

ON PARENTAL
SELF-EFFICACY IN
FAMILIES WITH
PAEDIATRIC CATARACT
AND ITS CLINICAL
IMPLICATIONS

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Gothenburg, Sweden, 2020



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GOTHENBURG

Cover illustration: Gyllén family photo 2008.

On parental self-efficacy in families with paediatric cataract and its clinical implications

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ISBN 978-91-8009-046-9 (PRINT)

ISBN 978-91-8009-047-6 (PDF)

Printed by Stema Specialtryck AB, Borås, Sweden 2020



Till min ängel Allie och finaste Edith,
ni ger mening.

Love is.

ABSTRACT

Introduction: Parent participation is implicit in all paediatric care. Responsibility for the treatment after cataract surgery (contact lenses, patching, eye drops and being alert to signs of complications) mainly lies with parents. However, little is known about what parents go through and what kind of support they need from the ophthalmological team.

Aims: To understand (I-III), explore (I and IV) and explain (IV) self-efficacy among parents of a child with paediatric cataract in order to promote self-management and improve clinical care.

Patients and methods: Study I: questionnaires with open and closed questions were sent to parents of children (n=72) registered in the PECARE (Pediatric cataract register) and to paediatric ophthalmologists (n=30). In studies II and III, in-depth interviews with 23 parents were analysed using directed content analysis (II) and the grounded theory method (III). In Study IV, questionnaires were sent to all parents of children registered in the PECARE since 2006 (n=506). Four scales were selected: sense of coherence, fatigue, perceived social support and family self-efficacy, and on patient-reported experience measures (PREM).

Results: Studies I and IV revealed what prerequisites the parents have – or do not have. In Study I, they wanted something they did not get, i.e. more information, both written and online. In Study IV, fatigue emerged as an obstacle to the sense of coherence, especially among parents of children with bilateral cataract. Studies II and III provided an in-depth understanding of the parents' self-efficacy and what strategies they use to adapt to a life in balance with a child with cataract. A model was developed in Study III to describe the parents' situation.

Conclusion: Parents go through a profound adaptation process throughout their child's visual trajectory. As they are a self-evident part of the care of their child, the team needs to acknowledge that fatigue and uncertainty constitute a barrier to self-efficacy. This could be achieved by using a digital app, introducing a key worker and including other professions to the ophthalmological team in order to implement family centred care.

KEYWORDS

Paediatric cataract, self-efficacy, parents, family, caregiver, self-management

SAMMANFATTNING

Att bli förälder till ett barn med katarakt (grå starr) är en omvälvande och ibland chockartad upplevelse. Katarakt är vanligt bland äldre människor, där en operation gör att synen återställs, ofta helt. Barnkatarakt är den tredje vanligaste orsaken till blindhet i världen. I Sverige och övriga världen är det numera dessbättre en mycket ovanlig sjukdom; det drabbas ca 40 barn om året. Anledningen till detta är bland annat de nationella vaccinationsprogrammen. Sjukdomen kan finnas på ett öga eller båda. Ofta upptäcks den synhotande katarakten redan på BB, där barnläkaren inte kan se in till ögats bakre delar. Det beror på att katarakt gör att linsen blir grumlig (helt eller delvis) och hindrar insyn. Detta betyder att ögat inte får den stimulans av ljus som behövs för att barnet ska kunna utveckla seendet. Efter remiss till ögonkliniken, ser ögonläkaren hur mycket katarakt barnet har. Om linsen är lite eller delvis grumlig går det att avvakta eller helt avstå från operation. Regelbundna kontroller görs då för att följa om katarakten ändras. Om linsen är så grumlig på det nyfödda barnet att inget ljus kommer in i ögat, görs oftast operationen före 3 månaders ålder. Operationerna utförs antingen på Sahlgrenska Universitetssjukhuset i Göteborg eller St Eriks Ögonsjukhus i Stockholm, som innehar uppdraget för Högspecialiserad vård, utfärdat av Socialstyrelsen. Alla operationer registreras i PECARE (Pediatric cataract register), som är ett kvalitetsregister och som syftar till att optimera screening och effektivare behandling av barnkatarakt. Vid operationen tas den grumliga linsen bort och ersätts oftast med en lins av plast. Den inopererade linsen har styrkan som passar när barnet är äldre, vilket innebär att barnet behöver ha kontaktlins fram till dess för att brytningen ska bli optimal och barnet ska kunna se tydligt. Föräldrarna lär sig sätta i och ta ur linsen. Ibland behövs lappbehandling, då man sätter en lapp på det ögat som inte är opererat. Detta görs för att det opererade ögat ska stimuleras och utveckla aktiviteten mellan ögat och hjärnan.

Under de första åren krävs täta kontroller på ögonkliniken för att följa synutvecklingen, korrigera kontaktlinser och senare även glasögonstyrka och upptäcka komplikationer. Familjen träffar då barnögonteamet, som består av läkare, sjuksköterskor, ortoptister och optiker. Kontrollerna glesas ut men fortsätter regelbundet tills barnet är 10 år, men trycket i det opererade ögat bör

följas hela livet, åtminstone vart femte år. Det ligger ett stort ansvar hos föräldrarna för behandlingen av katarakt, som i allra högsta grad kan ses som medvårdgivare.

Self-efficacy innebär tilltron till sig själv att klara av en uppgift. För föräldrar till barn med katarakt kan det vara de krav som ställs på dem efter deras barns operation, t ex att klara av behandling med lapp, kontaktlinser och ögondroppar. För att uppnå optimal synförmåga krävs motiverade föräldrar som har basala kunskaper om syn och optik, där deras self-management är avgörande. Begreppet self-management inbegriper en persons förmåga att klara av symptom, behandlingar, psykosociala konsekvenser och förändring i livsstilen som hör samman med att leva med en sjukdom. Ansvaret är stort, förutom behandlingen efter operation måste föräldrarna även vara observanta på tecken på komplikationer och lotsa och stötta det växande och utvecklande barnet i sin omgivning (förskola, skola och med vänner).

Det övergripande syftet med denna avhandling var att förstå (I-III), utforska (I och IV) och förklara (IV) self-efficacy hos föräldrar till barn som har katarakt för att främja self-management och därigenom förbättra värden.

I studie I undersöktes hur föräldrar och ögonläkare ville få/ge information. Enkäter skickades ut till 72 föräldrar och 30 ögonläkare runt om i Sverige som tar hand om familjerna på hemorten. Studien visade att både föräldrar och ögonläkare såg det personliga mötet som det viktigaste, därefter information i skrift och på internet. Föräldrarna efterfrågade även mer och specifikt anpassad information, vilket speciellt gäller för dem som har de yngsta barnen (som behöver opereras inom 3 månader från födelsen). Förbättrad kommunikation med vårdteamet var också något föräldrarna efterfrågade, liksom kontakt med andra föräldrar i samma situation. Studien ledde till att en informationsbroschyr togs fram, både i pappersformat och tillgänglig på internet.

Studien gav svar på frågor men skapade även nya. Hur är det att vara förälder till ett barn med katarakt? Vad är det som är utmanande? Vad fungerar och vad fungerar inte i det dagliga livet? Vad vill föräldrarna ha från vårdteamet? Hur kan vårdteamet lotsa dem i behandlingen av sitt barn som har katarakt?

Intervjuer i studie II och III med 23 föräldrar gav ökad förståelse av hur det är att vara förälder till ett barn med katarakt; att vara i ovisshet från det att barnet får sin diagnos till dess att acceptans kan uppnås. Ovissheten berodde på olika saker, t ex orsaken till varför barnet fick katarakt, behandling (t ex fungerar lappträningen?), uppföljning (hur ofta ska barnet kontrolleras, vad är planen?),

prognos, framtiden och känslan av att göra rätt saker för sitt barn för hens utveckling. På andra sidan vågskålen från ovisshet låg acceptans, då föräldrarna tyckte att det mesta går bra, kanske inte tänkte så mycket på barnets funktionsnedsättning och vardagslivet gick ihop.

Att minska gapet mellan ovisshet och acceptans var en ständig balansgång och påverkades av föräldrarnas self-efficacy, dvs tilltron till sig själva. Det är en ständig process. Vid varje händelse/bakslag/operation upplevde föräldrarna ovisshet. Denna hanterade de genom att bemästra. Därefter såg de att ett samarbete är nödvändigt och de underlättade för barnet och accepterade det som var och anpassade det dagliga livet. Det är vårdteamets uppgift att lotsa och ge support till föräldrarna under processen.

Intervjuerna gav även förståelse för de faser som föräldrarna genomgår under sitt barns behandling. En modell beskriver faserna bemästra, samarbeta, underlätta och anpassa. Vid en kris (som t ex en reoperation) hamnade föräldern i faser bemästra och därefter insåg att de måste samarbeta (med vårdteam, skola mm). I nästa fas underlättade föräldern för barnet och i den sista faser skedde en anpassning till situationen. För föräldrarna var det en ständig balansgång mellan att barnets förmåga/oförmåga (framgång/motgång). Det är ögonteamets uppgift att leda dem tillbaka till anpassning när det uppstår kriser. Modellen kan användas av teamet för att identifiera var i processen föräldrarna befinner sig.

För att undersöka om resultaten från studie I-III även gäller för andra föräldrar i samma situation, skickades fyra enkäter ut till alla föräldrar i hela Sverige, vars barn har opererats för katarakt sedan 2006 (506 föräldrar). Enkäterna innehöll frågor om känsla av sammanhang, fatigue (trötthet), socialt stöd och tillit till familjen. Dessutom inkluderades fyra frågor om nöjdhet med sin egen insats som förälder, nöjdhet med vården och om man känt att man blivit tagen på allvar. Totalt 231 föräldrar svarade och resultatet visade att 38% av dem led av svår fatigue, speciellt föräldrar som har barn med katarakt på båda ögonen. Det visade sig också att mammor var tröttare än pappor och att föräldrar som inte bor tillsammans med den andra föräldrar upplevde att de hade svårare att hantera sin situation. Det fanns ett samband mellan känslan av att bli tagen på allvar och nöjdhet med vården.

Sammanfattningsvis har studierna i denna avhandling visat att föräldrar till barn med katarakt genomgår en omfattande anpassningsprocess till ett liv där barnets synutveckling kommer främst. För att stödja denna behöver vården av

dessa barn förändras, där ögonteamet måste uppmärksamma även hur föräldrarna mår, inte bara deras barn. En förändring som har genomförts är framtagandet av en digital applikation, en så kallad app, för föräldrarna. Appen har två funktioner, dels en information, som innehåller fakta, råd och vägledning om barn med katarakt, dels en meddelandefunktion, där föräldrarna kan skicka meddelande, bilder eller ha videosamtal med ögonteamet. Detta möjliggör en direktkontakt med teamet och föräldrarna kan skriva sina frågor även utanför kontorstid. Andra förslag till förbättring är att föräldrarna får svara på en enkät om fatigue för att se hur trötta de är, och vid påtaglig trötthet särskilt ta hänsyn till detta och eventuellt även hänvisa till andra vårdgivare, för att minska risken för t ex utbrändhet. Att bli tagen på allvar är nyckeln till nöjdhet med vården och stödet från ögonteamet. Genom att införa familjecentrerad vård, där en koordinator har en central roll, stärks även familjens behov och inte bara barnet som har katarakt.

LIST OF PAPERS

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

- I. Gyllén J, Rosenberg A, Nyström A, Forsberg A, Magnusson G.
Important sources of information on self-management for families of children with pediatric cataracts – based on PECARE Sweden/Sahlgrenska University Hospital
International Journal of Ophthalmic Practice. 2015 Feb/March:23-29
- II. Gyllén J, Magnusson G, Forsberg A.
Uncertainty and self-efficacy in parents of a child with congenital cataract – New implications for clinical practice
Nursing Open. 6(3),799-807
- III. Gyllén J, Magnusson G, Forsberg A.
The Core of Parents' Main Concerns When Having a Child With Cataract and Its Clinical Implications
Journal of Pediatric Nursing. 2019; 44, e45-e51
- IV. Gyllén J, Magnusson G, Forsberg A.
Parents' reported experiences when having a child with cataract – important aspects of self-management obtained from the Pediatric Cataract Register (PECARE)
International Journal of Environmental Research and Public Health. 2020, 17, 6329

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ABBREVIATIONS

GF	General fatigue
GTM	Grounded theory method
IOL	Intraocular lens
JIA	Juvenile idiopathic arthritis
MF	Mental fatigue
MFI-19	Multidimensional fatigue inventory
MSPSS	Multidimensional scale of perceived social support
PECARE	Pediatric cataract register
PFCE	Perceived family collective efficacy scale
PhF	Physical fatigue
PREM	Patient-reported experience measures
RA	Reduced activity
RM	Reduced motivation
SOC-13	Sense of coherence scale
VA	Visual acuity

GLOSSARY

Adherence	the extent to which a person's behavior – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a healthcare provider
Amblyopia	the reduction of best-corrected visual acuity of one or both eyes that cannot be exclusively attributed to a structural abnormality of the eye
Cataract	a change in the crystalline lens affecting translucency
Epistemology	the theory of knowledge, especially with regard to methods, validity and scope, as well as the distinction between justified belief and opinion
Fatigue	an overwhelming feeling of physical and mental exhaustion not easily relieved by rest
Glaucoma	disease characterized by high intraocular pressure-related damage to the eye
Occlusion therapy	using an eye patch to cover the non-amblyopic eye for a couple of hours each day
Ontology	the study of the nature of being, becoming, existence, or reality
Self-efficacy	confidence to carry out a behaviour to achieve a desired goal
Self-management	the individual's ability to manage the symptoms, treatment, physical and psycho-social consequences and lifestyle changes inherent in living with a chronic condition
Visual acuity	the resolution capacity of the eye

INTRODUCTION

About 40 children are born each year with congenital cataract in Sweden, representing an occurrence of 36 per 100,000 births (Abrahamsson, Magnusson, Sjoström, Popovic, & Sjostrand, 1999). Thus, it is a rare but sight-threatening condition. Early detection of paediatric cataract is crucial, where screening on the maternity ward is preferable (Magnusson et al., 2013). The Swedish National Board of Health and Welfare has appointed two hospitals in Sweden to perform National Specialized Medical Care of children with paediatric cataract under the age of three years. PECARE (Paediatric Cataract Register), is a national quality register with the aim aimed at optimizing screening strategies and more effective treatment of paediatric cataract. All children under the age of eight years undergoing surgery for cataract are registered. The goal is to constitute a national base for quality assurance and to define and analyse unexpected treatment outcomes. Since the launch in 2006, the register has 95% coverage (from January 2007- December 2018) and has gathered data on 731 children (1,029 eyes on June 1, 2020) (PECARE).

A cataract is the loss of lens transparency due to opacification of the lens. Paediatric cataracts may be unilateral or bilateral and can be classified using age at onset, aetiology and morphology; partial or dense. Paediatric cataracts may be congenital if present within the first year of life, developmental if present after infancy, or traumatic (Medsing & Nischal, 2015). The only treatment of dense cataract is removal of the lens through surgery. The removal of the opaque lens is important, as afferent visual signals are necessary for the development of normal cortical pathways and to block infantile nystagmus (Brodsky & Dell'Osso, 2014). The timing of the surgery is critical for visual development and most surgeries in Sweden are performed within 5 weeks for unilateral cataract and within 3 months for bilateral cataract. The two major complications after cataract surgery are secondary glaucoma and visual axis opacification (Lambert, 2013; Solebo, Cumberland, & Rahi, 2018), which sometimes requires one or more re-operations.

Amblyopia is defined as the reduction of best-corrected visual acuity of one or both eyes that cannot be exclusively attributed to a structural abnormality of the eye. It can affect one or both eyes and is usually more responsive to treatment before the age of seven years (Holmes & Levi, 2018). Studies show that most children with amblyopia respond well to treatment, but more care, i.e. more parental education and

closer follow-up, may be needed in children who are non-adherent and have poorer initial visual acuity (VA) (Handa & Chia, 2019; Medsinghe & Nischal, 2015).

After the removal of the opacified lens, an intraocular lens (IOL) is often implanted. Regardless of whether or not an IOL is implanted, the child will need contact lenses after surgery, as the IOL is calculated to fit the child's refraction as an adult. Around the age of three, the child usually starts wearing spectacles. After surgery, the ophthalmological team expects the parents to take responsibility for handling contact lenses, eye drops and often laborious visual training with patches. The treatment also requires numerous visits to the clinic and the parents need to be observant to signs of complications, such as secondary glaucoma and infections. Typical follow-up frequency is as follows: postoperative day 1, week 1, week 2, week 4, week 7, month 3, every 2 months for the first year, every 3 months for the second year and thereafter every 4 months between the age of 2-7 years and every 6 months between the age of 8-10 years. After 10 years of age, regular check-ups are recommended throughout life because of the risk of developing secondary glaucoma. The parents are more or less involved during both childhood and adolescence. This calls for close collaboration with the ophthalmological team, the members of which need to be perceptive of parental self-management.

Research reveals that less than half of all children who have undergone bilateral surgery in western Sweden achieved sufficient visual acuity to obtain a driving licence (Magnusson, Abrahamsson, & Sjostrand, 2002). For unilateral cataracts, good visual acuity (0.2 or more) was only achieved in children who underwent cataract surgery before the age of three months and who adhered to children the occlusion therapy schedule (Allen, Speedwell, & Russell-Eggitt, 2010; Lundvall & Kugelberg, 2002). This raises the question of whether the outcome could be improved by closer collaboration with and better support for parents, who are the key players in the care process of children with cataract. The parents' self-efficacy, i.e. the belief in their ability to succeed in managing this new situation, is crucial for the child's visual outcome, as parents are expected to carry out much of the visual training. We have shown that parents are willing to do all they can, as their child's vision is at stake (Gyllen, Magnusson, & Forsberg, 2019; Gyllén, Magnusson, & Forsberg, 2019). However, despite this strong motivation inherent in parenthood, the care process is an extensive undertaking.

Self-management requires that the person with a chronic illness develops skills in order to achieve the different goals involved in managing the disease. In this context it is how the parents manage the treatment of their child's cataract. This includes; problem solving, decision making, resource utilization, the formation of a patient-

provider partnership and taking action (K. Lorig & González, 1992; K. R. Lorig & Holman, 2003). Knowledge about parental self-efficacy and an in-depth understanding of their struggle are a necessary foundation for quality person-centred care. There is a lack of scientific knowledge about self-management among parents of children with paediatric cataract. Thus, the focus of this thesis is to understand, explore and explain parental self-efficacy and the clinical implications of how they adapt to their situation with a child with cataract. This knowledge is a prerequisite for the provision of targeted and person-centred self-management support and might serve as a mediator for improved long-term treatment results in children with cataract.

BACKGROUND

PERSPECTIVES AND VIEWPOINTS

In this thesis, the ontological assumption is that a parent of a child with cataract is seen as a subjective human being within the healthcare setting and a partner in the ophthalmology team who can contribute knowledge to the care provided. Uncertainty is inherent in being a person. The parent is a person facing extensive self-management demands on behalf of her/his child and the caregiver role to a child with a chronic condition will give her/him substantial knowledge and experience complementary to that of healthcare professionals, a fact that must be taken into consideration when designing follow-up care. The meaning a parent attributes to the self-management process will influence the way she/he responds to recommendations from the ophthalmological clinic. How a parent of a child with cataract understands her/his situation is important for the ability to engage in healthcare-related self-management activities. Therefore, the studies on which this thesis is based stem from an inside perspective and describe the parents' experiences of their parental role as care givers, as well as their needs, challenges and expectations of support from healthcare providers.

The epistemological assumption in this thesis is that it is possible to obtain knowledge both by understanding and explanation, i.e., by interpreting and measuring a phenomenon of interest. This thesis adopted a mixed method approach including both quantitative and qualitative research methods. We used questionnaires and interviews in order to explore and understand parental self-efficacy. The point of departure was the parents' need for targeted information as a key prerequisite for parental self-management. In order to understand what it means to be a parent of a child born with cataract, two different qualitative methods were used. The inductive approach was motivated by the fact that this area had been quite poorly examined and there was a lack of scientific knowledge about the experiences of parents as caregivers to a child with impaired vision. In order to explain possible differences among unrelated groups, quantitative sampling by means of parent self-report questionnaires was performed and statistical analysis applied in Papers I and IV.

CHILDREN AND THEIR RIGHTS AND PERSPECTIVES

All children have a legal right to information and participation in healthcare decisions affecting them; this is stated in the Patient Act [In Swedish: Patientlagen] (SFS, 2014:821) (SFS, 2014). The purpose of the Patient Act is to protect patients' rights and interests. The law states that the patient is to be informed about her/his illness and the kinds of treatment available. She/he has the right to participate in all decisions about the care she/he will receive. The patient must also be told where she/he can obtain the care she/he needs.

Children must also be given the opportunity to state their point of view. The Patient Act emphasises children's right to freely express themselves and for their views to be given due weight in accordance with their age and maturity. This may be done together with the parents or legal guardians for a very young child, but the importance of participation by the child increases with age. In order to be active in her/his care and make decisions, it is important that the child understands the information. Coyne et al. concluded that children appeared content for adults to take responsibility for the major treatment decisions. However, they desired and valued the opportunity to receive information, voice their preferences and choose how treatments were administered to them (Coyne, 2006; Coyne, Amory, Kiernan, & Gibson, 2014). Feeling that they participated is a prerequisite for children's positive experiences of their care (Ambresin, Bennett, Patton, Sancı, & Sawyer, 2013; Gilljam, 2020).

To further strengthen children's rights in society, the Swedish government adopted the Convention on the rights of the child [in Swedish: Barnkonventionen] as law on January 1, 2020, (SFS 2018:1197) (SFS, 2020). The law is based on the 1989 United Nations Convention on the Rights of the Child (UN, 1989). Article 12 states that the views of the child should be given due weight in accordance with the child's age and maturity. Children's rights are also monitored by the Nordic network for children's rights and needs in healthcare [in Swedish: Nordiskt nätverk för barn och ungas rätt och behov inom hälso- och sjukvård, NOBAB]; the principles have been published and translated into six different languages (NOBAB, 1980). These principles are to be used as a guide for child-centred care and as a tool for creating equality for all children within the healthcare system. NOBAB is also a member of the European Association for Children in Hospital (EACH, 1993), which safeguards the welfare of children in hospital and other healthcare services.

Implicit in all paediatric care is the participation of the parents. The rationale from a family perspective is that the family is a constant element in a child's life. Important aspects include collaboration between family members and healthcare providers to

formulate care plans that take account of the family context the need for flexible policies and procedures and for patient, family and healthcare professional education (Kokorelias, Gignac, Naglie, & Cameron, 2019).

FAMILY AND FAMILY CENTRED CARE

Becoming a parent is a life changing event. If the child is born with a chronic condition, such as paediatric cataract, the unexpected and unfamiliar situation will throw the parent into a world of uncertainty and caregiver demands. The parenthood they had imagined turns into something else/completely different as healthcare providers/professionals immediately expect them to become a co-caregiver of specialized and demanding care. The parents must master this new situation, which affects the whole family. The lack of previous experience and therefore necessary self-management skills contribute to the challenge.

In this thesis, the family is seen as a unit. Family functioning can be defined as the ability of family members to interact, react and respond to each other (Ryan, Epstein, Keitner, Miller, & Bishop, 2005). A change in one family member will affect all family members (Benzein, Hagberg, & Saveman, 2012; Gyllen, Magnusson, & Forsberg, 2018; Wright & Bell, 2009; Wright & Leahey, 2013). The change can be both negative and positive (Wright & Bell, 2009). The child's cataract presents the family with a unique set of challenges. Parents are expected to master the demands of the treatment regimen, which may necessitate changes to usual family routines and patterns of interaction. The child with the condition is expected to cooperate with required treatment and accept possible changes to usual activities. Siblings are also expected to adjust to changes in the family's usual routine and may be asked to assume new responsibilities (Knafl et al., 2013).

We define family management as the efforts that family members make to incorporate the demands of the treatment regimen and their child's special needs into everyday family life (Knafl, Deatrick, & Havill, 2012; Knafl et al., 2013). The partnership between parents and professionals is consistent with a model of healthcare delivery known as family centred care (Salvador, Crespo, & Barros, 2019). The growing recognition of family centred care as the standard of care in paediatric units (Kuo et al., 2012) is supported by its potential benefits for the child and her/his family. Research in the field of paediatric conditions has provided evidence for the association between family centred care and the child's adaptation (Russell, Beckmeyer, & Su-Russell, 2018), parents' psychological adjustment (e.g., reduced anxiety, depression

and higher levels of well-being), and their satisfaction with care (King, Teplicky, King, & Rosenbaum, 2004).

SELF-EFFICACY

Self-efficacy is a complex phenomenon introduced by Bandura and defined as “confidence to carry out a behaviour to reach a desired goal” (A Bandura, 1997). Parental self-efficacy refers to parents’ beliefs about their ability to parent successfully (Jones & Prinz, 2005). In this context, parental self-efficacy is defined as the parent's belief in her/his ability to succeed in managing the care after their child’s cataract surgery. The fundamental hypothesis of the self-efficacy theory is that the personal expectations of succeeding in a task will predict how much effort the person will put into the task and how long the effort will be sustained despite obstacles or other threatening experiences.

The efficacy expectations are based on four major components; performance accomplishments, vicarious experience, verbal persuasion and emotional arousal (A Bandura, 1997). With regard to children with chronic illnesses, parents with higher parental self-efficacy are more likely to have positive attitudes towards the treatment and care of their child, to actively obtain information related to the disease and management of routine treatment and to engage in positive problem-solving when facing challenges in the process of caring for the child (Dai et al., 2020). As the ontological assumption is that uncertainty is inherent in being a person and a parent, uncertainty will be the main concept of this thesis and consequently parental self-efficacy will be illuminated from this perspective. As elaborated by Almgren et al. and Lindberg et al., we assume that uncertainty is a barrier to self-efficacy, which in turn is the driver of self-management. Thus, to promote self-management it is essential to explore and understand the magnitude of parental uncertainty in order to support self-efficacy and enhance parental self-management of their child’s everyday treatment (Almgren, Lennerling, Lundmark, & Forsberg, 2017a, 2017b; Almgren, Lundqvist, Lennerling, & Forsberg, 2020; Lindberg, Almgren, Lennerling, & Forsberg, 2020).

UNCERTAINTY IN ILLNESS

Uncertainty is defined as the inability to derive meaning from illness-related events and appears when the person is unable to form a cognitive schema for illness events. The goal is to construct meaning, thus reducing uncertainty, and through coping

strategies achieve adaptation in order to experience health (Mishel, 1988). The components of parental uncertainty include ambiguity, lack of clarity, lack of information, lack of availability and the unpredictability of the medical information pertinent to their child's treatment and care. Ambiguity refers to the absence of cues as it relates to the planning of care for the child. Lack of clarity refers to not receiving information or perceiving the information about a child's treatment and system of care as complex to grasp. Lack of information refers to the absence of information about the diagnosis and gravity of the condition. Unpredictability concerns the inability to make future predictions about symptoms and illness outcomes for the child (Eche & Aronowitz, 2018; Mishel, 1990; Mishel, Padilla, Grant, & Sorenson, 1991).

According to Mishel (Mishel, 1999), education as a structure provider can help patients and their families to know where and how to get health information, thus reducing illness-related uncertainty. However, continual uncertainty can be preferable because it may enable patients and their families to perceive various opportunities and have greater be more adaptable. This can happen when parents accept uncertainty as the normal rhythm of life and believe in a world without absolute certainty and predictability (Mishel, 1990, 1999).

PREVIOUS RESEARCH

When exploring the previous research in this area the following key words and databases were used: paediatric cataract, congenital cataract, child cataract, parents and children. The PubMed, CINAHL and Scopus databases were searched. Filters for language (English), date (2000-2020), abstract available and peer-reviewed were used. The research field of paediatric cataract mainly covers four different perspectives, where the medical aspect is by far the most dominant. Research relevant to the research questions in this thesis is briefly described below.

MEDICAL ASPECTS OF PAEDIATRIC CATARACT AND VISUAL IMPAIRMENT

Posterior capsule opacification and secondary glaucoma remain the major postoperative complications, necessitating long-term surveillance in children undergoing cataract surgery early in life. Successful management of paediatric cataracts depends on individualized care and experienced teamwork (Medsing & Nischal, 2015). Lambert et al. discuss the timing of surgery, and compare contact lenses and IOL correction (Lambert, 2016; Lambert et al., 2014; Lenhart et al., 2015). Several authors have published research on paediatric cataracts and genetic testing (Drack et al., 2019; Gillespie

et al., 2014; Lenassi et al., 2020), stating that next-generation DNA sequencing technologies are able to determine the precise genetic cause of congenital cataract in 75% of individuals.

The management, treatment and follow-up of children with cataract are demanding, requiring frequent hospital visits and repeated examinations and/or surgical procedures with general anaesthesia over many years, but mainly during the first year of life. It is important to convey this message to the parents at the onset of the disease (Al-Bakri et al., 2019). Holmes et al. discuss the fact that treatment of amblyopia after the age of 7 years tends to be, on average, less effective than in younger children (Holmes & Levi, 2018).

Three months after surgery, higher levels of parenting stress are associated with poorer adherence to patching (Drews-Botsch, Celano, Cotsonis, DuBois, & Lambert, 2019).

CHILDREN WITH CATARACT OR OTHER VISUAL IMPAIRMENTS

A study by Castenada et al. describe concerns reflecting the impact of cataracts in physical, emotional and social domains, which can be used for the development of questionnaires to quantify the quality of life and functional vision effects of cataracts (Castaneda et al., 2016).

Cataract has a marked long-term impact on functional visual ability and quality of life of children and young people, with health related quality of life affected to degrees similar to those reported in children with severe congenital cardiac defects or liver transplants (Tailor et al., 2017).

A child-centred approach to identifying the content for a self-report vision-related QoL questionnaire is feasible and it has been suggested that such an approach is critical for accurately capturing children and young peoples' subjective perspectives on the impact of living with impaired vision (Rahi, Tadic, Keeley, Lewando-Hundt, & Vision-related Quality of Life, 2011).

PARENTS OF CHILDREN WITH CATARACT OR OTHER IMPAIRMENTS

A life skills training programme can be used as an efficient, cost-effective and simple technique for managing parenting stress in these parents (Khooshab, Jahanbin, Ghadakpour, & Keshavarzi, 2016).

For the parents of children with complex health needs and disabilities, the sense that they make of their situation plays a pivotal role in determining how they experience the impact of the disability and the need for help and support (Whiting, 2014).

CAREGIVER AND SOCIETY

Findings by Rahi et al. support the implementation of programmes for information provision, support and liaison by key workers in all specialized centres for the assessment and diagnosis of children with serious visual problems (Rahi, Manaras, Tuomainen, & Hundt, 2004). In another study, the same authors emphasize the key role of ophthalmic professionals in directly improving parental education (Rahi, Manaras, & Barr, 2003).

The medical community may be better placed to support families dealing with childhood cataract by improving the detection of childhood cataract, building appropriate communication pathways and promoting social support with emphasis on empathetic, individualized care (Hamm, Boluk, Black, Dai, & Thompson, 2019).

In summary, there is a scarcity of scientific studies on parental concerns in paediatric ophthalmology and even more so when it comes to paediatric cataracts. This thesis will add to the knowledge and understanding of parental experiences and viewpoints and how to empower them as essential partners in the care team to enable as best possible visual function in the child.

RATIONALE

Paediatric cataract often requires long periods of amblyopic treatment and contact lenses, where parental involvement is instrumental as visual impairment or in a worst case scenario blindness is at stake. The children and their parents face many challenges involving recovery from the surgery, various complications as well as everyday life challenges and recommended restrictions. Outcome results reveal that less than half of all children who have undergone bilateral surgery achieve sufficient visual acuity to obtain a driving license and that for unilateral cataracts, good visual acuity

was only achieved in those children who underwent cataract surgery before the age of three months and who adhered to the occlusion therapy schedule. Health is a part of well-being and by pursuing health promotion the overall well-being of the child and presumably also her/his family will increase. One important aspect of health promotion is self-management support, as the major part of visual therapy takes place at home and requires advanced parental self-care skills. Today, there are great expectations on the part of ophthalmological professionals that the parents should be partners and co-actors in their child's care process in order to achieve good long-term visual results and overall health. Family centred care and child centred care have been thoroughly investigated among children with various disabilities and illnesses, but not, however, in relation to children with cataract. Very little is known about the context specific parental challenges and concerns involved in the care of children with cataract, in particular the pressure involved in being primarily responsible for the child's visual training in everyday life. Furthermore, it is difficult to develop targeted and person-centred self-management support without an understanding of the information and support needs, as well as how the process of adaption takes place in a family where a child has cataract. Therefore, the rationale behind this thesis was to explore and describe in-depth the main concerns among parents and how they deal with them. It is important to approach the parents from a basic caring science perspective as hardly anything is known about their inside perspective.

Thus, in Paper I a mixed method approach was adopted to explore the need for information to support self-management among the parents. The findings revealed that aspects that hindered self-management included contradictory information provided by different healthcare professionals and the fact that some healthcare professionals were considered to have limited knowledge about the parents' self-management needs. The families requested support from care teams to meet the demands of self-management and cope with the sense of loneliness. To build on these findings, a second rationale was to gain an in-depth understanding of the parents' self-management needs, for which we adopted an inductive approach in Paper II, using phenomenological hermeneutics to explore the meaning of uncertainty and self-efficacy among the parents. In Paper III the inductive approach continued by means of the Grounded Theory Method (GTM) to develop a model of the parents' adaptation process. Interviews with 23 parents generated the hypothesis that self-efficacy from the parents' perspective was about balancing between uncertainty and acceptance through performance accomplishment. Balancing expectations could minimise disappointments, which could also decrease distress and we found that fathers could be better equipped than mothers to adopt a salutogenic perspective due to less caregiver burden. It also generated a model of how parents balance their child's abilities and inabilities as a process towards adaptation.

The hypothesis generated needed further testing in a larger group of parents along with an investigation of the possible influence parents have on the visual outcome of their children. The understanding of the parental challenges derived from the first three papers (I-III) led to a need to explore whether they received enough support to continue their treatment tasks. Thus, the final rationale in Paper IV was to investigate the life situation of Swedish parental care givers of children with cataract by exploring and explaining their sense of coherence, family self-efficacy, perceived social support, fatigue and self-reported experiences of care. For this purpose, all parents registered in PECARE since 2006 were included. The outcome was that fatigue is the most important aspect to consider when meeting these parents, especially those with children who have bilateral cataract and other co-morbidities. In addition, being taken seriously is the key marker of satisfaction with care and support from professionals.

A key concern for ophthalmological healthcare professionals is how to affect and support health and self-care behaviours in order to engage parents in the extensive task of self-management and adhering to the recommendations from healthcare professionals. The basic assumption in this thesis is that how the parents perceive the treatment process and what they experience as their main concern are fundamental for the development of person-centred and targeted self-management support. The understanding of the parents' perspective and the knowledge about/ their adaptation process are essential aspects in promoting health and well-being for families with a child who has cataract. Distress and decreased well-being are all possible threats to adherence to prescribed recommendations but first and foremost to health. Therefore, an increased understanding of uncertainty, parental self-efficacy and self-management is perceived as vital for promoting long-term outcomes for children with cataract. The goal of this thesis is to narrow the knowledge-gap regarding how the child's treatment is affected by the parents' process of adaptation and develop evidence-based guidelines concerning how health promotion should be tailored to increase well-being, health and sustainable self-care activities for children with cataract and their families.

AIMS

The overall aim of this thesis was to understand (Papers I-III), explore (Papers I and IV) and explain (Paper IV) self-efficacy among parents of a child with paediatric cataract in order to promote self-management and improve clinical care.

Specific aims:

1. To improve concordance by investigating important sources of information on self-management for families of children with paediatric cataracts from the perspective of parents and ophthalmologists.
2. An in-depth exploration of uncertainty and self-efficacy among parents of a child with congenital cataract by means of two theoretical frameworks to re-design family care.
3. To investigate the main concerns associated with being a parent of a child with cataract and how the parents deal with these concerns.
4. To explore and explain sense of coherence, family self-efficacy, perceived social support, fatigue and parent reported experiences of care among parents with a child with paediatric cataract in order to improve clinical care.

PATIENTS AND METHODS

Paper	Design	Participants	Data collection	Analysis
I	Mixed method	Parents (n=47) and ophthalmologists (n=28)	Questionnaires with closed and open-ended questions	Factor analysis and manifest content analysis
II	Qualitative Explorative	Parents (n=23)	In-depth interviews	Directed content analysis
III	Theory generating	Parents (n=23)	In-depth interviews	Constructing grounded theory
IV	Quantitative Explorative	Parents of children who underwent cataract surgery, registered in PECARE (n=231)	Self-Report questionnaires	Factor analysis

TABLE 1. Research design overview.

SETTING

The participants in these studies were all recruited from PECARE and the two hospitals in Sweden appointed by the Swedish National Board of Health and Welfare to perform National Specialized Medical Care of children with a paediatric cataract. The participants were allowed to choose the setting for the interviews, which took place at their home, at the hospital, at their work or at a public location.

PAPER I

PARTICIPANTS

Parents of children registered in PECARE and operated at the Department of Ophthalmology at Sahlgrenska University Hospital between 2006 and 2011 were included (at the time of the study; $n=72$) (Table 1 in Paper I). Also included were the paediatric ophthalmologists ($n=30$) who monitored the patients upon their return to local healthcare facilities throughout Sweden.

DATA COLLECTION

Questionnaires with both closed and open-ended questions were sent to 69 families of children (34 girls and 35 boys) (Appendix 1). The remaining three families were excluded due to emigration in two cases and protected address status in the other. A similar questionnaire was sent to the 30 paediatric ophthalmologists. The response rate was 68% ($n=47$) for the parents and 93% ($n=28$) for the paediatric ophthalmologists.

DATA ANALYSIS

For descriptive purposes, means, standard deviation, median, minimum and maximum were calculated for continuous variables and frequency and percentage for categorical variables. Non-parametric tests were used for all statistical analyses. For comparison between two groups, the Mann-Whitney U-test was employed for continuous variables and Fisher's exact test for dichotomous variables. The sign test was used to compare percentage differences between the information requested and obtained by parents, and percentage differences in the information suggested and information given by ophthalmologists.

The open-ended responses were analysed based on manifest content analysis according to Graneheim and Lundman (Graneheim & Lundman, 2004).

PAPERS II AND III

PARTICIPANTS

Inclusion criteria were parents of a child diagnosed with paediatric cataract that had been operated on and had visited the paediatric ophthalmological clinic in 2016-2017. Other criteria were being able to verbally share their experiences and who were registered in the national quality register PECARE. An exclusion criterion was that the child did not have other systemic co-morbidities (Table 3 in Paper II and Table 1 in Paper III).

DATA COLLECTION

Data were collected between 2016 and 2017 in the form of interviews at a setting chosen by the parents. A total of 17 in-depth interviews were performed with 23 parents; 6 mothers, 5 fathers and 6 couples, which were recorded and transcribed verbatim for analysis. The open-ended interviews lasted on average 50 minutes (range 22-87 minutes) resulting in approximately 300 pages of transcribed text. An interview guide was used, starting with the question: Would you please tell me how it all started? After that, the areas covered were surgery, contact lenses, amblyopia training, self-efficacy, the future and quality of life.

DATA ANALYSIS PAPER II

A directed content analysis (Hsieh & Shannon, 2005) using Mishel's theory of uncertainty (Mishel, 1990) and Bandura's self-efficacy theory (A Bandura, 1997) was conducted retrospectively to deductively explore uncertainty and self-efficacy.

DATA ANALYSIS PAPER III

Data were analysed in accordance with Grounded Theory developed by Charmaz (Charmaz, 2014).

Following the recommendations of Hallberg (Hallberg, 2010) and Glaser (Glaser, 1998), we first established whether previous studies with a GTM approach and similar aim had been conducted within this particular context. No such study was found.

Secondly, initial line-by-line coding was performed to find words or phrases indicating important categories, qualities or contexts related to the research questions (Charmaz, 2014). Memos about each interview were logged, including reflections that emerged during the analysis and coding processes. In order to detect and perhaps explain the most significant codes, the third step involved focused coding, which illuminated the main concern. In turn, theoretical coding revealed relationships between the codes generated from the focused coding. The constant comparative-method (Charmaz, 2014) was used simultaneously on data, codes and categories. The interviewer who performed all interviews is a clinical nurse specialist at the paediatric ophthalmology clinic who had no care relationship with the participants. In line with Charmaz's constructivism (Charmaz, 2014), the categories and theory were developed from the patterns revealed by the researchers' theoretical constructions of the parents' subjective experiences.

PAPER IV

PARTICIPANTS

Inclusion criteria were all parents of children registered in PECARE since 2006 (n=506 at the time of the study start in March 2019) (Table 1 in Paper IV). Exclusion criteria were cataract caused by JIA, lens luxation and trauma.

DATA COLLECTION

A letter with information about the study and an invitation to participate was sent to all parents whose children met the inclusion criteria and were registered in the PECARE. When the informed consent form was returned, the questionnaires (Appendix 2) were sent out. If the questionnaires were not returned, two reminders were sent, which resulted in 231 returned questionnaires.

Four scales were selected to cover aspects of being a parent of a child with cataract.

SENSE OF COHERENCE (SOC-13)

Aaron Antonovsky introduced his salutogenic theory “sense of coherence” as a global orientation to view the world and the individual environment as comprehensible, manageable and meaningful, claiming that the way people view their life has a positive influence on their health (Antonovsky, 1987; Eriksson & Lindstrom, 2005). Antonovsky developed a questionnaire comprising 29 questions, and later a shorter version with 13 questions (used in this study), with 5 items measuring comprehensibility, 4 items measuring manageability and 4 items measuring meaningfulness. The response alternatives are a semantic scale of 1 point to 7 points and the questionnaire yields a summed score with a range from 13 to 91. Psychometrically, the SOC-13 scale has proved to be comparatively sound; the Cronbach’s alpha score is 0.7-0.92 (Eriksson & Lindstrom, 2005).

MULTIDIMENSIONAL FATIGUE INVENTORY SCALE (MFI-19)

MFI-19 is a self-report instrument designed to measure fatigue. In its original form it has 20 items, but item 19 was removed based on the recommendation by Hagelin et al., who performed a psychometric evaluation of the scale (Hagelin, Wengstrom, Runesdotter, & Furst, 2007). It covers the following dimensions: general fatigue (GF), physical fatigue (PhF), reduced activity (RA), reduced motivation (RM) and mental fatigue (MF). The questionnaire consists of 19 items for which the respondent must indicate the extent to which the statements relate to her/him on a five-point scale, ranging from “Yes, that is true” to “No, that is not true”. Each subscale is scored from 4 to 20, where a higher value indicates greater fatigue and the value 12 is the cut-off. Thus, a value of >12 on each subscale indicates fatigue. The MFI-20 has been translated and validated for Swedish conditions and has been found to have good validity and reliability; Cronbach's alpha for the various subscales was >0.70. (Ahsberg & Furst, 2001; Hagelin et al., 2007).

MULTIDIMENSIONAL SCALE OF PERCEIVED SOCIAL SUPPORT (MSPSS)

The scale used in this study was developed by Smets et al. (Smets, Garssen, Bonke, & De Haes, 1995). Perceived social support has different functions, i.e. emotional support, instrumental support (practical support), informative support and appraisal

support. It includes 12 items covering three dimensions; Family, Friends and Significant others. Each item is rated on a seven-point Likert-type response format (1 = very strongly disagree; 7 = very strongly agree). A total score is calculated by adding the results for all items. The possible score range is between 12 and 84, the higher the score the higher the perceived social support. In addition, separate subscales can be used by adding the responses from the items in each of the three dimensions. The possible score range for the subscales/dimensions is between 4 and 28. The scale has been translated and validated for Swedish use by Ekback et al. with sound psychometric properties. The internal consistency was good for all scales ($\alpha = 0.91-0.95$) (Ekback, Benzein, Lindberg, & Arestedt, 2013).

PERCEIVED FAMILY COLLECTIVE EFFICACY SCALE (PFCE)

The scale was developed by Caprara et al. to assess beliefs in the family's efficacy to operate as a integrated system in accomplishing tasks necessary for family functioning (Caprara, Regalia, Scabini, Barbaranelli, & Bandura, 2004). It focuses on the family's ability to manage daily routine operations, achieve consensus in decision-making and planning, cope together with adversities, promote reciprocal commitment, provide emotional support in difficult times and stressful situations, enjoy each other's company and relax together in spite of multiple obligations, and keep/maintain good relations with the community at large. The PFCE uses a 7-point response format, from 1 = Not well at all, to 7 = Very well. The scale has been translated into Swedish but not validated. Cronbach's alpha for the various subscales was 0.96 (Caprara et al., 2004).

The study also included four additional questions about PREM where the parents were asked to respond on a scale ranging from 1-100.

1. How satisfied are you with the treatment of your child's cataract?
2. How satisfied are you with your efforts as a parent of your child with a cataract?
3. Do you experience that you receive enough support from the clinic that treats your child's cataract?
4. Do you feel that you have been taken seriously by the clinic that treats your child's cataract?

DATA ANALYSIS

The SPSS for Windows version 25.0 (IBM Corp., Armonk, NY, USA) was used when analysing data, which were mainly ordinal. Descriptive statistics (parental demographics, socio-demographics etc.) were presented with frequencies. We chose to dichotomize parents' age, education, social status, occupation and native country and used an established cut off for general fatigue. For the children, we dichotomised age at the time of the study and age at surgery. When testing for differences between two unpaired groups we applied the Mann-Whitney U Test. In order to explore relationships between the different phenomena we employed Spearman's Rho. Hierarchical multiple regression was used to assess explanatory factors of family self-efficacy and satisfaction with care after controlling for the influence of age and gender. Differences with a p-value ≤ 0.05 were considered significant.

ETHICAL CONSIDERATIONS

All studies were conducted in accordance with the principles of the Declaration of Helsinki and the Medical Research Involving Human Subjects Act. (WMA, 2013). Approval was obtained for all studies; the Regional Ethical Review Board of Gothenburg approved studies I (610-11), II and III (746-14), while the Swedish Ethical Review Authority approved study IV (20 19-00836 /1234-18).

All participants were informed of the study's purpose, that participation was voluntary and that the information provided would be confidential. Informed consent was obtained from all participants. For studies I and IV, the pamphlet with the questionnaires was sent out after the return of the signed consent form. There is always a possibility of integrity intrusion when approached with the invitation to participate in the study. The parents may not want to be reminded about the difficulties of having a child with cataract and related experiences. However, it could also be beneficial to share and reflect upon one's experiences of being a parent of a child with cataract and the possibility of contributing more knowledge leading to enhanced treatment within the area could be positive. A social worker at the follow-up clinic was on hand to provide emotional support to the parents if the interview proved emotionally demanding for them. However, this need did not arise in any of the studies.

The results of the studies will probably lead to changes in the support strategies for families with children with cataract. From the perspective of the children, opportunity for individual care planning of prognoses of vision could be given, which will lead to reduced suffering. Results from the studies will benefit society in terms of fewer visits to the hospital and better use of hospital resources.

RESULTS

The overall results of this thesis reveal that being a parent of a child with paediatric cataract is a process of adaptation towards a life in balance that needs support from the ophthalmological team. A model was developed in Paper III to understand the parents' situation, which can be used for this. At the time of diagnosis, parents often find themselves in uncertainty due to shock, lack of information and the absence of support, all of which diminish their self-efficacy. The parents develop strategies through mastering, collaborating, facilitating and adapting to balance their child's inabilities and abilities. It is important to consider negative barriers, such as fatigue and low sense of coherence, when interacting with parents of children with cataract.

Papers I and IV reveal what prerequisites the parents have. In Paper I, they wanted something they did not receive, i.e. more information, both written and online. In Paper IV, fatigue emerged as an obstacle for sense of coherence, especially among the parents of children with bilateral cataract. In addition, being taken seriously by the ophthalmological clinic explained over 60 % of the variation in satisfaction with care.

Papers II and III provided an in-depth understanding of the parents' self-efficacy and what strategies they use to adapt to a life of balancing the abilities and disabilities of a child with cataract.

PAPER I

In total, 72% of the parents felt they had not received enough information. They expressed a desire for more written information, especially at the time of the diagnosis of their child's cataract. Parents also requested more information online and a desire for more contact with other parents in the same situation. Mothers and fathers of children under 3 months were more likely to prefer written information. Open-ended questions revealed that information could either promote or hinder self-management (Table 4 in Paper I). To meet the demands of self-management and cope with the sense of loneliness, the families requested support from care teams, social workers and other parents in the same situation.

In this study, the same questionnaire was sent to the ophthalmologists and the parents. Both families and ophthalmologists indicated that it is important to supplement oral information with written information and to include practical advice. Web-based information was also emphasised as essential for maintaining self-management. Aspects that prevented self-management included contradictory information provided by different health professionals. The need to provide preschools and public authorities with information was identified by most participants, as parents had experienced difficult situations, such as preschool staff refusing to administer eye drops.

PAPER II

Being a parent of a child with cataract means being in uncertainty from the moment the child is diagnosed until a state of acceptance is achieved.

Parental uncertainty involves many different aspects illustrated by several crucial questions as illustrated in Figure 1 in Paper II.

Causes; Where does the cataract stem from? Did I cause the cataract?

Treatment; Is the occlusion therapy working?

Follow-up; What will happen to my child? What is the plan for my child?

Prognosis; Will my child be visually impaired or even blind for the rest of her/his life?

Future; Will my child be able to drive a car, work and move out as an adult?

Parental performance; Am I doing the right thing and am I doing enough as a parent?

Self-efficacy means balancing between uncertainty and acceptance. A state of acceptance occurs when uncertainty is reduced (Figure 2 in Paper II). For parents, aspects of being in acceptance involved many different areas of life. "I look upon the future with hope that he will be able to drive." (Expectations), "She sees better now and that encouraged us." (Progress), "The teacher at the day-care centre handles the occlusion patch." (Adjustment), "My child has the other eye to see can see with the other eye, nothing wrong with it." (Positive), "He just jumps right off!" (Performance) and "For us, it's being together as a family." (Quality of life).

PAPER III

The core category *balancing the child's inability and ability* summarizes a process whereby the grounded theory generated contains the four main categories; *mastering*, *collaborating*, *facilitating* and *adapting* (Figure 1). Through this process a clear path of adaptation

is evident, starting with the child’s diagnosis and continuing for many years during the child’s growth and development.

Balance between inability and ability

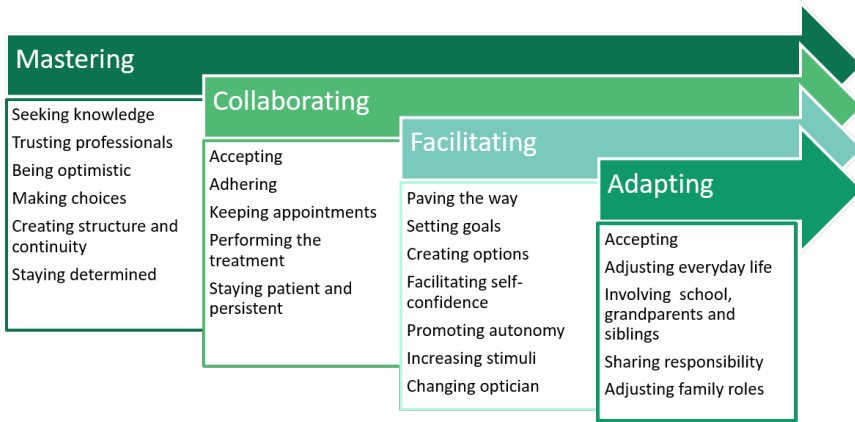


FIGURE 1. Model of the grounded theory Balancing the child’s inability and ability.

Parents do what is necessary to achieve the best treatment outcome and an everyday life that functions and promotes the child’s autonomy and abilities. The results show an overall salutogenic driver of this process to maintain a balance between the child’s inability and ability, i.e., the focus was more on health than illness and abilities rather than disabilities. The need to master emerged immediately on hearing the diagnosis, as the mothers and fathers were overwhelmed by the child’s visual inability and the unknown consequences. The treatment place many demands on parents and collaboration is essential. A functioning family life despite the child’s visual inability was important throughout the whole treatment process, demanding extensive parental facilitation. The parents finally balanced their expectations and learned how to adjust

their everyday life in order to achieve an optimum balance between the child's inability and ability at various developmental stages in her/his life.

PAPER IV

In the fourth study the percentage of responding participants was 46% (n= 231). The majority of respondents were mothers (80%) of Swedish origin (81.4%) with a university degree (69.7%). They were working or studying (87%). Furthermore, most of them were living in a conjugal family (87%). They also had a large support network, and consequently widely-spread positive resources for taking care of their affected child. The results of the measurements of self-efficacy as well as experiences of care are shown in Table 2 in Paper IV. Furthermore, 38% of the mothers and fathers scored as high as >12 in GF, showing that they suffered from severe fatigue.

PARENTS OF CHILDREN WITH BILATERAL CATARACT AND CO-EXISTING SYSTEMIC MORBIDITY

Parents of patients with bilateral cataract were considerably more tired compared to mothers and fathers of patients with unilateral cataract. When evaluating the results of four subscales of fatigue the parents of bilaterally operated cataracts reported significantly more fatigue than those parenting children with unilateral cataract; GF ($p=.007$), PhF ($p=.030$), RM ($p=.005$) and MF ($p=.019$). Despite this, the parents of these children were more satisfied with their own efforts ($p=.004$), the treatment ($p=.009$), the support from the ophthalmological clinic ($p=.000$) and felt that they were taken seriously ($p=.050$) compared to mothers and fathers of patients with unilateral cataract. When the patient had co-existing systemic morbidity, the mothers and fathers reported lower family self-efficacy ($p=.029$) as well as lower support from friends ($p=.049$) compared to when the patient had cataract without any systemic disorder.

LIFE SITUATION AND PARENT'S AGES

Mothers and fathers who were not living with the other parent of the child had more difficulties managing the situation ($p=.049$). Furthermore, they experienced less support from the eye clinic ($p=.024$) compared to parents living together in the same household. Not sharing the work with the other parent (9.5%) resulted in a lower

SOC in the manageability ($p=.038$) and meaningfulness ($p=.056$) subscales. In addition, the parents generally had more GF ($p=.003$), PhF ($p=.002$), RA ($p=.011$) and MF ($p=.015$). Furthermore, these mothers and fathers expressed lower support from their family ($p=.017$) and significant other ($p=.036$) and also lower family self-efficacy ($p=.023$).

Mothers and fathers older than 40 years showed less motivation due to fatigue ($p=.041$) compared to parents younger than 40 years of age. In addition, the older parents were less satisfied with the treatment ($p=.052$). Furthermore, lower support from their significant other ($p=.008$) was reported among the group of parents older than 40 years.

RELATIONSHIPS

Statistical analysis of relationships were performed and a strong relationship between the overall SOC total and PhF ($r_s=.516$), RA ($r_s=.510$), RM ($r_s=.508$) and MF ($r_s=.535$) was found. Consequently, a linear multiple regression was used testing the fatigue dimensions as independent variables in order to explain the variation in total SOC. This analysis resulted in the fact that only RM ($Beta=.239, p\leq.0001$) and MF ($Beta=.236, p\leq.0001$) contributed significantly to the variance in total SOC. Therefore, we proceeded with hierarchical multiple regression to assess the ability of RM and MF to predict levels of total SOC and this was done after controlling for the influence of parents' gender and age. Furthermore, preliminary analyses were performed to ensure no violation of the assumptions of normality, linearity, multicollinearity, and homoscedasticity. At step 1 age and gender were entered, explaining 12.6 % of the variance in the total SOC. Furthermore, after entry of RM and MF at step 2, the total variance explained by the model was 44.9%, $p\leq.0001$. RM and MF explained an additional 32.3 % of the variance in the total SOC, R -squared change=.323, F change (2, 224) =65.65, $p\leq.0001$. However, in the final model, MF recorded a slightly higher beta value ($beta=-.34, p\leq.0001$) than RM ($beta=-.33, p\leq.0001$).

PREM

When exploring parents' reported experience measures we found that satisfaction with the treatment at the eye clinic was moderately related to satisfaction with one's own effort as a parent ($r_s=.456$) but strongly related to support from the clinic ($r_s=.673$) and being taken seriously ($r_s=.507$). Furthermore, satisfaction with one's own effort was related to support from the eye clinic ($r_s=.447$) and it was weakly related to being taken seriously ($r_s=.256$). In addition, support from the eye clinic was strongly related to the sense of being taken seriously ($r_s=.718$). Therefore, we proceeded with hierarchical multiple regression to assess the ability of being taken seriously to predict experienced levels of support from the clinic, after controlling for the influence of parents' age and gender. Analyses were conducted to ensure no violation of the assumptions of normality, multicollinearity, linearity and homoscedasticity. At step 1, gender and age were entered, explaining only 1 % of the variance in experienced support from the eye clinic. In step 2, after entering being taken seriously, the total variance explained by the model was 60.3 %, $p \leq .0001$.

DISCUSSION

METHODOLOGICAL CONSIDERATIONS

This thesis has a mixed method design in order to explore and explain self-efficacy among parents of a child with paediatric cataract to promote self-management and improve clinical care. Papers I and IV used an explorative cross-sectional approach. A directed content analysis according to Hsieh & Shannon was employed in Paper II and the Grounded Theory approach was used in Paper III. The combination of different methods contributes to both a general and detailed picture of parental self-efficacy in families with paediatric cataract. Papers I and IV were assessed in relation to their validity and reliability (D. Polit & Beck, 2017). Trustworthiness can be assessed by a combination of credibility, dependability, confirmability and transferability (Y.S. Lincoln & Guba, 1985; D. Polit & Beck, 2017), which were evaluated in Papers II and III.

STUDY SAMPLE AND SAMPLING

All parents in this thesis have a child who is registered in PECARE. Paper I included parents of children who underwent cataract surgery at the Department of Ophthalmology at Sahlgrenska University Hospital between 2006 and 2011, which encompassed most children with cataracts in southern and western Sweden (n=72).

Triangulation was used in Papers I, II and III as a method of multiple reference to help capture a more complete, contextualized picture of the phenomenon (D. Polit & Beck, 2017). In Paper I, both parents and ophthalmologists answered the same questionnaire, which strengthened the results as several of the concerns expressed were the same. In Papers II and III, mothers, fathers and couples were interviewed, which provided an opportunity to validate the data on the phenomenon of being a parent of a child with cataract through multiple perspectives, i.e. gender differences.

VALIDITY AND RELIABILITY OF QUANTITATIVE DATA (PAPERS I AND IV)

INTERNAL VALIDITY

A strength of the study in Paper I was that it included both parents and the ophthalmologists who treated the children at their home clinic. It employed a mixed method approach, where closed and open-ended questions were answered simultaneously. One limitation is the design of the questionnaire, which was constructed by the research team but not scientifically validated in terms of reliability and validity (Boynnton & Greenhalgh, 2004). Also, the parents were not requested to provide demographical data on themselves or their child. Rattray et al. emphasize the need to adopt a logical, systematic and structured approach to questionnaire development and that a psychometric evaluation should be performed (Rattray & Jones, 2007). Failure to develop a questionnaire sufficiently may lead to difficulty interpreting the results, which in turn can impact on clinical or educational practice. Thus, it was not possible to determine whether demographic and social aspects influenced the results.

Construct validity in Paper IV concerns the choice of instruments, which were selected to cover aspects of being a parent of a child with cataract based on the findings in and hypothesis generated from Papers II and III. Sense of coherence, fatigue, social support and family self-efficacy were considered relevant, but other instruments might have given different insights and knowledge (D. F. Polit & Beck, 2006). Instruments covering aspects such as health related quality of life, health literacy, anxiety level, parental satisfaction and perceptions of the information provided may have revealed other aspects of parental self-efficacy, although ones that were not linked to the hypothesis in the thesis. All instruments in the study except PFCE were validated, but the latter was still considered to provide valuable and relevant knowledge as family functioning plays an important role in parental self-efficacy.

Another threat to internal validity is recall bias, which in this case concerns how long ago the child underwent surgery. As all parents in the PECARE were included, some of the children underwent surgery ten or more years ago and a few had already turned 18 years. The time span probably affected the ability to remember. In addition, some parents commented that fatigue was not related to the child with the cataract, but to being pregnant or suffering from depression at the time of the study.

EXTERNAL VALIDITY

External validity concerns whether relationships found among the study participants hold true for other people and in different settings (D. Polit & Beck, 2017). In Paper I, the response rate was high – 68% for the parents and 93% for the ophthalmologists. A threat to external validity is the fact that the informants just represented a part of the total population as the study only included parents from one of the hospitals operating on these children. However, generalizability was examined by comparing the descriptive statistics of the sample to those of the PECARE sample and the data were similar (PECARE). In Paper IV, the whole population was included, i.e. all parents whose children were registered in PECARE from its inception in 2006 were invited to participate, allowing us to generalize to other parents of children with cataract. The cohort was found to be representative when it was compared to the children of non-responders.

A strength of the study in Paper IV is that no violation of the assumptions of normality, multicollinearity, linearity and homoscedasticity were found, which allowed us to proceed with hierarchical multiple regression to assess the ability of RM and MF to predict levels of total SOC after controlling for the influence of parents' age and gender.

The response rate in Paper IV was 46%. In an attempt to ensure a high response rate (D. Polit & Beck, 2017), two reminders were sent in accordance with the recommendations of Polk et al. (Polk, Rasmussen, Brorson, & Olsen, 2013), which resulted in 231 participating parents. In a review article Badger et al. conclude that a 40% response rate is above-average for self-report postal questionnaires and 25-30% is common (Badger & Werrett, 2005). The reasons for declining participation in Paper IV are of course purely speculative. A combination of both traditional and online questionnaires may have increased the response rate (Horevoorts, Vissers, Mols, Thong, & van de Poll-Franse, 2015). Other contributory reasons could be the number of questions in the questionnaires (four different instruments with a total of 67 questions). Some parents agreed to participate but then did not return the questionnaire. The children of non-responders had a mean age of 9 years (range 0-20) - an age at which the visual system in normal children is fully developed and check-ups at the eye clinic are less frequent. This might have influenced the response rate.

TRUSTWORTHINESS (PAPER II)

According to Lincoln and Guba (Yvonna S. Lincoln & Guba, 1986), four criteria can be used for ensuring trustworthiness in qualitative research: credibility, dependability, confirmability and transferability.

Credibility means establishing confidence that the results (from the perspective of the participants) are true, credible and believable. Throughout the study, the research was scrutinized by highly experienced senior researchers, who served as mentors during the process.

A strength of the study is that all interviews were performed by the same researcher. The research team had previous knowledge of exploring both uncertainty and self-efficacy in a different medical context. This constituted a certain pre-understanding. In order to increase the credibility of the data and the subsequent results, an interview guide was used and probing questions such as: "how do you mean?" or "can you explain more?" were posed to deepen the understanding of the phenomenon. The 17 interviews lasted between 22-88 min and provided rich data. Another important issue of credibility concerns the number of participants. As content analysis emphasizes variation in content and multiplicity there must be sufficient data to cover significant variations (Hsieh & Shannon, 2005). During the data collection and analysis, theoretical saturation clearly emerged after 15 interviews and we found that in the end of the analysis process the subcategories and main categories merely confirmed the findings rather than adding any new data.

Dependability refers to the stability of data over time and conditions. Leaving a documented audit trail is one strategy. The interviews were recorded and transcribed verbatim and the researcher made field notes during the interviews. We measured the coding accuracy and intercoder reliability of the research team. This study was developed through extensive reading of the theories of uncertainty (Mishel et al., 1991) and self-efficacy (A Bandura, 1997) and a systematic search of the existing literature.

Confirmability refers to objectivity – the findings must reflect the participants' voice and the conditions of the inquiry and not the researchers' biases (Yvonna S. Lincoln & Guba, 1986). As the context was familiar to us, I and one of my co-authors attempted to bracket our pre-understanding during the data collection. However, one of the other co-authors was not at all familiar with the context. There was also a pre-understanding regarding knowledge of the concepts of uncertainty and self-efficacy, but in a different medical setting.

Transferability is analogous to generalizability and is the extent to which the findings are applicable to other settings or groups. The study was intended to be heterogenic and included mothers, fathers and couples whose children had either unilateral or bilateral cataract to generalize the findings to other families. As the majority of parents were Swedish, the transferability may be limited to families in Sweden and other

European countries. Transferability is also strengthened by rich descriptions of the findings, (Frambach, van der Vleuten, & Durning, 2013). This was done by including many quotations from the parents.

CRITERIA FOR ASSESSING GROUNDED THEORY (PAPER III)

Charmaz (Charmaz, 2014) suggests four criteria: credibility, originality, resonance and usefulness. A strong and credible originality increases resonance and the subsequent value of the contribution.

Credibility in this study is ensured by intimate familiarity with the setting. Following the recommendations of Hallberg (Hallberg, 2010) to investigate whether similar studies had been previously conducted with a GTM approach, we found that this was not the case. Another aspect concerns whether the data are sufficient to merit the claims of the research. In this study, six mothers, five fathers and six couples participated and after 15 interviews the data were rich and we achieved saturation in the emerging model. One mother, one father and one couple were invited to validate the model and to elaborate on our conceptualization of the findings, which is viewed as a major strength. Constant systematic comparisons were made between observations and between categories. However, the parents were recruited from one hospital, thus it is possible that the geographical location and hospital setting are a limitation.

Originality was ensured by the categories of this study, which offer new and valuable insights into the knowledge and understanding of parents of children with cataract, thus the originality is high. The GTM of achieving a balance between inability and ability has generated a new understanding of parents' process of adaptation and balance. The main categories constitute new condensed concepts of how parents manage the process of regaining balance and a functioning life.

Resonance is evident in the GTM, the main categories illustrate the richness of the informants' experiences, during the process of balancing the child's ability and inability.

Usefulness. The social implication of this theory is a more specific understanding of the strategies parents adopt to adjust to their new life situation. Although paediatric cataract is a very unusual condition, the insights and knowledge gained through this study can probably be applied to other parents who have children with a chronic condition.

GENERAL DISCUSSION OF THE RESULTS

The studies in this thesis all contribute to exploring and explaining self-efficacy among parents of children with paediatric cataracts in order to promote self-management by providing clinical implications. It might be possible to generalize the findings to other parents of children with visual impairments.

FROM THE PARENTS' PERSPECTIVE

This thesis revealed new findings as we became aware of the magnitude of uncertainty. From the parents' perspective self-efficacy is about balancing between uncertainty and acceptance through performance accomplishment. Reducing the gap between uncertainty and acceptance is a constant struggle, as in this process every setback/surgery/complication increases their uncertainty. As pointed out by Hinton et al., parents appear to interpret uncertainty as limiting their control over illness-related events, which relegates them to the position of powerless bystanders in their child's illness experience and causes them distress (Hinton & Kirk, 2017). Uncertainty drains the parents' energy levels when they are not piloted and guided through all the questions and worries they have and experience. The key to acceptance and well-being is confirmation of the parents' struggle, efforts and the child's progress by the ophthalmology team. Uncertainty must be understood a natural state that occurs when mothers and fathers witness their child suffering from a medical condition that shatters their world view. Events must be structured, ordered and predictable (Antonovsky, 1987). The sense of coherence is lost when the stimuli associated with illness, treatment and recovery are vague, ill-defined, probabilistic, ambiguous and unpredictable (i.e. uncertain). As Paper II revealed, fathers seem to prefer a salutogenic approach. This approach involves prioritizing health, goalsetting and progress. In contrast, mothers assume the role of primary caregiver more than fathers and subsequently worry about various aspects of the child's illness. According to Bandura's social learning theory (Bandura, 2001), it is also possible that children learn to adopt more positive problem-solving skills from their parents (A. Bandura, Barbaranelli, Caprara, & Pastorelli, 2001). Research shows that the sense of coherence of parents of children with chronic diseases or disabilities serves as a resource (Goldberg & Wiseman, 2014) and as a buffer, which facilitates moving people towards optimal outcomes in challenging situations (Hochwalder, 2015). This is consistent with the findings of Szulczewski et al., who argue that caregivers with positive reaction and coping styles experienced less uncertainty (Szulczewski, Mullins, Bidwell, Eddington, & Pai, 2017).

A fundamental understanding of the balance between uncertainty and acceptance, i.e., parental self-efficacy, will most probably be a useful tool for extensive improvements in the care of the families. s, interpreting parents' mastering as a lack of adherence. Higher parental self-efficacy is correlated with more effective management of children with chronic diseases and improves their health-related behaviour and outcomes (Lohan, Morawska, & Mitchell, 2017). The team must guide the parents towards acceptance, recognizing that uncertainty will always be present to some degree in parents of a child with cataract. We argue that stable and calm parents are more likely to have children who feel stable and calm.

The parents told that you do what you have to do, actually all parents, to reach a optimal visual development as their child's future is at stake. Through this process a clear path of adaptation was obvious. It started with the diagnosis of the child and continuing for several years during growth and development of the child. This adaptation is also true for the children, who gradually need to participate in the decisions about their own care in line with the principle of autonomy. For this to be possible, they need to participate in a positive healthcare environment, which includes being listened to and active participation. There are key elements of active participation embedded in Article 13 of the U.N. Convention that are integral to this research. These elements are that the child receives relevant information, is given an opportunity to freely express her/his own views and opinions and that her/his wishes and values are considered in the decision-making process (Quaye, Coyne, Söderbäck, & Hallström, 2019). Mårtenson et al. conclude that it is challenging to balance the power in the interaction between the child, parent and healthcare professional. However, when achieved, it gives rise to the possibility of children experiencing trust and making use of their self-determination. (Mårtenson & Fägerskiöld, 2008). This is a crucial step for the children in their own development and adaptation process towards autonomy.

FROM THE HEALTHCARE PERSPECTIVE

The findings in this thesis call for a re-design of the care of these families. As expected and in line with previous research, Paper I revealed that parents of children with cataracts desired more information and directed information. The main new finding in Paper IV showed that it is important to take fatigue into consideration when interacting with the parents, especially those who have children with bilateral cataract. Furthermore, paper IV also revealed that the key marker of satisfaction with care is being taken seriously. These findings call for a re-design of ophthalmological care for these families. Introducing family centred care highlight the family as actors and co-

providers of the health care and as (co-) decision makers and (co-) facilitators in relation to the treatment. Parental participation and care-by-parent acknowledge and utilize parental knowledge, skills and competence in the delivery of a child's treatment and hospital care (Uniacke, Browne, & Shields, 2018). Research shows that parents' perception of family centred care was associated with greater perceived self-efficacy, which in turn was linked to increased psychological well-being (Salvador et al., 2019). For the ophthalmological team, this could mean introducing a key worker and an individualized care plan for children in collaboration with the parents. This is also in agreement with the findings in Paper IV, where being taken seriously is the key marker of satisfaction with care and support from professionals. As concluded by Rantala, the need to be taken seriously can be fulfilled by listening to the parent, helping her/him to understand what has happened, what is about to happen and enabling her/him to feel safe and content in the present moment (Rantala, 2017). Consequently, for parents of a patient with a chronic health condition such as paediatric cataract, being taken seriously and listened to is imperative.

Another important and novel finding was the theoretical framework in Paper III, as we developed a grounded theory on the social process of mastering the balance between the inability and ability of the child. This model provides a comprehensive understanding of parental self-management that can be used by the ophthalmological team to pilot the parents through the process. The parents suggested how the ophthalmological team could assist them by facilitating their child's treatment in everyday life. They requested better ways of communication to improve the care. This could be provided through a mobile app, which would facilitate communication, partnership and support in an easy and accessible way. Today's parents use their mobile devices frequently and as pointed out by Wulfovich et al., mobile apps have the potential to enable better self-management and further improve the patients' general self-efficacy as well as the self-efficacy of activities contributing to chronic disease management (Wulfovich, Fiordelli, Rivas, Concepcion, & Wac, 2019). Mobile apps are used in many different healthcare settings, both in adult as well as paediatric care, for example breast cancer survivors, diabetes, paediatric leukaemia and parents of young people with bipolar disorder (Brzan, Rotman, Pajnikihar, & Klanjsek, 2016; Kapoor, Nambisan, & Baker, 2020; Nadkarni & Fristad, 2012; Wang et al., 2016).

CONCLUSIONS AND CLINICAL IMPLICATIONS

The main conclusions of this thesis are:

- Parents need more information and directed information, both personal and written, as well as online
- Parents are going through/undergoing a profound adaptation process
- Parental uncertainty constitutes a strong barrier to self-efficacy
- Parents constantly balance the inability and ability of their child in order to achieve the best possible visual and habitual outcome
- It is essential to take fatigue into consideration when interacting with parents of children with cataract, especially those with bilateral cataract and single parents
- Being taken seriously is the key marker of satisfaction with care and support from professionals/professional support
- All the parents emphasized that you do what you have to do to achieve a successful visual outcome for the child

Based on the findings in this thesis, ophthalmic care needs to be re-designed in order to support the adaptation process for families with paediatric cataract. It is a life-long adaptation, and for parents it continues for as long as the children are living at home. At the beginning it is the parent's adaptation until the child gradually becomes autonomous. Appointing a key worker to monitor the process and work in collaboration with the parents and the other members of the team would be a starting point for family centred care.

In addition, the inclusion of an occupational therapist in the ophthalmological team would further improve the care for these families. The findings in this thesis show that the parents struggle to maintain a balance in their lives and therefore occupational therapy interventions could be beneficial, as they are designed to facilitate the

performance of everyday tasks and adaptation of settings in which the person works, lives and socializes.

While the team members cannot reduce the cause of parents' fatigue, they can confirm it and perhaps alleviate it by assessing stressful factors. The team needs to develop and offer self-management support strategies to parents to help them cope despite fatigue. This could be done by screening parents with instruments to evaluate fatigue in order to identify those who might need referral for additional support.

During the work on this thesis, the research team developed a mobile app specially designed for parents of children with cataract with the tech company Kind (Kind, 2020) in close collaboration with parent representatives. The app has two main features; information from the ophthalmological team about paediatric cataract, contact lenses, the surgery and post-operative complications. The other feature is a chat function that enables parents to post questions or comments to the team 24/7. The parents using the app at the Department of Ophthalmology at Sahlgrenska Hospital particularly appreciate the possibility to chat at any time, knowing they will receive a response on the following working day. They also point out the possibility of sending photos when questions arise about, for instance, a red eye or a cloudy cornea after surgery, as well as when observing signs of complications such as glaucoma. After responding to the parent, a note is written in the child's journal. The advantage of using mobile apps instead of websites is that apps offer better personalization through tailored communication. Apps also provide the possibility of sending notifications to the user and can utilize features of a mobile device such as the camera and microphone for video calls. Furthermore, the app can be used as a tool in research, allowing anonymous questionnaires to be administered to the parents. In addition, it can also be used for sending forms for the parents to submit information on their child before a visit.

FUTURE PERSPECTIVES

We need to evaluate what digital support means for self-management support, which could be investigated with PREM-questionnaires. Furthermore, we need to develop health and care plans that are not only directed towards the children but also include the parents to ensure a family-centred care perspective. Parents not living with the other parent might need extra support with the demanding care of the child. Here, targeted assessments and efforts are necessary, i.e. a screening instrument, especially for fatigue and the causes of fatigue need further investigation. The model of balancing requires testing in a larger group of parents and should also be studied in other parental settings with children with chronic conditions to determine whether it is generic or very specific for children with cataracts. Furthermore, the comments from the 68 parents in Paper IV will be analysed and published in a future study. Finally, it is important to interview the children when they are older, preferably teenagers, to understand what it is like to have undergone this intensive treatment during a large part of their childhood.

ACKNOWLEDGEMENTS

Life is a journey and a possibility of constant learning. Some journeys you decide on your own, and some journeys are decided *for* you as life happens *to* you.

In 2014, I had the opportunity to start walking the academic journey. It has been an adventure and I am grateful for all the experience and knowledge I have gained during this time. This journey could not have been done without the amazing people surrounding me, so thank you:

All the fantastic parents participating in my studies. Thank you for opening your hearts to me and letting me see some of your world.

Gunilla Magnusson, my supervisor. For your kindness and gentle guidance in the academic world. For supporting me in my work, both as a researcher, in the clinic and as friend. I love it when you say: we have a plan! And we have many!

Anna Forsberg, my co-supervisor and role-model as a nurse. You have accomplished amazing things and I am grateful that you challenge me into thinking critically and with new eyes. Thanks for being there.

Madeleine Zetterberg, my co-supervisor, for making it possible for me start my research career and always giving valuable feedback and encouraging words.

The amazing team at Queen Silvia Children's Hospital, the paediatric ophthalmological clinic: Abbas, Adela, Alf, Annika, Carola, David, Eva A, Eva R, Grete, Johanna, Knattan, Linda, Lothar, Mariya, Marita, Mónica, Nuria, Shahnaz, Sofie, Stella, Susann, Ulrika (and Melanie, Jennifer and Nathalie) for constant encouragement and covering for me when I am off doing research. It's an honor and pleasure to work with you!

My great colleagues at the at the Department of Ophthalmology at Mölndal, where I started as an ophthalmic nurse in 2002. I enjoy working with you, even though I have now moved on to work with the children at Östra. Thanks for all the support through the years!

Anna Gyberg, my fabulous sister-in-arms on this journey as a PhD-student, I am so happy we met during that course. Thank you for your support and endless discussions on academic and methodological challenges, nursing, children, guitars and other important matters in life. I look forward to your dissertation!

Dragana Škiljić for excellent proofreading and Alf Nyström for valuable comments on the manuscript. Mathilda Selvén for practicalities with my dissertation.

The PECARE steering committee for the possibility to do research on the important data you have gathered through the years.

Annica Carnbring Belfrage for the giving me the opportunity to develop the app that supports the parents. Maria Strajnic and colleagues at Kind.app for close and rewarding collaboration with this ongoing project.

Ida Gunnarsson for the excellent work with the photos in my thesis. So, when are you bying a Vespa, dear?

My beloved sisters – not by blood but by heart. Annika, Anna-Karin, Greta, Karin HA, Karin E, Kicki, Lisanne and Matilda. You are always there when I need it, laughing with me, crying with me, encouraging and supporting me through thick and thin.

Kaisa and Hans Bergqvist, my loving parents, for teaching me the importance of knowledge and education. For supporting me on my journey and being there.

Sanjoa James, my dear and loving friend, for always having my back. For supporting me with English translations, our discussions on important matters in research and life and for your amazing cooking.

Bosse Sörensson, my very much missed Lambretta Senior Mechanic, who always encouraged me to be curious and learn new things. One day I will drive my Giovanni from -57 on the streets of Gothenburg again, in a new coat of paint!

Lalla Fondin, you are *always* there for me – thanks for also being my computer wizard!
Life is good with you by my side.

Allie, my angel. Your strength and love still reminds me do extraordinary things and encourages me to be loving and giving.

Edith, my strong, brave and brilliant daughter. As I travel each day with you, I am thankful, encouraged and blessed. You are amazing!

Love is.

Financial support was provided by:

Kungl och Hvitfeldtska stiftelsen
De Blindas Vänner



Sunbeams shine down on the earth.
Without the sunshine we would be ice.
Our bodies need sunshine and animals and flowers.
I am happy when the sun shines.
I am happy as the sun.

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