yes						***
11	Shaw et al., (2022)											Yes	Can't tell	Yes	No	Yes						***
12	Tang et al., (2022)						Yes	Yes	No	Can't tell	Yes											***
13	Wong-Gibbons et al., (2009)											Yes	Yes	Can't tell	Yes	Can't tell						***
14	Dangsomboon & Jirapaet (2017)	Yes	Yes	Yes	Yes	Yes																*****
15	Kuta et al., (2020)	Yes	Yes	Yes	Yes	Yes																*****
16	Letourneau et al., (2003)	Yes	Can't tell	Can't tell	Yes	Can't tell																**
17	Netherton et al., (2021)	Yes	Yes	Yes	Yes	Yes																*****
18	Saydam et al., (2021)	Yes	Yes	Yes	Yes	Yes																*****
19	Zerpe et al., (2020)	Yes	Yes	Yes	Yes	Yes																*****
20	Zerpe et al., (2022)	Yes	Yes	Yes	Yes	Yes																*****

5***** meets 100% quality criteria
4 **** meets 80% quality criteria
3 *** meets 60% quality criteria
2 ** meets 40% quality criteria
1 * meets 20% quality criteria

Author(s) and year	Overall quality score	Reasons
Costa et al., (2022)	*****	Well reported, including table with exemplar quotes that clearly illustrates the interpretation of data. Extensive use of measures, albeit not all standardised, and plenty of detail presented regarding sample and analysis. Authors had made good use of service-user/expert by experience involvement in the research design.
Coulter et al., (1991)	***	Appropriate sampling strategy to answer the research question, although limited numbers meant study could not assess for confounding variables. There are reasons given as to why some people did not wish to participate however, no demographics reported for those who took part.
Gray et al., (2015)	*****	Adequate information reported, including clear description of sample, inclusion/exclusion criteria and analysis. Appears methodologically sound with relatively low dropout rates. Use of measure that has been widely used in craniofacial literature to investigate stress.
Kilipiris et al., (2022)	**	Authors acknowledge a low response rate, and the sample obtained is heavily weighted toward non-syndromic population. Measure was devised for the study and the questions included are outlined in the paper, although there is no reference to analysing any qualitative data from the open-ended question.
Kim et al., (2008)	****	Measure appropriate and full outcome data reported, with consideration of confounding variables. No explanation as to why some eligible participants might not have participated, especially as recruitment was over several years.
Kluba et al., (2016)	**	Measure devised for study but no indication as to if this had been trialled or pilot tested. Response rate to measure stated but lacks detail about non-responders. There is an emphasis on descriptive statistics and there may have been potential to further analyse results (e.g., thinking of specific diagnosis, mother/father).
Nieroba & Larysz (2020)	****	Clear research aims and appropriate methodology undertaken. Measure had been tested in a pilot study. Sample is largely non-syndromic. Some query about nonresponse bias, as drop-out is not explicitly specified and so it can only be assumed that all participants completed the measure.
Rosenberg et al., (2011)	*****	Appears methodologically sound and coherent. Good use of validated measure and thoroughly explained. Large sample size.
Rotimi et al., (2021)	****	Good explanation of participants. Measure deemed suitable to answer research question, although not validated and unclear if it had been used before and/or piloted.
Sarimski (1998)	***	Clear use of previously validated measures. Significant percentage of potential participants contacted did not wish to participate but no further justification of this. Also no inclusion/exclusion criteria specified.
Shaw et al., (2022)	***	Varying levels of information given, such as brief demographics presented for children but not parents. Measure brief and devised by staff, although appropriate to answer research questions.
Tang et al., (2022)	***	Well reported, including patient enrolment flow diagram. Unfortunately, not complete outcome data as there was drop outs although this is well explained. Inadequate study numbers for some variables (such as patients with syndromic diagnoses) created 'flooring' effect in regression analysis. Although questionnaires of stress have been previously used within craniofacial literature, measures are not yet validated for caregivers of patients with craniosynostosis.
Wong-Gibbons et al., (2009)	***	Evolved from a larger study. Limited consideration of confounders and methodology unclear without supplementary material, such as interview questions/measures used and what responses can be given. Clear description of sample although specifically focused on mothers, so not necessarily entirely representative of all parents. Demographics could have been clarified in further detail. Data analysis used descriptives and indicated quantitative design, although manuscript read as qualitative study.
Dangsomboon & Jirapaet (2017)	*****	Methodology and data analysis appear appropriate. Full analysis process explained and supported by data.
Kuta et al., (2020)	*****	Appropriate methodology to answer research aims with analysis information clearly derived from data and explained.
Letourneau et al., (2003)	**	Research questions clearly stated and methodology was suitable to answer these. Some quotes given to evidence that interpretation was substantiated by data. However would have benefitted from further clarification – quotes not given pseudonym/participant numbers so unclear if all voices are equally heard. "Particularly eloquent" participants were invited back for third focus group –perhaps introducing bias? No further detail or justification given on these participants.
Netherton et al., (2021)	*****	Clear research aims and appropriate methodology undertaken. Well reported with awareness of limitations and strengths. The reader can follow the journey from data collection – interpretation easily.
Saydam et al., (2021)	*****	Qualitative approach well justified and adequate to answer research aims. Coherence between data collection and interpretation. Well reported with clinical implications, limitations and directions for further research.
Zerpe et al., (2020)	*****	Seems methodologically sound with analysis information clearly derived from data and good amount of information offered about analysis approach.
Zerpe et al., (2022)	*****	As above. Appropriate methodology selected and findings/interpretation appear relevant to collected data. Good visual illustration of steps within analysis process, which signified coherence between data collection and interpretation.

5***** meets 100% quality criteria
4 **** meets 80% quality criteria
3 *** meets 60% quality criteria
2 ** meets 40% quality criteria
1 * meets 20% quality criteria

Appendix 7. Ethical Approval.

Health Research Authority

North West - Greater Manchester East Research Ethics Committee

3rd Floor, Barlow House
4 Minshull Street
Manchester
M1 3DZ

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

27 June 2022

Dr Laura Soulsby
Senior Lecturer
University of Liverpool
University of Liverpool
Department of Psychology
Eleanor Rathbone Building
L69 7ZQ

Dear Dr Soulsby

Study title:	**Exploring the Experience of Parents Whose Children Have Been Diagnosed with Single-Suture Craniosynostosis (SSC): A Focus on Protective & Risk Factors for Psychological Wellbeing**
REC reference:	**22/NW/0190**
Protocol number:	**UoL001687**
IRAS project ID:	**311913**

Thank you for your response of 24 June 2022, to the Research Ethics Committee's (REC) request for further information on the above research and for submitting revised documentation.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Good practice principles and responsibilities

The UK Policy Framework for Health and Social Care Research sets out principles of good practice in the management and conduct of health and social care research. It also outlines the responsibilities of individuals and organisations, including those related to the four elements of research transparency:

1. registering research studies
2. reporting results
3. informing participants
4. sharing study data and tissue

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All research should be registered in a publicly accessible database and we expect all researchers, research sponsors and others to meet this fundamental best practice standard.

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database within six weeks of recruiting the first research participant. For this purpose, 'clinical trials' are defined as:

- clinical trial of an investigational medicinal product

- clinical investigation or other study of a medical device
- combined trial of an investigational medicinal product and an investigational medical device
- other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice.

Failure to register a clinical trial is a breach of these approval conditions, unless a deferral has been agreed by the HRA (for more information on registration and requesting a deferral see: Research registration and research project identifiers).

If you have not already included registration details in your IRAS application form you should notify the REC of the registration details as soon as possible.

Publication of Your Research Summary

We will publish your research summary for the above study on the research summaries section of our website, together with your contact details, no earlier than three months from the date of this favourable opinion letter.

Should you wish to provide a substitute contact point, make a request to defer, or require further information, please visit:
https://www.hra.nhs.uk/planning-and-improving-research/application-summaries/research-summaries/

N.B. If your study is related to COVID-19 we will aim to publish your research summary within 3 days rather than three months.

During this public health emergency, it is vital that everyone can promptly identify all relevant research related to COVID-19 that is taking place globally. If you haven't already done so, please register your study on a public registry as soon as possible and provide the REC with the registration detail, which will be posted alongside other information relating to your project. We are also asking sponsors not to request deferral of publication of research summary for any projects relating to COVID-19. In addition, to facilitate finding and extracting studies related to COVID-19 from public databases, please enter the WHO official acronym for the coronavirus disease (COVID-19) in the full title of your study. Approved COVID-19 studies can be found at:
https://www.hra.nhs.uk/covid-19-research/approved-covid-19-research/

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

After ethical review: Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol

- Progress and safety reports
- Notifying the end of the study, including early termination of the study
- Final report
- Reporting results

The latest guidance on these topics can be found at https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/.

Ethical review of research sites

NHS/HSC sites

The favourable opinion applies to all NHS/HSC sites taking part in the study, subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS/HSC sites

I am pleased to confirm that the favourable opinion applies to any non-NHS/HSC sites listed in the application, subject to site management permission being obtained prior to the start of the study at the site.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Advertisement/Leaflet]	2	22 February 2022
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance Certificate]	1	26 July 2021
Interview schedules or topic guides for participants [Interview Topic Guide]	4	22 April 2022
IRAS Application Form [IRAS_Form_25052022]		25 May 2022
Letter from funder [Letter from University]	1	14 December 2021
Letter from sponsor [Sponsor Approval]	1	10 March 2022
Other [Consent to Contact form]	1	24 June 2022
Other [Response to REC]	1	24 June 2022
Participant consent form [Consent Form V4]	4	24 June 2022
Participant information sheet (PIS) [Participant Information Sheet V4]	4	24 June 2022
Research protocol or project proposal [Research Protocol]	4	04 May 2022
Summary CV for Chief Investigator (CI) [Dr Laura Soulsby CV]	1	18 April 2022
Summary CV for student [Doctoral Student CV]	1	05 May 2022
Summary CV for supervisor (student research) [Dr Laura Soulsby CV]	1	18 April 2022
Summary CV for supervisor (student research) [Dr Anna Kearney CV]	1	20 April 2022
Summary CV for supervisor (student research) [Dr Jo Horton CV]	1	19 April 2022

Summary CV for supervisor (student research) [Dr Louise Roper CV]	1	19 April 2022

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities– see details at:
https://www.hra.nhs.uk/planning-and-improving-research/learning/

IRAS project ID: 311913 Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

On behalf of Mr Simon Jones
Chair

Email:gmeast.rec@hra.nhs.uk

Enclosures: List of names and professions of members who were present at the meeting and those who submitted written comments
"After ethical review – guidance for researchers" *[SL-AR2]*

Copy to: Miss Karen Wilding

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Laura Soulsby
Senior Lecturer
University of Liverpool
University of Liverpool
Department of Psychology
Eleanor Rathbone Building
L69 7ZQ

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

30 June 2022

Dear Dr Soulsby

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

Study title:	**Exploring the Experience of Parents Whose Children Have Been Diagnosed with Single-Suture Craniosynostosis (SSC): A Focus on Protective & Risk Factors for Psychological Wellbeing**
IRAS project ID:	**311913**
Protocol number:	**UoL001687**
REC reference:	**22/NW/0190**
Sponsor	**University of Liverpool**

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

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

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report

(including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

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

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

What are my notification responsibilities during the study?

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

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

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

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **311913**. Please quote this on all correspondence.

Yours sincerely,
Amber Slack

Approvals Specialist

Email: approvals@hra.nhs.uk

Copy to: *Miss Karen Wilding*

List of Documents

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

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Advertisement/Leaflet]	2	22 February 2022
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Insurance Certificate]	1	26 July 2021
Interview schedules or topic guides for participants [Interview Topic Guide]	4	22 April 2022
IRAS Application Form [IRAS_Form_25052022]		25 May 2022
Letter from funder [Letter from University]	1	14 December 2021
Letter from sponsor [Sponsor Approval]	1	10 March 2022
Organisation Information Document [Organisation Information Document]	2	09 May 2022
Other [Consent to Contact form]	1	24 June 2022
Other [Response to REC]	1	24 June 2022
Participant consent form [Consent Form V4]	4	24 June 2022
Participant information sheet (PIS) [Participant Information Sheet V4]	4	24 June 2022
Research protocol or project proposal [Research Protocol]	4	04 May 2022
Schedule of Events or SoECAT [Schedule of Events]	2	09 May 2022
Summary CV for Chief Investigator (CI) [Dr Laura Soulsby CV]	1	18 April 2022
Summary CV for student [Doctoral Student CV]	1	05 May 2022
Summary CV for supervisor (student research) [Dr Laura Soulsby CV]	1	18 April 2022
Summary CV for supervisor (student research) [Dr Anna Kearney CV]	1	20 April 2022
Summary CV for supervisor (student research) [Dr Jo Horton CV]	1	19 April 2022
Summary CV for supervisor (student research) [Dr Louise Roper CV]	1	19 April 2022

Appendix 8. Consent Form.

NHS Birmingham Women's and Children's NHS Foundation Trust

UNIVERSITY OF LIVERPOOL

NHS Alder Hey Children's NHS Foundation Trust

Participant Consent Form

Title of project: Exploring the experience of parents whose children have been diagnosed with single-suture craniosynostosis: A focus on protective and risk factors for psychological wellbeing

Researcher(s): Gemma Hall, Dr Laura Soulsby, Dr Anna Kearney, Dr Louise Roper, Dr Jo Horton

Participant Identification Number:

	Please initial box
1. I confirm that I have read and have understood the information sheet (V4 dated 24/06/2022) for the above study, or it has been read to me. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	☐
2. I understand that taking part in the study involves participating in an interview with a researcher.	☐
3. I understand and agree that my participation will be audio recorded and I am aware of and consent to your use of these recordings for the following purposes: transcription and analysis.	☐
4. I understand that my child's current and historical medical records may be accessed for the purpose of this study by members of the NHS Trust or regulatory authorities.	☐
5. I understand that my participation is voluntary and that I am free to stop taking part and can withdraw from the study at any time prior to anonymisation without giving any reason and without my rights being affected. In addition, I understand that I am free to decline to answer any particular question(s).	☐
6. I understand that I can ask for access to the information I provide and I can request the destruction of that information if I wish at any time prior to anonymisation. I understand that following anonymisation I will no longer be able to request access to or withdrawal of the information I provide.	☐
7. I agree that my anonymised information can be quoted in the publication of this research.	☐

NB: TWO COPIES OF CONSENT FORM TO BE SIGNED - Original to be securely stored at the University of Liverpool and retained by the research team, and a copy provided to participants.

NHS Birmingham Women's and Children's NHS Foundation Trust | UNIVERSITY OF LIVERPOOL | NHS Alder Hey Children's NHS Foundation Trust

8. I understand that my responses will be kept strictly confidential. I give permission for members of the research team to have access to my fully anonymised responses. I understand that my name will not be linked with the research materials, and I will not be identified or identifiable in any publications that result from the research. ☐

9. I understand that other authorised researchers may use my words in publications, reports, webpages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form. ☐

10. I understand that the information I provide will be held securely and in line with data protection requirements at the University of Liverpool. At the end of the study, the data custodian (primary supervisor – Dr Laura Soulsby) will be responsible for the data for ten years after which it will be destroyed. ☐

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

Name of participant	Date	Signature
Gemma Hall		G.Hall
Name of person taking consent	Date	Signature

Principal Investigator
Dr Laura Soulsby
University of Liverpool
Department of Psychology
Eleanor Rathbone Building
Bedford St South
Liverpool
L69 7ZQ
l.k.soulsby@liverpool.ac.uk

Principal Investigator
Dr Anna Kearney
Alder Hey Children's Hospital
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Student Investigator
Gemma Hall
University of Liverpool
Clinical Psychology
Eleanor Rathbone Building
Bedford St South
Liverpool
L69 7ZQ
gemma.hall@liverpool.ac.uk

NB: TWO COPIES OF CONSENT FORM TO BE SIGNED - Original to be securely stored at the University of Liverpool and retained by the research team, and a copy provided to participants.

Appendix 9. Distress Protocol.

Distress Protocol

Modified from: Draucker et al., 2009 Developing Distress Protocols for research on Sensitive Topics. *Archives of Psychiatric Nursing, 23*(5), pp. 343-350.

Mental health (distress) is a potential adverse effect of the study. Participants who are asked to take part in interviews relating to wellbeing and diagnosis may disclose distress, upset, or have questions that they would like answered. Participants may become distressed reflecting on their experience and the impact of their experience on their well-being.

Consent

We will ensure all participants are informed of the potential for distress to occur via consent form. Informed consent will be obtained from all participants before interviews commence. This will consist of essential information on the participant's right to withdraw from the research at any point up to the point of data anonymisation, with clear contact details for the researcher provided should participants wish to withdraw their data. All participants will be given a detailed explanation of what the research entails so they can make a clear judgement on whether they wish to participate, considering all aspects of the study and the expectation on them.

This information will be reiterated verbally and on a copy of the Participant Information Sheet (PIS), which will also include the supervisory team's contact information. During the recruitment period, the researcher will meet with the supervisory team regularly to discuss how recruitment and data collection is going.

Distress Protocol

Researchers will follow a distress protocol, should participants become distressed.

1. Participants will be given the opportunity to take a break from the interview, or withdraw their participation, without having to give a reason.
2. If the researcher notices clear indications of distress during the interview such as crying, indications of flashbacks, incoherent speech etc, then the interview will be stopped.
3. Attempt to assess mental status of the participant – can you tell me how you are feeling? Do you feel you are able to go on about your day? Do you feel safe?
4. If a participant discloses sensitive information that indicates they may be at risk of harm, either from themselves or another person, the researcher will encourage the participant to contact a relevant support group or their regular health provider (staff member, GP, specialist nurse, consultant for example), and will inform the participant that they will have to relay this information to an appropriate authority for their safety.
5. In the event that a participant has experienced emotional distress, the researcher will contact the supervisory team to determine the most appropriate course of action, dependent on the nature of information disclosed.

6. In the event that a participant has experienced emotional distress, the participant will be invited to supply their phone number, explaining that with permission, the researcher or a member of the supervisory team will follow the participant up and contact them the next day to see how they are and if further action needs to be taken. In the case of no number being provided, the participant will be encouraged to contact the researcher or the supervisory team (contact details on PIS) if they experience increased distress in the hours/days following interview.
7. In the instance where a participant feels unsafe or risky (for example expresses suicidal thoughts), there will be a Mental Health Crisis team referral and assessment or the participant will be asked to attend A&E. The patient's GP will also be informed of the risk (as per Clinical Psychology department protocol).

Appendix 10. Researcher Safety Protocol.

Researcher Safety Protocol

This document details standard operating procedures designed to protect researcher safety when conducting qualitative data collection. Safety issues for researchers are paramount. All researchers supervised by Dr Laura Soulsby in the Department of Psychology, University of Liverpool, will be trained in these standard operating procedures to protect their safety. This includes a 'Buddy system', which is designed so that the researcher checks in and out of their interview with a designated 'buddy', usually the research supervisor, who has access to full contact details for the researcher and data collection site.

Researcher training

All researchers will receive training to ensure they are equipped to **confidently and competently collect qualitative data**. In most cases, doctoral-level researchers will have knowledge of the principles of qualitative research and study design, and experiences of using qualitative research methods through their taught programme. To extend this, researchers will attend a series of qualitative training workshops focused on qualitative design, data collection, and data analysis as part of their Doctorate in Clinical Psychology course. All researchers will be trained on how to deal with difficult situations relevant to their study.

Researchers will receive training on how to manage the risk of **participant distress** including forewarning participants of their ability to pause or stop data collection at any point, without explanation. Researchers will familiarise themselves with the *Distress Protocol (V1 dated 05/01/2022)* which outlines what to do if they notice a participant becoming distressed.

Researchers will be reminded of **boundaries and their position as researcher**, ensuring they are clear from the start as to what their position is, how they present themselves, and the boundaries they will put in place should participants request information and support. Researchers will be reminded that data collection is not an opportunity for therapeutic intervention.

Researchers will also receive training on the **mechanisms of support available to them as researchers** and how to access appropriate guidance and support when conducting their research study. Training will include the potential impacts of exposure to sensitive, challenging or difficult material and researchers will be encouraged to engage with continued reflection to identify the impact of their research activity on their own mood and behaviour. Use of a reflective diary may be encouraged to support this.

Regular supervision meetings will provide an opportunity to ensure that the researcher is supported in their work and that onward referral for additional support, where required, is facilitated. Researchers will have the opportunity to discuss with a member/member(s) of their supervisory team any issues or concerns following a data collection event, including experiences of participant or researcher distress.

Appropriate scheduling of data collection

Researchers are advised against scheduling too many data collection events in one day or per week. This is particularly important for research studies involving a sensitive, challenging, or difficult subject matter. Researchers will be advised to conduct a maximum of two interviews in a single day; to avoid scheduling data collection on more than three consecutive days; and to not undertake more than six per week, unless there are exceptional circumstances.

Where possible, data collection events should not take place in the evenings or at weekends. Where it is necessary for data collection to occur outside of normal working hours, there must be sufficient notice for the research supervisor(s) to ensure that they can provide the same level of support as they would during working hours, if and/or when needed.

The 'Buddy System'

For all data collection, virtual and face to face, researchers will engage with the 'Buddy System'. The Buddy System relies on there being an effective, and pre-planned telephone chain. That is, one member of the research team (usually the primary research supervisor) will be a nominated point of contact for the researcher collecting data in the field. A second buddy (usually the secondary research supervisor) will be nominated for when the primary buddy is otherwise occupied, ill and/or on annual leave.

For every individual data collection event (virtual or face-to-face), the field researcher should:

1) **Ahead of the data collection event**, provide the buddy with the date, time (virtual and face-to-face) and location (face-to-face) of the interview via a text message, phone call, or email.
2) **Just before the start of the data collection event**, make contact with the buddy via a text message or phone call, not email, to state the interview or focus group is about to start.
3) During the interview, the interviewer must **keep the phone switched on and close by**, but on silent. Ideally, for face-to-face interviews, researchers should ensure they have unrestricted access to an exit.
4) Make contact with the buddy **when the interview has ended and they are back in a place of safety**, via a text message or phone call, not email. Requests can be made for researcher support/debrief.

After a pre-determined amount of time has lapsed since the first contact commencement message (i.e., 2.5 hours for interviews), the nominated buddy is responsible for contacting the field researcher to ensure they have finished/returned from the data collection location safely. If they cannot get in touch with the field researcher, the buddy must make every attempt to locate and contact the researcher, including alerting the police if necessary.

Appendix 11. Leaflet.

WE ARE LOOKING FOR YOUR HELP!

Who?

Parents/guardians of children who have recently been diagnosed with craniosynostosis (such as metopic, sagittal, coronal).

Why?

We are interested in hearing your experiences as a parent/guardian, to try and understand what helps when receiving this diagnosis. We hope this information would also help us support other families.

What?

You would be invited to take part in an interview, lasting about 60 minutes. This could take place in person or virtually, at a time convenient for you.

How?

If you think you are interested in taking part, or would like more information about the project, please contact:
Gemma.Hall@liverpool.ac.uk

Appendix 12. Information Sheet.

NHS Birmingham Women's and Children's NHS Foundation Trust

UNIVERSITY OF LIVERPOOL

NHS Alder Hey Children's NHS Foundation Trust

Participant Information Sheet

Title of project: Exploring the experience of parents whose children have been diagnosed with single-suture craniosynostosis: A focus on protective and risk factors for psychological wellbeing

Researcher(s): Gemma Hall, Dr Laura Soulsby, Dr Anna Kearney, Dr Louise Roper, Dr Jo Horton

Invitation to Take Part

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

What is the purpose of the study?

The aim of the study is to explore the experience of receiving a craniosynostosis diagnosis from the perspective of the parent/guardian. More specifically, we want to learn more about what helps and what can hinder psychological wellbeing when your child receives this diagnosis. We hope to use this knowledge to support other families in the early stages of their craniofacial journey and help guide what the craniofacial service offer.

One member of the research team (Gemma Hall) is currently enrolled on a Doctorate in Clinical Psychology (qualification that allows someone to practice as a Clinical Psychologist) and this study will be undertaken in conjunction with completing this programme.

Why have I been chosen to take part?

You have been asked to take part because you have been identified as a parent/guardian of a child who has recently received a diagnosis of single-suture craniosynostosis (could be known as metopic, sagittal, coronal or non-syndromic craniosynostosis). We are seeking participants from two nationally designated craniofacial centres; Alder Hey Children's Hospital and Birmingham Children's Hospital.

Do I have to take part?

Taking part is completely voluntary and you are under no obligation to take part. Your decision to take part will have no impact upon the treatment you receive. You are free to withdraw at any time during the study without giving a reason. Once your data has been anonymised, we will not be able to identify your data and we are unable to withdraw you from the study. The data will be anonymised approximately one month after the interview date.

What will happen if I take part?

If you choose to take part, then you will be asked to sign a consent form which outlines you have read all this information and understand what is involved in taking part. This will ask for your consent for the research team to access your child's medical records. This will only be used to help the researcher understand what diagnosis and treatment your child has received.

You will then be asked to take part in an interview with a Trainee Clinical Psychologist. This can take place in person or via online methods (i.e., Zoom or MS Teams). Questions will focus on your craniofacial journey, family and friends and your wellbeing. The interview will last around 60 minutes.

The interview will be audio-recorded, but only with your permission. The audio-recordings will be transferred via a secure online platform to a professional transcription service, where all identifying information (e.g., names, places) will be

anonymised during transcription. Only people involved in the research and transcribing process will have access to the recordings.

Expenses and / or payments

Each parent/guardian who completes the interview will receive a £10 Amazon voucher, as reimbursement for taking part in the study. Travel costs up to the value of £15 will be covered for each family.

Are there any risks in taking part?

There are no anticipated risks to taking part in this study. If you experience distress or are upset, you are free to stop or pause the interview at any time you wish and you can withdraw from the study at any time, without explanation, up until the data has been anonymised. If you do experience any discomfort or disadvantage as a result of the study, please let the researchers know immediately. You may also wish to discuss this with your GP and/or specialists involved in your child's care. You will not have to answer any questions that you do not feel comfortable with.

Are there any benefits in taking part?

There are no known direct benefits from taking part, though we hope that the findings will help to improve the support we can offer families when their child receives a diagnosis of craniosynostosis.

How will we use information about you?

We will need to use information from you for this research study. This information will include your name and contact details, and possibly your child's medical records. People will use this information to do the research or to check your records to make sure that the research is being done properly. People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

What will happen to the results of the study?

The results of the study will be written up as part of a Doctorate in Clinical Psychology (qualification that allows someone to practice as a Clinical Psychologist) and will be published in a peer reviewed journal (where research is published and can be accessed). Participants will not be identifiable from any of the results indicated in either of these formats. All the information collected during the course of the study will be kept strictly confidential.

All who take part are welcome to hear about the results of the study if they would like and can receive a copy of the paper or brief overview of the results. If you would be interested in this please contact Gemma Hall (gemma.hall@liverpool.ac.uk) who will be able to send you this information when it is available.

What are your choices about how your information is used?

You can stop being part of the study at any time, without giving a reason, until the point of data anonymisation. After that time, we will keep information about you that we already have. We need to manage your records in specific ways for the research to be reliable. This means that we won't be able to let you see or change the data we hold about you.

Where can you find out more about how your information is used?

You can find out more about how we use your information

- at www.hra.nhs.uk/information-about-patients/
- our leaflet available from www.hra.nhs.uk/patientdataandresearch
- by asking one of the research team via details below

- by sending an email to sponsor@liverpool.ac.uk

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

If you are unhappy or if there is a problem, please let us know by contacting the Principal Investigators: Dr Laura Soulsby (l.k.soulsby@liverpool.ac.uk) or Dr Anna Kearney (anna.kearney@alderhey.nhs.uk) and we will try to help. If you remain unhappy or have a complaint which you feel you cannot come to us with then you should contact Research Integrity at the University on 0151 794 8290 or at ethics@liverpool.ac.uk. When contacting them, please provide details of the name or description of the study (so that it can be identified), the researcher(s) involved, and the details of the complaint you wish to make.

Who can I contact if I have further questions?

Principal Investigator
Dr Laura Soulsby
University of Liverpool
Department of Psychology
Eleanor Rathbone Building
Bedford St South
Liverpool
L69 7ZQ
l.k.soulsby@liverpool.ac.uk

Principal Investigator
Dr Anna Kearney
Alder Hey Children's Hospital
Clinical Health Psychology
The Institute, 2nd floor
Eaton Rd
Liverpool
L12 2AP
anna.kearney@alderhey.nhs.uk

Student Investigator
Gemma Hall
University of Liverpool
Clinical Psychology
Eleanor Rathbone Building
Bedford St South
Liverpool
L69 7ZQ
gemma.hall@liverpool.ac.uk

Appendix 13. Consent to Contact

NHS Birmingham Women's and Children's NHS Foundation Trust

UNIVERSITY OF LIVERPOOL

NHS Alder Hey Children's NHS Foundation Trust

Consent to Contact for Research Purposes

Title of project: Exploring the experience of parents whose children have been diagnosed with single-suture craniosynostosis: A focus on protective and risk factors for psychological wellbeing

Researcher(s): Gemma Hall, Dr Laura Soulsby, Dr Anna Kearney, Dr Louise Roper, Dr Jo Horton

You are being invited to give consent for a qualified member of the study team to contact you at some time in the future to invite you to participate in a research study.

Are you willing to learn more about the above study? (Circle one)

YES NO

If yes, you will be contacted at a later date. Please include contact information below.

☐ ***Email***: _____

☐ ***Telephone***: _____

Please indicate the best time to reach you by placing an **X** in the relevant boxes:

	Morning	Afternoon	Evening
Monday			
Tuesday			
Wednesday			
Thursday			
Friday			
Saturday			
Sunday			

Signature: _____

Date: _____

Clinician's Name: _____

Appendix 14. Topic Guide.

NHS Birmingham Women's and Children's NHS Foundation Trust

UNIVERSITY OF LIVERPOOL

NHS Alder Hey Children's NHS Foundation Trust

Interview Topic Guide

Title of project: Exploring the experience of parents whose children have been diagnosed with single-suture craniosynostosis: A focus on protective and risk factors for psychological wellbeing

Start of interview: I would like to learn more about your experiences as a parent/guardian during the diagnosis process; I'm particularly interested in your views about what may have helped, or not helped, how you felt during this process. This is just about your experiences – no right or wrong answers.

The interview will take about 60 minutes and I will record the interview with your permission.

I remind everyone that I speak to that it's up to you if you want to pause or stop the interview at any time. All interviews for this study will be used to write a final report which will be submitted as part of a Doctoral thesis and also used with the Craniofacial team to see how we can support parents/guardians. Interviews will be transcribed but all identifying information will be anonymised. I will include some quotes to illustrate what people have said in these reports but your name will not be used; everything you say will remain confidential.

Is that ok? Do you have any questions?

Before we begin, can I check that you are happy with me to turn the recorder on?

1) Demographics

"A few questions now on demographics so we can ensure our research covers a range of participants…"

a. How old are you?
b. What is your gender?
c. What is your ethnicity? (Or you can choose 'prefer not to say')
d. Which part of the UK do you live in? Postcode?
e. What is your highest education level?
f. How would you describe your job role or current employment status?
g. How would you describe your current family set-up? (i.e. spouse/partner, other children)

h. How old is your child under the Craniofacial team?

i. What is your child's gender?

j. What is your relationship to your child? (e.g. mother, father)

k. In which hospital did your child receive a diagnosis?

l. Has your child had any surgery related to diagnosis?

m. Was genetic testing offered to you?

n. Do you have a family history of craniosynostosis? (i.e. others in your family now or in the past)?

o. Does your child have any other health conditions?

p. Are there any concerns about your child's development?

2) Journey to diagnosis

"If I could now just ask you about your journey up until this point. I will start by asking you…"

a. Without going into specific detail, what has been your personal experience of illness/hospital prior to your child's diagnosis?

Prompts
- *Own diagnoses*
- *Past medical experiences (extending to family)*

b. Can you tell me about when your child was diagnosed and what it was like for you?

Prompts
- *When did your child get a diagnosis*
- *Child's specific diagnosis*
- *How did this come about*
- *How long did it take*
- *How did it feel*
- *Who diagnosed and communicated*
- *How well was condition explained*
- *Particular staff groups who facilitated or hindered the process of diagnosis*
- *Where you listened to*
- *Key milestones in journey so far*
- *Was there any other stressful life events happening at the same time (e.g. major changes, bereavement)*
- *What was your (cultural) understanding of the diagnosis*

- *Other people's understanding of diagnosis e.g. mother, father, grandparents…*
- *What did you think of the future when you got diagnosis, what do you think of the future now*

"I'm curious about the support you've received during this time you've just described. We can break support down into formal and informal support. Formal tends to be from professionals like in the hospital where they are paid for their work, whereas informal is more about your social network and community. I will start asking about formal, so…"

c. Following diagnosis, can you tell me about the formal support you received?

Prompts
- *What was helpful/unhelpful*
- *Where you provided with the information about diagnosis and/or surgery that you needed*
- *What format did you receive this information*
- *Have you received any psychological support*
- *Satisfaction with formal support*

"Now I'm going to ask you about support that wasn't from the hospital or any other professionals…"

d. Following diagnosis, can you tell me about the informal support you received?

Prompts
- *What was helpful/unhelpful*
- *Who is your support network and who are you closest to*
- *Family/friends*
- *Do you enjoy spending time with these people*
- *Satisfaction with informal support*

3) Adjustment and wellbeing

"I would like to know how you really felt during this time-frame of diagnosis. I'm trying to get a sense of how, or even if, this experience may have impacted you. Let's start with…"

a. How would you describe yourself as a person and your feelings before child was diagnosed?

Prompts

- *Emotional wellbeing*
- *When faced with uncertain times, how would you have described your general outlook on things (your expectations, would you expect the best/worst)*
- *How did you adapt with changes and stressors in the past*
- *How often were you stressed/upset*
- *History of mental health condition*

b. How would you describe yourself and how you feel now?

Prompts

- *Same prompts as above, but applied to now*
- *Has your child's condition had an impact on your wellbeing*
- *Mood*
- *Do worrying thoughts go through your mind and frequency of these*
- *How able do you feel to control your worries and feelings*
- *How easy is it to relax*

c. Can you describe any impact the diagnosis has had on yourself?

Prompts

- *Any impact on daily life or things you used to enjoy*
- *Would you describe any impact on your relationships*
- *Practical factors such as distance, travel, finances, childcare*
- *Can you tell me about the positives in your life or anything for the better that has happened since diagnosis*

d. How able are you to access the support you need now?

Prompts

- *Both formal and informal*
- *Who still supports and doesn't support and how*
- *Why do/don't they support*
- *What is the effect of this*
- *Has the understanding of your family/friends of diagnosis changed*

e. How well do you feel you have adjusted? What has helped/hindered this?

Prompts
- *Most important thing about the support received*
- *Coping strategies, what do you do when you feel a certain way (optimistic viewpoint, keeping busy)*

4) Improving Services

"I'm wondering if there's any way we can make a difference to other families…"

a. How could your experiences of diagnosis have been improved and/or how could it be improved in the future for other people?

b. To what extent do you feel the way you received the diagnosis influenced how you have adjusted?

c. Based on your personal experiences, what advice would you give someone with a similar diagnosis?

5) Ending

"Thanks very much for answering those questions…"

a. Are there any things not covered so far you would like to add to this conversation?

b. Would you like to be sent a copy of the study findings when they are published? If so, in what format?

c. Finally, do you have any questions for me?

6) Debrief

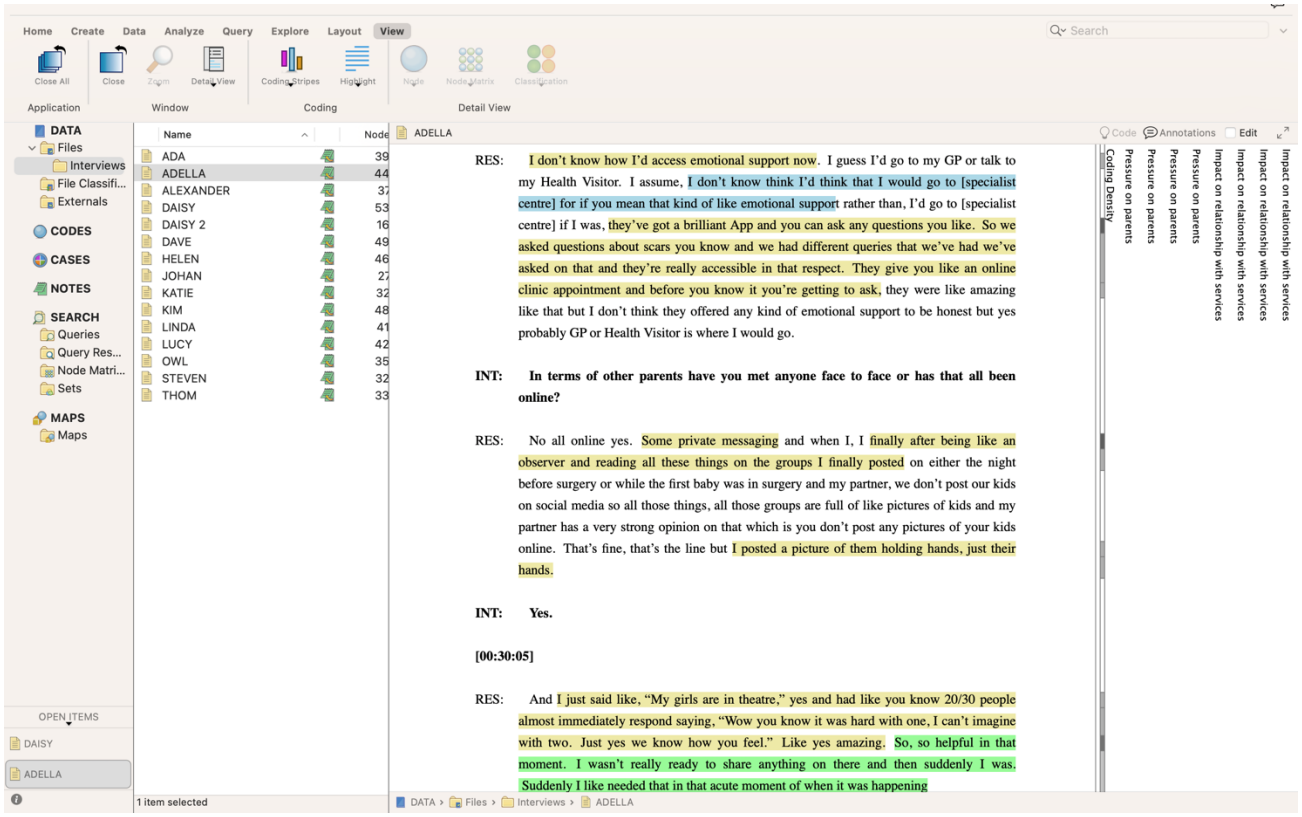
a. Debrief sheet

b. Choose a name to be known by in the research?

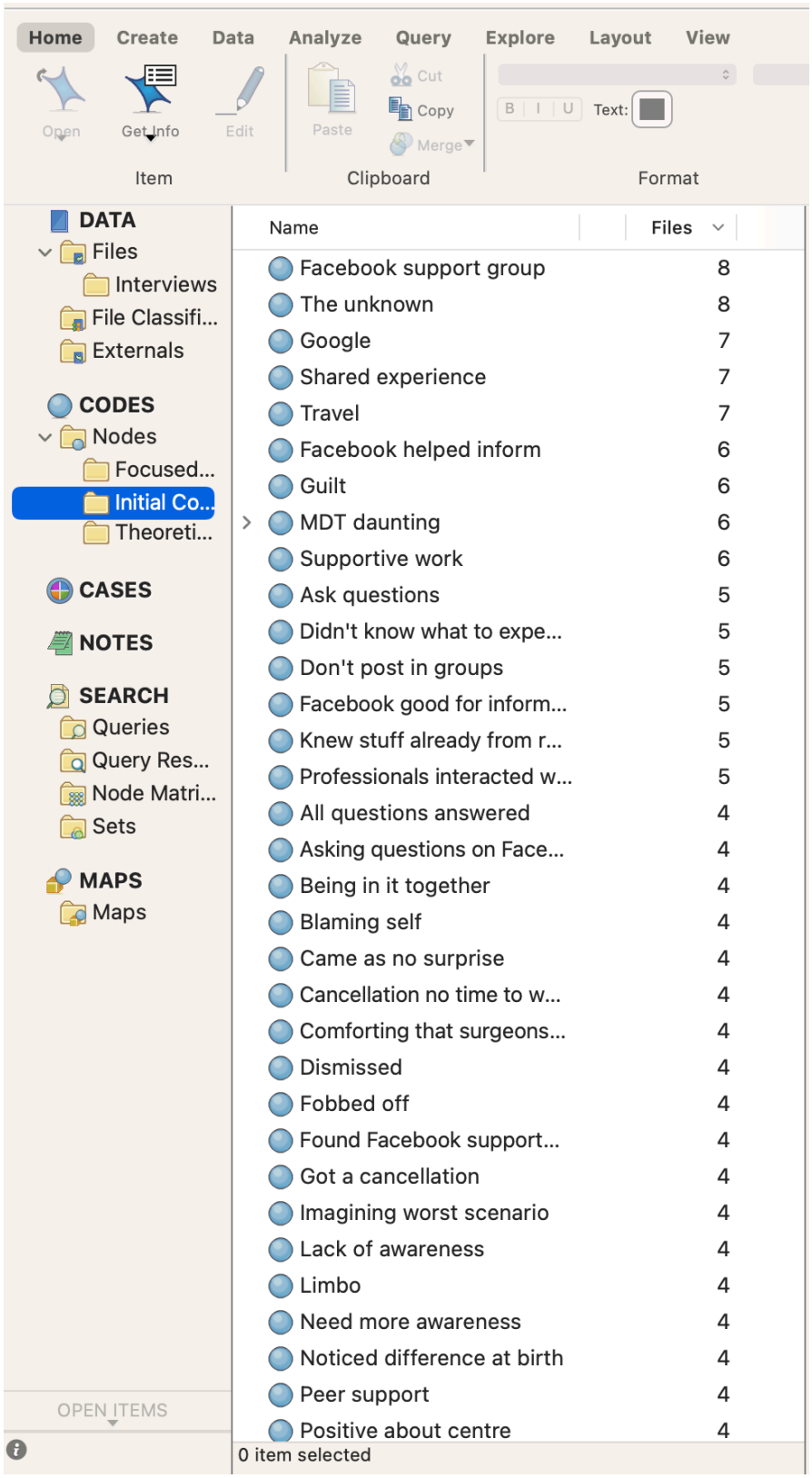
Appendix 15. Topic Guide Refinement.

After interview number	Refinement
2	Prompted about the support of workplaces when considering formal/informal support
4	Moved the question “Without going into specific detail, what has been your personal experience of illness/hospital prior to your child’s diagnosis?” to later in the interview (if deemed appropriate) as was felt to be too invasive as an introductory question
5	Deleted question “To what extent do you feel the way you received the diagnosis influenced how you have adjusted?” as was generating no new insights and was unclear

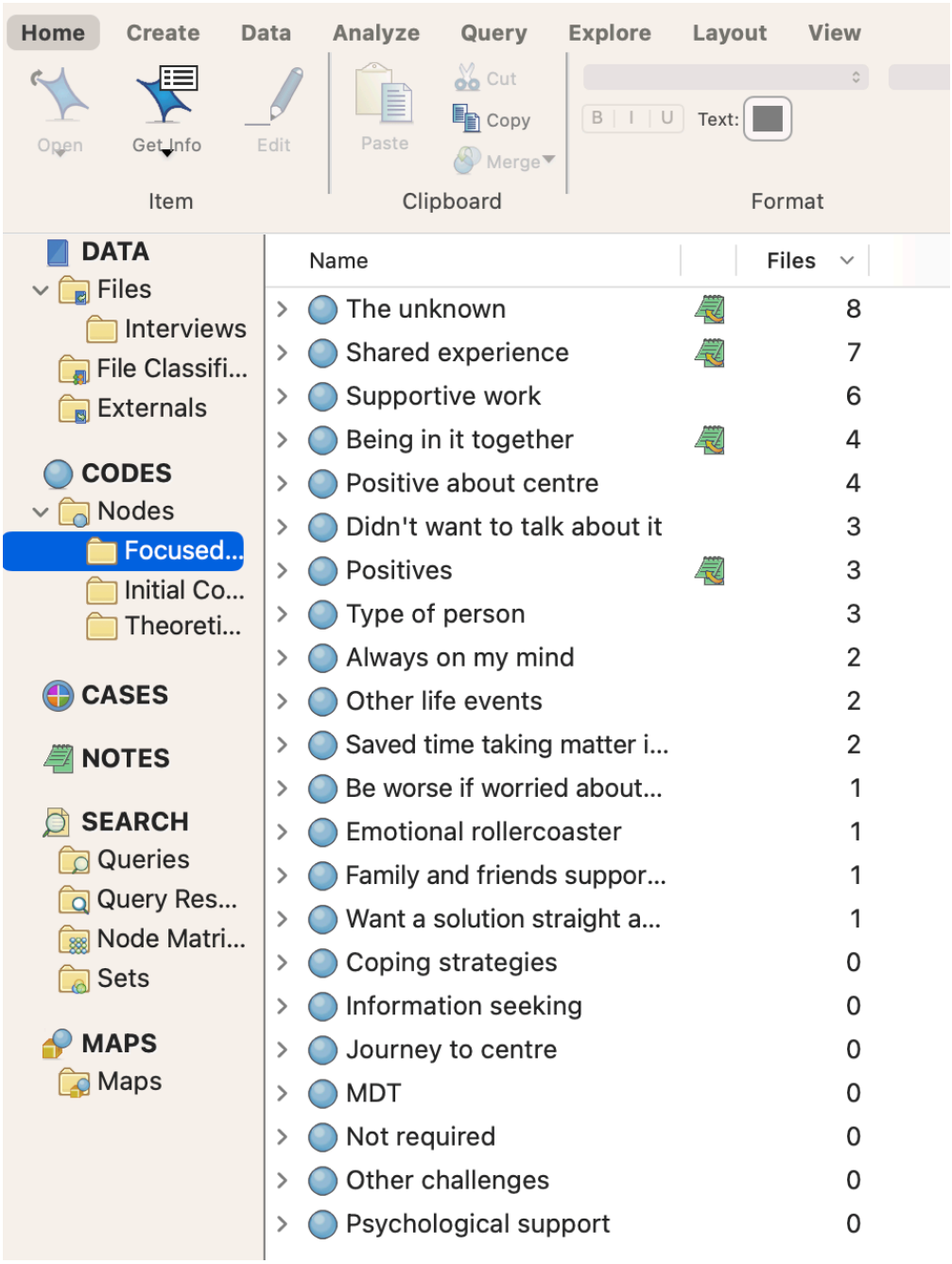
Appendix 16. Example Transcript.



Appendix 17. Example of NVivo Initial Coding.



Appendix 18. Example of NVivo Focused Coding.



Appendix 19. Mapping Focused Codes onto Theoretical Codes.

Key:
Theoretical codes
Focused codes
Initial code examples

Predisposing Influences
Ways of coping
Distraction
Keeping busy
Type of person
Making things feel 'normal'

Prior life experiences
Mental health
Previous hospital experiences
Other stressors
Life events

Starting Point
Fobbed off
Dismissed
Just squished
Told nothing to worry about
Delay

Lack of awareness
Numerous professionals interacted with
Never heard of it before
Need more awareness

The Unknown
A lack of answers
Worst part
In the dark
More information needed

Waiting
Life on hold
Wait impacts surgery options
Limbo
Hard to plan for future
Long process

Information Seeking
The driving force
Pushed for referral
Started reading straight away
Had to chase
Feel responsible
Need to advocate
Parent sped up process
Being prepared

Web of misery
Didn't know what to trust
Took as gospel
Posts can make you second guess
Cautious of information source
Tried to avoid reading

Support
Formal
Sensitive support
Interpersonal skills
It's the people
Approachable
Welcoming staff

Trust and faith
Experts
With the right people
Experienced team

Responsive
Calm
All questions answered
Speed of reply
Point of contact

Informal
In it together
Appointments together
Talk things through
Solo parent
Family and friends
In it as a family
Two sets of ears

Shared experience
Facebook
Learn from others
Peer support
Informal questions
WhatsApp group
Spoke to other parents
Reading others' stories

Accessing Craniofacial Service
Other challenges
Travel
Parking
Expense
Hotel
Other children

Appendix 20. Summary Sent to Participants.

NHS Birmingham Women's and Children's NHS Foundation Trust

UNIVERSITY OF LIVERPOOL

NHS Alder Hey Children's NHS Foundation Trust

Research summary so far - Exploring the experience of parents whose children have been diagnosed with single-suture craniosynostosis

Researcher(s): Gemma Hall, Dr Laura Soulsby, Dr Anna Kearney, Dr Louise Roper, Dr Jo Horton

Once again, thank you for taking part in the study exploring your experiences of the diagnosis process. Offering your time and sharing your experiences was really appreciated as it made the study possible and helped us discover some key findings. The study included fifteen parents in total – 10 mothers, 5 fathers - who all offered a valuable contribution to the research. Once interviews were completed, they were transcribed and examined to understand patterns in peoples' experiences and to help develop a model that summarised experiences overall. This document presents a summary of the key findings and a draft version of the model that has been developed. We welcome your thoughts on this. Your feedback will be used to help refine the final write-up.

All participants referred to different periods or stages of their journey to diagnosis. The diagram showing the model (on next page) has been designed with these different stages in mind. The length of time that each parent was within any given stage varied. For example, some participants spent longer than others trying to access support from a craniofacial centre. The model shown here is focused on the journey to diagnosis so any information shared about surgical procedures has not been included for now.

In most cases, participants spoke of noticing a difference at birth. However, raising concerns or asking questions at this point was **not always taken seriously** and led to a sense of feeling **dismissed or 'fobbed off'**. Parents often interacted with **many** healthcare professionals such as paediatricians, GP, health visitors and consultants to explore what their concerns could be. A professionals' awareness and understanding of craniosynostosis was crucial here. Most participants spoke of a **lack of awareness** of the condition which potentially impacted how quickly a child was diagnosed. In a couple of cases, either the parents or a professional were already aware of craniosynostosis which arguably sped up the process. This period of time, whilst parents **wait in 'the unknown' for answers**, was often described as the most challenging time.

To try and help move things along, there was an emphasis throughout the interviews about **gathering and seeking out information**. This could have been Googling, researching online, reading books or articles, watching videos or social media. Information seeking tended to start quite early and there was a real sense that parents felt a responsibility or pressure to be **the driving force** and find answers themselves. For some participants, this is how they found out about the specialist craniofacial centres (e.g., Alder Hey, Birmingham, Oxford, Great Ormond St) and even contacted them directly. Gathering information was also spoken about positively as parents found it helped **prepare and inform them** for what might happen next. Although, there was a downside to looking for information as it could be overwhelming.

There was a sense of relief for parents when a referral was made and accepted to a specialist centre as this signified finally **being in the right place with people who could help**. In this way, parents spoke of the importance of feeling able to **trust** and have **faith** in professionals. Parents described the first time meeting the craniofacial team in a multidisciplinary setting which could be perceived as daunting and/or intense. There tended to be many staff members in the room and information was presented in quite a factual, clinical way. For some, this direct approach was preferable but others found it overwhelming and had not known what to expect.

A significant finding was the importance of **feeling supported** throughout the journey to a diagnosis. This was both formal support, with the likes of the specialist centres, and also informal support relating to family/friends and parents in a similar position. Feedback about staff, particularly clinical nurse specialists, at the centres was glowing. This was often less to do with *what* they did, but *how* they did it. For instance, parents valued the **interpersonal skills** embodied by staff such as being warm, welcoming, approachable and responsive.

Outside of the hospital, support was provided by family members, friends, partners/spouses and peers. Participants spoke of how their experiences could be isolating at times so there was a focus on what helped parents **not feel alone** during their journey; whether this was having someone else to attend appointments with or speaking to parents going through the same process. Facebook was mentioned on numerous occasions as a place of **shared experience** and even if parents did not wish to post themselves, reading the stories of others online was felt to be reassuring. Whilst **family and friends** could provide practical support such as childcare arrangements or words of encouragement, participants found connecting with others who were in their shoes (i.e., their partner or other parents) of added value.

Logistics such as childcare, in addition to time off work, were especially important when considering attending appointments. Many parents had a long commute to access a craniofacial centre so had **additional challenges** to navigate such as travel, parking, finances and accommodation.

The model proposes that the extent to which a parent may adjust to this process is impacted by **how they cope generally**, **their personal outlook or attitude** and **prior life experiences**. For instance, some participants described how they do not like to overthink or worry about things in advance whereas others described a desire to quickly find a solution. In addition, many participants shared previous life events that had influenced their views about hospitals, their perspective on difficult times and/or contributed to their current stress levels and emotional state.

Appendix 21. Feedback from Participants.

1

Thank you very much for sharing this. No particular comments or thoughts, just that I think it definitely reflects what we spoke about.

I also just wanted to let you know that we used the Amazon vouchers to buy things from the wish list that the cranio ribbons Facebook page has, they send out hampers full of helpful things to parents before their children go through the surgery. So thank you again for the vouchers - they went to a very good cause!

Good luck with the rest of your research, looking forward to reading the full thing.

2

It has been lovely to hear from you again, I hope you are well!

I had a read through your draft and think it is exceptional! The only thing I might add is a mention of the apps like 'MyChart' that XX use so parents can message the nurses and staff directly anytime with questions or for reassurance. I felt that made a huge difference in feeling supported by XX and the team despite how far away the hospital is from us.

I look forward to seeing the final write up when it's ready!

3

Thanks Gemma, this is a really interesting read. I think as XX issue was picked up in the post birth paediatric check, and dealt with swiftly and comprehensively, I am probably not in the majority, so much of this does not ring true for me. I'm an outlier! But I wouldn't expect any edits due to that.

4

Looks great!

5

Hope you're keeping well and thanks for getting in touch with your draft findings. Myself and my wife had a read through over the weekend and we were really moved by the findings in your report and we felt a sense of reassurance that many of the things that we had felt and been through during the process were experienced by others and I think you have captured the process really well in the model.

I just have a few points that really stood out to me....

I think the lack of awareness of the condition from professionals outside of the craniofacial centres is a really important point and I wonder if this is a feedback point that could be somehow fed back into the system either by the craniofacial specialist units or through the work that Headlines do. I would also suggest that this lack of awareness goes beyond just recognition of the condition but the process that should

be applied by hospitals, GPs etc when the condition is suspected. For example in our case, the Consultant Paediatrician who dealt with XX when he was born actually recognised the condition straight away but was unaware of the process for referral. Initially we were told that a CT scan would be done prior to XX being released from hospital, then we were told it would be as an outpatient, then we had to chase this up ourselves and were told that no scan would be done but we needed to see another Paediatrician. By this point we had done our own research and insisted that a Paediatrician at XX refer us to XX which was eventually done.
The Paediatrician then suggested that it would be helpful for XX if they did an x-ray at XX which was done. As it happens XX did not need this to diagnose and in fact only a CT scan was required at the point of operation. So even after diagnosis, the process followed was confusing and incorrect and led to an unnecessary x-ray being done and an appointment with a second Paediatrician that wasn't required.

I think the point about the downside to information gathering is particularly important as well. One of the main stressors for myself was the overwhelming amount of information regarding the large scale of variety of craniofacial conditions. To an extent that anxiety has remained with me even now. XX provided lots of reassurance once we were finally in their system but I cannot unread what I read if that makes sense and so I hold concerns over speech and language and other aspects of development and even with the reassurance offered by XX, who I trust, I struggle to overcome the previous research that was done while in 'The unknown' area of the model. I think this is another reason why, in my opinion, information and communication in the whole diagnosis process needs to be clearer.

Finally, I just want to comment on the part after the diagnosis and appreciate this is probably outside the scope of your research, but I just thought I would mention it. I think the next big challenge in the 'accessing craniofacial services' is deciding what procedure to go for. Most hospitals now or will soon offer a variety of procedures for single suture craniosynostosis and we found decision making around procedures extremely difficult. We opted for the endoscopic followed by helmet therapy, a decision that we made based on our own perceived benefits to XX of dealing with this problem early and at a young age. Since choosing that option we felt that there were many unanswered questions regarding the helmet process, and we felt we fell into a small gap between the care of XX who managed the operation side of things and Technology in Motion who managed the helmet. This led to some confusion and frustration over trying to get answers to questions about the helmet process. I think this just adds to my point above that, in my opinion, the whole process from observation of a problem to treatment selection and action needs just a little bit clearer communication and information.

Hope this all makes sense.

Thanks again for sending this over and well done with the research, I think some really key points have been identified and your model is certainly very representative of our experience.

6

Thank you very much for sharing your findings. They are great findings which I can certainly relate to. Reading through the starting point evoke memories in me and nearly made me cry. Oh well… it's great there is such research out there, and I hope there will be more awareness of single-suture craniosynostosis among medical practitioners so that families receive help on time.

It was a great pleasure to take part in this study. All the best wishes with it and with writing it up!

7

My initial reaction is you've perfectly encapsulated our feelings during the period and slide 2 sets this out in a really concise manner. I'll have another read during the week to see if there's anything I can add on reflection to help you refine but it's really good and my wife thought so too!

8

Thanks for your email and apologies it took me this long to reply. The summary seems like a really accurate reflection of our own conversation and there's a comfort in that we all share really similar experiences/knowing it's not just me.

Appendix 22. Reflexive Statement.

I am a 28-year-old White British female, training to be a Clinical Psychologist. I have previous paid clinical experience of working with families who have received a diagnosis of craniosynostosis. This experience, plus academic teaching on the DClin programme about physical health in paediatric settings, has allowed me to consider some of the challenges families may face and influenced my choice of research topic.

Whilst working as an Assistant Psychologist in a paediatric craniofacial team, I was struck by how families were invited into a room full of professionals and told that their child (often only a few months old) had craniosynostosis. I recall wondering how this must have felt as often appointments were brief and left little time to digest what had just been said. Certainly before starting this job role, I had no idea what craniosynostosis was so I expected many parents may have felt the same. This is an expectation I held for my research study too. It may be important to note that I am not a parent myself and therefore my assumptions come from my previous clinical experience and not first-hand knowledge of parenting.

As part of my Assistant role, I met with families throughout the child's lifespan to see how things were going and to provide regular reviews. These appointments were mainly concentrated on the child – how they felt, how they were getting on at school and how they performed on a cognitive assessment. There was less time given to asking parents how they were and how they were coping. Instead, only a small proportion of parents/family members received regular, dedicated psychological support. I remember this is something that was discussed in supervision and later prompted the development of this research project. It raised questions of: who should support be offered to? Do some parents/families require increased psychological support compared to others? How can we identity these? And what do parents/families wish for support with at the time of diagnosis? As a Trainee Clinical Psychologist, these are questions that spring to my mind naturally but often in a medical setting these seemed to be less commonplace.

There has been more psychology research being conducted into the craniofacial population in recent years but the main focus seems to be on cleft lip and/or palate. Out of the research available on craniosynostosis, there is some interesting new papers (published 2020/2021) exploring parents' experiences however there is no model developed to encapsulate these findings. Consequently, I felt a grounded theory methodology would be helpful to build on previous research and also build a model. I have had prior experience of

conducting qualitative research projects and have always valued the rich data and meaningful narratives that the methodology provides. I feel my previous research work has equipped me with the skills and confidence to conduct participant interviews in a semi-structured way.

My expectations for the research are that parents/families will have endured a long process to receive a diagnosis and that they will not have heard of craniosynostosis before. I assume they will feel shocked and emotional, with questions about what the diagnosis means for their child's future. I feel like recruitment and the process of generating interest in the study will be slow as advertising for a study around the time of diagnosis may feel too overwhelming for some people. I expect more mothers than fathers will wish to take part, although I would hope to recruit an equal number of each as I wonder if there are any differences between maternal and paternal experiences. I expect social networks and being satisfied with the help and support of staff will be a protective factor for parental distress and/or adjustment. Previous literature seems to suggest that pre-existing mental health will be a risk factor.

It will be important to revisit these initial expectations and assumptions as the research project evolves, to make sure data analysis and the model is grounded in participant narratives. Utilising memos, research supervision, reflective diary and encouraging feedback from my participants will help with this also.

Appendix 23. Excerpts from Research Diary.

April 2022
Seeking parent involvement

I spoke with a parent today who has a son with a diagnosis of craniosynostosis. She had looked over my research materials and was happy to give me some feedback. She mentioned that there is a WhatsApp group with other parents on that she has found the most supportive thing of all. I didn't know such a thing existed so it will be interesting to see if this or anything similar comes up in the interviews.

It struck me that her day job as a social worker might help her challenge things, so more likely to ask questions. For example, she described herself as being vocal. Maybe this would differ to parents who don't feel confident?

November 2022
Interview 4

She got really upset during the interview. This made me feel sad because this time frame of not knowing what's going to happen at next appointment/surgery must be so tough. I can only imagine.

November 2022
Interview 5

Found this interview much harder and challenging to conduct. He was less open in his answers than my previous ones. This makes me wonder whether it was my interviewing style and whether I prompted sufficiently enough for detailed answers – transcribing and paying attention to the flow of the interview will allow me to reflect more on this. Alternatively I was wondering what not being open in an interview could represent – difficult content? Difficult to speak about?

January 2023
Research supervision

Spoke about what themes were coming up in the interviews so far. I reflected on how some people who have had surgery said it was hard to get them back to discussing that time frame between diagnosis and surgery. Thought about how this is like a coping strategy, distancing self from the memory of how difficult that time was, otherwise they would get really upset?

January 2023
Undergoing analysis

A theme of not being alone seems to be coming up a lot. This can be both metaphorically and literally as most people valued going to appts with someone else, and also valued having someone else there emotionally. Another theme that seems to be occurring is the value of shared experience – perhaps even more so from other parents than friends and family? Why could this be? Because other parents are in same shoes?

February 2023
Recruitment complete!

I managed to interview 15th participant today, which marks the end of recruitment. It has actually been faster in terms of recruitment than I imagined. I had expected to struggle. I find myself wondering why this is, why people were so keen to participate immediately? I think it is has something to do with helping other parents and raising awareness at all points. Many parents spoke of the lack of awareness of craniosynostosis and how they try to actively try to raise awareness, e.g., fundraising, writing to local hospitals, GP, being active on Facebook to help other parents, taking part in research…

March 2023
Devising model

I have sketched out an initial draft for the model. It follows the form of a timeline as participants tended to talk about their experiences in terms of checkpoints or specific milestones. I am happy with it, although it feels complex at the moment due to the many different components and I wonder if I could simplify it a bit more. I'd like the model to make sense as a 'stand-alone' visual, as well as offering a narrative in the final write-up.

Appendix 24. Example Narrative Summary.

Interview Summary
Participant 14 – Owl

Individual

This participant was a 34-year-old mother. Child was diagnosed with sagittal craniosynostosis at around 2 months old. No family history of the diagnosis and child had no other health conditions.

Overview

This parent had a child who had already undergone surgery at [specialist centre]. She described the "torturous" bit was the wait between suspecting something was "wrong" with child and the formal diagnosis itself. They got rang with a surgery cancellation, which meant they had less time to worry. Child's father had diagnosed baby at 3 days old from researching online and googling 'strange head shape'.

They were positive about nursing staff. Nurses went to first helmet appointment with them – interpersonal moments described as professional balanced with sensitivity and personalisation. She mentioned how the MDT clinic was like being a fly on the wall to surgical conversations, that they probably shouldn't have been privy too, but appreciated the directness of this environment.

This family had suffered two bereavements and were undergoing a house renovation when baby was diagnosed.

Points to consider

- This was an interview post-surgery. Be interesting to see if this differs to any that I've done pre-surgery
- The notion of having less time to worry and ruminate
- How early researching begins
- Appreciating the directness of the clinical environment – does this represent something more? Such as being transparent in communication? Knowing the risks? Feeling like you can trust professionals?
- Other life stressors
- The importance of staff relationships

Reflections

- Felt like I could feel emotions at time during the interview but didn't let herself get upset. I wonder if this was protective?
- Asked me questions before the interview about why I was interested in parents. How she had assumed research was always about the child. This is reflective in the evidence base? Shows a rationale for the project?
- I am due to interview this participant's partner so will be interesting to hear their perspective. I want to know more about process of researching online