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Understanding Families' Experiences Following a Diagnosis of Non-Syndromic Craniosynostosis: A Qualitative Study

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ABSTRACT

Objectives: Craniosynostosis is typically diagnosed and surgically corrected within the first year of life. The diagnosis and surgical correction of the condition can be a very stressful experience for families. Despite this, there is little research exploring the impact that craniosynostosis has on families, especially in the period immediately following diagnosis and correction. In this study, the authors aimed to qualitatively examine the psychosocial experience of families with a child diagnosed with craniosynostosis.

Design: Qualitative study.

Setting: Tertiary care paediatric health centre.

Participants: Parents of children newly diagnosed with single-suture, non-syndromic craniosynostosis

Intervention: Semistructured interviews regarding parental experience with the initial diagnosis, their decision on corrective surgery for their child, the operative experience, the impact of craniosynostosis on the family and the challenges they encountered throughout their journey.

Primary and Secondary Outcome Measures: Interpretive phenomenological analysis, a type of qualitative analysis that provides an in-depth account of participant's experiences and their meanings, was used to analyze the interview data.

Results: Over a four-year period, twelve families meeting eligibility criteria completed the study. Three main themes (6 subthemes) emerged from the pre-operative interviews: frustration with diagnostic delays (parental intuition and advocacy, hope for improved awareness), understanding what to expect (healthcare supports, interest in connecting with other families), and justifying the need for corrective surgery (influence of the surgeon, struggle with cosmetic indications). Two main themes (4 subthemes) were drawn from the post-operative interviews: overcoming fear (the

role of healthcare professionals, transition home) and relief (reduction in parental anxiety, cosmetic improvements).

Conclusions: Overall, the diagnosis of craniosynostosis has a profound impact on families, leading them to face many struggles throughout their journey. A better understanding of these experiences will help to inform future practice, with a hope to improve this experience for other families moving forward.

ARTICLE SUMMARY

Strengths and limitations of this study

- The prospective, qualitative study involving semi-structured interviews allowed parents of children with craniosynostosis to richly describe their journey from the point of diagnosis through to the post-operative period.
- Major pre-operative themes included frustration with diagnostic delays, understanding what to expect, and justifying the need for corrective surgery. Major post-operative themes included overcoming fear and relief.
- The themes of concern identified provide a helpful guide to both primary care physicians and members of craniofacial teams involved in the care of families with craniosynostosis.
- Given the small, homogeneous group of participants included, it is unclear whether the results accurately represent the experience of other populations.

INTRODUCTION

Craniosynostosis, a congenital anomaly involving abnormal fusion of calvarial sutures, affects 1 in every 2,000 to 3,000 live births[1-4]. It is traditionally classified as either syndromic or non-syndromic. Non-syndromic synostosis is not associated with other dysmorphisms outside the abnormal craniofacial morphology, and typically involves only a single suture. The most common subtypes include sagittal, metopic, unicoronal, bicoronal and lambdoidal. Non-syndromic craniosynostosis is classically treated with corrective surgery within the first year of life, with inconclusive evidence that earlier intervention may be beneficial for certain subtypes[5].

While still controversial, there is increasing evidence that non-syndromic craniosynostosis may be associated with long term neurodevelopmental deficits, including difficulties with visuospatial skills, memory, speech and language, and learning disorders[5]. Further studies have suggested that these impairments will persist and cannot be prevented with corrective surgery[6-9]. Despite this inconclusive evidence, most parents opt for corrective surgery to remodel the skull and allow for normal head growth in their child.

Although the impact of non-syndromic craniosynostosis on neurocognitive development remains in question, children with this congenital anomaly may be faced with social and psychological barriers that negatively impact their self-esteem and social function owing to their abnormal appearance[10-11]. While many reports document the psychosocial aspects of craniosynostosis from the perspective of the patient, they do not detail the experience of the family. Because corrective surgery is typically performed when patients are infants, parents are responsible for making proxy decisions and are actively involved in patient care. Thus, to obtain a true understanding of early experiences with craniosynostosis, it is important to expand our scope, and study not the just the patient, but the family.

Previous studies that attempted to quantify parental stress levels found no difference in the level of stress experienced by parents of children with and without single-suture craniosynostosis before corrective surgery[12-16]. Other studies have examined parental satisfaction with their child's postoperative results, with high satisfaction with surgical outcomes generally reported[16-18].

The aim of the current study is to provide an in-depth qualitative description of families' experiences with craniosynostosis. By adopting a qualitative approach involving semi-structured interviews, we allowed families to richly describe their journey and freely communicate personally meaningful topics. This study prospectively explored the experience of families beginning at the time of diagnosis and continuing to the postoperative period. We aim to use our findings to inform future research and practice, with the hope of improving the overall experience for families facing this diagnosis in the future.

METHODS

Interpretive phenomenological analysis (IPA) was selected as the qualitative methodology. IPA is designed to examine, in detail, participants' experiences of an event and its meaning through an interpretive process of examining the information they provide[19,20]. The role of the researcher in the interpretation is also recognised[21]. Rather than testing a specific hypothesis, this method allows for flexible exploration of a topic in a small, homogeneous sample of respondents for whom the topic is particularly relevant. IPA informed both the data collection and the analysis for the current study. Neither patients nor the public were involved in the study design. The interviewer received IPA technique training under one of the senior authors (J.M.C). The interviewer was not directly involved in the management of patients.

Patient and public involvement

The research question was developed based on comments expressed to the corresponding author by several families with a diagnosis of craniosynostosis regarding the need for timelier referral to the craniofacial program and a need for additional teaching resources to primary care providers on the diagnosis. While these families were the impetus for the research question, they did not directly participate in the design or conduct of the study. As stated below, participants were given the opportunity to review a summary of the themes and provide feedback following data analysis.

Study sample

Institutional research ethics approval was obtained for this study from the IWK Health Centre Research Ethics Board. All families presenting to the IWK Health Centre with a child who received a new diagnosis of non-syndromic craniosynostosis were eligible for this study. These families were identified prospectively by participating surgeons between February 15, 2016 and February 15, 2018. Eligible families were informed of the study by one of the participating surgeons during their initial consult, after receiving a diagnosis. Families were then consented to have their contact information provided to the principle investigator of the study.

Data collection

Participants completed two phone interviews. The first interview was completed within a month of receiving the initial diagnosis. The second interview was completed three months post-operatively, or three months after the initial interview if the family decided not to proceed with surgery. All interviews were completed by the first author. Verbal consent was obtained over the phone before initiating the interviews. Interviews were semistructured using an interview guide

based on the team's clinical experience and a scoping literature review. The initial interview guide contained questions on the diagnostic experience as well as the decision on corrective surgery. The second interview guide investigated the surgical experience and the recovery period.

Analysis method

Interviews were recorded and transcribed verbatim by a professional transcriptionist with subsequent deidentification. Transcripts were divided into pre-operative and post-operative categories. Initial coding was completed by the first author using accepted IPA methods. Transcripts were reviewed multiple times to assign codes to the main topics discussed by the participant. The codes identified in earlier transcripts informed the coding of later transcripts. New codes identified in later transcripts prompted earlier transcripts to be reviewed again to determine if these codes were also present in these. The organization of themes followed an iterative process aiming to identify the meaning behind participants' statements rather than solely the prevalence of topics discussed. Themes were then clustered, allowing for superordinate themes to be generated based on subsumption and abstraction techniques.

To ensure rigour, theme development was reviewed and discussed between the first and senior authors to confirm that the interpretations accurately represented the transcript data. A second author reviewed the transcripts independently to assess for representativeness. Member checking was also performed, where participants were given the opportunity to review a summary of the themes and provide feedback.

RESULTS

Twelve eligible families were identified and enrolled into the study over the enrollment period. This sample size is typical for IPA studies to reach thematic saturation[20]. Participant demographics are presented in Table 1.

Themes were organised into preoperative and postoperative categories. Three main themes emerged from the preoperative interviews: frustration with diagnostic delays, understanding what to expect, and justifying the need for corrective surgery. Two main themes emerged from the postoperative interviews: overcoming fear, and relief. Representative quotes are included throughout the text.

Frustration with diagnostic delays

Most participants expressed some frustration around diagnostic delays, excepting two participants whose child was born at the tertiary care hospital and received a diagnosis immediately post-partum. Two subthemes emerged: parental intuition/advocacy and hope for improved awareness.

<u>Parental intuition and advocacy</u>

Ten families noticed the abnormal shape of their child's head at birth and expressed concerns (Participant 12 - "I knew something was wrong, but I couldn't prove it"). They were frequently offered the explanation that it was a result of the birthing process and were told it would resolve spontaneously (Participant 2 - "The day he was born at the hospital we started noticing that one of his eyes would not open, and his nose was crooked a bit and the opening in one of his nostrils was very narrow. We were told it was because of what they call a traumatic birth, and it would fix as he grows.")

Over time, when no aesthetic improvement was observed, families began seeking medical advice. One family requested an x-ray; however, the diagnosis of craniosynostosis was missed. Other families resorted to taking their child to the emergency room or requesting a referral to a paediatrician after feeling their concerns were not adequately addressed by their family physician. One family expressed feelings of guilt around not pushing for the referral to a specialist earlier (Participant 2 - "I started doing my own research online and that's when I realised something should have been done when he was younger. I was a bit frustrated with my doctor. I felt like I should have pushed for it sooner when he was younger").

Hope for improved awareness

Overall, families describe a lack of awareness among community family physicians around craniosynostosis. One mother explains her surprise that the craniosynostosis wasn't picked up by her family physician despite regular exams (Participant 5 - "At every doctor's appointment they are always doing measurements of his head and looking for his soft spot"). Another mother describes her own physical findings that she felt were discounted (Participant 6 - "I also noticed a ridge along the top of his skull that I brought up to my GP and he kind of passed if off as not a big deal"). When asked how their overall experience could be improved, many parents suggested efforts to increase craniosynostosis awareness to allow for earlier detection (Participant 10 – "Being able to have more education for family doctors, nurse practitioners, that sort of thing, around what is normal and what's not normal"; Participant 2 - "I think it's something they should be more educated on.")

Understanding what to expect

Participants described the importance of being informed on what to expect and how this helped them to feel more comfortable during the whole experience. Two subthemes emerged: health care supports, and interest in connecting with other families.

Health care supports

Participants described feeling overwhelmed during their initial consultation, and most were unaware that surgery would be recommended for their child. Many had come mentally prepared with questions, but were then unable to recall these during the consult (Participant 1 -"So when he said 'do you have any questions' I was like 'no' because I was just trying to take it all in"). Other families chose to write down their questions ahead of time, which proved to be a more successful strategy. One participant commented on too many learners being present in the room- a comment that nursing staff later agreed with. This added to the overwhelming nature of the consult and hindered this participant's ability to express themselves. All participants described receiving verbal information; however, many suggested that additional written resources could have been provided for review once they have had time to process things (Participant 10 - "So I would say having a cheat sheet of something, where it's already written down that you leave with. Because in the moment, you're listening and not thinking of writing it down yourself"). Skull models used during the consult were helpful for participant education. As many participants were doing their own research, they requested references to reputable resources for further information. Additionally, participants appreciated having access to a specialised nurse after the consultation with the surgeons who they could email or call with additional questions. All participants spoke very highly of this support system and felt that it significantly reduced their anxiety (Participant

10 - “It was so helpful to know that if we did [have questions], we had a way to get a hold of [the clinic nurse]”).

Interest in connecting with other families

While all of the participants felt their consultation visits were informative, they expressed a strong interest in connecting with other families who have been through a similar experience (Participant 8 – “The doctor told me what I could expect, what I’m going to see after the surgery and all this, but hearing it from a parent’s perspective is a whole different story”). Participants felt it was important to hear other local success stories and mentioned that they would like access to pre- and post-operative photos from other families (Participant 7 - “As a mom and dad you really need to see that other children have risen through it”). Many participants reached out to other families through craniosynostosis support groups on social media platforms. They described the support and hope provided through these online chats (Participant 10 - “Those connections are important, I think, just to see that there are other people who are going through it and have made it through to the other side”). While most participants thought these types of communication would be helpful, one participant describes her emotional struggle after meeting with a family who experienced complications (Participant 12 - “I was scared. I’m even more scared now than I was then, because now that we’re in support groups and see what’s going to happen, we are scared about the surgery. It’s always hard when you have a small sample size too. It can make things look like they are in different proportions than they are”).

Justifying the need for corrective surgery

Participants describe the difficult process of reaching a decision on corrective surgery for their child. Two subthemes emerged: influence of the surgeon, and struggle with cosmetic indications.

Influence of the surgeon

All participants decided to proceed with corrective surgery for their child. This decision was reached during the initial consultation. The families described the importance of the information they received during this surgical visit and stated there were no outside influences factored into their decision. This speaks to the magnitude of the influence held by the surgeons. Many participants describe their positive relationship with the surgeons, and how it gave them confidence to consent to surgery during the first visit (Participant 7 - "I feel really confident with the doctors, I feel good with them, which I definitely think is a part of it"). Both bedside manner and the communication style of the surgeons were noted to help participants feel more comfortable with their decision on surgery (Participant 11 - "They talk to you like you're a human being. They talk to you in a fashion that, you know, we know what you're actually telling us. But it's easing my mind that we have such a great team"). Participants also appreciated surgeons speaking in lay terms during the consultation and consenting processes. Other families focused primarily on the evidence and risks communicated by the surgeon (Participant 4 - "When he told us there are 10-15% that have pressure build-up in their brain and it can affect development and also his vision, […] I don't think we had to think very long to decide that we do not want to take that risk if we can definitely prevent it by doing a surgery"). While many families deliberated on this difficult decision, some families describe the feeling of not having a choice, that surgery was the only option (Participant 1 -"He was talking about how the shape of his head would just continue to grow

this way, and his brain would be squeezed and there would be pressure. But yeah, I felt like this was our only option").

Struggle with cosmetic indications

The participants discussed their struggle weighing the importance of cosmetic indications, with most families stating that the decision would be much more difficult if the surgery was for aesthetic purposes alone (Participant 4 -"If it was just cosmetic it definitely would have taken us more time to think about it."). While most families identified potential neurological risks as their primary motivator, it seems that aesthetic concerns were still present, even if not directly vocalised (Participant 11 – "So we know that it's not a decision that we're being selfish and trying to fix her look. It needs to be completed"). Other families were more direct in voicing their cosmetic concerns and were worried about potential psychosocial difficulties later in life, especially after learning about the potentially progressive nature of the condition. This included concerns around future bullying, depression, and the even the risk of suicide if surgery was not performed (Participant 8 – "No child should grow up and develop that head shape"; Participant 12 – "When she was first diagnosed, I would have said no, but now, the asymmetry is so much that it wouldn't be fair to her not to repair it. She would always look very different from other children"; Participant 10 – "If we don't do the surgery, he's going to hate us later in life because we didn't fix this. He would probably be teased and picked on"). One family related their cosmetic concerns to the sex of their child, describing the gender-biased aesthetic standard they have experienced in society (Participant 12 - "[My husband] keeps saying specifically because she's a girl, and we live in a society where what a girl looks like is important").

Overcoming fear

The participants describe fear at various stages of their journey and shared what helped them cope with this emotion. Two subthemes emerged: the role of healthcare professionals, and the transition home.

The role of healthcare professionals

Participants discussed at length how health care providers helped reduce their fear and anxiety while in hospital. Firstly, although parents found it very difficult to hand over their child for surgery, the were comforted by regular updates throughout the procedure (Participant 4 - "You're just waiting for that nurse to come and give us the news that everything is going well, and like it's supposed to. And she did, every time. That was great"). There was only one family who did not receive regular updates throughout the operation. This participant describes feeling extremely nervous in the waiting room after not being informed about a delay in the surgical start time (Participant 5 - "I would have liked to know that they started later than think something bad happened"). Overall, regardless of the stage in their journey, parents described feeling much calmer when they were kept informed. In addition, families commented on the importance of empathy in healthcare. For example, one participant (Participant 10) spoke of the impact that small gestures can have on a family during a difficult time: "They brought us out the bag of his hair. One of them had written on it 'baby's first haircut'. It let you know that they care about your child, that they see that it's not just another patient."

Transitioning Home

Most participants were very surprised with the short recovery time after discharge home (Participant 5 – "You don't think they are going to recover that quick […], but within 2 or 3 days

they're their normal self"). This introduced a new fear for parents. Many families described difficulty allowing their child to return to regular activities out of fear they would hurt themselves (Participant 8 - "We're still really scared, like if he falls and bangs his head or something, we're like 'Ooohh!'"). These concerns were heightened if the child had young siblings (Participant 1- "And even now, it's hard, because [my other children] are so young, and he still has the soft spot on his head, but they don't understand"). When asked what helped ease their transition home, families stated that were very grateful to be given contact information to reach their healthcare team with questions after discharge. They felt comfortable emailing or calling members of the team with post-operative questions. Ultimately, the ongoing support for parents helped to reduce feelings of fear and anxiety after discharge.

Relief

All twelve participants expressed a sense of relief post-operatively, feeling confident they had made the right decision regarding corrective surgery. Two subthemes emerged: reduction in parental anxiety and cosmetic improvements.

Reduction in parental anxiety

Participants described significant anxiety leading up to the operation, despite feeling very well informed. Many families feared that they would regret their decision regarding corrective surgery and felt a substantial amount of pressure to make the right choice (Participant 7 -"My fear was that he would be changed for the worse and that we would forever regret the decision to do it"). All participants felt their anxiety subside post-operatively after a successful operation.

Parents also described significant anxiety around the potential for neurological deficits associated with craniosynostosis, worrying that irreversible effects would occur before surgery (Participant 1 - "I was always making sure he could focus on me, and if he couldn't focus on me I'd think 'oh no, is he going blind'"). Post-operatively, participants no longer worried about neurological deficits, and felt they were no longer anxious about their child meeting developmental milestones. Many families also described positive behavioural changes in their child that they attributed to the surgery (Participant 7 – "He is happier and a little more relaxed. He is able to play more"; Participant 12 – "She was almost, I would say, mute leading up to surgery. Within a week of surgery she started making sounds and now, three months later, has a full vocabulary").

Cosmetic improvements

Although most families claimed neurological deficits were their primary motivation for surgery, the cosmetic improvements were heavily commented upon in the post-operative interviews (Participant 3 - "The best part would be how he looked after surgery. Like three weeks after, how good he looked. He looked like a total different baby"). Participants expressed relief with the aesthetic success of the operation (Participant 4 – "It did really change the way that his face and features look. It wasn't the main reason for us to do the surgery, but it was definitely, like, 'oh wow!'"). One mother commented on the practical aspect of her child's new head shape (Participant 6 - "I appreciate being able to put a hat on him now"). Another reflected on the progressive nature of craniosynostosis, describing what she felt her child would have looked like now without the operation (Participant 8 - "If we never would have done that surgery […], his head would be so much like a football right now"). Overall, parents seemed very satisfied that their child would no longer stand out due to a cranial deformity (Participant 6 - "He looks like a completely normal 8-month old now, besides the really faint scarring").

DISCUSSION

The diagnosis and treatment of craniosynostosis has a significant impact on families. This qualitative analysis provides a rich description of families' experiences with craniosynostosis, from the point of diagnosis through to the period of surgical recovery.

In the pre-operative interviews, most families described frustration around diagnostic delay, acknowledging the importance of advocating for their child and their hope for improved craniosynostosis awareness in community practice. They stressed the importance of knowing what to expect, and the value in both healthcare supports and making connections with other families. They also discussed the struggle to decide on corrective surgery, acknowledging the influence of the surgeon and their difficulties weighing functional and cosmetic indications.

In the post-operative interviews, families discussed their journey of overcoming their fear. They highlighted the contribution of healthcare professionals and emphasised the challenges of transitioning home. There was also a very different tone to the second round of interviews, one of relief. All families were happy with their decision to proceed with corrective surgery, and felt their anxiety was reduced in the post-operative period. They also commented on their satisfaction with the cosmetic improvement.

For most forms of non-syndromic craniosynostosis the prevention of elevated intracranial pressure and associated neurocognitive deficits is the principal indication for surgery[22]. Sagittal craniosynostosis may be an exception, as compensatory growth along patent sutures largely prevents elevated intracranial pressure but produces a stigmatising head shape. To this end, aesthetic concerns may be a greater motivation for surgical correction of sagittal craniosynostosis[23]. A recent health utility outcome study found relatively high utility scores for

sagittal craniosynostosis, suggesting that the cosmetic burden of this condition as perceived by the general population is low[24]. This aligns with our findings, where most participants stated that the cosmetic indications for corrective surgery were secondary to the neurological ones. Despite this, all families decided to proceed with corrective surgery, including those who received a diagnosis of sagittal craniosynostosis. While families may find it difficult to choose a potentially morbid corrective surgery for aesthetic indications alone, it is important to remind parents of the potential psychosocial consequences of living with an uncorrected craniofacial abnormality[25]. Interestingly, despite the difficulty justifying aesthetic indications pre-operatively, the satisfaction with cosmetic improvements was heavily commented on in the postoperative interviews.

Many of the themes developed in our study align with those reported by previous studies examining the experiences of families with children diagnosed with other craniofacial deformities. For example, parents with children diagnosed with cleft lip/palate described their anxiety around surgery and their need for emotional support throughout treatment, for both themselves and their child[26-28]. Furthermore, families of children diagnosed with craniofacial abnormalities have expressed fears that their child will be bullied and ostracised later in life[26,28]. Previous studies have emphasised the importance of parental support in healthcare, suggesting that the emotional state of caregivers significantly influences the emotional development of children with craniofacial abnormalities[28-30]. Elevated caregiver stress was found to have long-lasting, negative psychosocial effects on children who received corrective surgery for craniofacial abnormalities and was also associated with increased levels of anxiety and depression among patients during childhood[31]. By better understanding the experience of craniosynostosis by families, supports can be appropriately tailored to address current areas of concern and improve the overall experience.

While many of our themes supported previous research, frustration with diagnostic delays was a key theme in our study and has not previously been reported for craniosynostosis. While there are currently no guidelines outlining the optimal age for surgical correction of craniosynostosis, much research has focussed on this issue. A systematic review by Mandela *et al.* found no conclusive evidence that earlier surgery may be beneficial to patients with sagittal synostosis, for example. There is no evidence that later surgery is beneficial for any of the craniosynostosis subtypes[5]. This speaks to the importance of early detection, as the age of diagnosis will affect when and what type of surgery is offered. One family in our study received the diagnosis of craniosynostosis early in the post-partum period. Due to the young age at detection, the child was eligible for less invasive endoscopic correction and helmet therapy. This option would not have been offered had the family experienced a diagnostic delay like that experienced by most families in our series.

In addition to improving craniosynostosis awareness, families also suggested that it would have been helpful to have received printed material during the initial consult to complement the information that was provided verbally. They expressed interest in receiving written pamphlets as well as a list of reliable and recommended internet sources where they could review the information further. The provision of these decision aids has previously been found to increase both comprehension and risk recall[32]. In addition to improving informed consent, these interventions may increase overall satisfaction with the decision-making process[33]. This is especially relevant to the craniosynostosis patient population, as parents described significant anxiety associated with therapeutic decision-making.

This study is not without limitations. Firstly, given the small, homogeneous group of participants included, it is unclear whether our results accurately represent the experience of other

populations. All twelve families elected to have the patient's mother complete the interviews rather than the father, regardless of marital status. While this may reflect the traditional distribution of caregiver responsibilities, it prevents us from identifying potential meaningful gender differences in the family experience of craniosynostosis. For example, parental stress has previously been reported to be higher in mothers with children diagnosed with single suture craniosynostosis when compared to their paternal counterparts[34]. Secondly, because our study design assigned recruitment responsibilities to the participating surgeons, the surgeons were not blinded to which families were enrolled. Although participants were ensured anonymity, it is unclear whether this influenced the interviews, potentially making participants more reluctant to identify points of dissatisfaction around their interactions with the surgeons. Despite these limitations, our study offers important insights for physicians caring for children with craniosynostosis and helps health care providers better understand the needs of families during the pre-, peri- and post-operative periods.

This study also suggests future avenues of research and development. Despite the fear expressed by parents in the pre-operative period, all families were ultimately pleased with their decision to proceed with corrective surgery. Future studies aim to explore the opinion of the patients themselves, and their views on their parents' decision regarding surgical correction of their craniosynostosis. Additionally, our findings speak to the importance of lifelong learning in the medical field and identify the need for additional craniosynostosis teaching among general practitioners to allow for earlier detection in the community.

CONCLUSION

The diagnosis of craniosynostosis has a significant impact on families. This study offers a detailed look into the experiences of families from the point of diagnosis through to the post-

operative recovery and transition home. Participants provide rich descriptions of their frustrations, accomplishments, supports and their suggestions for improvement. A better understanding of this experience will identify where further supports are needed and inform future practice, with the goal of improving the overall experience for other families moving forward.

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CONTRIBUTORSHIP STATEMENT

Dr. Victoria Kuta: Literature review, ethics submission, interviews, primary thematic analysis, manuscript preparation and submission

Dr. Lauren Curry: Secondary reviewer of transcripts and themes

Dr. P. Daniel McNeely: Patient recruitment, surgery, manuscript preparation

Dr. Simon Walling: Patient recruitment, surgery, manuscript preparation

Dr. Jill M. Chorney: Ethics submission, study design, manuscript preparation

Dr. Michael Bezuhly: Primary supervisor, ethics submission, study design, patient recruitment, surgery, manuscript preparation and submission

TABLE 1. Participant Demographics (n=12)

Variable	n (%)
Age, y*	32.4 ± 6.3 (range, 19-42)
Relationship to patient	
Mother	12 (100)
Father	0 (0)
Location	
Home address < 50 km from hospital	9 (75)
Home address > 50 km from hospital	3 (25)
Sex of Child	
Male	10 (83.3)
Female	2 (16.6)
Craniosynostosis type	
Sagittal	6 (50.0)
Coronal	4 (33.3)
Metopic	2 (16.7)

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Standards for Reporting Qualitative Research (SRQR)*

http://www.equator-network.org/reporting-guidelines/srqr/

	Page/line no(s).
Title and abstract	
Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	1-2
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	47-75
Introduction	
Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	114-139
Purpose or research question - Purpose of the study and specific objectives or questions	140-146
Methods	
Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	149-155; 181-188
Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	155-157
Context - Setting/site and salient contextual factors; rationale**	160-162
Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**	162-166;
Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	160-161; 179-180
Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**	163-164; 182-193;197

Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	173-176; 179-180
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	196-198;600
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	160-161; 179-181
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	182-188
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	189-193

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	199-393
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	199-393

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	396-459
Limitations - Trustworthiness and limitations of findings	460-473

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	27-28
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	30-32

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

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Understanding Families' Experiences Following a Diagnosis of Non-Syndromic Craniosynostosis: A Qualitative Study

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ABSTRACT

Objectives: Craniosynostosis is typically diagnosed and surgically corrected within the first year of life. The diagnosis and surgical correction of the condition can be a very stressful experience for families. Despite this, there is little research exploring the impact that craniosynostosis has on families, especially in the period immediately following diagnosis and correction. In this study, the authors aimed to qualitatively examine the psychosocial experience of families with a child diagnosed with craniosynostosis.

Design: Qualitative study.

Setting: Tertiary care paediatric health centre.

Participants: Mothers of children newly diagnosed with single-suture, non-syndromic craniosynostosis

Intervention: Semistructured interviews regarding parental experience with the initial diagnosis, their decision on corrective surgery for their child, the operative experience, the impact of craniosynostosis on the family and the challenges they encountered throughout their journey.

Primary and Secondary Outcome Measures: Thematic analysis, a type of qualitative analysis that provides an in-depth account of participant's experiences, was used to analyze the interview data.

Results: Over a four-year period, twelve families meeting eligibility criteria completed the study. Three main themes (6 subthemes) emerged from the pre-operative interviews: frustration with diagnostic delays (parental intuition and advocacy, hope for improved awareness), understanding what to expect (healthcare supports, interest in connecting with other families), and justifying the need for corrective surgery (influence of the surgeon, struggle with cosmetic indications). Two main themes (4 subthemes) were drawn from the post-operative interviews: overcoming fear (the

role of healthcare professionals, transition home) and relief (reduction in parental anxiety, cosmetic improvements).

Conclusions: Overall, the diagnosis of craniosynostosis has a profound impact on families, leading them to face many struggles throughout their journey. A better understanding of these experiences will help to inform future practice, with a hope to improve this experience for other families moving forward.

ARTICLE SUMMARY

Strengths and limitations of this study

- The prospective, qualitative study involving semi-structured interviews allowed mothers of children with craniosynostosis to richly describe their journey from the point of diagnosis through to the post-operative period.
- This study examined both major pre-operative and post-operative themes within the same cohort, allowing for identification of how maternal concerns change over the course of the treatment of their child.
- The themes of concern identified provide a helpful guide to both primary care physicians and members of craniofacial teams involved in the care of families with craniosynostosis.
- Given the small, homogeneous group of participants included, it is unclear whether the results accurately represent the experience of other populations.

INTRODUCTION

Craniosynostosis, a congenital anomaly involving abnormal fusion of calvarial sutures, affects 1 in every 2,000 to 3,000 live births[1-4]. It is traditionally classified as either syndromic or non-syndromic. Non-syndromic synostosis is not associated with other dysmorphisms outside the abnormal craniofacial morphology, and typically involves only a single suture. The most common subtypes include sagittal, metopic, unicoronal, bicoronal and lambdoidal. Non-syndromic craniosynostosis is classically treated with corrective surgery within the first year of life, with inconclusive evidence that earlier intervention may be beneficial for certain subtypes[5].

While still controversial, there is increasing evidence that non-syndromic craniosynostosis may be associated with long term neurodevelopmental deficits, including difficulties with visuospatial skills, memory, speech and language, and learning disorders[5]. Further studies have suggested that these impairments will persist and cannot be prevented with corrective surgery[6-9]. Despite this inconclusive evidence, most parents opt for corrective surgery to remodel the skull and allow for normal head growth in their child.

Although the impact of non-syndromic craniosynostosis on neurocognitive development remains in question, children with this congenital anomaly may be faced with social and psychological barriers that negatively impact their self-esteem and social function owing to their abnormal appearance[10-11]. While many reports document the psychosocial aspects of craniosynostosis from the perspective of the patient, they do not detail the experience of the family. Because corrective surgery is typically performed when patients are infants, parents are responsible for making proxy decisions and are actively involved in patient care. Thus, to obtain a true understanding of early experiences with craniosynostosis, it is important to expand our scope, and study not the just the patient, but the family.

Previous studies that attempted to quantify parental stress levels found no difference in the level of stress experienced by parents of children with and without single-suture craniosynostosis before corrective surgery[12-16]. Other studies have examined parental satisfaction with their child's postoperative results, with high satisfaction with surgical outcomes generally reported[16-18].

The aim of the current study is to provide an in-depth qualitative description of families' experiences with craniosynostosis. By adopting a qualitative approach involving semi-structured interviews, we allowed families to richly describe their journey and freely communicate personally meaningful topics. This study prospectively explored the experience of families beginning at the time of diagnosis and continuing to the postoperative period. We aim to use our findings to inform future research and practice, with the hope of improving the overall experience for families facing this diagnosis in the future.

METHODS

Thematic analysis was used as the qualitative methodology. Thematic analysis is a method for identifying, analyzing and reporting, in detail, patterns within participants' experiences of an event[19,20]. The role of the researcher in the interpretation is also recognised[21]. Rather than testing a specific hypothesis, this method allows for flexible exploration of a topic in a small, homogeneous sample of respondents for whom the topic is particularly relevant. Thematic analysis informed both the data collection and the reporting for the current study. Neither patients nor the public were involved in the study design. The interviewer received thematic analysis training under one of the senior authors (J.M.C) using previously described methodology[20]. The interviewer was not directly involved in the management of patients.

Patient and public involvement

The research question was developed based on comments expressed to the corresponding author by several families with a diagnosis of craniosynostosis regarding the need for timelier referral to the craniofacial program and a need for additional teaching resources to primary care providers on the diagnosis. While these families were the impetus for the research question, they did not directly participate in the design or conduct of the study. As stated below, participants were given the opportunity to review a summary of the themes and provide feedback following data analysis.

Study sample

Institutional research ethics approval was obtained for this study from the IWK Health Centre Research Ethics Board. All families presenting to the IWK Health Centre with a child who received a new diagnosis of non-syndromic craniosynostosis were eligible for this study. These families were identified prospectively by participating surgeons between February 15, 2016 and February 15, 2018. Eligible families were informed of the study by one of the participating surgeons during their initial consult, after receiving a diagnosis. Families were then consented to have their contact information provided to the principle investigator of the study.

Data collection

Participants completed two phone interviews. The first interview was completed within a month of receiving the initial diagnosis. The second interview was completed three months post-operatively, or three months after the initial interview if the family decided not to proceed with surgery. All interviews were completed by the first author. Verbal consent was obtained over the phone before initiating the interviews. Interviews were semistructured using an interview guide

based on the team's clinical experience and a scoping literature review. The initial interview guide contained questions on the diagnostic experience as well as the decision on corrective surgery. The second interview guide investigated the surgical experience and the recovery period.

Analysis method

Interviews were recorded and transcribed verbatim by a professional transcriptionist with subsequent deidentification. Transcripts were divided into pre-operative and post-operative categories. Initial coding was completed by the first author using thematic analysis methods. Transcripts were reviewed multiple times to assign codes to the main topics discussed by the participant. The codes identified in earlier transcripts informed the coding of later transcripts. New codes identified in later transcripts prompted earlier transcripts to be reviewed again to determine if these codes were also present in these. The organization of themes followed an iterative process aiming to identify the meaning behind participants' statements rather than solely the prevalence of topics discussed. Themes were then clustered, allowing for superordinate themes to be generated based on subsumption and abstraction techniques.

To ensure rigour, theme development was reviewed and discussed between the first and senior authors to confirm that the interpretations accurately represented the transcript data. A second author reviewed the transcripts independently to assess for representativeness. Member checking was also performed, where participants were given the opportunity to review a summary of the themes and provide feedback.

RESULTS

Twelve eligible families were identified and enrolled into the study over the enrollment period. This sample size is typical for thematic analysis studies to reach thematic saturation[20]. Participant demographics are presented in Table 1. All participants were mothers and were interviewed individually.

Themes were organised into preoperative and postoperative categories. Three main themes emerged from the preoperative interviews: frustration with diagnostic delays, understanding what to expect, and justifying the need for corrective surgery. Two main themes emerged from the postoperative interviews: overcoming fear, and relief. Representative quotes are included throughout the text.

Frustration with diagnostic delays

Most participants expressed some frustration around diagnostic delays, excepting two participants whose child was born at the tertiary care hospital and received a diagnosis immediately post-partum. Two subthemes emerged: parental intuition/advocacy and hope for improved awareness.

Parental intuition and advocacy

Ten families noticed the abnormal shape of their child's head at birth and expressed concerns (Participant 12 - "I knew something was wrong, but I couldn't prove it"). They were frequently offered the explanation that it was a result of the birthing process and were told it would resolve spontaneously (Participant 2 - "The day he was born at the hospital we started noticing that one of his eyes would not open, and his nose was crooked a bit and the opening in one of his

nostrils was very narrow. We were told it was because of what they call a traumatic birth, and it would fix as he grows.")

Over time, when no aesthetic improvement was observed, families began seeking medical advice. One family requested an x-ray; however, the diagnosis of craniosynostosis was missed. Other families resorted to taking their child to the emergency room or requesting a referral to a paediatrician after feeling their concerns were not adequately addressed by their family physician. One family expressed feelings of guilt around not pushing for the referral to a specialist earlier (Participant 2 - "I started doing my own research online and that's when I realised something should have been done when he was younger. I was a bit frustrated with my doctor. I felt like I should have pushed for it sooner when he was younger").

Hope for improved awareness

Overall, families describe a lack of awareness among community family physicians around craniosynostosis. One mother explains her surprise that the craniosynostosis wasn't picked up by her family physician despite regular exams (Participant 5 - "At every doctor's appointment they are always doing measurements of his head and looking for his soft spot"). Another mother describes her own physical findings that she felt were discounted (Participant 6 - "I also noticed a ridge along the top of his skull that I brought up to my GP and he kind of passed if off as not a big deal"). When asked how their overall experience could be improved, many parents suggested efforts to increase craniosynostosis awareness to allow for earlier detection (Participant 10 – "Being able to have more education for family doctors, nurse practitioners, that sort of thing, around what is normal and what's not normal"; Participant 2 - "I think it's something they should be more educated on.")

Understanding what to expect

Participants described the importance of being informed on what to expect and how this helped them to feel more comfortable during the whole experience. Two subthemes emerged: health care supports, and interest in connecting with other families.

Health care supports

Participants described feeling overwhelmed during their initial consultation, and most were unaware that surgery would be recommended for their child. Many had come mentally prepared with questions, but were then unable to recall these during the consult (Participant 1 -"So when he said 'do you have any questions' I was like 'no' because I was just trying to take it all in"). Other families chose to write down their questions ahead of time, which proved to be a more successful strategy. One participant commented on too many learners being present in the room- a comment that nursing staff later agreed with. This added to the overwhelming nature of the consult and hindered this participant's ability to express themselves. All participants described receiving verbal information; however, many suggested that additional written resources could have been provided for review once they have had time to process things (Participant 10 - "So I would say having a cheat sheet of something, where it's already written down that you leave with. Because in the moment, you're listening and not thinking of writing it down yourself"). Skull models used during the consult were helpful for participant education. As many participants were doing their own research, they requested references to reputable resources for further information. Additionally, participants appreciated having access to a specialised nurse after the consultation with the surgeons who they could email or call with additional questions. All participants spoke

very highly of this support system and felt that it significantly reduced their anxiety (Participant 10 - "It was so helpful to know that if we did [have questions], we had a way to get a hold of [the clinic nurse]").

Interest in connecting with other families

While all of the participants felt their consultation visits were informative, they expressed a strong interest in connecting with other families who have been through a similar experience (Participant 8 – "The doctor told me what I could expect, what I'm going to see after the surgery and all this, but hearing it from a parent's perspective is a whole different story"). Participants felt it was important to hear other local success stories and mentioned that they would like access to pre- and post-operative photos from other families (Participant 7 - "As a mom and dad you really need to see that other children have risen through it"). Many participants reached out to other families through craniosynostosis support groups on social media platforms. They described the support and hope provided through these online chats (Participant 10 - "Those connections are important, I think, just to see that there are other people who are going through it and have made it through to the other side"). While most participants thought these types of communication would be helpful, one participant describes her emotional struggle after meeting with a family who experienced complications (Participant 12 - "I was scared. I'm even more scared now than I was then, because now that we're in support groups and see what's going to happen, we are scared about the surgery. It's always hard when you have a small sample size too. It can make things look like they are in different proportions than they are").

Justifying the need for corrective surgery

Participants describe the difficult process of reaching a decision on corrective surgery for their child. Two subthemes emerged: influence of the surgeon, and struggle with cosmetic indications.

Influence of the surgeon

All participants decided to proceed with corrective surgery for their child. This decision was reached during the initial consultation. The families described the importance of the information they received during this surgical visit and stated there were no outside influences factored into their decision. This speaks to the magnitude of the influence held by the surgeons. Many participants describe their positive relationship with the surgeons, and how it gave them confidence to consent to surgery during the first visit (Participant 7 - "I feel really confident with the doctors, I feel good with them, which I definitely think is a part of it"). Both bedside manner and the communication style of the surgeons were noted to help participants feel more comfortable with their decision on surgery (Participant 11 - "They talk to you like you're a human being. They talk to you in a fashion that, you know, we know what you're actually telling us. But it's easing my mind that we have such a great team"). Participants also appreciated surgeons speaking in lay terms during the consultation and consenting processes. Other families focused primarily on the evidence and risks communicated by the surgeon (Participant 4 - "When he told us there are 10-15% that have pressure build-up in their brain and it can affect development and also his vision, […] I don't think we had to think very long to decide that we do not want to take that risk if we can definitely prevent it by doing a surgery"). While many families deliberated on this difficult decision, some families describe the feeling of not having a choice, that surgery was the only option (Participant 1 -"He was talking about how the shape of his head would just continue to grow

this way, and his brain would be squeezed and there would be pressure. But yeah, I felt like this was our only option").

Struggle with cosmetic indications

The participants discussed their struggle weighing the importance of cosmetic indications, with most families stating that the decision would be much more difficult if the surgery was for aesthetic purposes alone (Participant 4 -"If it was just cosmetic it definitely would have taken us more time to think about it."). While most families identified potential neurological risks as their primary motivator, it seems that aesthetic concerns were still present, even if not directly vocalised (Participant 11 – "So we know that it's not a decision that we're being selfish and trying to fix her look. It needs to be completed"). Other families were more direct in voicing their cosmetic concerns and were worried about potential psychosocial difficulties later in life, especially after learning about the potentially progressive nature of the condition. This included concerns around future bullying, depression, and the even the risk of suicide if surgery was not performed (Participant 8 – "No child should grow up and develop that head shape"; Participant 12 – "When she was first diagnosed, I would have said no, but now, the asymmetry is so much that it wouldn't be fair to her not to repair it. She would always look very different from other children"; Participant 10 – "If we don't do the surgery, he's going to hate us later in life because we didn't fix this. He would probably be teased and picked on"). One family related their cosmetic concerns to the sex of their child, describing the gender-biased aesthetic standard they have experienced in society (Participant 12 - "[My husband] keeps saying specifically because she's a girl, and we live in a society where what a girl looks like is important").

Overcoming fear

The participants describe fear at various stages of their journey and shared what helped them cope with this emotion. Two subthemes emerged: the role of healthcare professionals, and the transition home.

The role of healthcare professionals

Participants discussed at length how health care providers helped reduce their fear and anxiety while in hospital. Firstly, although parents found it very difficult to hand over their child for surgery, the were comforted by regular updates throughout the procedure (Participant 4 - "You're just waiting for that nurse to come and give us the news that everything is going well, and like it's supposed to. And she did, every time. That was great"). There was only one family who did not receive regular updates throughout the operation. This participant describes feeling extremely nervous in the waiting room after not being informed about a delay in the surgical start time (Participant 5 - "I would have liked to know that they started later than think something bad happened"). Overall, regardless of the stage in their journey, parents described feeling much calmer when they were kept informed. In addition, families commented on the importance of empathy in healthcare. For example, one participant (Participant 10) spoke of the impact that small gestures can have on a family during a difficult time: "They brought us out the bag of his hair. One of them had written on it 'baby's first haircut'. It let you know that they care about your child, that they see that it's not just another patient."

Transitioning Home

Most participants were very surprised with the short recovery time after discharge home (Participant 5 – "You don't think they are going to recover that quick […], but within 2 or 3 days

they're their normal self"). This introduced a new fear for parents. Many families described difficulty allowing their child to return to regular activities out of fear they would hurt themselves (Participant 8 - "We're still really scared, like if he falls and bangs his head or something, we're like 'Ooohh!'"). These concerns were heightened if the child had young siblings (Participant 1- "And even now, it's hard, because [my other children] are so young, and he still has the soft spot on his head, but they don't understand"). When asked what helped ease their transition home, families stated that were very grateful to be given contact information to reach their healthcare team with questions after discharge. They felt comfortable emailing or calling members of the team with post-operative questions. Ultimately, the ongoing support for parents helped to reduce feelings of fear and anxiety after discharge.

Relief

All twelve participants expressed a sense of relief post-operatively, feeling confident they had made the right decision regarding corrective surgery. Two subthemes emerged: reduction in parental anxiety and cosmetic improvements.

Reduction in parental anxiety

Participants described significant anxiety leading up to the operation, despite feeling very well informed. Many families feared that they would regret their decision regarding corrective surgery and felt a substantial amount of pressure to make the right choice (Participant 7 -"My fear was that he would be changed for the worse and that we would forever regret the decision to do it"). All participants felt their anxiety subside post-operatively after a successful operation.

Parents also described significant anxiety around the potential for neurological deficits associated with craniosynostosis, worrying that irreversible effects would occur before surgery (Participant 1 - "I was always making sure he could focus on me, and if he couldn't focus on me I'd think 'oh no, is he going blind'"). Post-operatively, participants no longer worried about neurological deficits, and felt they were no longer anxious about their child meeting developmental milestones. Many families also described positive behavioural changes in their child that they attributed to the surgery (Participant 7 – "He is happier and a little more relaxed. He is able to play more"; Participant 12 – "She was almost, I would say, mute leading up to surgery. Within a week of surgery she started making sounds and now, three months later, has a full vocabulary").

Cosmetic improvements

Although most families claimed neurological deficits were their primary motivation for surgery, the cosmetic improvements were heavily commented upon in the post-operative interviews (Participant 3 - "The best part would be how he looked after surgery. Like three weeks after, how good he looked. He looked like a total different baby"). Participants expressed relief with the aesthetic success of the operation (Participant 4 – "It did really change the way that his face and features look. It wasn't the main reason for us to do the surgery, but it was definitely, like, 'oh wow!'"). One mother commented on the practical aspect of her child's new head shape (Participant 6 - "I appreciate being able to put a hat on him now"). Another reflected on the progressive nature of craniosynostosis, describing what she felt her child would have looked like now without the operation (Participant 8 - "If we never would have done that surgery […], his head would be so much like a football right now"). Overall, parents seemed very satisfied that their child would no longer stand out due to a cranial deformity (Participant 6 - "He looks like a completely normal 8-month old now, besides the really faint scarring").

DISCUSSION

The diagnosis and treatment of craniosynostosis has a significant impact on families. This qualitative analysis provides a rich description of families' experiences with craniosynostosis, from the point of diagnosis through to the period of surgical recovery.

In the pre-operative interviews, most families described frustration around diagnostic delay, acknowledging the importance of advocating for their child and their hope for improved craniosynostosis awareness in community practice. They stressed the importance of knowing what to expect, and the value in both healthcare supports and making connections with other families. They also discussed the struggle to decide on corrective surgery, acknowledging the influence of the surgeon and their difficulties weighing functional and cosmetic indications.

In the post-operative interviews, families discussed their journey of overcoming their fear. They highlighted the contribution of healthcare professionals and emphasised the challenges of transitioning home. There was also a very different tone to the second round of interviews, one of relief. All families were happy with their decision to proceed with corrective surgery, and felt their anxiety was reduced in the post-operative period. They also commented on their satisfaction with the cosmetic improvement.

For most forms of non-syndromic craniosynostosis the prevention of elevated intracranial pressure and associated neurocognitive deficits is the principal indication for surgery[22]. Sagittal craniosynostosis may be an exception, as compensatory growth along patent sutures largely prevents elevated intracranial pressure but produces a stigmatising head shape. To this end, aesthetic concerns may be a greater motivation for surgical correction of sagittal craniosynostosis[23]. A recent health utility outcome study found relatively high utility scores for

sagittal craniosynostosis, suggesting that the cosmetic burden of this condition as perceived by the general population is low[24]. This aligns with our findings, where most participants stated that the cosmetic indications for corrective surgery were secondary to the neurological ones. The observation that concerns regarding cognitive sequelae were the main motivation for corrective surgery underscores the need for ongoing clinical research into functional aspects of craniosynostosis management. Of note, all families decided to proceed with corrective surgery, including those who received a diagnosis of sagittal craniosynostosis. While families may find it difficult to choose a potentially morbid corrective surgery for aesthetic indications alone, it is important to remind parents of the potential psychosocial consequences of living with an uncorrected craniofacial abnormality[25]. Interestingly, despite the difficulty justifying aesthetic indications pre-operatively, the satisfaction with cosmetic improvements was heavily commented on in the postoperative interviews.

Many of the themes developed in our study align with those reported by previous studies examining the experiences of families with children diagnosed with other craniofacial deformities. For example, parents with children diagnosed with cleft lip/palate described their anxiety around surgery and their need for emotional support throughout treatment, for both themselves and their child[26-28]. Furthermore, families of children diagnosed with craniofacial abnormalities have expressed fears that their child will be bullied and ostracised later in life[26,28]. Previous studies have emphasised the importance of parental support in healthcare, suggesting that the emotional state of caregivers significantly influences the emotional development of children with craniofacial abnormalities[28-30]. Elevated caregiver stress was found to have long-lasting, negative psychosocial effects on children who received corrective surgery for craniofacial abnormalities and was also associated with increased levels of anxiety and depression among patients during childhood[31].

While it is conceivable that surgical "normalisation" of appearance may have a salutary effect on parent-child interactions, the evidence for this is conflicting, with some demonstrating that mothers may be more protective of children with craniofacial differences thus leading to stronger attachment[32-34]. By better understanding the experience of craniosynostosis by families, supports can be appropriately tailored to address current areas of concern and improve the overall experience.

While many of our themes supported previous research, frustration with diagnostic delays was a key theme in our study and has not previously been reported for craniosynostosis. While there are currently no guidelines outlining the optimal age for surgical correction of craniosynostosis, much research has focussed on this issue. A systematic review by Mandela *et al.* found no conclusive evidence that earlier surgery may be beneficial to patients with sagittal synostosis, for example. There is no evidence that later surgery is beneficial for any of the craniosynostosis subtypes[5]. This speaks to the importance of early detection, as the age of diagnosis will affect when and what type of surgery is offered. One family in our study received the diagnosis of craniosynostosis early in the post-partum period. Due to the young age at detection, the child was eligible for less invasive endoscopic correction and helmet therapy. This option would not have been offered had the family experienced a diagnostic delay like that experienced by most families in our series.

In addition to improving craniosynostosis awareness, families also suggested that it would have been helpful to have received printed material during the initial consult to complement the information that was provided verbally. They expressed interest in receiving written pamphlets as well as a list of reliable and recommended internet sources where they could review the information further. The provision of these decision aids has previously been found to increase

both comprehension and risk recall[35]. In addition to improving informed consent, these interventions may increase overall satisfaction with the decision-making process[36]. This is especially relevant to the craniosynostosis patient population, as parents described significant anxiety associated with therapeutic decision-making.

This study is not without limitations. Firstly, given the small, homogeneous group of participants included, it is unclear whether our results accurately represent the experience of other populations. All twelve families elected to have the patient's mother complete the interviews rather than the father, regardless of marital status. While this may reflect the traditional distribution of caregiver responsibilities, it prevents us from identifying potential meaningful gender differences in the family experience of craniosynostosis. For example, parental stress has previously been reported to be higher in mothers with children diagnosed with single suture craniosynostosis when compared to their paternal counterparts[37]. Secondly, because our study design assigned recruitment responsibilities to the participating surgeons, the surgeons were not blinded to which families were enrolled. Although participants were ensured anonymity, it is unclear whether this influenced the interviews, potentially making participants more reluctant to identify points of dissatisfaction around their interactions with the surgeons. Despite these limitations, our study offers important insights for physicians caring for children with craniosynostosis and helps health care providers better understand the needs of families during the pre-, peri- and post-operative periods.

This study also suggests future avenues of research and development. Despite the fear expressed by parents in the pre-operative period, all families were ultimately pleased with their decision to proceed with corrective surgery. Future studies aim to explore the opinion of the patients themselves, and their views on their parents' decision regarding surgical correction of their craniosynostosis. Additionally, our findings speak to the importance of lifelong learning in

the medical field and identify the need for additional craniosynostosis teaching among general practitioners to allow for earlier detection in the community.

CONCLUSION

The diagnosis of craniosynostosis has a significant impact on families. This study offers a detailed look into the experiences of families from the point of diagnosis through to the post-operative recovery and transition home. Participants provide rich descriptions of their frustrations, accomplishments, supports and their suggestions for improvement. A better understanding of this experience will identify where further supports are needed and inform future practice, with the goal of improving the overall experience for other families moving forward.

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CONTRIBUTORSHIP STATEMENT

Dr. Victoria Kuta: Literature review, ethics submission, interviews, primary thematic analysis, manuscript preparation and submission

Dr. Lauren Curry: Secondary reviewer of transcripts and themes

Dr. P. Daniel McNeely: Patient recruitment, surgery, manuscript preparation

Dr. Simon Walling: Patient recruitment, surgery, manuscript preparation

Dr. Jill M. Chorney: Ethics submission, study design, manuscript preparation

Dr. Michael Bezuhly: Primary supervisor, ethics submission, study design, patient recruitment, surgery, manuscript preparation and submission

TABLE 1. Participant Demographics (n=12)

Variable	n (%)
Age, y*	32.4 ± 6.3 (range, 19-42)
Relationship to patient	
Mother	12 (100)
Father	0 (0)
Location	
Home address < 50 km from hospital	9 (75)
Home address > 50 km from hospital	3 (25)
Sex of Child	
Male	10 (83.3)
Female	2 (16.6)
Craniosynostosis type	
Sagittal	6 (50.0)
Coronal	4 (33.3)
Metopic	2 (16.7)

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Standards for Reporting Qualitative Research (SRQR)*

http://www.equator-network.org/reporting-guidelines/srqr/

	Page/line no(s).
Title and abstract	
Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	1-2
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	47-75
Introduction	
Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	114-139
Purpose or research question - Purpose of the study and specific objectives or questions	140-146
Methods	
Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	149-155; 181-188
Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	155-157
Context - Setting/site and salient contextual factors; rationale**	160-162
Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**	162-166;
Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	160-161; 179-180
Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**	163-164; 182-193;197

Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	173-176; 179-180
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	196-198;600
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	160-161; 179-181
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	182-188
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	189-193

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	199-393
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	199-393

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	396-459
Limitations - Trustworthiness and limitations of findings	460-473

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	27-28
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	30-32

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. **Standards for reporting qualitative research: a synthesis of recommendations.** *Academic Medicine*, Vol. 89, No. 9 / Sept 2014
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Understanding Families' Experiences Following a Diagnosis of Non-Syndromic Craniosynostosis: A Qualitative Study

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Data Sharing

Extra data can be accessed via the Dryad data repository at http://datadryad.org/ with the doi:10.5061/dryad.fr9305r

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ABSTRACT

Objectives: Craniosynostosis is typically diagnosed and surgically corrected within the first year of life. The diagnosis and surgical correction of the condition can be a very stressful experience for families. Despite this, there is little research exploring the impact that craniosynostosis has on families, especially in the period immediately following diagnosis and correction. In this study, the authors aimed to qualitatively examine the psychosocial experience of families with a child diagnosed with craniosynostosis.

Design: Qualitative study.

Setting: Tertiary care paediatric health centre.

Participants: Mothers of children newly diagnosed with single-suture, non-syndromic craniosynostosis

Intervention: Semistructured interviews regarding parental experience with the initial diagnosis, their decision on corrective surgery for their child, the operative experience, the impact of craniosynostosis on the family and the challenges they encountered throughout their journey.

Primary and Secondary Outcome Measures: Thematic analysis, a type of qualitative analysis that provides an in-depth account of participant's experiences, was used to analyze the interview data.

Results: Over a four-year period, twelve families meeting eligibility criteria completed the study. Three main themes (6 subthemes) emerged from the pre-operative interviews: frustration with diagnostic delays (parental intuition and advocacy, hope for improved awareness), understanding what to expect (healthcare supports, interest in connecting with other families), and justifying the need for corrective surgery (influence of the surgeon, struggle with cosmetic indications). Two main themes (4 subthemes) were drawn from the post-operative interviews: overcoming fear (the

role of healthcare professionals, transition home) and relief (reduction in parental anxiety, cosmetic improvements).

Conclusions: Overall, the diagnosis of craniosynostosis has a profound impact on families, leading them to face many struggles throughout their journey. A better understanding of these experiences will help to inform future practice, with a hope to improve this experience for other families moving forward.

ARTICLE SUMMARY

Strengths and limitations of this study

- The prospective, qualitative study involving semi-structured interviews allowed mothers of children with craniosynostosis to richly describe their journey from the point of diagnosis through to the post-operative period.
- This study examined both major pre-operative and post-operative themes within the same cohort, allowing for identification of how maternal concerns change over the course of the treatment of their child.
- Given the small, homogeneous group of participants included, it is unclear whether the results accurately represent the experience of other populations.

INTRODUCTION

Craniosynostosis, a congenital anomaly involving abnormal fusion of calvarial sutures, affects 1 in every 2,000 to 3,000 live births[1-4]. It is traditionally classified as either syndromic or non-syndromic. Non-syndromic synostosis is not associated with other dysmorphisms outside the abnormal craniofacial morphology, and typically involves only a single suture. The most common subtypes include sagittal, metopic, unicoronal, bicoronal and lambdoidal. Non-syndromic craniosynostosis is classically treated with corrective surgery within the first year of life, with inconclusive evidence that earlier intervention may be beneficial for certain subtypes[5].

While still controversial, there is increasing evidence that non-syndromic craniosynostosis may be associated with long term neurodevelopmental deficits, including difficulties with visuospatial skills, memory, speech and language, and learning disorders[5]. Further studies have suggested that these impairments will persist and cannot be prevented with corrective surgery[6-9]. Despite this inconclusive evidence, most parents opt for corrective surgery to remodel the skull and allow for normal head growth in their child.

Although the impact of non-syndromic craniosynostosis on neurocognitive development remains in question, children with this congenital anomaly may be faced with social and psychological barriers that negatively impact their self-esteem and social function owing to their abnormal appearance[10-11]. While many reports document the psychosocial aspects of craniosynostosis from the perspective of the patient, they do not detail the experience of the family. Because corrective surgery is typically performed when patients are infants, parents are responsible for making proxy decisions and are actively involved in patient care. Thus, to obtain a true understanding of early experiences with craniosynostosis, it is important to expand our scope, and study not the just the patient, but the family.

Previous studies that attempted to quantify parental stress levels found no difference in the level of stress experienced by parents of children with and without single-suture craniosynostosis before corrective surgery[12-16]. Other studies have examined parental satisfaction with their child's postoperative results, with high satisfaction with surgical outcomes generally reported[16-18].

The aim of the current study is to provide an in-depth qualitative description of families' experiences with craniosynostosis. By adopting a qualitative approach involving semi-structured interviews, we allowed families to richly describe their journey and freely communicate personally meaningful topics. This study prospectively explored the experience of families beginning at the time of diagnosis and continuing to the postoperative period. We aim to use our findings to inform future research and practice, with the hope of improving the overall experience for families facing this diagnosis in the future.

METHODS

Thematic analysis was used as the qualitative methodology. Thematic analysis is a method for identifying, analyzing and reporting, in detail, patterns within participants' experiences of an event[19,20]. The role of the researcher in the interpretation is also recognised[21]. Rather than testing a specific hypothesis, this method allows for flexible exploration of a topic in a small, homogeneous sample of respondents for whom the topic is particularly relevant. Thematic analysis informed both the data collection and the reporting for the current study. The interviewer received thematic analysis training under one of the senior authors (J.M.C) using

previously described methodology[20]. The interviewer was not directly involved in the management of patients.

Patient and public involvement

Neither patients nor the public were involved in the study design. The research question was developed based on comments expressed to the corresponding author by several families with a diagnosis of craniosynostosis regarding the need for timelier referral to the craniofacial program and a need for additional teaching resources to primary care providers on the diagnosis. While these families were the impetus for the research question, they did not directly participate in the design or conduct of the study. As stated below, participants were given the opportunity to review a summary of the themes and provide feedback following data analysis.

Study sample

Institutional research ethics approval was obtained for this study from the IWK Health Centre Research Ethics Board. All families presenting to the IWK Health Centre with a child who received a new diagnosis of non-syndromic craniosynostosis were eligible for this study. These families were identified prospectively by participating surgeons between February 15, 2016 and February 15, 2018. Eligible families were informed of the study by one of the participating surgeons during their initial consult, after receiving a diagnosis. Families were then consented to have their contact information provided to the principle investigator of the study. Twelve eligible families were identified and enrolled into the study over the enrollment period. This sample size is typical for thematic analysis studies to reach thematic saturation[20]. Participant

demographics are presented in Table 1. All participants were mothers and were interviewed individually.

Data collection

Participants completed two phone interviews. The first interview was completed within a month of receiving the initial diagnosis. The second interview was completed three months post-operatively, or three months after the initial interview if the family decided not to proceed with surgery. All interviews were completed by the first author. Verbal consent was obtained over the phone before initiating the interviews. Interviews were semistructured using an interview guide based on the team's clinical experience and a scoping literature review. The initial interview guide (supplementary file) contained questions on the diagnostic experience as well as the decision on corrective surgery. The second interview guide investigated the surgical experience and the recovery period.

Analysis method

Interviews were recorded and transcribed verbatim by a professional transcriptionist with subsequent deidentification. Transcripts were divided into pre-operative and post-operative categories. Initial coding was completed by the first author using thematic analysis methods. Transcripts were reviewed multiple times to assign codes to the main topics discussed by the participant. The codes identified in earlier transcripts informed the coding of later transcripts. New codes identified in later transcripts prompted earlier transcripts to be reviewed again to determine if these codes were also present in these. The organization of themes followed an iterative process aiming to identify the meaning behind participants' statements rather than solely

the prevalence of topics discussed. Themes were then clustered, allowing for superordinate themes to be generated based on subsumption and abstraction techniques.

To ensure rigour, theme development was reviewed and discussed between the first and senior authors to confirm that the interpretations accurately represented the transcript data. A second author reviewed the transcripts independently to assess for representativeness. Member checking was also performed, where participants were given the opportunity to review a summary of the themes and provide feedback.

RESULTS

Themes were organised into preoperative and postoperative categories. Three main themes emerged from the preoperative interviews: frustration with diagnostic delays, understanding what to expect, and justifying the need for corrective surgery. Two main themes emerged from the postoperative interviews: overcoming fear, and relief. Representative quotes are included throughout the text.

Frustration with diagnostic delays

Most participants expressed some frustration around diagnostic delays, excepting two participants whose child was born at the tertiary care hospital and received a diagnosis immediately post-partum. Two subthemes emerged: parental intuition/advocacy and hope for improved awareness.

Parental intuition and advocacy

Ten families noticed the abnormal shape of their child's head at birth and expressed concerns (Participant 12 - "I knew something was wrong, but I couldn't prove it"). They were

frequently offered the explanation that it was a result of the birthing process and were told it would resolve spontaneously (Participant 2 - "The day he was born at the hospital we started noticing that one of his eyes would not open, and his nose was crooked a bit and the opening in one of his nostrils was very narrow. We were told it was because of what they call a traumatic birth, and it would fix as he grows.")

Over time, when no aesthetic improvement was observed, families began seeking medical advice. One family requested an x-ray; however, the diagnosis of craniosynostosis was missed. Other families resorted to taking their child to the emergency room or requesting a referral to a paediatrician after feeling their concerns were not adequately addressed by their family physician. One family expressed feelings of guilt around not pushing for the referral to a specialist earlier (Participant 2 - "I started doing my own research online and that's when I realised something should have been done when he was younger. I was a bit frustrated with my doctor. I felt like I should have pushed for it sooner when he was younger").

Hope for improved awareness

Overall, families describe a lack of awareness among community family physicians around craniosynostosis. One mother explains her surprise that the craniosynostosis wasn't picked up by her family physician despite regular exams (Participant 5 - "At every doctor's appointment they are always doing measurements of his head and looking for his soft spot"). Another mother describes her own physical findings that she felt were discounted (Participant 6 - "I also noticed a ridge along the top of his skull that I brought up to my GP and he kind of passed if off as not a big deal"). When asked how their overall experience could be improved, many parents suggested efforts to increase craniosynostosis awareness to allow for earlier detection (Participant 10 – "Being able to have more education for family doctors, nurse practitioners, that

sort of thing, around what is normal and what's not normal"; Participant 2 - "I think it's something they should be more educated on.")

Understanding what to expect

Participants described the importance of being informed on what to expect and how this helped them to feel more comfortable during the whole experience. Two subthemes emerged: health care supports, and interest in connecting with other families.

Health care supports

Participants described feeling overwhelmed during their initial consultation, and most were unaware that surgery would be recommended for their child. Many had come mentally prepared with questions, but were then unable to recall these during the consult (Participant 1 - "So when he said 'do you have any questions' I was like 'no' because I was just trying to take it all in"). Other families chose to write down their questions ahead of time, which proved to be a more successful strategy. One participant commented on too many learners being present in the room- a comment that nursing staff later agreed with. This added to the overwhelming nature of the consult and hindered this participant's ability to express themselves. All participants described receiving verbal information; however, many suggested that additional written resources could have been provided for review once they have had time to process things (Participant 10 - "So I would say having a cheat sheet of something, where it's already written down that you leave with. Because in the moment, you're listening and not thinking of writing it down yourself"). Skull models used during the consult were helpful for participant education. As many participants were doing their own research, they requested references to reputable resources for further information. Additionally, participants appreciated having access to a

specialised nurse after the consultation with the surgeons who they could email or call with additional questions. All participants spoke very highly of this support system and felt that it significantly reduced their anxiety (Participant 10 - "It was so helpful to know that if we did [have questions], we had a way to get a hold of [the clinic nurse]").

Interest in connecting with other families

While all of the participants felt their consultation visits were informative, they expressed a strong interest in connecting with other families who have been through a similar experience (Participant 8 – "The doctor told me what I could expect, what I'm going to see after the surgery and all this, but hearing it from a parent's perspective is a whole different story"). Participants felt it was important to hear other local success stories and mentioned that they would like access to pre- and post-operative photos from other families (Participant 7 - "As a mom and dad you really need to see that other children have risen through it"). Many participants reached out to other families through craniosynostosis support groups on social media platforms. They described the support and hope provided through these online chats (Participant 10 - "Those connections are important, I think, just to see that there are other people who are going through it and have made it through to the other side"). While most participants thought these types of communication would be helpful, one participant describes her emotional struggle after meeting with a family who experienced complications (Participant 12 - "I was scared. I'm even more scared now than I was then, because now that we're in support groups and see what's going to happen, we are scared about the surgery. It's always hard when you have a small sample size too. It can make things look like they are in different proportions than they are").

Justifying the need for corrective surgery

Participants describe the difficult process of reaching a decision on corrective surgery for their child. Two subthemes emerged: influence of the surgeon, and struggle with cosmetic indications.

Influence of the surgeon

All participants decided to proceed with corrective surgery for their child. This decision was reached during the initial consultation. The families described the importance of the information they received during this surgical visit and stated there were no outside influences factored into their decision. This speaks to the magnitude of the influence held by the surgeons. Many participants describe their positive relationship with the surgeons, and how it gave them confidence to consent to surgery during the first visit (Participant 7 - "I feel really confident with the doctors, I feel good with them, which I definitely think is a part of it"). Both bedside manner and the communication style of the surgeons were noted to help participants feel more comfortable with their decision on surgery (Participant 11 - "They talk to you like you're a human being. They talk to you in a fashion that, you know, we know what you're actually telling us. But it's easing my mind that we have such a great team"). Participants also appreciated surgeons speaking in lay terms during the consultation and consenting processes. Other families focused primarily on the evidence and risks communicated by the surgeon (Participant 4 - "When he told us there are 10-15% that have pressure build-up in their brain and it can affect development and also his vision, […] I don't think we had to think very long to decide that we do not want to take that risk if we can definitely prevent it by doing a surgery"). While many families deliberated on this difficult decision, some families describe the feeling of not having a choice, that surgery was the only option (Participant 1 -"He was talking about how the shape of

his head would just continue to grow this way, and his brain would be squeezed and there would be pressure. But yeah, I felt like this was our only option").

Struggle with cosmetic indications

The participants discussed their struggle weighing the importance of cosmetic indications, with most families stating that the decision would be much more difficult if the surgery was for aesthetic purposes alone (Participant 4 -"If it was just cosmetic it definitely would have taken us more time to think about it."). While most families identified potential neurological risks as their primary motivator, it seems that aesthetic concerns were still present, even if not directly vocalised (Participant 11 – "So we know that it's not a decision that we're being selfish and trying to fix her look. It needs to be completed"). Other families were more direct in voicing their cosmetic concerns and were worried about potential psychosocial difficulties later in life, especially after learning about the potentially progressive nature of the condition. This included concerns around future bullying, depression, and the even the risk of suicide if surgery was not performed (Participant 8 – "No child should grow up and develop that head shape"; Participant 12 – "When she was first diagnosed, I would have said no, but now, the asymmetry is so much that it wouldn't be fair to her not to repair it. She would always look very different from other children"; Participant 10 – "If we don't do the surgery, he's going to hate us later in life because we didn't fix this. He would probably be teased and picked on"). One family related their cosmetic concerns to the sex of their child, describing the gender-biased aesthetic standard they have experienced in society (Participant 12 - "[My husband] keeps saying specifically because she's a girl, and we live in a society where what a girl looks like is important").

Overcoming fear

The participants describe fear at various stages of their journey and shared what helped them cope with this emotion. Two subthemes emerged: the role of healthcare professionals, and the transition home.

The role of healthcare professionals

Participants discussed at length how health care providers helped reduce their fear and anxiety while in hospital. Firstly, although parents found it very difficult to hand over their child for surgery, the were comforted by regular updates throughout the procedure (Participant 4 - "You're just waiting for that nurse to come and give us the news that everything is going well, and like it's supposed to. And she did, every time. That was great"). There was only one family who did not receive regular updates throughout the operation. This participant describes feeling extremely nervous in the waiting room after not being informed about a delay in the surgical start time (Participant 5 - "I would have liked to know that they started later than think something bad happened"). Overall, regardless of the stage in their journey, parents described feeling much calmer when they were kept informed. In addition, families commented on the importance of empathy in healthcare. For example, one participant (Participant 10) spoke of the impact that small gestures can have on a family during a difficult time: "They brought us out the bag of his hair. One of them had written on it 'baby's first haircut'. It let you know that they care about your child, that they see that it's not just another patient."

Transitioning Home

Most participants were very surprised with the short recovery time after discharge home (Participant 5 – "You don't think they are going to recover that quick […], but within 2 or 3 days they're their normal self"). This introduced a new fear for parents. Many families described difficulty allowing their child to return to regular activities out of fear they would hurt themselves (Participant 8 - "We're still really scared, like if he falls and bangs his head or something, we're like 'Ooohh!'"). These concerns were heightened if the child had young siblings (Participant 1-"And even now, it's hard, because [my other children] are so young, and he still has the soft spot on his head, but they don't understand"). When asked what helped ease their transition home, families stated that were very grateful to be given contact information to reach their healthcare team with questions after discharge. They felt comfortable emailing or calling members of the team with post-operative questions. Ultimately, the ongoing support for parents helped to reduce feelings of fear and anxiety after discharge.

Relief

All twelve participants expressed a sense of relief post-operatively, feeling confident they had made the right decision regarding corrective surgery. Two subthemes emerged: reduction in parental anxiety and cosmetic improvements.

Reduction in parental anxiety

Participants described significant anxiety leading up to the operation, despite feeling very well informed. Many families feared that they would regret their decision regarding corrective surgery and felt a substantial amount of pressure to make the right choice (Participant 7 -"My fear was that he would be changed for the worse and that we would forever regret the decision to do it"). All participants felt their anxiety subside post-operatively after a successful operation.

Parents also described significant anxiety around the potential for neurological deficits associated with craniosynostosis, worrying that irreversible effects would occur before surgery (Participant 1 - "I was always making sure he could focus on me, and if he couldn't focus on me I'd think 'oh no, is he going blind'"). Post-operatively, participants no longer worried about neurological deficits, and felt they were no longer anxious about their child meeting developmental milestones. Many families also described positive behavioural changes in their child that they attributed to the surgery (Participant 7 – "He is happier and a little more relaxed. He is able to play more"; Participant 12 – "She was almost, I would say, mute leading up to surgery. Within a week of surgery she started making sounds and now, three months later, has a full vocabulary").

Cosmetic improvements

Although most families claimed neurological deficits were their primary motivation for surgery, the cosmetic improvements were heavily commented upon in the post-operative interviews (Participant 3 - "The best part would be how he looked after surgery. Like three weeks after, how good he looked. He looked like a total different baby"). Participants expressed relief with the aesthetic success of the operation (Participant 4 – "It did really change the way that his face and features look. It wasn't the main reason for us to do the surgery, but it was definitely, like, 'oh wow!'"). One mother commented on the practical aspect of her child's new head shape (Participant 6 - "I appreciate being able to put a hat on him now"). Another reflected on the progressive nature of craniosynostosis, describing what she felt her child would have looked like now without the operation (Participant 8 - "If we never would have done that surgery […], his head would be so much like a football right now"). Overall, parents seemed very

satisfied that their child would no longer stand out due to a cranial deformity (Participant 6 - "He looks like a completely normal 8-month old now, besides the really faint scarring").

DISCUSSION

The diagnosis and treatment of craniosynostosis has a significant impact on families. This qualitative analysis provides a rich description of families' experiences with craniosynostosis, from the point of diagnosis through to the period of surgical recovery.

In the pre-operative interviews, most families described frustration around diagnostic delay, acknowledging the importance of advocating for their child and their hope for improved craniosynostosis awareness in community practice. They stressed the importance of knowing what to expect, and the value in both healthcare supports and making connections with other families. They also discussed the struggle to decide on corrective surgery, acknowledging the influence of the surgeon and their difficulties weighing functional and cosmetic indications.

In the post-operative interviews, families discussed their journey of overcoming their fear. They highlighted the contribution of healthcare professionals and emphasised the challenges of transitioning home. There was also a very different tone to the second round of interviews, one of relief. All families were happy with their decision to proceed with corrective surgery, and felt their anxiety was reduced in the post-operative period. They also commented on their satisfaction with the cosmetic improvement.

Overall, the pre-operative themes centered around feelings of uncertainty and illustrate the struggle families experience in the early stages of this process. Parents were often left questioning their decisions and wondering if they were making the right choice for their child. In this period of uncertainty, parents tended to place significant weight on the opinion of their child's surgeon and draw confidence from this interaction. In contrast, the post-operative period

was characterized by themes illustrating strength and independence. Although uncertainty still existed throughout the recovery period, parents appeared well equipped to handle these challenges as a family unit, needing less reassurance from healthcare professionals. All parents included in this study described a positive change in their child after surgery. Parents appeared to draw strength from the fact that their decision to proceed with surgery was what led to this positive outcome. Also, while parents acted as advocates for their own children in the preoperative period, they advocated for the craniosynostosis community at large in the post-operative period, once again illustrating their personal growth and confidence.

For most forms of non-syndromic craniosynostosis the prevention of elevated intracranial pressure and associated neurocognitive deficits is the principal indication for surgery[22]. Sagittal craniosynostosis may be an exception, as compensatory growth along patent sutures largely prevents elevated intracranial pressure but produces a stigmatising head shape. To this end, aesthetic concerns may be a greater motivation for surgical correction of sagittal craniosynostosis[23]. A recent health utility outcome study found relatively high utility scores for sagittal craniosynostosis, suggesting that the cosmetic burden of this condition as perceived by the general population is low[24]. This aligns with our findings, where most participants stated that the cosmetic indications for corrective surgery were secondary to the neurological ones. The observation that concerns regarding cognitive sequelae were the main motivation for corrective surgery underscores the need for ongoing clinical research into functional aspects of craniosynostosis management. Of note, all families decided to proceed with corrective surgery, including those who received a diagnosis of sagittal craniosynostosis. While families may find it difficult to choose a potentially morbid corrective surgery for aesthetic indications alone, it is important to remind parents of the potential psychosocial consequences of living with an

uncorrected craniofacial abnormality[25]. Interestingly, despite the difficulty justifying aesthetic indications pre-operatively, the satisfaction with cosmetic improvements was heavily commented on in the postoperative interviews.

Many of the themes developed in our study align with those reported by previous studies examining the experiences of families with children diagnosed with other craniofacial deformities. For example, parents with children diagnosed with cleft lip/palate described their anxiety around surgery and their need for emotional support throughout treatment, for both themselves and their child[26-28]. Furthermore, families of children diagnosed with craniofacial abnormalities have expressed fears that their child will be bullied and ostracised later in life[26,28]. Previous studies have emphasised the importance of parental support in healthcare, suggesting that the emotional state of caregivers significantly influences the emotional development of children with craniofacial abnormalities[28-30]. Elevated caregiver stress was found to have long-lasting, negative psychosocial effects on children who received corrective surgery for craniofacial abnormalities and was also associated with increased levels of anxiety and depression among patients during childhood[31]. While it is conceivable that surgical "normalisation" of appearance may have a salutary effect on parent-child interactions, the evidence for this is conflicting, with some demonstrating that mothers may be more protective of children with craniofacial differences thus leading to stronger attachment[32-34]. By better understanding the experience of craniosynostosis by families, supports can be appropriately tailored to address current areas of concern and improve the overall experience.

While many of our themes supported previous research, frustration with diagnostic delays was a key theme in our study and has not previously been reported for craniosynostosis. While there are currently no guidelines outlining the optimal age for surgical correction of

craniosynostosis, much research has focussed on this issue. A systematic review by Mandela *et al.* found no conclusive evidence that earlier surgery may be beneficial to patients with sagittal synostosis, for example. There is no evidence that later surgery is beneficial for any of the craniosynostosis subtypes[5]. This speaks to the importance of early detection, as the age of diagnosis will affect when and what type of surgery is offered. One family in our study received the diagnosis of craniosynostosis early in the post-partum period. Due to the young age at detection, the child was eligible for less invasive endoscopic correction and helmet therapy. This option would not have been offered had the family experienced a diagnostic delay like that experienced by most families in our series.

In addition to improving craniosynostosis awareness, families also suggested that it would have been helpful to have received printed material during the initial consult to complement the information that was provided verbally. They expressed interest in receiving written pamphlets as well as a list of reliable and recommended internet sources where they could review the information further. The provision of these decision aids has previously been found to increase both comprehension and risk recall[35]. In addition to improving informed consent, these interventions may increase overall satisfaction with the decision-making process[36]. This is especially relevant to the craniosynostosis patient population, as parents described significant anxiety associated with therapeutic decision-making.

This study is not without limitations. Firstly, given the small, homogeneous group of participants included, it is unclear whether our results accurately represent the experience of other populations. All twelve families elected to have the patient's mother complete the interviews rather than the father, regardless of marital status. While this may reflect the traditional distribution of caregiver responsibilities, it prevents us from identifying potential

meaningful gender differences in the family experience of craniosynostosis. For example, parental stress has previously been reported to be higher in mothers with children diagnosed with single suture craniosynostosis when compared to their paternal counterparts[37]. Secondly, because our study design assigned recruitment responsibilities to the participating surgeons, the surgeons were not blinded to which families were enrolled. Although participants were ensured anonymity, it is unclear whether this influenced the interviews, potentially making participants more reluctant to identify points of dissatisfaction around their interactions with the surgeons. Despite these limitations, our study offers important insights for physicians caring for children with craniosynostosis and helps health care providers better understand the needs of families during the pre-, peri- and post-operative periods.

This study also suggests future avenues of research and development. Despite the fear expressed by parents in the pre-operative period, all families were ultimately pleased with their decision to proceed with corrective surgery. Future studies aim to explore the opinion of the patients themselves, and their views on their parents' decision regarding surgical correction of their craniosynostosis. Additionally, our findings speak to the importance of lifelong learning in the medical field and identify the need for additional craniosynostosis teaching among general practitioners to allow for earlier detection in the community.

CONCLUSION

The diagnosis of craniosynostosis has a significant impact on families. This study offers a detailed look into the experiences of families from the point of diagnosis through to the post-operative recovery and transition home. Participants provide rich descriptions of their frustrations, accomplishments, supports and their suggestions for improvement. A better

understanding of this experience will identify where further supports are needed and inform future practice, with the goal of improving the overall experience for other families moving forward.

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CONTRIBUTORSHIP STATEMENT

Dr. Victoria Kuta: Literature review, ethics submission, interviews, primary thematic analysis, manuscript preparation and submission

Dr. Lauren Curry: Secondary reviewer of transcripts and themes

Dr. P. Daniel McNeely: Patient recruitment, surgery, manuscript preparation

Dr. Simon Walling: Patient recruitment, surgery, manuscript preparation

Dr. Jill M. Chorney: Ethics submission, study design, manuscript preparation

Dr. Michael Bezuhly: Primary supervisor, ethics submission, study design, patient recruitment, surgery, manuscript preparation and submission

TABLE 1. Participant Demographics (n=12)

Variable	n (%)
Age, y*	32.4 ± 6.3 (range, 19-42)
Relationship to patient	
Mother	12 (100)
Father	0 (0)
Location	
Home address < 50 km from hospital	9 (75)
Home address > 50 km from hospital	3 (25)
Sex of Child	
Male	10 (83.3)
Female	2 (16.6)
Craniosynostosis type	
Sagittal	6 (50.0)
Coronal	4 (33.3)
Metopic	2 (16.7)

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Understanding Families' Experiences Following a Non-Syndromic Craniosynostosis Diagnosis: A Qualitative Study - Parent Interview Guide ID # _____

Interview Guide #1

Thank you for agreeing to this first interview to discuss your child's craniosynostosis diagnosis.

Pre Diagnosis – When you first learned that your child might have something wrong with the development of their skull. Thinking back to *before your visit with the surgeon* here today;

1. Can you tell me when and how you noticed there was an issue for your child and who was the first person to notice it?

2. After realizing something was wrong, did you have any main concerns or worries?

3. Can you tell me who referred you and your child to the health centre? Do you recall how long you waited before you received this appointment?

4. Can you tell me about what you were feeling before the consultation with your child's surgeon?
    a. Did you do any research on your own?
    b. Did you know what craniosynostosis was before the consultation?
    c. What did you want to address at the consultation today?

5. Were you aware of the potential need for surgery?
    a. If so, what were your thoughts on corrective surgery going into the consultation?
    b. If not, what did you think the option(s) may be for your child?

Post Consultation- Once the diagnosis was made;

6. Please tell me about the consultation with your surgeon.
    a. What were your thoughts and feelings during the consult?
    b. Did you have questions for the surgeon? Was the surgeon able to provide you with the information that you needed?
    c. What type of information did you receive during the consult? What format did you receive the information (written, verbal)?
    d. Do you have access to other resources should you have additional questions before the surgery?

7. Please tell me about how you made the decision to have corrective surgery for your child.
    a. Did the consultation have any influence on your decision?
    b. Were there any factors that influenced your decision?
    c. Do you believe this will impact the quality of life of your child?

8. In closing, is there anything else you would like to include?

Understanding Families' Experiences Following a Non-Syndromic Craniosynostosis Diagnosis: A Qualitative Study - Parent Interview Guide ID # _____

Interview Guide #2

Thank you for meeting with me for a second interview. It's been about three months since your child's surgery; today we will discuss your experience with your child's surgery and recovery.

Following Surgery

1. Please tell me about the days leading up to the surgery.
    a. How were you feeling?
    b. Did you have a thorough understanding of what the surgery entailed?
    c. Were you comfortable with your decision to go ahead with surgery?

2. Please tell me what your experience with your child's surgery was like.
    a. How did you feel in the days and weeks following the surgery?
    b. How did others respond to you during your child's recovery?
    c. Were there any particularly challenges in the recovery?
    d. How did you deal with the challenges that arose?

3. Please tell me how you feel now about your decision to have reconstructive surgery for your child?
    a. Is there anything you would change about the experience?
    b. If you were making the decision now, would you do it again?

4. Has this experience impacted your life for you and your child?

5. What do you think health professionals should know about parents' experiences with craniosynostosis?

6. In closing, is there anything else you would like to include?

Standards for Reporting Qualitative Research (SRQR)*

http://www.equator-network.org/reporting-guidelines/srqr/

	Page/line no(s).
Title and abstract	
Title - Concise description of the nature and topic of the study Identifying the study as qualitative or indicating the approach (e.g., ethnography, grounded theory) or data collection methods (e.g., interview, focus group) is recommended	1-2
Abstract - Summary of key elements of the study using the abstract format of the intended publication; typically includes background, purpose, methods, results, and conclusions	47-75
Introduction	
Problem formulation - Description and significance of the problem/phenomenon studied; review of relevant theory and empirical work; problem statement	114-139
Purpose or research question - Purpose of the study and specific objectives or questions	140-146
Methods	
Qualitative approach and research paradigm - Qualitative approach (e.g., ethnography, grounded theory, case study, phenomenology, narrative research) and guiding theory if appropriate; identifying the research paradigm (e.g., postpositivist, constructivist/ interpretivist) is also recommended; rationale**	149-155; 181-188
Researcher characteristics and reflexivity - Researchers' characteristics that may influence the research, including personal attributes, qualifications/experience, relationship with participants, assumptions, and/or presuppositions; potential or actual interaction between researchers' characteristics and the research questions, approach, methods, results, and/or transferability	155-157
Context - Setting/site and salient contextual factors; rationale**	160-162
Sampling strategy - How and why research participants, documents, or events were selected; criteria for deciding when no further sampling was necessary (e.g., sampling saturation); rationale**	162-166;
Ethical issues pertaining to human subjects - Documentation of approval by an appropriate ethics review board and participant consent, or explanation for lack thereof; other confidentiality and data security issues	160-161; 179-180
Data collection methods - Types of data collected; details of data collection procedures including (as appropriate) start and stop dates of data collection and analysis, iterative process, triangulation of sources/methods, and modification of procedures in response to evolving study findings; rationale**	163-164; 182-193;197

Data collection instruments and technologies - Description of instruments (e.g., interview guides, questionnaires) and devices (e.g., audio recorders) used for data collection; if/how the instrument(s) changed over the course of the study	173-176; 179-180
Units of study - Number and relevant characteristics of participants, documents, or events included in the study; level of participation (could be reported in results)	196-198;600
Data processing - Methods for processing data prior to and during analysis, including transcription, data entry, data management and security, verification of data integrity, data coding, and anonymization/de-identification of excerpts	160-161; 179-181
Data analysis - Process by which inferences, themes, etc., were identified and developed, including the researchers involved in data analysis; usually references a specific paradigm or approach; rationale**	182-188
Techniques to enhance trustworthiness - Techniques to enhance trustworthiness and credibility of data analysis (e.g., member checking, audit trail, triangulation); rationale**	189-193

Results/findings

Synthesis and interpretation - Main findings (e.g., interpretations, inferences, and themes); might include development of a theory or model, or integration with prior research or theory	199-393
Links to empirical data - Evidence (e.g., quotes, field notes, text excerpts, photographs) to substantiate analytic findings	199-393

Discussion

Integration with prior work, implications, transferability, and contribution(s) to the field - Short summary of main findings; explanation of how findings and conclusions connect to, support, elaborate on, or challenge conclusions of earlier scholarship; discussion of scope of application/generalizability; identification of unique contribution(s) to scholarship in a discipline or field	396-459
Limitations - Trustworthiness and limitations of findings	460-473

Other

Conflicts of interest - Potential sources of influence or perceived influence on study conduct and conclusions; how these were managed	27-28
Funding - Sources of funding and other support; role of funders in data collection, interpretation, and reporting	30-32

*The authors created the SRQR by searching the literature to identify guidelines, reporting standards, and critical appraisal criteria for qualitative research; reviewing the reference lists of retrieved sources; and contacting experts to gain feedback. The SRQR aims to improve the transparency of all aspects of qualitative research by providing clear standards for reporting qualitative research.

**The rationale should briefly discuss the justification for choosing that theory, approach, method, or technique rather than other options available, the assumptions and limitations implicit in those choices, and how those choices influence study conclusions and transferability. As appropriate, the rationale for several items might be discussed together.

Reference:

O'Brien BC, Harris IB, Beckman TJ, Reed DA, Cook DA. **Standards for reporting qualitative research: a synthesis of recommendations.** *Academic Medicine*, Vol. 89, No. 9 / Sept 2014
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