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from the Faculty of Medicine 2083*

Having a child with non-syndromic craniosynostosis

*Parents' experiences of care, need of support and
perceived stress*

ANNA STENSON ZERPE



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Abstract

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Objective: Being a parent of a child with a diagnosis of craniosynostosis that requires surgery can lead to anxiety and emotional distress. The general aim was to explore parents' experiences when their child is diagnosed with and treated for non-syndromic craniosynostosis. This was accomplished by investigating how parents perceived treatment, information, and participation in care, as well as their parental stress, symptoms of anxiety and depression, and health-related quality of life.

Methods: Study I investigated Swedish parents' (n=20) experiences of having a child with craniosynostosis and their perceptions of the care provided by conducting interviews and thematic analysis. Study II explored parents' (n=19) experiences of the time at the hospital and the year after discharge by conducting interviews and content analysis. Study III explored parents' satisfaction with hospital care and factors that influenced their perception of quality of care using questionnaires (n=98) and interviews (n=19), employing a mixed method. Study IV assessed parents' perceived parental and psychological stress and health-related quality of life before and one year after surgery, using questionnaires (n=29).

Results: Parents rarely had previous knowledge about craniosynostosis. For this reason, the craniofacial team was highlighted as the most important source of information and support. Parents described the time in the hospital and after discharge as challenging but ultimately good, and support from family, peers, and the expert team was considered essential. Parents were generally satisfied with hospital care, and factors existed that either facilitated or impeded their experience of quality of care. No differences regarding parental stress, health-related quality of life, and psychological distress before and one year after the child's surgery were found, but there was an association between parental stress and symptoms of depression both before and one year after surgery.

Conclusions: This thesis provides an understanding of parents' experiences when having a child undergoing craniosynostosis surgery. Most parents were satisfied with the care provided, but areas of improvement were described. There appears to be an association between parental stress and symptoms of depression. The findings highlight that healthcare professionals should be more responsive to parents' different care needs.

Keywords: Craniosynostosis, parents, family, parental stress, support, surgery, hospital care

Anna Stenson Zerpe, Department of Surgical Sciences, Plastic Surgery, Akademiska sjukhuset, Uppsala University, SE-75185 Uppsala, Sweden.

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Little blue, be my shelter

Be my cradle, be my womb

Be my boat, be my river

Be the stillness of the moon

If I could, I'd go with you

To a place, I never knew

In your eyes, so dark and open

There's a light that leads me back to you

Jacob Collier

To Juno & Ilon

List of Papers

This thesis is based on the following papers, which are referred to in the text by their Roman numerals.

- I. Zerpe, A. S., Nowinski, D., Ramklint, M., & Oster, C. (2020). Parents' Experiences of Their Child's Craniosynostosis and the Initial Care Process. *The Journal of Craniofacial Surgery*, 31(1), 251-256.
- II. Zerpe, A. S., Nowinski, D., Ramklint, M., & Oster, C. (2022). "When the surgery was over, I felt like the worst part had passed" experiences of parents of children with craniosynostosis. *Journal for Specialists in Pediatric Nursing*, 27(2)
- III. Zerpe, A. S., Ramklint, M., Nowinski, D., & Öster, C. (2024). Parental satisfaction with hospital care for children with non-syndromic craniosynostosis: A mixed-method study. *Journal of Pediatric Nursing*, 77, e465-e473.
- IV. Zerpe, A. S., Ramklint, M., Nowinski, D., & Öster, C. Assessing parents' perceived stress, psychological distress and health-related quality of life before and one year after their child's craniosynostosis surgery (Manuscript).

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Abbreviations

CT	Computer Tomography
ICU	Intensive Care Unit
ICD-10	International Classification of Diseases, Tenth Revision
SPSQ	Swedish Parenting Stress Questionnaire
EQ-5D-3L	A standardized measure of health-related quality of life
HSCL-25	Hopkins Symptom Checklist 25
VAS	Visual Analogue Scale

Preface

In my work as a clinical nurse coordinator for craniosynostosis and craniofacial syndromes, a significant part of my role involves meeting families who have recently received their child's diagnosis. I follow them through the process from diagnosis to surgery, as well as during the aftercare, and continue to provide advice and support for several years. Early on during my time as a nurse coordinator, I noticed that the concept of craniosynostosis is usually unknown in the population and that this group of parents often experiences anxiety about treatment and future prospects when their child is diagnosed.

Over the past 10 years in this profession, a lot has changed. Nowadays, almost every first-time visitor to the craniofacial team comes in already well-informed about the condition, thanks to easy access to information on the internet. In my experience, parents have also become more adept at connecting online and forming communities for support through forums and social media. Despite this, I realize that parents still have the same worries, and that I receive the same questions today as I did ten years ago.

When I was given the opportunity to take on this PhD project, I had some doubts about its necessity and whether it would be of interest to anyone else outside craniofacial teams, since the group of patients and their parents whom I am focusing on is relatively small. But over time, I have concluded: Why shouldn't we make an effort to understand the parents' experiences simply because their child belongs to a group with an uncommon diagnosis? The knowledge we can gain is no less valuable just because it is useful to a smaller group of parents. Perhaps research on this group of parents can bring attention to other groups of parents whose children have uncommon diagnoses and whom may not receive as much visibility.

I also believe that parents of children who need surgery often share similar concerns and uncertainties regardless of their child's diagnosis. If this project can shed light on and provide an understanding of the experiences of parents of children with craniosynostosis, it may improve care not only for this specific group, but also for others navigating hospital care with a child in need of surgery.

This thesis could not have been completed without the generous contribution of all the parents who shared their stories. I am incredibly grateful to them all.

Introduction

Craniosynostosis

Craniosynostosis is a congenital malformation occurring when the suture(s) between two or more of the cranial bones fuse prematurely (I. M. J. Mathijssen, 2021). This results in abnormal head shape and growth and a risk of developing raised intracranial pressure (Dias et al., 2020; Ursitti et al., 2011). The incidence of craniosynostosis and craniosynostosis syndromes in Sweden is 7.7 cases per 10,000 live births (Tarnow et al., 2022), which means approximately 80-100 children per year.

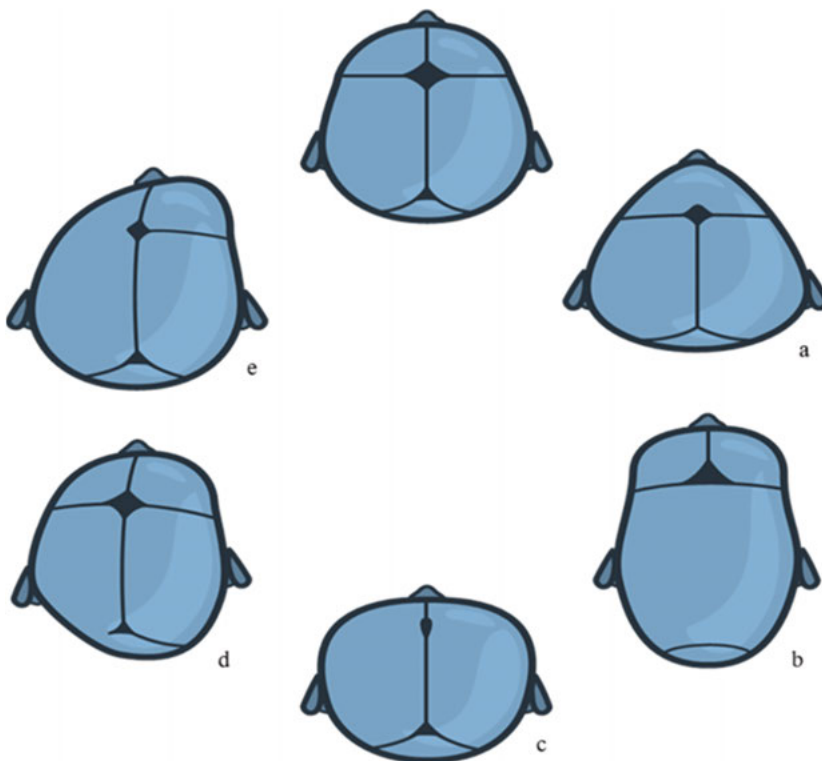


Figure 1. Shapes of the skull due to different types of craniosynostosis (the normally shaped skull with open sutures, at the top center), a) metopic, b) sagittal, c) bicoronal, d) lambdoid, e) unicoronal. *Illustrations by Lina Cabal Romero.*

A majority of craniosynostosis cases involve only one fused suture (Dempsey et al., 2019), the diagnosis is usually apparent after birth or within a few months. Single suture craniosynostosis produces a predictable head shape, can usually be diagnosed on clinical examination of the child (Dias et al., 2020) and be confirmed with computer tomography (CT) scans (Ursitti et al., 2011) or ultrasonography (Rozovsky et al., 2016). The most common forms of non-syndromic, single-suture craniosynostosis are (in order of prevalence): Sagittal craniosynostosis, which results in an elongated head shape with frontal bossing, parietal narrowing, and occipital bossing (scaphocephaly); metopic craniosynostosis, which leads to a triangular head shape with narrow-set eyes and a keel-shaped forehead (trigonocephaly); unicoronal craniosynostosis, which gives a trapezoidal head shape, flattening the forehead with raised eyebrow and the nasal bone deviated to the affected side and temporal bossing on the unaffected side (anterior plagiocephaly); unilateral lambdoid craniosynostosis, which results in a trapezoidal head shape, with the ear on the affected side placed lower, more posterior, and compensatory growth on top of the head on the unaffected side (posterior plagiocephaly) (Dias et al., 2020; I. M. J. Mathijssen, 2021). Bicoronal craniosynostosis accounts for a small percentage of non-syndromic, and most syndromic craniosynostoses, and results in a shortened skull with a flat, tall, and wide forehead (brachycephaly) (Boulet, Rasmussen, & Honein, 2008). Non-syndromic craniosynostosis is coded Q75.0 according to ICD-10, and craniofacial syndromes with craniosynostosis are coded Q75.1 or Q87.0.

Treatment

The treatment for craniosynostosis is almost always surgery, which is performed to prevent harmful effects on the brain, correct deformities, and allow for normal brain growth. One transcranial operation, preferably performed during the first year of life, is sufficient for most children with single-suture craniosynostosis (Warren et al., 2012). In the case of a metopic bone ridge and mild trigonocephaly, spontaneous improvement can be expected with growth, and only the most distinct form of metopic craniosynostosis has an undisputed indication for surgery (I. M. J. Mathijssen, 2021). As a result, there is an increasing discussion about the criteria for surgery and a trend towards operating on fewer patients based on stricter indications.

Referral to the specialist team

Treatment of patients with craniosynostosis requires a comprehensive approach and should be provided in a multidisciplinary setting at craniofacial centres (I. M. J. Mathijssen, 2021). Early referrals are encouraged to allow

early identification and surgical intervention (Dias et al., 2020). Multidisciplinary craniofacial teams comprise various specialists, each bringing their own expertise. Most craniofacial teams have a care coordinator who acts as a liaison between the patient and the treating team (Buchanan, Xue, Xue, Olshinka, & Lam, 2017). The care coordinator, usually a nursing specialist, is responsible for coordinating care, acts as the main point of contact for patients, parents, and external co-treatment providers (I. M. J. Mathijssen, 2021) and provides the family with a single point of contact to coordinate all their care needs (Buchanan et al., 2017).

Since 2012, Sweden has centralized craniofacial surgical care to two licensed national Craniofacial Centres: Uppsala University Hospital and Sahlgrenska University Hospital in Gothenburg. The licensed Centres are monitored by yearly data reporting, to the National Board of Health and Welfare, on surgically treated craniosynostosis and craniofacial syndromes in Sweden. The two national craniofacial units register diagnostic, surgical, and follow-up data in a national quality register. The register aims to ensure and improve care for children with craniosynostosis and craniofacial syndromes and has been produced by the teams at Sahlgrenska University Hospital and Uppsala University Hospital in collaboration



Figure 2. Meeting with the craniofacial team. *Photograph by Ola Lundström*

Care programme

Of the 80-100 children born with craniosynostosis annually in Sweden, 30-40 are treated at Uppsala University Hospital. The following paragraph describing the care pathway applies to families treated in Uppsala.

At the first appointment, the family meets with members of the craniofacial team, typically a plastic surgeon, a neurosurgeon, a clinical nurse coordinator, a psychologist, and an ophthalmologist. During this meeting, the family receives information about the diagnosis and the upcoming surgery. The surgery is planned at the most favourable time depending on which suture is closed and the age of the child at diagnosis.

At the time of surgery, the child and parent(s) are admitted to a paediatric ward. During the day of enrolment, the child is examined by a paediatrician, an anaesthetist, and a clinical geneticist. The surgical team repeats information about the surgical procedure. The family is shown around the children's ward and receives information about the ward's routines.

The surgery is performed in the neurosurgical operation unit; it usually takes between one and three hours, depending on the specific procedure. Including preparation time, parents and the child may be separated for up to six hours before reuniting in the intensive care unit (ICU). After the surgery, the child and parents stay in the ICU for approximately 24 hours. Afterward, the family returns to the paediatric ward for further postoperative care. The length of a hospital stay ranges from four to seven days, depending on the type of surgical procedure performed. Upon discharge, families receive verbal and written care instructions concerning the surgical wound and how to recognize signs of infection.

The first follow-up appointment is scheduled one month after the surgery. This appointment can be held as an on-site or a video meeting with the clinical nurse coordinator. During this meeting, the nurse coordinator assesses wound healing, provides advice on continued wound care if necessary, and addresses any questions about the surgery, the treatment in the paediatric ward, and future planning. On this occasion, complications that may have occurred within 30 days are registered in the national quality register.

After this initial follow-up, the child will be followed by the craniofacial team with meetings scheduled at specific time points: One year after the surgery, at three, five, and eight years of age. In addition, the child is followed up at their local hospital with ophthalmological examination at set times, as well as a follow-up CT at the age of three years. An extra appointment with the craniofacial team is offered for children born with unicoronal craniosynostosis when they reach the age of 16.

Background

Parents of children with a condition that requires surgery

Parent and caregiver

In Sweden, a caregiver is a legal term and could refer to one or both parents or a person appointed by the court. In many contexts, such as in previous studies, the concepts of parent and caregiver are not defined. In this thesis and the included studies, the term parent is used to refer to all caregivers.

A condition that requires surgery

Being a parent of a child with a diagnosis or an illness that requires surgery can lead to anxiety and emotional distress (Charana et al., 2018; Hinton, Locock, Long, & Knight, 2018; Pomicino, Maccacari, & Buchini, 2018). Diagnosis with a chronic condition can cause parents to feel overwhelmed by the new situation, and support from healthcare professionals can be essential in handling emotional reactions as well as practical tasks (Iversen, Graue, Haugstvedt, & Råheim, 2018). The unexpected experience of having a young child undergo surgery is added to the transition of becoming a parent, and when parents face their child's need for surgery, the entire family goes through a stressful time (Sjostrom-Strand & Terp, 2019). Worries about the child's treatments and prospects as well as long-term consequences are common (Rosenberg, Kapp-Simon, Starr, Cradock, & Speltz, 2011).

Being in the hospital

Every child has the right to be accompanied by parents or significant others during hospitalization. The family's presence has been seen to reduce stress and anxiety in the child and improve their state of health during the hospital stay (Coyne, O'Neill, Murphy, Costello, & O'Shea, 2011). The surgical period in an unfamiliar hospital environment can be challenging in many respects for both paediatric patients and their parents (Gabriel et al., 2018), and parents have reported concerns about potential complications, anaesthesia-related side effects, and surgical outcomes (Hui et al., 2020).

When a child is admitted to a paediatric ICU, parents experience a wide range of emotions (Dahav & Sjöström-Strand, 2018), and parents have reported carrying with them vivid memories and being affected by the experience years later (Terp & Sjöström-Strand, 2017). Parents who experience anxiety, stress, and exhaustion during their child's hospitalization and surgery may further distress their child (Gabriel et al., 2018), and parental anxiety levels have also been found to correlate with levels of pre- and postoperative pain in children (Yayan, Zengin, Düken, & Suna Dağ, 2020). Parents need to be prepared for the perioperative care so that they can best support their child (Chorney & Kain, 2010; Chorney, Tan, & Kain, 2013).

Participating in care

The term parental participation refers to actively including the parents in all caregiving activities (Hill, Knafl, & Santacroce, 2018). It aims to keep parents and children together and to enable parents to play an active role in their child's care (Aarthun, Øymar, & Akerjordet, 2019; Coyne, Amory, Kiernan, & Gibson, 2014). Additionally, it is a fundamental aspect of modern paediatric nursing. Families in healthcare should receive accurate information about the child's condition, and parents should have the right to participate in decisions concerning the child's care and treatment. By being involved, parents can feel they are being helpful and contributing (Coyne et al., 2011), and their participation in their child's healthcare decisions can impact their ability to cope with the parental role at the hospital (Aarthun et al., 2019).

Parents have reported feeling confused about the expectations of healthcare professionals, which can lead to trust issues and dissatisfaction (Romaniuk, O'Mara, & Akhtar-Danesh, 2014). They seek guidance on how to participate in their child's care and hope that the staff will recognize their expertise as parents when assessing the child's needs (Ames, Rennick, & Baillargeon, 2011). To optimize hospital care for children, clinical practices regarding parental involvement need to be established (Gustavsson, Gremyr, & Kenne Sarenmalm, 2016). Healthcare professionals must help parents feel welcome in the hospital environment and assist them in maintaining their parental role (Galvin et al., 2000). The fact that parents' needs for information and support may vary during hospitalization should be considered (Kirk, Fallon, Fraser, Robinson, & Vassallo, 2015).

Coming home from the hospital

After being discharged from the hospital, the transition to the home environment can be difficult. Some parents are overwhelmed by feelings of uncertainty (Simeone et al., 2018), they may worry about their infant's safety, not knowing what to do if something happens, and not having anyone to contact for advice. This can affect parents' ability to adjust and adapt to caring for their

fragile child at home (Gaskin, 2018), and many parents have difficulty managing the instructions they receive when their child is discharged from the hospital (Glick et al., 2017). Parents need to be provided with information that prepares them for discharge and informs them about who to contact when they have questions regarding their child's care (Galvin et al., 2000).

Parents' satisfaction with hospital care

The concept of quality in healthcare can be defined in various ways. While many quality dimensions concentrate on the contributions of healthcare staff, quality is also related to the value perceived by patients. Parental satisfaction has been used successfully to measure the quality of paediatric care, as it is closely tied to the adequacy of children's treatment and the performance of the staff (Bradley, 2013; Tsironi & Koulierakis, 2019).

Parents generally express high levels of satisfaction with paediatric inpatient care, although there is a need to establish clinical practices for parental involvement to improve hospital care for children (Matziou et al., 2011). Collaboration among healthcare professionals and involvement of parents in the care process emerge as important factors for ensuring parental satisfaction with the care provided (Matziou et al., 2011). Additionally, lower parent anxiety and higher social functioning in children are reported as predictors of higher parental satisfaction (Shafer et al., 2018).

Parents of children with craniofacial conditions

Psychological distress

Having a child who receives a diagnosis of a congenital craniofacial condition can have a significant impact on the psychological and social functioning as well as the well-being of the family unit as a whole (I. M. J. Mathijssen, 2021; Nelson, Kirk, Caress, & Glenny, 2012). Knowledge about the condition can sometimes evoke feelings of disappointment and sadness (Pope, Tillman, & Snyder, 2005), and parents have reported complex emotional responses, including anger, shock, grief, guilt, and worry (Nelson et al., 2012). Parents have described feelings of powerlessness and the weight of being responsible for their child's care, but also reported that their perspective on life had changed, characterized by growing self-confidence and an increased capacity for empathy for others as a result of their experiences (Feragen, Stock, Myhre, & Due-Tønnessen, 2020). Parents of children with craniosynostosis sometimes experience significant anxiety before their child's surgery despite feeling well-informed (Kuta et al., 2020; Rosenberg et al., 2011). Parents' anxiety and emotional distress may be reduced if they can be helped to feel confident in their ability to support their child (McCarthy et al., 2012). Support from healthcare

professionals can contribute to improved psychological and social functioning for all family members (I. M. J. Mathijssen, 2021). Many parents report feelings of relief after surgery (Kuta et al., 2020), but also worry that their child will have neurological deficits or developmental problems during childhood (Kuta et al., 2020; Rosenberg et al., 2011).

Parental stress

Parental stress is related to stressors caused by the responsibilities that come with being in the parental role (Holly et al., 2019). It is a construct that includes both psychological and physiological factors related to the role as a parent or caretaker of a child. In previous research on parental stress related to an infant's initial diagnosis of craniosynostosis, some studies have shown that parents of children with craniosynostosis experience higher stress levels before surgery (Rosenberg et al., 2011). However, other research suggests that parents are not significantly affected by increased stress over time (Gray et al., 2015). Tang and colleagues found that caregivers of children with craniosynostosis experienced lower stress levels three months after the surgical interventions. However, this decrease was not sustained during the 6-month follow-up visits (Tang et al., 2022).

Parents' need for support

Support from an expert team and accessibility to information have been described as key to mitigating anxiety and stress among parents (Myhre, Agai, Dundas, & Feragen, 2019). Parents suggested written information and recommended internet sources to improve the possibility of reducing anxiety and worries. Internet-based peer support has become more common, making it possible to share parents' experiences in similar situations (I. M. Mathijssen, 2015; Sweeney et al., 2013). This has been reported as beneficial by parents of children with craniosynostosis (Kuta et al., 2020).

Satisfaction with craniofacial care

Parents of children with craniosynostosis have previously reported high satisfaction with interdisciplinary craniofacial care (Kluba et al., 2016; Kuta et al., 2020). Mothers in the US reported satisfaction with craniosynostosis-related information and medical care support, and access to treatment and care was also rated as satisfactory (Wong-Gibbons et al., 2009).

Theoretical framework

A theory applicable to this thesis is Swanson's theory of caring, which was developed from empirical phenomenological studies (Swanson, 1991). The theory aims at helping nursing personnel deliver care that promotes dignity, respect, and empowerment. The theory has previously often been used in research as a reference and, to a lesser extent, used as a connecting theory in practice (Andershed & Olsson, 2009). However, in studies of parents' experiences of hospital care, the theory has been used as a guiding theory for the analysis and to suggest caring interventions (Enskär, Darcy, Björk, Knutsson, & Huus, 2020; Roscigno, 2016; Wei, Roscigno, & Swanson, 2017). Swanson's theory encompasses five caring processes that, when applied to nursing practice, could influence the caregiver's attitude and improve the ability to meet the caring needs of parents. This model was developed to ensure consistent caring behaviours, ultimately leading to improved satisfaction with care. The five caring processes in Swanson's caring theory are:

Maintaining Belief. Having faith in parents' capacity and trusting them to care for their child, thus fostering hope and optimism. Maintaining belief involves holding others in esteem and having faith in their ability to achieve their goals. It involves accepting others with respect and, importantly, with a hopeful attitude. At the same time, the nurse also helps individuals regain a positive perspective on their experiences. The foundation of caring starts with a basic belief in people and their ability to navigate through challenges and changes and to approach their future with meaning.

Knowing. Care providers need to seek to understand parents and actively listen if they are to understand parents' experiences and needs, taking time to learn about their cultural background, personal preferences, and values. This information helps in addressing their unique needs and desires. When the process of knowing occurs, a bond of empathy and understanding develops between the care provider and the care recipient. Knowing includes having a humanistic view of the parent, understanding their situation, showing compassion and empathy, using academic knowledge, and communication skills, showing respect for individual differences, and recognizing the parent as a significant individual.

Being With. It is important to accompany parents physically and emotionally by offering emotional support, comfort, and companionship by staying present, offering reassurance, and providing words of comfort. The qualities of being with include mutual trust, availability, faithfulness, patience, and compliance. Being emotionally present involves calming fears through the action of being present. Being with, as well as being emotionally present, conveys the message that the parents and their experiences are important to the nurse.

Doing For. Doing for means helping parents by providing physical care and assistance to meet their needs. The unique function of a nurse is to assist

the individual, whether sick or well, in the performance of those activities that contribute to health or recovery and to do this in such a way as to help them gain independence as rapidly as possible. This includes comforting them, anticipating their needs, performing procedures skilfully, protecting them from harm, and ultimately preserving their human dignity. Doing for refers to the activities a nurse engages in with parents, to help them do what they normally would themselves but what they cannot do at present.

Enabling. Supporting parents to be the best parents they can be by empowering them to engage in care and decision-making, promoting independence. An important part of enabling is regular and empathetic communication with patients and their families qualified by sensitivity to family dynamics, cultural and religious beliefs, and previous experience. Communication also includes providing information and explanations about the given care as well as the overall condition of the patient. Written materials, phone calls, and counseling could also be encouraged in the process of enabling the parents. Here, parents are partners who have knowledge and self-management skills.

Rationale for the studies

This thesis is part of a larger multidisciplinary research programme concerning different factors of importance for the course of craniosynostosis and the result of current treatment.

When a small child is going through skull surgery, it often leads to worries and anxiety for the parents. In craniosynostosis surgery the risk of more severe complications is low, but serious complications that can lead to permanent damage do occur. It is also important to acknowledge the fact that the indication for surgery is not lifesaving. The risks associated with surgery and anaesthesia as well as the parents' own stress and psychological distress do affect their experiences and their satisfaction with the care provided.

To further develop and improve care for children with craniosynostosis and their parents, it is essential to assess and evaluate the current practices and to identify what parents require to feel safe and satisfied with their care experience. Parents' stress, psychological distress, and health-related quality of life when experiencing a beloved child going through craniosynostosis surgery have not been investigated comprehensively. Parents' opinions about the care provided deserve more attention from health professionals. Their opinions can be used to optimize the treatment, but also as an instrument for quality control.

Aims

The general aim of this thesis was to explore parents' experiences when their child is diagnosed with and treated for non-syndromic craniosynostosis, to investigate how parents perceived the received information, treatment and participation in care, as well as their parental stress, symptoms of psychological distress and health-related quality of life.

The specific aim of each study is listed below.

Study I

The aim of the study was to investigate Swedish parents' experiences of having a child with craniosynostosis and their perceptions of the care provided early in the diagnosis process as well as throughout the treatment process.

Study II

The aim of the study was to explore parents' experiences of the time at the hospital and the year after discharge, and their perceived support needs after their child's craniosynostosis surgery.

Study III

The aim of the study was to explore how satisfied parents of children with craniosynostosis were with hospital care at the time of their child's craniosynostosis surgery and to identify factors that influenced parents' perception of quality of care.

Study IV

The aim of the study was to assess parents' perceived parental and psychological stress and health-related quality of life before their child's craniosynostosis surgery and one year after surgery.

Methods

Table 1. Study design of the four studies

Paper	Design	Participants	Data Collection	Analysis
I	Descriptive Qualitative	Parents of children with craniosynostosis (n=20)	Semi-structured telephone interview, one month after surgery	Thematic analysis
II	Descriptive Qualitative	Parents of children with craniosynostosis (n=19)	Semi-structured telephone interview, one year after surgery	Content analysis
III	Mixed Method	Parents of children with craniosynostosis Interview (n=19) Questionnaire (n=98)	Semi-structured telephone-interview Questionnaire	Content analysis Descriptive statistics
IV	Descriptive Quantitative	Parents of children with craniosynostosis (n=29)	Questionnaires before and one year after surgery	Nonparametric statistics

Setting

All four studies were conducted at Uppsala Craniofacial Centre, where, each year, 30-40 children with craniosynostosis receive care. In these studies, all participating families have children diagnosed with non-syndromic craniosynostosis (Q75.0). Patients are typically referred to the multidisciplinary craniosynostosis team by local paediatricians upon suspicion of craniosynostosis. Surgery is preferably performed during the first year of life, but the timing depends on the type of craniosynostosis and the age at which the patient is referred. All primary and outpatient clinical visits take place in the plastic surgery outpatient clinic and during hospitalization, the child is treated at the paediatric ward for neurology, orthopaedics, surgery, and urology. Following surgery, the child spends about 24 hours in either the neurosurgical ICU or the paediatric ICU, before returning to the paediatric ward for further postoperative care. The healthcare professionals at the wards include paediatricians, physicians with various competencies, specialized nurses (registered

generalist nurses with a 1-year master-level education in childcare), RNs (generalist nurses with a bachelor's degree), and nursing assistants.

Participants

Study I

The participants were parents of children with non-syndromic craniosynostosis scheduled for surgery at the Uppsala Craniofacial Centre. Twenty participants were consecutively recruited at the first appointment with the craniofacial team. They were given written and verbal information about the study and asked to be interviewed separately. All the invited parents agreed to participate; their ages ranged from 27 to 43 years (mean 35).

Study II

A purposive sample was used, and participants were parents of children with non-syndromic craniosynostosis who had undergone surgery at the Uppsala Craniofacial Centre. From August 2016 to October 2016, seven children had undergone surgery, and parents of these children were invited by post to participate in the study. Six mothers and three fathers of the seven children agreed to participate. In addition, we included parents who had participated in Study I; from that cohort, parents were contacted consecutively, by phone, 12 months after surgery. In all, 19 parents, 11 mothers, and eight fathers agreed to participate; age ranged from 26 to 40 years (mean 35).

Study III

All families of children who had surgery for non-syndromic craniosynostosis from January 2018 to August 2022 (n=146) received a questionnaire by regular mail between one and four weeks after the child's surgery. In addition, interviews from Study I with 20 parents were used.

Study IV

This longitudinal study included parents of children diagnosed with non-syndromic craniosynostosis who were scheduled to undergo surgery at the Uppsala Craniofacial Centre. Data collection took place between May 2016 and February 2024. During that period, a total of 243 patients underwent surgery, among these patients 146 met the inclusion criteria. Out of the eligible patients, 96 (66%) chose to participate, and 29 (30%) provided data at both time-points.

Data Collection

Study I

The interviews were conducted over the telephone approximately one month after the child's surgery, from June 2017 to January 2018. An interview guide was used that included open-ended questions about the parents' experiences during the interval between the suspected diagnosis and the time of surgery. The interviewer was a social worker not connected to the craniofacial team involved in treatment. Socio-demographic information was collected prior to the research interview. One of the participants was interviewed twice due to a technical failure during the first interview. All interviews were digitally recorded and transcribed verbatim.

Study II

All interviews, except two, were conducted by phone, approximately one year after the child's surgery, from September 2017 to August 2018. Two interviews took place at the 1-year follow-up with the craniofacial team based on participants' requests. The same interviewer as in Study I conducted the interviews. An interview guide was used that included open-ended questions about the parents' experiences and feelings regarding support needs and information, both in the postoperative period at the hospital and after discharge. Prior to the interview, socio-demographic information was collected. The interviews were recorded digitally and transcribed verbatim.

Study III

The questionnaire

The study used The Swedish Pyramid Questionnaire for Treatment (Ygge & Arnetz, 2001) to assess parental satisfaction with quality of care. Twenty-five items were selected, inspired by Willebrand and colleagues (Willebrand, Sjöberg, Huss, & Sveen, 2018). The first 24 items had four response options ("yes, to a great degree", "yes to a certain degree", "no, not especially", and "no, not at all"), and these items were grouped into six quality domains: *Information Illness*, *Information Routines*, *Medical Treatment*, *Processes*, *Staff Attitudes*, *Participation*. In addition, the questionnaire included a visual analogue scale (VAS) question, where participants were asked to rate their overall perception of the care on a scale from 1 to 10 (very negative to very positive). The questionnaire also provided a free-text area for comments and suggestions for improvement. The questionnaire was sent by post, completed anonymously, and did not request personal information.

Interviews

The interviews were conducted by phone from June 2017 to January 2018 (see Study I for detailed information). The interviewer used an interview guide that included open-ended questions about factors that influenced parents' perception of quality of hospital care.

Study IV

The parents received information about the study during their first appointment to the Craniofacial Centre. Written information and pre-stamped reply envelopes were given to the parents along with the first round of questionnaires. The follow-up questionnaires were mailed to the patient's home one year after the surgery and a reminder by phone was made a few weeks later. Those who failed to respond received an additional package of questionnaires by regular mail. At the start of the study, the participants were asked to complete the questionnaires digitally by logging on to a secure website. However, due to software problems, some parents were unable to complete the questionnaires. Data from 15 of the 96 families were collected from the website, and questionnaires were henceforth sent as hard copies via regular mail.

Parental Stress

Parental stress was assessed using The Swedish Parenthood Stress Questionnaire (SPSQ) (Ostberg, Hagekull, & Wettergren, 1997) based on the Parenting Stress Index (PSI) (Abidin, 1990), which focuses on parents' experience of being a parent, with no reference to specific child behaviours. The questionnaire (SPSQ) consists of 34 items within five subscales: *incompetence regarding parenthood*; *restriction of roles*; *social isolation*; *spousal relationship strain* and *parents' physical health*. Parents responded on a 5-point Likert scale indicating to what extent they agree or disagree with the statements, from strongly disagree (1) to strongly agree (5), with a total possible score of 170. Higher scores indicate higher stress. The SPSQ has been validated in several studies (Lagerberg, Magnusson, & Sundelin, 2011; Ostberg et al., 1997) with Cronbach's alpha values for the total score of 0.89 and 0.90, respectively. In this study, the internal consistency (Cronbach's alpha) for the SPSQ total score was 0.89 before surgery and 0.93 one year after. The alpha value for the subscales ranges between 0.57 (physical health) to 0.83 before surgery and 0.68 (physical health) to 0.86 after one year.

Psychological distress

Psychological distress was assessed using the Hopkins Symptom Checklist (HSCL-25) (Derogatis, Lipman, Rickels, Uhlenhuth, & Covi, 1974), validated in a Swedish sample (Lundin, Hallgren, & Forsell, 2015). The self-report questionnaire encompasses 25 statements of symptoms of depression (15

items) and anxiety (10 items) in two subscales. Participants were asked to think of the past week and how bothersome the symptoms had been, reporting from “Not at all” (1) to “Extremely” (4). The subscale score was calculated by summing the item scores and dividing by the number of items. A mean score of ≥ 1.75 in either of the two subscales was used as a cut-off for clinical significance (Lundin et al., 2015). In this study, the internal consistency for the HSCL-25 total score was 0.92 before surgery and 0.96 one year after. The alpha value for the subscale symptoms of depression was 0.89 before surgery and 0.94 one year after, while the alpha for the subscale symptoms of anxiety was 0.77 before and 0.89 one year after the surgery.

Health-Related Quality of Life

Health-related quality of life was assessed using the EQ VAS from the EQ-5D-3L questionnaire (Rabin & de Charro, 2001). EQ VAS is a vertical 20-cm line graded from “worst possible health state” (0) to “best possible health state” (100), on which respondents are asked to mark their current state of health. The reference score used was from a Swedish study where participants scored “good health” and at the same time a mean EQ VAS of 83.4 (Burström et al., 2014).

Analysis

Study I

The thematic data analysis used was outlined by Braun and Clarke (Braun & Clarke, 2006). The transcripts were read multiple times to get an overall understanding of the material, and during this step, two of the authors looked for patterns of meaning and formed preliminary themes. In the next step, words were highlighted and sentences relevant to the research questions and codes were created. These codes were then categorized into preliminary themes. After reviewing and refining the themes, subthemes were identified and were then formulated as a final definition for each theme. Finally, all authors considered the material under each theme and named the themes. Last, illustrative citations for some of the themes were chosen to highlight the meaning of the themes.

Study II

The interviews were analysed using content analysis with an inductive approach, following the process described by Elo and Kyngäs (Elo & Kyngäs, 2008). The analysis commenced with two of the authors reading the transcripts several times to become familiar with the data and overview the material. The authors then discussed the contents and their understanding of the text in

relation to the aim. Keywords and phrases were identified from the transcripts, and open coding was conducted. The codes were compared to find commonalities and variations, and codes with similar meanings were grouped into categories to capture the parents' experiences (Elo & Kyngäs, 2008). Then the categories were divided into subcategories based on dissimilarities within the categories. The analysis continued until all categories and subcategories were considered clearly defined and distinct from one another. All authors discussed the categorization until consensus was achieved.

Study III

A mixed-methods approach was chosen with a convergent, parallel design, which involved collecting, analysing, and integrating both quantitative and qualitative data (Creswell & Plano Clark, 2017). This approach gives equal weight to both types of data and enhances the potential to triangulate findings. With this approach, the idea is to combine the strengths of quantitative methods, such as large sample sizes and the ability to generalize, with those of qualitative methods, such as enriched meaning from small sample sizes (Patton, 2004). The quantitative and qualitative data were collected and analysed separately.

Questionnaires

Questionnaire data were analysed using descriptive statistics. According to Ygge and Arnetz (Ygge & Arnetz, 2001), mean values were calculated for each domain, as well as for the overall level of satisfaction, and converted to a percentage, representing the maximum achievable domain score (from 0 to 100%). The free-text responses were coded, and codes with similar meanings were grouped into six categories (Elo & Kyngäs, 2008).

Interviews

The interviews were analysed using qualitative content analysis, using an inductive approach and following the steps described by Elo and Kyngäs (Elo & Kyngäs, 2008). Transcripts were read several times to get an overview of the material; parts of the text related to the study aim were highlighted, open coding was conducted; codes with similar meanings were grouped into categories (Elo & Kyngäs, 2008). The authors agreed on eight categories and two overarching themes.

Study IV

In some of the returned packages of questionnaires, entire questionnaires were missing or only partially filled out. To our knowledge, there is no recommendation for which imputation method is best applied for these questionnaires, therefore we chose to use mean imputation for single missing items in HSCL-

25 and SPSQ (4 and 2 questionnaires, respectively). The mean of the observed values for each variable was computed and the missing values for that item variable were imputed by this mean. Questionnaires with more than one missing item were excluded. All analyses were performed with the statistical package IBM SPSS Statistics (Version 28.0). Wilcoxon signed ranked test was used for differences between the two time points and Spearman rank correlation for associations between variables.

Ethical considerations

In the interview studies, the participants signed a consent form before the interviews were conducted. Participants were informed that participation was anonymous and voluntary and that they could withdraw their participation at any time without explanation or consequences. The interviews began with an assurance that the participant was aware the interview would be recorded. The participants were anonymized and given code numbers in the transcripts to protect their anonymity and encourage accurate reporting. Only the main author had access to the recorded interviews, and coded transcripts were used during the analysis and in discussions among the researchers.

In Study III, filling in the Swedish Pyramid Questionnaire for Treatment and sending it in an addressed and prepaid envelope was equated with consenting to participate. The questionnaires did not obtain any personal information or identification. In Study IV, all participants signed a consent form collected by the researchers.

All questionnaires, as well as recorded interviews, were stored in a locked cabinet at the clinic.

The studies were approved by the Ethical Review Board of Uppsala University (Reg. no. 2014/396/1).

Results

Study I

Six themes with subthemes were identified.

Table 2. Parents' experiences of having a child with craniosynostosis and their perceptions of the care provided early in the diagnosis and treatment process

<u>THEMES</u>					
Detection of the abnormal skull shape	Thoughts and feelings before the appointment with the craniofacial team	Appointment with the craniofacial team	Searching the internet and social media	Waiting for surgery	Suggestions for improvement
<u>SUBTHEMES</u>					
Detection and possible causes	Referral	Treatment and accessibility	Searching	Worries and thoughts	The care
Information and parents' previous knowledge	Worries and thoughts	Thoughts after the appointment	What was found	Support	Information
	Parents' previous knowledge			Handling emotions	How the information is communicated

It was either a physician or a parent who first suspected the child had craniosynostosis. Concerns were raised due to the shape of the skull or a ridge over one of the sutures. Information from primary caregivers varied and was not always accurate or complete. Parents were sometimes told that the skull was compressed after birth and that the malformation would resolve with time, they reported a lack of effort on the part of primary caregivers, as well as feeling they were not being listened to.

Most parents had no previous knowledge about craniosynostosis. There were complaints that the doctor diagnosed their child but did not inform them about the diagnosis, only referring them to the craniofacial team. Communication between the maternity ward and the childcare centre was found to be inadequate at times. Delayed referrals and incomplete information led to feelings of dissatisfaction and frustration. Information about the diagnosis was sometimes received from the primary care provider, but parents also described turning to the internet to find information about craniosynostosis. Others chose not to search the internet before obtaining information from the craniofacial team. Parents' concerns often stemmed from a lack of understanding about their child's diagnosis.

There were descriptions of a positive experience from their initial appointment with the craniofacial team. Parents were confident in the team's ability, and they highlighted the support and accessibility provided by the psychologist and team nurse as key factors for this positive experience. Some felt calm after the initial meeting with the team, while other parents' concerns were heightened.

After this first meeting, parents found it beneficial to search for information online, and they also used social media to connect with others who had similar experiences. During this time, concerns were raised about the surgery and the potential side effects of anaesthesia, as well as worries about the impact on their child's development. Parents expressed their need for support in preparing for what was to come, and they considered support from friends, relatives, the specialist team, or a psychologist from primary care to be essential.

Study II

Analysis of the interviews yielded six categories, with subcategories.

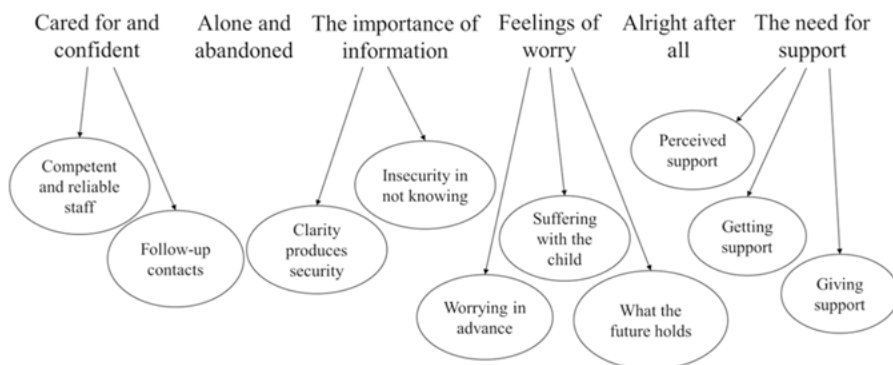


Figure 3. Parents' experiences of the time at the hospital, the year after discharge, and their perceived support needs after their child's craniosynostosis surgery

The parents thought the hospital stay was positive, and their experiences of the staff were good. Returning home was a relief for most. The parents appreciated having easy access to the craniofacial team or the paediatric ward after discharge, but there were descriptions of parents feeling reluctant to call. Follow-up appointments were appreciated, and a desire for more frequent visits during the first year after surgery was mentioned.

The time at the hospital was sometimes hard to cope with, and practical issues could intensify feelings of insecurity. Parents described the time spent in the ICU as the most difficult, mentioning feelings of abandonment in a room with unfamiliar equipment. After being discharged, some parents found it difficult to find the right support at the local hospital.

During hospitalization, parents expressed satisfaction with the information they received. They valued the well-established structure in the paediatric ward and the direct and honest answers provided by the physicians. However, it could be challenging not knowing exactly how long the surgery would take. At discharge, parents felt that staff were not fully informed, which delayed their journey home. Some would have wanted more information about the child's development as well as information about the risk of persistent deformity even after surgery.

During their child's hospital stay, parents stated that they felt surprisingly calm, and that expecting the worst in advance sometimes made the problems during hospitalization appear minor. The worst experience was described as feeling helpless while witnessing their child being sad and in pain. The first weeks after discharge could be challenging, as new worries emerged. Parents expressed concerns about their children's long-term development and potential complications, which sometimes led to overprotective behaviour.

The hospital stay in the paediatric ward was found to be good after all. Knowing that their child was not the first to go through this kind of surgery provided a sense of security. After discharge, everything gradually returned to normal, and there were descriptions of how the children's development accelerated after the surgery.

For families who could stay together at the hospital, their own family members were often considered the best source of support. Support from the craniofacial team and hospital staff was often appreciated, but parents mentioned that the support would have been of better help if it had been given before, rather than after, the surgery. An ongoing contact with the team psychologist after discharge was appreciated, but just calling the team nurse if needed was also described as sufficient.

Peer support, both in person and through social media, was said to be important, and seeking advice from others with similar experiences was considered highly valuable. The will to continue to support other parents of children with craniosynostosis on social media and online "parenting forums" after their own child had completed treatment was mentioned as a way to "pay it forward".

Study III

Satisfaction with hospital care

The questionnaire was completed and returned by 95 parents (65%). Parents rated the overall quality grade on a VAS scale, with a high mean value of 87% (SD 16) and range from 10-100%. Parents were generally most satisfied in the dimension related to *staff attitudes* and less satisfied in the dimensions *information routines* and *participation*.

Factors that influenced parents' perception of quality of care.

The analysis of the free-text comments yielded six categories: *Information and planning*; *Being disturbing*; *Deficiencies in care*; *Continuity in staffing*, *Staff competence and attitude*, and *Facilities*.

Comments described the information as incomplete and unnuanced, and some stated they would have appreciated more detailed information. Others were satisfied with the information provided. Parents wished they had received notice well in advance of any changes in the scheduled care. Some also wanted better routines around meals.

Some brought up that staff were stressed and mentioned being pushed aside by staff and feeling bothersome when asking questions. There were concerns about sharing a room, worrying about disturbing other patients.

Some comments described that staff lacked knowledge on certain occasions, leading to forgotten or delayed administration of pain relief, inadequately taped intravenous catheters, and insufficient food supplies for the baby. Some stated that the given psychological support was unsatisfactory, while other parents stated they had not received any support even though it was promised.

In their comments, parents mentioned wanting a “staff member of their own” who was responsible for their child's care and that rounds with the surgeons were appreciated during the hospital stay. Some comments also mentioned uninformed staff and unclarity about who was responsible.

The staff were also described as skilful and competent, and parents stated that staff members were supportive and caring. However, other comments described some staff members as careless or unpleasant.

There were descriptions of dissatisfaction with the facilities, such as unsatisfactory cleaning, an uncomfortable or no bed for parents, and insufficiently replenished patient rooms.

Analysis of the interviews yielded eight categories (in italics) and two overarching themes.

Theme 1. Factors that parents experienced as facilitating good quality of care.

Parents described the importance of *feeling supported by communication*, i.e., how being listened to by staff felt supportive. Parents expressed a need for more extensive dialogue with the staff and some wished to be accompanied by staff when they moved between examinations. Parents felt safe handing over their child to the anaesthetists before surgery, and the support offered by the anaesthetist and the psychologist were described as a most welcome resource in care.

Parents felt involved in care when they received a great deal of information, even if they recognized it was sometimes difficult to take all the information in. It was helpful to get regular updates and clear information about the child's condition. Parents described *being a team around the child* and that the staff cooperated with them. Parents wanted to be asked for their opinion, but not to be responsible for decision-making about their child's medical treatment.

Parents emphasized the importance of *continuity for both parent and child*; it was helpful for them to see a clear structure where staff were well-informed. Having available and approachable staff was important, and parents felt safe when they could discern good planning and a clear allocation of duties.

Parents reported feeling like they were *in good hands* and treated with kindness and support during hospitalization. Staff were described as professional, competent, and obviously used to this type of patient. Some parents also mentioned how efficiently they worked.

Theme 2. Factors that parents experienced as impeding good quality of care

There were descriptions of how *conflicting information and lack of dialogue left parents unsatisfied*. It was frustrating to be responsible for the child without knowing how they could contribute to the care. Parents reported feeling scolded by staff for not meeting their expectations, which left them feeling inadequate as caregivers.

In the paediatric ward, unclear planning and a *lack of routines made parents feel insecure*. The intense day of admission was described as stressful and tiring for the child. Parents sometimes felt that staff made unnecessary mistakes, and some even reported observing carelessness and neglect.

Parents noticed that there was a lack of consistency when different employees performed tasks in different ways. They felt that there were *too many people involved* and expressed the desire for staff to be more synchronized. Parents wanted to have the same staff day after day; some wished there had been a designated person who was responsible for their child. Some parents felt that the doctors only came sporadically, and it was difficult for them to keep track of who was responsible for what. With too many doctors involved, it was sometimes difficult to reach a decision, according to parents.

At times, parents perceived the staff to be uninterested and lacking in understanding for the parents' situation. The parents acknowledged that when the *staff are uninformed*, it is probably due to their high workload.

Study IV

Out of the 96 families agreeing to participate, there were 72 (75%) who returned fully or partially completed questionnaires before the surgery, and 57 (59%) one year after the surgery. The questionnaires were filled in by one of the parents or both together; 29 (30%) participants completed all three questionnaires at both time points.

Parental Stress

There was no difference in the total parental stress score before surgery and one year after surgery. In the analysis of each subscale of the SPSQ, the parents rated their feelings of incompetence higher one year after the surgery than before. The other subscales did not differ between the time points.

Psychological distress

There was no difference in the total score of HSCL-25 or the scores of the subscales of anxiety or depression before and one year after the surgery. Of the 29 participants who answered both before and after the surgery, symptoms of anxiety >1.75 were reported by 4 (14%) participants before surgery and 5 (17%) after surgery. Symptoms of depression ≥ 1.75 were reported by 3 (10%) participants both before and after surgery.

Health-Related Quality of Life

There was no difference in EQ VAS scores before and one year after surgery (Md 80, 90, $z = -1.38$, $p = 0.17$). Mean value at both time points, 79.0, is somewhat lower than the Swedish population value that was used as a reference (Burström, Johannesson, & Diderichsen, 2001).

Associations between SPSQ and HSCL-25

Before the surgery, the subscales in SPSQ that were associated with symptoms of depression were *incompetence*, *role restriction*, and *spousal relationship strain*. There were no associations between any subscales in SPSQ and symptoms of anxiety.

One year after the surgery, the subscales in SPSQ that were associated with symptoms of depression were *incompetence*, *role restriction*, and *social isolation*. There was also an association seen between symptoms of anxiety and the subscale *role restriction*.

Discussion

In the studies, parents shared their experiences of healthcare and reported estimated psychological distress in relation to having a child with craniosynostosis in need of surgery. The work follows the parents' journey: from detection of the abnormal head shape, through the diagnostic process, to the surgical procedure, and one year after the surgery. The results provide valuable knowledge when considering the maintenance or development of caring processes, both in outpatient clinics and hospital care.

The need for accurate information

Parents concern about a congenital malformation was typically prompted by an unusual head shape or a bone ridge over one of the cranial sutures. In some cases, parents were given alternative explanations for the shape in the maternity ward or the childcare centre. Some families were referred directly to the craniofacial centre, while others underwent preliminary diagnostic procedures before being referred. Most parents wished they have been given more information about craniosynostosis immediately after it was detected. The information provided by primary caregivers to parents about the suspected diagnosis varied and was unfortunately not always accurate. Paediatricians and general practitioners need to be able to identify abnormalities in head shape by combining their understanding of normal calvarial growth with a thorough physical examination (Cunningham & Heike, 2007). Uncertainty about the condition can lead to unnecessary or delayed referrals, inaccurate or incorrect diagnostics and inconsistent or incomplete information. These situations may cause stress and leave parents feeling frustrated and dissatisfied (I. M. Mathijssen, 2015). In a Dutch study, parents of children with craniosynostosis raised concerns about the lack of care and information in primary care (I. M. Mathijssen, 2015). On the other hand, studies from the US (Wong-Gibbons et al., 2009) and Germany (Kluba et al., 2016) have reported that parents were very satisfied with the information they received about craniosynostosis, as well as the support and encouragement given by healthcare professionals at the time of diagnosis. These differences may be due to varying levels of competence and procedures in different healthcare systems or regions. To help reduce uncertainty, ensure earlier referrals, and provide accurate information,

there is a need for additional education for caregivers involved in identifying craniosynostosis in Sweden. According to McCarthy and colleagues (McCarthy et al., 2012), families need to prepare themselves and their children for surgery and postsurgical care. Parental confidence in their own ability to provide care and support for their child will decrease emotional distress. However, the information has to be given in a way that is specially tailored to the individual (I. M. Mathijssen, 2015). Parents' need for information and support also changes over time and should be evaluated repeatedly (Kirk et al., 2015). We found that many parents wanted to fully understand their child's condition, surgery, and prognosis, while others intentionally avoided information due to fear of its potential negative impact. Both approaches may be coping strategies – one seeks to protect against unexpected surprises, and the other seeks to shield themselves from information they may not be ready for or capable of handling. As unique individuals, parents have varying information needs, which are not always understood by healthcare professionals (Hummelinck & Pollock, 2006). With an abundance of information readily available online, many parents come to the first appointment with the craniofacial team well-informed. However, it can be difficult for parents to determine which pieces of information they can trust. While accurate and complete information does exist, it may be challenging and time-consuming to find, as the ranking system on the internet does not necessarily prioritize information quality (Lloyd et al., 2016). As a result, important information related to the child's diagnosis could be overlooked. While much of the information available online is relevant and valuable, it may not be easily understood by non-professionals. Furthermore, craniosynostosis is treated differently across medical centres, meaning that information about surgeries and treatment methods may vary, leading to potential misunderstandings. Parents asked for easy-to-find websites with relevant and accurate information authored by specialist healthcare professionals.

When parents were left alone and uninformed during hospitalization, they described feeling insecure and abandoned, indicating that providing continuous information is essential to comforting parents (Kuta et al., 2020). Nevertheless, information can lose its effectiveness if it is provided at the wrong time or in excessive amounts (Hinton et al., 2018).

According to the questionnaire data, parents showed low satisfaction with the dimension *information routines*, which was further emphasized in the free text comments, particularly in the category of *information and planning*. Parents described experiencing a lack of routines in the ward. The interview data also supported these findings, as parents reported feeling dissatisfied and insecure due to unclear planning and a lack of routines. Parents have also expressed that they value receiving advance notice about any changes. They further described experiencing anxiety when they were uncertain about the expected duration of the surgery; a prolonged surgery could be misinterpreted as a sign that something had gone wrong. According to Kuta and colleagues (Kuta et al., 2020), parents found it difficult to hand over their child for

craniofacial surgery. However, they were comforted by receiving regular updates throughout the procedure.

At discharge, most parents were happy with the information provided, which is in accordance with previous studies on craniosynostosis (Kluba et al., 2016; Wong-Gibbons et al., 2009). However, parents mentioned that they would have liked more information about the long-term consequences and risks. It is challenging to find the perfect time to talk about risks, but based on the parents' feedback, these concerns should be addressed early in the care process. Some parents requested more information about the risk of abnormal head shape persisting even after surgery. It appears that these parents may not have fully understood the limitations of surgery. To prevent disappointment, it is crucial for surgeons to clearly explain the goals and limitations of surgical treatment (Kluba et al., 2016).

Most parents expressed satisfaction with the treatment, information, provided care, and hospitalization. Most described the meetings with the craniofacial team members and the staff in the paediatric ward as well as those at the ICU as satisfactory. This was further supported by the questionnaire in Study III, which found high overall parental satisfaction. This is consistent with previous research on parents' satisfaction with hospital care (Kruszecka-Krówka et al., 2019) and with care provision around craniosynostosis surgery (Kluba et al., 2016; Kuta et al., 2020).

Highly trained experts

It was considered beneficial that both the team nurse and psychologist were present when the child and parents had their first meeting with the craniofacial team. This observation is supported by studies showing that having easy access to a member of a craniofacial team – one who can handle not only coordination issues but also medical, psychosocial, and emotional concerns – is of considerable value (Burokas, 2013; I. M. Mathijssen, 2015; Sweeney et al., 2013). Overall, this emphasizes the importance of having an interdisciplinary team of specialists when developing care parameters for craniosynostosis (I. M. Mathijssen, 2015; McCarthy et al., 2012). Parents reported being concerned about the surgery, but it was not clear whether the parents expressed a clear preference for or against surgery for their child. However, other studies have shown that parents face a difficult decision when choosing whether or not to have surgery when their child is diagnosed with craniosynostosis, and that parents spend a great deal of time thinking about and gathering information prior to taking their decision (Letourneau, Neufeld, Drummond, & Barnfather, 2003). For example, some parents did not want their child to undergo early surgery because they did not think the cosmetic improvement was worth the risk (Ozgur et al., 2006). These studies are somewhat dated, and there may be a difference across countries and regions. Based on the studies in this

thesis, we found nothing to indicate that parents felt hesitant about surgery or that they opposed the surgeon's recommendation for surgery. Another reason for not discussing the pros and cons of surgery may be parents' confidence in healthcare and skilled professionals. Something that was important for parents was their trust that their child was being cared for by highly trained experts.

Participating in care

The goal of parental participation is to facilitate children and their parents being together throughout the care process, enabling parents to play an active role and be highly involved in providing care (Aarthun et al., 2019; Coyne et al., 2014). Research indicates that involving parents in the planning process and care provision can reduce anxiety and enhance parental satisfaction with healthcare (Çamur & Sarıkaya Karabudak, 2021). Parents need to be well-informed about their child's condition, and they value having comprehensive knowledge about the disease from various perspectives. When healthcare professionals fail to fully understand what parents find important, they may not be able to meet their needs, leading to parental insecurity (Melo, Ferreira, Lima, & Mello, 2014). It is important for healthcare professionals to consistently provide clear and sufficient information and to actively involve parents in decision-making. This approach aims to empower parents and increase their engagement (Aarthun et al., 2019).

Parents expressed a low level of satisfaction with participation; they described feelings of being in the way or disturbing staff when they wanted to ask questions. The interviews revealed that some parents felt that hospital staff scolded them for not behaving as expected during their child's hospitalization. Romaniuk and colleagues emphasized that, for parents in hospitals, it can be difficult to understand what healthcare professionals expect them to do (Romaniuk et al., 2014). However, in the interviews, parents also reported feeling involved in, or at least not being left out of, their child's care when they received a great deal of information. This is consistent with a study by Aarthun and colleagues (Aarthun et al., 2019), who stressed that parents need substantial amounts of information about their child's disease, health condition, and the healthcare system if they are to participate in decisions related to their child's healthcare. Parents felt they were part of their child's care team when staff cooperated with them and when the teamwork around the child was perceived as well-functioning. It is important that staff assume responsibility for building a trusting relationship (Wei et al., 2017), and that communication be ongoing throughout the child's hospitalization (Rennick, St-Sauveur, Knox, & Ruddy, 2019). Research has indicated that parents often want to be more involved than they can be, because healthcare professionals tend to be dominant in the decision-making process (Aarthun & Akerjordet, 2014). Several participants expressed a desire for both parents to be able to stay in the hospital with

their child during the entire period of hospitalization. They wanted not only to participate in the care, but also to support each other during the stressful time in the hospital. For parents, it is considered a rather simple practical arrangement to put an extra bed in the room (Lernevall, Moi, Cleary, Kornhaber, & Dreyer, 2020), but parents' requests were often denied with reference to the limited premises.

Some parents mentioned that they may have received more assistance at the paediatric ward if they had asked for it, but they found it difficult to initiate contact with the staff during hospitalization. The paediatric ward staff often have a heavy workload and may therefore seem stressed, which could have caused parents to be hesitant about asking for help, feeling like they were bothering the staff. It is important to recognize and address individual parents' needs during a child's hospitalization. Supporting parents may increase their participation in care while also reducing psychological distress (Jones, Nowacki, Greene, Traul, & Goldfarb, 2017). Parents also emphasized the importance of continuity in care. They requested a dedicated staff member responsible for their child's care. This suggests that parents expect the staff to be well-informed and prepared to help without having to ask for help.

Parents' perception of staff attitudes

The actions of healthcare professionals can either make it easier or harder for parents to navigate the care experience for their children. This suggests that caring for patients and their parents is just as important to well-being as are the clinical actions undertaken (Swanson, 1991). The results of these studies indicate that parents' views of staff members' attitudes influenced their opinions about the quality of care. Parents stated that the staff were caring, supportive and knowledgeable; they also emphasized the importance of being able to communicate with staff. Having a positive relationship with professionals may help to reduce stress during the hospital stay for both the child and family, and it can have a positive impact on communication (Tsironi & Koulierakis, 2019). A similar description was seen in a study from paediatric care, where parents reported having received too little kindness, attention, and time with the staff during their hospital stay. They wished the staff had supported them more by listening and being more available and present (Sarajarvi, Haapamaki, & Paavilainen, 2006).

Parents suggested that staff members appear stressed due to their heavy workload, and that they were not in sync with each other, resulting in conflicting information and tasks being performed differently. One can speculate whether the high workload is simply due to being understaffed or whether it is because of an inadequate, unreasonable or unclear distribution of tasks. Perhaps the nurses are spending too much time on illegitimate tasks that, in the long run, negatively affect their ability and motivation to perform high-quality

care. Ahlstedt and colleagues (Ahlstedt, Moberg, Brulin, & Nyberg, 2023) found an association between higher levels of illegitimate tasks and lower work engagement, fewer opportunities to provide high-quality care, lower employer satisfaction, and decreased intention to remain at the workplace. In a hospital ward, much of the daily work relies on cooperation, and many tasks do not have a clear designated performer. According to the present findings, parents appreciate it when staff have clear role definitions and when each task is carried out by a designated person.

Some parents mentioned that some of the staff seemed young and inexperienced and that they may not have received adequate training for their roles. As a result, they seemed stressed and did not perform tasks in the way the parents expected. When nurses are new to their jobs, it can be challenging to prioritize among the many tasks involved in nursing and care, as many of the tasks are time-consuming before they become routine. In Swanson's theory of caring (1991), the process *doing for* refers to the activities that nurses perform to promote health and that parents would do themselves if they could and if they had the necessary knowledge. This includes performing procedures skilfully and ensuring the child's safety. Parents may perceive staff as untrained or uninformed due to brief introductions or variations in how tasks are performed by different staff members. While parents may not necessarily know the correct way of performing different tasks, they do notice when tasks are done differently, leading to a potential lack of trust.

Although the questionnaire data showed high ratings for satisfaction with staff attitudes, some parents' interview statements suggested that the staff were negligent, unjust, and even unpleasant. The contrasting feedback from the interviews highlights the fact that staff competence and attitudes are decisive factors in shaping parents' perception of the quality of hospital care. Previous research has shown that the attitudes, knowledge, and skills of medical and nursing staff significantly impact parents' satisfaction with paediatric hospital care (Xenodoxidou, Theodorou, Karagianni, Intas, & Platis, 2022).

Identifying needs

Healthcare professionals must be responsive to the kind of support parents need at different stages in the care process. Earlier research from paediatric intensive care has shown that targeted support should be provided to mitigate the adverse emotional and psychological effects parents may experience (Suleman, Evans, & Manning, 2019).

Parents were generally satisfied with the support offered, though some found it difficult to initiate contact with the professionals. There were suggestions that the craniofacial team should provide routine support from psychologists rather than only offering such support upon request. Parents in the stressful situation constituted by a beloved child's surgery may not be able to

identify their own support needs. Integrating psychological support into the care programme may make it easier for parents to accept help from healthcare professionals, as some may be hesitant about asking for psychological support because they feel it reflects on their mental health.

Having access to support from healthcare after discharge was important. Being able to easily get in contact with the craniofacial team or the paediatric ward was perceived as comforting and made parents feel well taken care of. It is important to encourage interaction between parents and caregivers, as some parents are worried about the risks during recovery and have concerns about potential additional health issues and their child's development. Parents found it reassuring to have contact information to a nurse in the craniofacial team or the paediatric ward, so they could easily get in touch if necessary. In previous interviews, parents emphasized the importance of having access to healthcare support after leaving the hospital and caring for the child during the postoperative period (Hinton et al., 2018). This is consistent with earlier research on craniofacial surgery, which has demonstrated that it is crucial to have easy access to a team member who can address medical, psychosocial, or emotional issues and coordinate care (Burokas, 2013; Kuta et al., 2020; I. M. Mathijssen, 2015).

Parents mentioned having received support from family and friends, which in past research has been found to be associated with less negative family impact and lower psychological stress (Baker, Owens, Stern, & Willmot, 2009). Some considered the support of peers to be the most valuable, such support often being received through internet-based social media and forums for parents of children with craniosynostosis. Interacting with others who have had similar experiences was considered soothing. In previous studies, parents have described support from other parents who have gone through similar situations as crucial to helping them cope with and comprehend their child's diagnosis (Hinton et al., 2018). Parents enjoy interacting with each other and receive practical and emotional support and information through online forums (Jacobs, Boyd, Brennan, Sinha, & Giuliani, 2016; Niela-Vilén, Axelin, Salanterä, & Melender, 2014), and internet-based peer support is described as a unique form of support for parents, not replacing but supplementing support from healthcare professionals (Niela-Vilén et al., 2014). It offers parents the opportunity to share their experiences with others in a similar situation (I. M. Mathijssen, 2015; Sweeney et al., 2013).

Parents also continued to be active in social media and forums for a long time after their child's surgery. This was described as a way of paying forward the valuable support they themselves had received. Kuta and colleagues (Kuta et al., 2020) found that, while parents acted as advocates for their own children in the preoperative period, they advocated for the craniosynostosis community at large in the postoperative period. A study from 2016, investigating peer support in groups and online forums, found that most participants would appreciate the active participation of healthcare practitioners in their group or forum (Jacobs et al., 2016).

Worries and concerns

As a parent, daily life with a young child can be tough and even more challenging when the child has a craniofacial condition that requires surgery at a young age. Several factors in addition to the surgery can influence parents' feelings and health. Parents of children with craniosynostosis often expressed worries stemming from a limited understanding of and uncertainty about the implications of the diagnosis. After the first appointment with the craniofacial team, parents expressed feeling calm and confident. Yet one group of parents described feeling equally or even more stressed, because the new knowledge made the current situation more real to them. Burokas (Burokas, 2013) suggested that meeting all the specialists in a craniofacial team and having to process large amounts of information can be overwhelming for parents

Despite the descriptions provided by parent in interviews of their feelings of anxiety and stress during the period before and around the child's surgery, there was no difference in the reporting of symptoms of depression and anxiety before and after surgery. The findings also indicated that, while some participants showed symptoms of depression and anxiety, only a few exceeded the recommended cut-off value for clinically relevant symptoms. These findings of low psychological stress are consistent with previous research on parents of children with craniofacial anomalies (Cloonan, Collett, Speltz, Anderka, & Werler, 2013; Gray et al., 2015; Rosenberg et al., 2011). On the contrary, in another study, parents of children with non-syndromic and syndromic craniosynostosis reported higher levels of stress and more symptoms of anxiety compared to the general population. They also reported more symptoms of depression, although the scores remained within the normal range according to a clinical cut-off point (Costa, Edwards, Wilkinson-Bell, & Stock, 2023).

Parents of children with craniosynostosis do not seem to experience higher levels of parental stress than Swedish parents in general (Ostberg et al., 1997), either before or after surgery. In other studies as well, parents' stress prior to craniofacial surgery has been reported to be relatively low (Rosenberg et al., 2011). This has been discussed as a possible result of the multidisciplinary, family-centred care approach, which can moderate the stressors associated with the diagnosis and treatment of craniosynostosis (Gray et al., 2015).

There appears to be an association between parental stress and symptoms of depression both before and one year after the surgery, and it is important to note that interviews have indicated that parents of children with non-syndromic craniosynostosis do worry about surgery and anaesthesia before the operation, which has been previously reported (Rosenberg et al., 2011; Zhang et al., 2018). Furthermore, Rosenberg and colleagues reported that mothers of infants with single-suture craniosynostosis expressed increased stress if the condition was more visible (Rosenberg et al., 2011).

Parents described feeling relieved immediately after their child's surgery, others expressed worries concerning their child's development and potential comorbid disorders linked to craniosynostosis upon returning home after surgery. In previous research, parents in focus group interviews voiced concerns about the potential negative impact of craniosynostosis on their child's development (I. M. Mathijssen, 2015). Some parents talked about feeling that they needed to protect their child from being injured during recovery, which had caused them to be overprotective. It appears that apprehension and distress concerning surgery and worries about the child's long-term prognosis are still evident despite the accessibility of information and support from the team.

The results indicate that there are areas for improvement for children with craniosynostosis. Swanson defined nursing as informed caring for the well-being of others and a nurturing way of relating to others towards whom one feels a personal sense of responsibility (Swanson, 1991). The structure of caring in Swanson's theory enlightens nursing caregivers as to the significance of caring. The findings in this thesis can be linked to Swanson's five caring processes, and this connection will be further clarified in the section on clinical implications.

Methodological considerations

A strength of this thesis was that qualitative and quantitative approaches were combined to explore and describe the experiences of parents of children with craniosynostosis. Another strength is the intentional selection of parents of children with non-syndromic craniosynostosis. This deliberate selection is important, as patients with craniosynostosis syndrome may differ from non-syndromic craniosynostosis patients in terms of treatment plan, type of surgery, length of hospitalization and most importantly, in terms of prognosis. This thesis also has limitations that need to be mentioned.

In Study I, the interview with each parent was conducted one month after their child's surgery. Thus, for some parents several months had transpired from detection of craniosynostosis to the date of the interview. In Study II, the interviews were held one year after the child's surgery, thus there is a risk that memories had faded or been altered. Some of the interviews were shorter than one would wish for in a qualitative interview study. Nonetheless, these interviews contained valuable information that contributed to the data collection. Parents' willingness to participate and the detailed stories they provided can be seen as reflecting their perception of the research topic's importance.

To establish methodological rigor, the concepts of trustworthiness from Lincoln and Guba (Lincoln & Guba, 1985) were applied: *credibility, dependability, confirmability, and transferability*. To strengthen credibility, the sampling was purposive in nature, designed to ensure the participation of both fathers and mothers with experiences of their child's surgery; moreover, the parents were interviewed at approximately the same time after the surgery. To further ensure credibility, researcher triangulation was used in the sense that two authors conducted the analysis together. The interviewer used an interview guide, was experienced in conducting interviews, and was not involved in treatment of the children; this was done to ensure dependability.

The results of the analysis must be considered in the light of my experiences as a clinical nurse coordinator in a craniofacial team. In all analysis of the interviews, my theoretical and clinical knowledge about this group of patients and their parents may have influenced my understanding of their statements.

To provide confirmability, the analysis was constantly discussed by the two authors performing the analysis. After creating categories and themes, all authors – with different expertise and knowledge as physicians and nurses in plastic surgery, child and adolescent psychiatry, and paediatric care –

discussed the results in light of the interviews. The aim of the studies was not to generalize the results, but to gain a deeper understanding of the parents' experiences. We sought to ensure transparency by clearly describing the context, participants, data collection, and analysis to enable transferability.

A relatively large number of individuals were involved in Study III. The questionnaire data comprise 65% completed surveys, which can be considered a reasonable response rate. The use of items and dimensions from a recently used questionnaire (Willebrand et al., 2018) without controlling for internal consistency is a limitation. Neither demographic data nor information about which of the parents filled out the questionnaire was collected, which reduces the generalizability of the results. Questionnaires are an important tool for gathering data quickly and inexpensively. However, when using questionnaires with fixed response options, we may not be able to fully understand why or how care might be lacking, according to the parents. The fact that half of the parents chose to provide free-text comments suggests they were actively engaged in the research topic.

In Study IV, the attrition rate was high, but only 30% of participants reported at both time points, which is unfortunate and limits the ability to make better comparisons. The participants were recruited from one of two national craniofacial centres in Sweden. Our findings are limited in representativity due to the small sample size and data being collected from only one centre. One of the strengths of the study is that measurements were taken before and one year after surgery using standardized questionnaires that have been partly compared to population values.

The Swedish Parenting Stress Questionnaire is a general measure of parenting stress in typical families, without specific considerations of the medical conditions or disorders affecting infants. SPSQ may therefore not fully address the challenges faced by parents of children with conditions requiring major surgery, such as craniosynostosis.

We must consider that the results of all four studies may have been different if the inclusion criteria had been different, as only parents who could fluently read and understand Swedish were invited to participate in Study I, II and IV and in the interviews in Study III.

Conclusions

- The findings indicate a lack of knowledge among caregivers who see children with craniosynostosis, leading to concern in parents as a result of incorrect or insufficient information and delayed referrals.
- Parents of children with craniosynostosis are generally satisfied with the care provided. There are factors that either facilitate or impede parents' perceptions of good quality of care.
- Parents emphasized the importance of information and support from a multidisciplinary team, but still expressed concerns about surgery and their infants' long-term prognosis, highlighting the need for increased parent-caregiver interaction.
- Parents appreciate seeing the same staff day after day, and they feel well cared for and safe when there is continuity in care.
- Parents' perceptions of the staff's attitudes influence their perception of quality of care.
- Parents whose children undergo surgery for craniosynostosis do not report higher levels of parental stress than Swedish parents in general, nor do the reports differ between those made before and one year after the surgery.
- There appears to be an association between parental stress and parental symptoms of depression both before and one year after the surgery. Meanwhile, an association between parental stress and anxiety is only seen one year after.

Clinical implications

Based on the results of the studies and Swanson's theory of caring (Swanson, 1991), certain measures have been identified. These measures should be taken to improve the care of children undergoing surgery for craniosynostosis. As a consequence, the measures could also increase parents' well-being and satisfaction during the hospital stay. In health and medical care, nurses are responsible for nursing and for ensuring good quality of care. This means that, although all professionals involved in craniofacial care need to know the parents of children with craniosynostosis and their individual care needs, most of the proposed measures are nursing measures.

Parents experience good quality of care when they feel they are understood, actively listened to, and when their input is acknowledged. The process of *Knowing* includes the ability to listen and understand the needs of the parents. It all starts with creating a relationship, and nurses need to, together with the parent, identify needs and at the same time prioritize and sort out what is possible and what is most important in a given situation.

For parents, their child's surgery is a traumatic event, even if the surgery is considered routine by professionals. Caregivers should keep this in mind when talking to parents and preparing them for the surgery. They should ensure that parents feel reassured about the surgeons' experience and competence and that they do not come across as indifferent or casual about their work.

There were parents who reported symptoms of depression, anxiety, and parental stress before, and one year after. The knowledge that there are parents who may require more support than routine information and follow-ups underlines the importance of identifying parents' individual needs.

If, in the process of *Knowing*, the nurse and the parents together have identified and prioritized individual needs, the process of *Being with* could partly be more anticipated. At the ward, parents' feelings of being abandoned in the room and sometimes also hesitating to initiate contact or ask for help can be avoided if this has been brought up in dialogue with the nurse. Parents valued continuity of staffing and continuous visits from the surgeons. Routine updates during the operation but also in the aftercare could be part of the care planning.

All professionals need to be aware that their attitudes and behaviour strongly influence the wellbeing of parents. It is crucial for parents to feel comfortable with professionals, especially in an environment where much is

unknown and when they are worrying about their child. Providing continuity in the care, such as allowing parents to meet with the same staff every day, implies that there can exist a relationship between the nurse and the parent, which is the main purpose of the process of *Being with*.

Parents feel valued when they are recognized as capable decision-makers and allowed to participate in their child's care. Swanson's process of *Enabling* is vital when developing care from parents participating in care to parents really being part of their child's care team. More transparency and detailed information can facilitate parents' ability to be involved in care. For example: Staff should invite parents to be part of the team and parents' wishes to be together as a family in the hospital must be respected. The routines in the hospital ward must be clear and transparent for parents, and the craniofacial team should provide searchable websites with accurate information.

Maintaining belief and helping parents to maintain their own capacity as parents is a process that sometimes is demanding when parents put all their trust in professional experts. To be confident that the child will be in good hands during critical procedures, such as anaesthesia and surgery, is essential for parents, and the results from the studies indicate that parents do have trust in the experts. Parents need to be provided with detailed and correct information, along with regular updates, to increase their likelihood of trusting that they can be capable parents.

The process *Doing for* is likely easier for nurses to interpret than the other of Swanson's processes, due to the common professional identity as a "doer". Still, there must be a balance between doing things for the parents and at the same time helping them maintain their trust in themselves as parents. It can sometimes be easier to list nursing interventions adapted to specific routines and guidelines instead of using processes such as *Knowing* and *Enabling*. In these processes, the nurse, together with the parents, identifies individual needs and enables parents to be team members in the care, in that way sorting out what parents need help with and what they can deal with on their own. Having the parents' needs and wishes documented makes it easier to prioritize and hopefully free up time, so that the nurse can be available in situations that cannot be foreseen – on occasions when parents need nurses to do for them what they would do for themselves if only they could or knew how.

Future directions

This thesis focused solely on non-syndromic craniosynostosis. There is a lack of longitudinal studies following the experiences of families with children affected by craniofacial syndromes over time. Conducting a study to explore these parents' experiences over many years of caring contacts and measuring their subjective reporting of parental stress and psychological distress could provide important insights into the experiences of this particularly vulnerable group of parents.

In this thesis, the results of the studies and Swanson's theory of caring were used to suggest certain interventions that should be considered in an effort to improve care for children going through craniosynostosis surgery and support their families. In future research, it would be interesting to investigate whether care can be structured so that it facilitates the five nursing processes and to observe how parents perceive nursing based on Swanson's theory. It may also be of interest to compare parents' and nurses' perspectives on caring for children with craniosynostosis, with a view to identifying similarities and differences in care processes.

Svensk sammanfattning

Icke-syndromal kraniosynostos är en medfödd missbildning som innebär att en eller flera av skallens tillväxtzoner sluts för tidigt vilket ger skallen en avvikande form. Behandlingen är en operation som oftast utförs under barnets första levnadsår. Operationen syftar till att normalisera huvudets form och minska risken för ett förhöjt intrakraniellt tryck på sikt. Barn med kraniosynostos behandlas med fördel av multiprofessionella team. Att vara förälder till ett barn med en diagnos som kräver operation och sjukhusvård kan leda till känslomässig stress. För föräldrar till barn med kraniosynostos innebär diagnosen ofta mycket oro, dels för kirurgi och anestesi och dels för eventuell påverkan på barnets utveckling och mående.

Det övergripande syftet med avhandlingen var att undersöka föräldrars upplevelser av att ha ett barn som diagnostiseras med och behandlas för icke-syndromal kraniosynostos i ung ålder, bemötande, information och delaktighet i vården. Ytterligare att mäta föräldrarnas egenskattade föräldrastress, symtom på ångest och depression samt hälsorelaterade livskvalitet. Avhandlingen består av fyra delarbeten där både kvalitativa och kvantitativa metoder har använts.

Studie I undersökte föräldrars upplevelser av att ha ett barn med kraniosynostos och deras uppfattningar om den vård som gavs initialt, detta genom intervjuer som analyserades med tematisk analys.

Studie II undersökte föräldrars upplevelser av vården på sjukhuset och året hemma efter utskrivning, genom intervjuer som analyserades med innehållsanalys.

Studie III undersökte föräldrars tillfredsställelse med sjukhusvården och faktorer som påverkade deras uppfattning om vårdkvalitet genom enkäter och intervjuer som analyserades med en mixad metod.

Studie IV bedömde föräldrars upplevda föräldrastress, symtom på depression och ångest samt hälsorelaterad livskvalitet vid tidpunkten för operation och ett år efter genom frågeformulär som analyserades med deskriptiv och jämförande statistik.

Resultaten visar att föräldrar till barn med icke-syndromal kraniosynostos, överlag är positiva till omhändertagandet under hela vårdförloppet; från diagnos, operation och eftervård till fortsatt vårdkontakt efter operationen. Det finns dock områden där föräldrarna har önskemål och förslag till förbättringar.

Det verkar finnas en kunskapsbrist hos vårdgivare i primärvården som träffar barn med misstänkt kraniosynostos vilket skapar oro hos föräldrarna på grund av otillräcklig information och, i vissa fall, försenad remittering. Föräldrarna hade sällan någon kunskap om kraniosynostos innan barnet fick sin diagnos och de uttryckte oro över operationen och sitt barns långsiktiga prognos. Detta trots att de ansåg sig ha blivit väl informerade av det kraniofaciala teamet, vilket talar för att det finns ett behov av utökad kontakt mellan föräldrar och vårdgivare.

Stöd uppskattas av föräldrarna, en del vill ha mycket professionellt stöd medan andra föredrar att få och stöd inom den egna familjen eller från andra föräldrar som varit i samma situation. Resultaten visar att vårdpersonal behöver vara medveten om den oro som ofta följer med en kraniosynostos-diagnos och ha beredskap för att anpassa information och stöd för den aktuella föräldern då behoven varierar, både mellan olika individer och under tiden från diagnos till utskrivning.

Föräldrar till barn med kraniosynostos är överlag nöjda med den vård som ges. Det finns faktorer som antingen underlättar eller försvårar föräldrars uppfattning av god kvalitet i vården. Föräldrar känner trygghet och upplever god vårdkvalitet när det är kontinuitet i personalen, när de får tillräcklig information och när de känner sig inbjudna att delta i vården av sitt barn. God kommunikation med personalen och tydliga rutiner ger också en positiv upplevelse av vården. Å andra sida är det stressande för föräldrar när de inte får veta om ändringar i planeringen i god tid, när de känner att de stör personalen med sina frågor och när personalen upplevs stressad. Otillräcklig och felaktig information, brist på rutiner och motstridig information är också faktorer som gör att föräldrarna upplever vårdkvaliteten som mindre god. Föräldrars uppfattning om personalens attityder påverkar även deras uppfattning om vårdens kvalitet.

Föräldrar vars barn opereras för kraniosynostos rapporterar inte högre nivåer av föräldrastress än svenska föräldrar i allmänhet. Resultaten visar inga skillnader gällande föräldrastress, hälsorelaterade livskvalitet och psykiska symtom före och ett år efter barnets operation. Det verkar finnas ett samband mellan föräldrastress och depressionssymtom både vid tidpunkten för operation och ett år efter, medan ett samband mellan föräldrastress och ångestsymtom ses först ett år efter operationen.

Utifrån studiens resultat och Swansons omvårdnadsteori (Swanson 1991) har vissa åtgärder identifierats, som bör vidtas för att förbättra vården av barn som genomgår operation för kraniosynostos. Dessa teoribaserade åtgärder skulle kunna öka föräldrarnas välbefinnande och nöjdhet med vården,

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