Improving family communication and disorder knowledge: An intervention study with siblings of children with chronic disorders

Therese Händel Waggestad

Thesis for the professional study
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Improving family communication and disorder knowledge: An intervention study with siblings of children with chronic disorders
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Abstract

Author: Therese Händel Waggestad

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Supervisor: Krister Fjermestad

Siblings of children with chronic disorders are highly underrepresented in research. Living with a child with a chronic disorder affects the whole family, including siblings. Research has revealed negative impact and several risk factors for family and siblings. However, two risk factors have been emphasized as especially important, namely poor family communication and the lack of disorder knowledge. This thesis is based on an open trial of an intervention for siblings and their parents. The research questions are investigating if 1) the quality of family communication, and disorder knowledge in siblings will increase from pre-to post intervention, and 2) quality of communication and level of knowledge are related to each other. Method. This thesis is based on a one group pre-post design intervention study. The Parent-Child Communication Scale was used to measure siblings-rated family communication, and the Sibling Knowledge Interview was used to measure disorder knowledge in siblings, both at pre-and post intervention. The sample consist of 101 siblings and their parents. The age of siblings ranged from 8 to 16 years \((M = 11.5, SD = 2.06)\) and 54.7 % of siblings were girls. Results. The results showed that quality of communication increased significantly from pre-to post intervention, but only for sibling-rated communication with the mother. The level of sibling disorder knowledge also increased significantly from pre- to post intervention. Increase in the quality of communication was significantly related to increase in disorder knowledge, but only for communication with the mother, not the father. Conclusions. These findings support the use of this intervention for siblings of children with chronic disorders. Furthermore, the findings represent an important step in exploring who will benefit more from the intervention. The findings show that both communication and knowledge improves. Thus, siblings with poorer family communication and lower disorder knowledge could be in greater need of the intervention than siblings with better family communication and more disorder knowledge.
Sammendrag

Forfatter: Therese Hændel Waggestad

Tittel: Bedrìng av familiekommunikasjonskvalitet og kunnskapsnivå om diagnose. En intervensjonsstudie med søsken til barn med kroniske lidelser.

Veileder: Krister Fjermestad

Preface

This thesis is part of a larger project, the Sibling Study, which is a collaboration project between Frambu Resource Centre for Rare Disorders, and the department of Psychology at the University of Oslo. I took part in the data collection used in this thesis by coding 108 Sibling Knowledge Interviews at pre- and post intervention, tested for reliability, and computed the data into variables. The hypotheses were chosen by me, as were the measurements to investigate these hypotheses. I did all the analyses in this thesis, the drafting, and completion of the text.

I would really like to thank my supervisor Krister Fjermestad for the good advice and support through the whole process of data collection and writing. Thank you! I would also thank the Sibling Study group that included me in their project and inspired me to explore such an interesting and important field of science.

I would also like to thank my sisters and friends for taking the time to read through my thesis and give me feedback and valuable encouragement. Thank you so much! Finally, a big thanks to my fabulous Mæggi, for forcing me out for walks every day. Keeps me sane.

Oslo, October 17th, 2017

Therese Händel Waggestad
## Concepts and abbreviations in the thesis

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<thead>
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<th>Abbreviation</th>
<th>Description</th>
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<tbody>
<tr>
<td>Child</td>
<td>Child is used about the ages 0-18 years if nothing else is specified.</td>
</tr>
<tr>
<td>CD</td>
<td>Chronic Disorder</td>
</tr>
<tr>
<td>Pre</td>
<td>The time of the first collection of data, at start-up of the intervention.</td>
</tr>
<tr>
<td>Post</td>
<td>The time of the second collection of data, 3 months after the intervention.</td>
</tr>
<tr>
<td>Communication with mother</td>
<td>The sibling-evaluated communication with mother.</td>
</tr>
<tr>
<td>Communication with father</td>
<td>The sibling-evaluated communication with father</td>
</tr>
<tr>
<td>The PCCS</td>
<td>The Parent-Child Communication Scale. Questionnaire for measuring the quality of communication between the sibling and the parent, reported by the sibling.</td>
</tr>
<tr>
<td>The SKI</td>
<td>The Sibling Knowledge Interview. A structured interview with siblings to assess the level of their disorder knowledge about the diagnosis of their brother or sister.</td>
</tr>
<tr>
<td>The Affect RTPB</td>
<td>The affect revised Theory of Planned Behaviour. Theoretical model.</td>
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<td>The TPB</td>
<td>The Theory of Planned Behaviour. Theoretical model.</td>
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1 Introduction

Between 10 and 15% of children in Norway have a chronic disorder that affects their everyday functioning (Norwegian Institute of Public Health, 2010). Chronic disorders (CD) in childhood affect these children’s parents and siblings, who are also at increased risk of mental health problems (Haukeland, Fjermestad, Mossige, & Vatne, 2015; Karst & Van Hecke, 2012; Metcalfe, Coad, Plumridge, Gill, & Farndon, 2008). Studies have shown that the consequences of growing up in a family where a child has a CD can be severe, also for typically developing siblings. Siblings of children with CD (referred to hereafter as siblings) evaluate their subjective wellbeing and quality of life lower than siblings of typically developing children (Emerson & Giallo, 2014; Fullerton, Totsika, Hain, & Hastings, 2017; Rana & Mishra, 2015).

Risk factors in families with a child with CD have been explored, and two of the most important risk factors have been found to be the quality of family communication and lack of disorder knowledge (Jackson, Bijstra, Oostra, & Bosma, 1998; Metcalfe et al., 2008). Poor quality in communication and low levels of disorder knowledge have been linked to challenges in sibling adjustment and mental health (Carpenter & Levant, 1994; Jackson et al., 1998; Metcalfe et al., 2008; Strohm, 2008; Vermaes, van Susante, & van Bakel, 2011).

This thesis is based on a pilot intervention study that is a part of a larger project. The project will use the data from the pilot, and advance the pilot intervention to provide municipal health care services with a novel and user-tailored intervention. This will allow health providers to offer evidence-based health services to siblings. The pilot intervention in this thesis entails focus on family communication and disorder knowledge, and seeks to improve both quality of communication and level of disorder knowledge in siblings.

A recent review of studies involving siblings of children with neurodevelopmental disorders emphasized that being a sibling is not a diagnosis (Tudor & Lerner, 2015). The review suggests that future research should investigate different individual features in siblings and families that separate high and low risk, and explore what individual attributes lead to increased benefit from the intervention. This thesis responds to this call and aims to take a first step in mapping out who the intervention can be most beneficial for.

1.1 Aims
This thesis is based on an exploratory open trial of a manual-based group intervention for siblings and parents of children with CD. The intervention targets parent-sibling communication, family challenges, and siblings’ disorder knowledge.

The aim is to investigate if the quality of parent-sibling communication and the level of disorder knowledge improves from before the intervention to three months after the intervention. The hypotheses are that both sibling-rated communication and level of disorder knowledge will improve from pre- to post intervention. Furthermore, the relationship between sibling-rated communication and level of disorder knowledge will be explored. The hypotheses are that sibling-rated communication are related to the level of disorder knowledge in siblings at pre- and post intervention. Additionally, the change in the sibling-rated communication are related to the change in sibling disorder knowledge from pre-to post. The assumption is that better quality of communication is associated with higher level of disorder knowledge.
2 Empirical background

The literature investigating the impact and perspective of being a sibling emphasize the need for focusing on siblings. Through several quantitative and qualitative studies, focus on different outcomes for siblings and family risk factors are revealed. Additionally, studies describe what it can be like to live with a brother or sister with a CD, and give valuable information needed to build the optimal interventions or support systems for siblings.

2.1.1 Quantitative studies

An early meta-analysis found negative impacts on being a sibling on cognitive functioning, peer activities, and psychological functioning (Sharpe & Rossiter, 2002). More recently, another meta-analysis found that being a sibling was associated with less positive self-attributes and more internalizing and externalizing problems (Vermaes et al., 2011). The most recent review found that younger siblings often displayed externalising behaviour and that the presence of internalizing behaviour increased with age (Tudor & Lerner, 2015). Several single studies have also found that being a sibling is associated with higher levels of emotional and behavioural difficulties (Fullerton et al., 2017; Giallo, Gavidia-Payne, Minett, & Kapoor, 2012; Goudie, Havercamp, Jamieson, & Sahr, 2013; Lovell & Wetherell, 2016; O’Neill & Murray, 2016; Saban & Arıkan, 2013). A single study aimed at exploring siblings’ attitudes toward their brother or sister with CD found that siblings described the child with CD as both a limitation for the family, as well as a strain. At the same time, siblings reported feeling a sense of personal fulfilment when the child with CD accomplished new goals (Caroli & Sagone, 2013).

Individual risk factors related to being a sibling, has not been investigated to a large degree. However, some family risk factors have been identified, such as being a single parent, having a low socioeconomic status, lack of social support, parental stress, and especially maternal stress (Mulroy, Robertson, Aiberti, Leonard, & Bower, 2008; O’Brien, Duffy, & Nicholl, 2009; Taylor, Fuggle, & Charman, 2001). There is a connection between the functioning of all the members of the families of children with CD. Sibling adjustment has been linked to how satisfied parents were with their role as caregivers to the child with CD (Hesse, Danko, & Budd, 2013). The more satisfied parents were, the less problems the sibling had with adjustment (Hesse et al., 2013). The need for support to all family members is emphasized by the fact that functioning of family members are intertwined, and also because
families of children with CD report lower level of quality of life compared to families with typically developing children (Juhássová, 2015).

Families with a child with CD demonstrated higher levels of hostile, intrusive, and/or withdrawn communication compared with families of healthy, typically developing children (Murphy, Murray, & Compas, 2017). This recent review by Murphy et al. (2017) included 14 studies, all of which investigated the quality of communication in families with a child with CD using direct observation of communication. The studies included different CDs (asthma, epilepsy, type 1 diabetes, cystic fibrosis, and spina bifida), and sample sizes ranging from 20 to 68 participants. Murphy et al. (2017) pointed out that it is particularly important for future research to explore the relationship between family communication and different family and child variables.

2.1.2 Qualitative studies

Siblings and parents’ experiences of living with a child with Downs syndrome or autism spectrum disorder was explored in an integrated review of 28 qualitative studies (Mandleco & Webb, 2015). The authors found from sibling reports that the parents paid more attention to the child with CD than the sibling. The review also found that family function can be affected by having a child with CD, with parents reporting high levels of stress, depression, disruption in family functioning, and caregiver burden (Mandleco & Webb, 2015).

Siblings were found to struggle with feelings of loss, separation anxiety, loneliness, neglect, and rejection as a consequence of the parental attention being unevenly distributed, or a perceived differential implication of the house rules (Knecht, Hellmers, & Metzing, 2015). These findings came from a recent review of literature exploring siblings’ perspectives, and the authors found that due to these difficult feelings that siblings report, they are at risk of developing internalizing strategies to cope. For example, siblings describe a tendency to “withdraw into themselves”, “fall silent” and “stay in the shadows”. The review found that although internalizing strategies were most common, there was also a tendency of displaying externalizing behaviour, and feelings of jealousy, injustice, and envy were reported. (Knecht et al., 2015).

Many siblings denote feeling isolated in their experience when growing up with a brother or sister with CD (Jacobs & MacMahon, 2017) and report unfair household responsibilities and reduced parental attention (Cridland, Jones, Stoyles, Caputi, & Magee, 2015). Studies exploring siblings’ self-report about the impact of having a brother or sister
with CD reveal both positive and negative experiences (Luijkx, van der Putten, & Vlaskamp, 2016). In a study where siblings of children with autism spectrum disorder were asked how it was like to grow up with their affected sibling, siblings reported a sense of responsibility, concern about the future and a desire to talk about their experiences (Corsano, Musetti, Guidotti, & Capelli, 2017). Families where a child has a CD represent a heterogeneous group and the challenges in everyday life vary from family to family. Some challenges can be stressful for the families and hard to handle, such as frequent and long-lasting visits to the hospital, or challenging behaviours in the child with CD. A recent study explored parents’ experience of their child’s disorder, through 33 semi-structured interviews. The study found four subgroups of how parents related to the disorder (i.e., possibility, focus on illness, denial, and anger). The grouping was found to give meaning to how the families related to the health services and corresponded to the different illness progressions in the children. The authors state that the groupings emphasize a need for the whole family to receive support, and the possibility that the coping mechanism of the families might contribute to the trajectory of the illness of their child (Cipolletta, Marchesin, & Benini, 2015).

The challenges met by siblings and their families emphasize the importance of tailoring a support system to optimize family functioning. Interventions are a common way to give these families support and a platform where they can meet others in the same situation and discuss challenges. Both communicating about problems and emotions, and discussing the child’s diagnosis, are important factors in the intervention this thesis explores. The relationship between communication about the disorder, and disorder knowledge is not much investigated, but a lack of communication or information have been found to cause misconceptions or uncertainty about the diagnosis (Vatne, Helmen, Bahr, Kanavin, & Nyhus, 2015).
3 Theoretical background

Family functioning has been hypothesized to be crucial for siblings’ coping (Sommerschild, 1998). Sommerschild (1998) developed a model for children’s coping based on research on resilient children. Several protective factors that led some children to not be as strongly affected by stress were identified, and these protective factors were related to resilience (Rutter, 1985). The most important factors related to resilience are included in the model and divided into two main conditions, namely closeness, and competence. The model in Figure 1 illustrates how closeness and competence lead to coping through self-worth and resilience. Self-worth is also an important part of the model, because more resilient children tend to have a higher assessment of their self-worth than less resilient children (Sommerschild, 1998). More resilient children were described as taking more initiative, believing they could accomplish things, being independent and curious, describing themselves in a more positive manner, being prouder of own accomplishments and withstanding disappointment, criticism and change better than their less resilient peers (Harter, 1990; Sommerschild, 1998). Sommerschild emphasise that the model can also indicate possible areas for intervention, to help the children whose coping is impaired.

<table>
<thead>
<tr>
<th>Closeness</th>
<th>Competence</th>
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<tbody>
<tr>
<td>The Dyad:</td>
<td>Being able</td>
</tr>
<tr>
<td>At least one close relationship with a significant other</td>
<td>Being of use</td>
</tr>
<tr>
<td>The Family:</td>
<td>Receiving and taking responsibility</td>
</tr>
<tr>
<td>Predictability, confirmation, belongingness</td>
<td>Displaying love for another</td>
</tr>
<tr>
<td>The Network:</td>
<td>Encountering and handling adversity</td>
</tr>
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<td>Common values, social support</td>
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![Figure 1. Conditions for Coping (Sommerschild, 1998).](image)

Sommerschild’s (1998) model of the conditions for coping works as a theoretical foundation for linking communication and knowledge to outcomes such as better adjustment. In the model, adjustment is represented by coping, as an outcome of closeness and competence. Closeness and competence in this model are viewed to be representations of...
communication and knowledge. That is, communication is one of the tools for accomplishing closeness. Through the development of the reciprocal dynamic of the communication between a child and the main provider, the significant other and the family can give the child confirmation of value, a sense of belonging, and support. Communication is also one of the tools for accomplishing competence. In the model, knowledge is defined as a part of Competence.

The Sommerschild model illustrates that communication and knowledge are important for coping, but the model does not illustrate how communication and knowledge relate to each other. The affect model of the Revised Theory of Planned Behaviour (RTPB) can be used to illustrate the relationship between communication and knowledge (Ajzen, 1988, 2002; Leng, Lee, & Lim, 2016). The affect RTPB model (see Figure 2) entails several determinants for performing the behaviour of sharing knowledge. The principal determinant is thought to be the intention to do the behaviour. The intention is influenced by four factors; an attitude towards the behaviour, and the subjective norm (i.e. the normative views on the behaviour that the individual is impacted by), together with the controllability of knowledge sharing practices, and perceived knowledge sharing self-efficacy. The last two factors also influence the behaviour itself.

![Figure 2. The affect RTPB model (Ajzen, 1988, 2002; Leng et al., 2016) to explain how Social/ Family Communication Behaviour influences Knowledge Sharing Behaviour.](image-url)
If parents believe they have a good understanding of the child’s disorder, and believe they can share it with the sibling, they will share it. Or, to rephrase it with the RTPB model, the more you believe that you know what you share, and the more you believe you will be able to share, the more likely it is that you will share the knowledge. Several versions of the Theory of Planned Behaviour (TPB) models on knowledge sharing behaviour were tested, and the revised version of the TPB was found to best predict knowledge sharing behaviour (Kuo & Young, 2008). The RTPB added to the TPB because of the strong significant relationship that was found between perceived knowledge sharing self-efficacy, and knowledge sharing behaviour. The study by Kuo and Young (2008) was based on a web-based survey for teachers (n = 460), and used questionnaires and measures of online knowledge sharing behaviour. The RTPB represents both an argument for discussing diagnostic knowledge in the intervention for these families, to strengthen their knowledge sharing self-efficacy, and for including parents in interventions to increase their perception of their ability to share knowledge.

The second addition to the TPB, the affect dimension, was included in the model following a study investigating the determinants of knowledge sharing intention (Leng et al., 2016). The affect dimension included social communication behaviour, social collaboration behaviour, and positive and negative affect. The hypothesis was that poor communication and collaboration in social settings, such as a family, led to poor knowledge sharing culture. The extended model is still being empirically explored. However, some empirical support of the link between communication behaviour and knowledge sharing exists already. It was found that training in communication skills led to more efficient information sharing (Cabrera & Cabrera, 2005). The same study also found that face-to-face communication is very important for establishing trusting relationships and for facilitating knowledge sharing. Some empirical support for this hypothesis comes from computer-mediated communication research where it was found that a culture of constructive and supportive communication positively influenced knowledge sharing (Van Den Hooff & De Ridder, 2004). It has also been found that a more agreeable style of communication (examples; patient and kind), was related to more willingness to share knowledge (de Vries, van den Hooff, & de Ridder, 2006). Furthermore, an extroverted style of communication (examples; articulate and energetic), was related to more eagerness and willingness to share knowledge. Measures were based on questionnaires, and the participants were asked to rate their own willingness and eagerness of knowledge-sharing, and their groups style of communication (de Vries et al., 2006). These findings were
important for the intervention in this thesis because they support the hypothesized relationship between communication and knowledge, and the findings also add more information as to how to optimize the intervention to increase communication and knowledge.

By using the affect RTPB model in this thesis to explain the relationship between communication and knowledge, a premise is set that the measured level of disorder knowledge in the thesis represents knowledge sharing behaviour. An increase in measured disorder knowledge in the thesis is accepted as a result of a change in Knowledge Sharing Behaviour. Furthermore, Social/ Family Communication Behaviour can be measured by siblings’ subjective rating of communication with mother and father. In sum, a change in the sibling-evaluated communication with mother and father will represent change in family communication behaviour, and change in siblings’ level of disorder knowledge will represent change in knowledge sharing behaviour.
4 Focus of the thesis

The focus of this thesis is family communication and disorder knowledge. These factors are some of the most important outcome variables in the intervention in this thesis. The literature point out the importance of focusing on both variables and emphasizes the benefits of improving them.

4.1 Family Communication

Family communication can be defined as the process through which “each member of the family engages in communicative behaviour and exchanges verbal and nonverbal messages and information with other family members” (Stamp, 2004, p.18). Stamp (2004) also points out that relationships between the individuals in the family are created and maintained through this communication, thus making communication an important factor for family functioning.

Family communication has been found to be an important part of family functioning and problem-solving (Barnes & Olson, 1985). Better communication between adolescents and parents, such as more problem-free and more open communication, was associated with higher scores on family cohesion, adaptability and satisfaction. Better family communication was also related to better individual and overall quality of life. The sample consisted of 426 families, and data was collected from one adolescent, their mothers and fathers. The age of the adolescents ranged from 12 to 20 years ($M = 16.4$). Data were based on the Parent-Adolescent Communication Scale (PACS; Barnes & Olson, 1985), which is composed of two scales, one that measures the level of openness of communication in the family, and one that measures the degree of problems in communication. The study also used scales for measuring the family adaptability, cohesion, satisfaction, and life satisfaction.

Quality of communication is important for families and for the adjustment of the individual family member. Jackson et al. (1998) used the PACS together with multiple scales to examine adolescents’ individual adjustment. The study found a strong relationship between “open” or “problematic” communication in the family, and the decision-making, coping and self-esteem of the adolescent (Jackson et al., 1998). Thus, to focus on communication in families is important, both for family functioning, as well as the individual functioning of siblings.
4.1.1 Family communication when a child has a chronic disorder

Quality of communication has been found to be especially challenging in families in which a child has a CD (Mulroy et al., 2008). Families face several challenges to achieve the open and problem-free communication Barnes and Olsen (1985) found to be related to better family functioning and individual coping. The most typical challenges identified in the communication between parents and siblings was that both parents and siblings find it difficult to communicate about the diagnosis (Metcalfe et al., 2008). Parents may find it hard to talk about the diagnosis with the sibling because of the emotional activation, including anxiety associated with the memory of their own first reaction to the information about the diagnosis (Sullivan & McConkie-Rosell, 2010). Siblings may find it difficult to talk about the diagnosis with their parent because of fear of hurting their feelings (Chesler, Allswede, & Barbarin, 1992). Parents might also find it difficult to notice that siblings have some of the same feelings as the diagnosed child, and to notice the siblings’ need to talk about the diagnosis (Metcalfe et al., 2008). Parents report that they are often waiting for the sibling to ask a question, or just avoid talking about it at all (Metcalfe et al., 2008; Plumridge, Metcalfe, Coad, & Gill, 2011). Little, or lack of, communication in general is found to be an especially crucial risk factor for sibling adjustment, and the absence of communication can make siblings feel isolated and confused (Strohm, 2008).

The importance of an open family communication between siblings and parents was the focus of a qualitative meta-synthesis by Metcalfe et al. (2008). The meta-synthesis, consisting of 17 studies, investigated communication between siblings and parents about genetic conditions. Three factors were considered especially important when conveying knowledge about the diagnosis; providing information, checking understanding, and explaining and managing the emotions that arose in siblings. The study pointed out that in addition to how difficult the communication involving CD can be, the communication about genetic or inherited conditions can entail an extra stressor because of the level of possible future consequences for the sibling. Openness of communication was emphasized by some parents and explicitly holding back information to protect siblings was emphasized by others. When parents withheld information about the genetic component of the diagnosis, siblings were found more frequently to react in a resentful way when they were finally told, and felt that this should have been verbalised earlier. In families with a more closed style of communication, siblings reported becoming more upset and frustrated with family secrecy. The families who were defined as having a more open communication tended to report an
initial struggle when dealing with the information, but were described as more supporting in the long run, and siblings showed more emotional resilience. This indicates the importance of an open communication within the family, and the need for appropriate help to constructively communicate openly about the CD, and the challenging situations that arise when living with a child with CD (Metcalfe et al., 2008).

The challenges to family communication are multiple, and the consequence of low family communication quality can be serious, so an important focus is to explore the best way for the intervention to help these families overcome the obstacles to constructive communication.

4.2 Disorder Knowledge

Lack of disorder knowledge has been found to be an influential risk factor for the development of sibling adjustment problems and emotional distress (Carpenter & Levant, 1994; Metcalfe et al., 2008). Siblings have been found to have a lack of knowledge about the diagnoses of their brothers and sisters (Lobato & Kao, 2002). A study found that 64% of siblings asked had little or no knowledge about the nature of the diagnosis of their brother or sister (Rana & Mishra, 2015). Siblings of children with CD have been found to seldom discuss challenges with other siblings, or seek out information outside the family, like extended family or friends (Plumridge et al., 2011). Siblings can therefore be at risk of standing alone in dealing with the diagnosis if the parents don’t recognise the need for information in their healthy children.

4.2.1 Factors influencing sibling disorder knowledge

How much disorder knowledge children have, depend on different factors of the diagnosis. One of the factors is the incidence rate. Children are found to have more knowledge about more common diagnoses than less common diagnoses (Perrin & Gerrity, 1981). Knowledge also depends on the characteristics of the diagnosis, like the level of chronicity (Myant & Williams, 2005), or complexity (Vatne et al., 2015). Also, siblings of children with mental disorders, such as autism spectrum disorders, have been found to have less information about the diagnosis than siblings of children with primarily somatic diagnoses (Glasberg, 2000; Lobato & Kao, 2002).

A review by Mandleco and Webb (2015) found that siblings of children with autism
knew more about the disorder than siblings of children with Downs Syndrome. Furthermore, siblings learned what they knew from their parents, but the knowledge depended on the ethnicity of the family and sibling age. The study pointed out for healthcare workers and future practice that improving sibling knowledge was essential. The authors highlighted that when dealing with a family where one of the children have a diagnosis, it is important to be aware of sibling knowledge by providing information, clearing up misunderstandings and answering questions honestly (Mandleco & Webb, 2015). Additionally, an important task is to support parents in informing about the diagnosis in order to make sure that the information is age- and developmentally appropriate. It is also important to help make parents more comfortable in talking with siblings about the diagnosis.

The age of the sibling also seem to affect disorder knowledge. Age has been found to significantly predict understanding of the diagnosis (Myant & Williams, 2005) and children’s conception of complex diagnoses is found to develop with age (Glasberg, 2000; Mandleco & Webb, 2015; Paterson, Moss-morris, & Butler, 1999). Disorder knowledge can be divided into five subgroups or categories; i.e., 1) identity, name and symptoms, 2) cause, 3) treatment, 4) duration and timeline, and 5) consequences of the disorder (Vatne et al., 2015). It has been found that siblings develop an understanding of the consequences of the disorder later in life than the other categories (Glasberg, 2000). Vatne et al. (2015) found that 16.4% of siblings’ statements about rare disorders involved misconceptions. However, age did not explain any of the variance. The sample included ages ranging from 6 to 17 years. The authors speculated that based on what is known about the development of diagnosis understanding, rare diagnoses could consist of so many abstract and complex concepts that the understanding develops even later in life.

Glasberg (2000) interviewed 63 siblings of children with autism spectrum disorders using a measure that investigated siblings’ knowledge about the disorder. Parents were given the same interview, but were asked to predict how the sibling responded. Parents correctly predicted the siblings’ response for definition and etiology of the disorder, but overestimated siblings’ understanding of the implications of the disorder. Parents also predicted that the sibling would respond at a level more advanced than what would be expected based on the siblings age.

A recent review called for more research on the outcomes that are associated with knowledge and psychoeducation about diagnoses (Tudor & Lerner, 2015). Lack of knowledge, together with the fact that siblings do not discuss the diagnosis with anyone, can
lead to siblings misunderstanding, or confusion around the disorder (Vatne et al., 2015). Siblings learn about the disorder from their parents (Mandleco & Webb, 2015), and when explanations are not provided by parents, children tend to make up their own explanations (Tanaka, Uchiyama, & Endo, 2011; Vatne et al., 2015). The child-created explanations may be more stressful than the truth (Lobato, 1993).

In summary, the knowledge of siblings may depend upon the diagnosis, the information presented by the parent, or ethnicity of the family, and many other possible variables not yet investigated. It is therefore important to measure and include disorder knowledge in interventions to all siblings and further explore the relationships between knowledge and adjustment. Furthermore, it is important to include knowledge in the interventions because an increase in disorder knowledge has been found to lead to more positive outcomes for siblings, such as better coping (Meyer & Vadasy, 1994), higher perceived control, and less anxiety (Houtzager, Grootenhuis, & Last, 2001).
5 Intervention for siblings

The process of developing and optimizing interventions for siblings has begun, but several aspects of interventions have yet to be explored. Intervention studies include a wide variety in measured outcomes and included components. Outcomes like reduced anxiety, improved mood, and behavioural adjustment was found in a systematic review of studies with interventions for siblings (Hartling et al., 2014). However, the review emphasized that the results were not consistent across studies. Another review also note that the conclusions from studies of outcomes after intervention have been diverse (Tudor & Lerner, 2015), and calls for more systematic aproaches. One study found that siblings were found to have significantly lower score on the Strengths and Difficulties Questionnaire (Goodman, Meltzer, & Bailey, 1998) than the control group after intervention (Roberts et al., 2015). However, another study found no difference between the treatment group and control group from pre- to post intervention on outcomes like behaviour problems and self-concept (McLinden, Miller, & Deprey, 1991). Note that the study’s sample was very small (N=11).

Findings concerning what components should be included in the intervention are also under investigation, but in general, siblings has been found to benefit from information, support and involvement (McCaffrey, 2016). An intervention study with a randomized three group design, found that the group that included psychoeducational and psychosocial components was found to be more effective than the group that only focused on recreational activities (Williams et al., 2003).

When it comes to interventions and knowledge, it has been found that interventions for siblings increased sibling disorder knowledge, and increased perception of siblings own competence (Lobato & Kao, 2002, 2005). Lobato and Kaos (2002) Siblings Knowledge Interview (SKI), was used in the intervention SibLinks (Lobato & Kao, 2002). SibLinks was an integrated siblings-parent intervention, and the goal was to increase siblings’ understanding and adjustment. SibLinks entailed a focus on knowledge, managing emotions, problem solving and managing individual needs. SibLinks was offered to families at an outpatient service at a children’s hospital. The intervention focused on 8-13-year old siblings and included 54 siblings and their parents. The SibLinks consisted of 6 sessions over 6-8 weeks. The study found a significant increase in knowledge, competence and connectedness, and decreases in internalizing behaviour, externalizing behaviour, and negative adjustment in siblings (Lobato & Kao, 2002).
5.1 Including parents in interventions

According to the theoretical models presented in this thesis, the family is an important factor for sibling coping (Ajzen, 1988, 2002; Leng et al., 2016; Sommerschild, 1998). Sommerschild’s (1998) model links the closeness of the dyad and the family, directly to the self-worth and resilience of the sibling. Following this model, strengthening the family will therefore strengthen the sibling. The intervention in this thesis includes parents and siblings, and they have joint dialogue-sessions on disorder knowledge and sibling challenges. Parents are found to be siblings’ main source of knowledge, and therefore it is important that parents and sibling get to practice communication about knowledge and challenges. This could be some of the reason as to why outside sources of support, like sibling support programs are found to be a strong protective factor for siblings (Strohm, 2008).

The affect RTPB model illustrated that Family Communication Behaviour leads to Knowledge Sharing Behaviour. The family communication was an important determinant in increased knowledge, and following the affect RTPB model, the family, through family communication, supplies the knowledge about the diagnosis. Studies supports this, and find parents to be the main source of information about the diagnosis (Mandleco & Webb, 2015). In a study targeting siblings of children with a rare disorder (22q11.2 deletion syndrome), 87% of 23 siblings answered that they only got information about the diagnosis from their parents. Further inquiry revealed that although the sibling had a lot of information about the diagnosis, there were topics that were never discussed (Okashah, Schoch, Hooper, Shashi, & Callanan, 2015).

When children are given the information about a diagnosis it is also crucial that giving the information is considered a process and that the information given must be customized. The level and content of information needs to consider the developmental level of the child and the course of the illness (Plumridge et al., 2011). The inclusion of parents in the intervention also give parents support in conveying the information as a process to siblings, and could therefore be valuable.

Interventions have previously included only siblings in interventions, but there are multiple arguments for including parents in the interventions with siblings. Several studies emphasize the need for a parent to take part in the intervention (Lobato, 1990; Lobato & Kao, 2005; Tsao, Davenport, & Schmiege, 2012; Williams et al., 2003). Lobato and Kao (2002) suggested the family as the most important context for managing the diagnosis in the family, because the study found that parent and child adjustment are interrelated. Individual features
in parents such as higher levels of stress and more emotional difficulties predict more sibling adjustment problems (Baker, Blacher, Crnic, & Edelbrock, 2002; Baker et al., 2003). The fact that individual features in parents influence the sibling adjustment, indicates that including a parent or parents in the intervention is important. A review found that parents of children with CD was found to underestimate the needs of all other members of the family aside from the child with CD, including themselves (Verhaeghe, Defloor, Van Zuuren, Duynstee, & Grypdonck, 2005). Therefore, parents may also need support to acknowledge that the sibling has needs that are not met. Furthermore, they might need support in how to communicate, how to share knowledge, and how to talk about sibling challenges. The parents in the Lobato and Kao (2002) intervention also reported high satisfaction and had a 100% completion rate. The authors associated the satisfaction and completion rate with the reported great need for these interventions, and suggested that the interventions are of good assistance for the families (Lobato & Kao, 2002).

An intervention focusing on sibling knowledge and sibling relationship that did not include parents, found that scores on both knowledge and relationship still increased significantly from pre- to post intervention (Granat, Nordgren, Rein, & Sonnander, 2012). The theoretical foundations for arguing that interventions should still include both siblings and parents might be strong, but the empirical arguments are still being explored. Some of the empirical support might be in the fact that parents are found to pay more attention to the ill child than the sibling (Lobato, Miller, Barbour, Hall, & Pezzullo, 1991), and this can in turn be distressing for the sibling (McHale & Gamble, 1989). Getting more information about the individual risk-factors in siblings and parents, will be important for targeting the families most in need of intervention programs.

An additional argument for including parents in the intervention is the need for multi-informant reports. In a systematic review of literature concerning the impact on siblings of children with CD, the authors found that parents rated sibling health-related quality of life higher than the siblings’ self-reports. The authors stress that this emphasizes the importance of including siblings’ subjective perspective in studies with families with a child with CD (Limbers & Skipper, 2014). Similar results were found in a study where sibling self-report of problems was higher than the parent report of siblings problems, and this was also the case in reporting peer-problems (Rankin, Tomeny, & Barry, 2017). The literature reveals that parents and siblings do not have the exact same perspective on the CD, and that there is a discrepancy in the way sibling and parents report how the diagnosis affect the family (Guite, Lobato, Kao,
& Plante, 2004). Thus, it is important to ask both sibling and parent for their subjective assessment. A study used Strengths and Difficulties Questionnaire and had parents report siblings, and siblings report themselves (Rankin et al., 2017). The siblings that were considered “at risk” from the parent and sibling reports, where not identified as “at risk” by both siblings and parents. When siblings considered themselves as “at risk”, parents did not agree, and parents who considered their sibling “at risk”, were not supported by the siblings. The authors stress that this emphasizes the need for multi-informant reports.

5.2 The need for interventions

In June 2017, the Norwegian Parliament passed a proposal from the Government to amend the Law for Health Personnel (§10a) to require health providers to address siblings’ health needs (The Norwegian Parliament, 2017). The new law (§10a) states that health personnel shall contribute in safeguarding the need for information and necessary follow-up care that minors might need as a consequence of being a child with a parent, or sibling, who are a patient with mental illness, drug addiction or serious somatic illness or injury. This means Norwegian health providers are in urgent need of effective interventions for siblings. Unfortunately, there are no evidence-based sibling interventions (Tudor & Lerner, 2015). The intervention in this thesis is an attempt to meet this need, and the findings of this pilot-intervention will be informing the next step, the randomized controlled prevention intervention trial (RCT) in the Preventing Mental Health Problems in Siblings of Children with Neurodevelopmental Disorders (SIBS) project.

This thesis is based on a collaboration between Frambu Resource Centre for Rare Disorders and the Department of Psychology, University of Oslo. In addition, several patient user organizations are involved; the Autism Union, the Norwegian association for children with congenital heart disease (NACCHD), the Norwegian CP Union, and Ups and Downs (Union for Down syndrome), as well as the National Competence Centre for Neurodevelopmental Disorders and Hypersomnias (NevSom).
6 Research questions and hypotheses

**Research question 1**: Does the communication and knowledge increase from pre- to post intervention? It is hypothesized that the quality of communication will increase from pre- to post intervention. Secondly, it is hypothesized that the level of disorder knowledge in siblings will increase from pre- to post intervention. The model by Sommerschild (1998) is used as a theoretical basis for the hypothesized benefits of the intervention in this thesis. An increase in communication and knowledge would support the intervention explored in this thesis.

**Research question 2**: Does the quality of communication and level of disorder knowledge relate to each other? It is hypothesized that the quality of communication and level of knowledge are related to each other at pre- and post intervention. Secondly, it is hypothesized that the improvement in communication quality are related to the increase in disorder knowledge. A relationship between communication and knowledge would indicate a link between Sommerschild’s (1998) two main areas of coping, closeness and competence. A relationship would additionally support the affect RTPB model which is used as a model for how Family Communication Behaviour is related to Knowledge Sharing.

The analyses in this thesis is controlled for age of sibling. The thesis investigates how age relates to communication and knowledge. The literature on knowledge in siblings often show a strong relationship between knowledge and age (Myant & Williams, 2005).
7 Method

7.1 Participants

The sample comprised 101 siblings of a child with CD from 98 families. The age of siblings ranged from 8 to 16 years ($M = 11.5, SD = 2.06$) and 54.7% of siblings were girls. In the sample, 75.5% of siblings lived with both parents.

The inclusion criteria were: a) being aged between 8 and 16 years, b) having a brother or sister with a rare disorder; and c) absence of developmental disorder. See Table 2 for overview of the different diagnosis in the thesis.

Table 2

<table>
<thead>
<tr>
<th>Sibling diagnosis</th>
<th>n</th>
</tr>
</thead>
<tbody>
<tr>
<td>Smith-Magenis syndrome</td>
<td>5</td>
</tr>
<tr>
<td>Angelman syndrome</td>
<td>2</td>
</tr>
<tr>
<td>Duchenne muscular dystrophy</td>
<td>8</td>
</tr>
<tr>
<td>Autism spectrum disorders</td>
<td>26</td>
</tr>
<tr>
<td>Spinal muscular atrophy</td>
<td>3</td>
</tr>
<tr>
<td>Congenital heart disease</td>
<td>12</td>
</tr>
<tr>
<td>Congenital muscular dystrophy</td>
<td>4</td>
</tr>
<tr>
<td>Neurodegenerative diseases</td>
<td>3</td>
</tr>
<tr>
<td>47 XXY syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Cerebral palsy</td>
<td>3</td>
</tr>
<tr>
<td>Velocardiofacial syndrome</td>
<td>7</td>
</tr>
<tr>
<td>Fragile X syndrome</td>
<td>4</td>
</tr>
<tr>
<td>Limb-girdle muscular dystrophy</td>
<td>1</td>
</tr>
<tr>
<td>Neuronal migration</td>
<td>1</td>
</tr>
<tr>
<td>Chromosome 5q deletion syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Progressive central nervous system deterioration</td>
<td>1</td>
</tr>
<tr>
<td>Osteogenesis imperfecta</td>
<td>1</td>
</tr>
<tr>
<td>Neurofibromas type 1</td>
<td>1</td>
</tr>
<tr>
<td>Cri-du-chat syndrome</td>
<td>1</td>
</tr>
<tr>
<td>Rett syndrome</td>
<td>1</td>
</tr>
<tr>
<td>22q11.2 duplication</td>
<td>1</td>
</tr>
<tr>
<td>Humoral immune deficiency</td>
<td>1</td>
</tr>
<tr>
<td>Noonan syndrome</td>
<td>2</td>
</tr>
<tr>
<td>Down syndrome</td>
<td>7</td>
</tr>
<tr>
<td>Hereditary ataxias</td>
<td>1</td>
</tr>
</tbody>
</table>
7.2 Procedures

This thesis is part of an open pre-post trial of a manualized sibling-parent intervention. Participants were recruited from Frambu Resource Centre for Rare Disorders, the National Competence Centre for Neurodevelopmental Disorders and Hypersomnias (NevSom), or through one of the collaborative nation user organizations (i.e., the Autism Union, the Norwegian association for children with congenital heart disease (NACCHD), the Norwegian CP Union (Union for Cerebral Palsy), and Ups and Downs (Union for Down syndrome)). Families who had applied and been accepted for one of these courses or camps, were approached and invited to take part in the study. Families provided written consent. In total, 198 siblings and their parents were approached to take part in the study. Out of these, 57.6% accepted, 13.1% declined and 26.8% did not respond to the invitation. In six cases (2.5%), it was not registered if the families declined or did not answer. Of the 114 families that accepted, 16 families did not partake in the intervention for different reasons, which left a total of 98 families who attended the intervention.

Only one parent from each family took part in the intervention with the sibling, and there were two reasons for this. One reason was the fact that there was a parallel family program at the intervention-sites, so one parent had to take part in the intervention and the other parent had to take part in the family program with the child with CD. Secondly, the experience from the pre-pilot study was that the power balance could be uneven with both parents present with the sibling in the joint sessions. The only exception was if two siblings from the same family participated. In these cases, both parents were encouraged to participate. The participating parent was encouraged to discuss and share the content of the intervention with the non-participating parent, but it was not examined to what extent this was complied with. The families decided which parent would participate in the intervention with the sibling. The same parent had to participate throughout the intervention, meaning that the parents could not switch in between sessions.

Data included both audio recorded standardized interviews and self-report questionnaires. Disorder knowledge was measured with the recorded standardized interviews and communication was measured with questionnaires. Both outcome variables in this thesis were measured before the intervention (pre), as well as three months after intervention (post). Data was collected from 2014 to 2016, coding of the SKI from 2016 to 2017, and the analysis was performed in 2017.
## 7.3 The intervention

The intervention in this thesis targets disorder knowledge, child and parent communication about the diagnosis, and the challenges faced by the siblings. The intervention comprises five group sessions, of which three are parallel sessions in which parents and siblings are in separate groups, and two are joint sessions in which parents and siblings interact. See Table 1 for a detailed description of the intervention.

### Table 1

**Overview of the intervention and session aims**

<table>
<thead>
<tr>
<th>Session 1 Introduction (20mins) – sibling and parents in separate groups</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>S:</strong> Warm-up exercises. Psychoeducation about the intervention. Establish group rules.</td>
</tr>
<tr>
<td><strong>P:</strong> Warm-up exercises. Psychoeducation about the intervention.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Session 2 Knowledge Module (60 mins) – siblings and parents in separate groups</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>S:</strong> Explore children’s knowledge of their siblings’ disorder. Use of a semi-structured group interview on characteristics, prognosis, cause, and impact of the CD. Group selects one question about the disorder to present for the parent group. Each participant writes individual questions to present for their parent in session 3.</td>
</tr>
<tr>
<td><strong>P:</strong> Communication training in how to talk with children about the diagnosis. Use of video examples and the question from the sibling group.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Session 3 Sibling-Parent Dialogue on Knowledge (20-60 mins) * - sibling and parent joint</th>
</tr>
</thead>
<tbody>
<tr>
<td>A sibling-parent dialogue based on the questions posed by the sibling</td>
</tr>
<tr>
<td>Individual feedback from the group leader</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Session 4 Emotion and Coping Module (60 mins) – sibling and parents in separate groups</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>S:</strong> Semi-structured interview exploring children’s emotions, coping strategies, and challenges regarding their sibling and family life. Group selects one challenge to be presented for the parent group. Each participant writes individual challenges to present for their parent in session 5.</td>
</tr>
<tr>
<td><strong>P:</strong> Continued training in communicating with children about the diagnosis using video examples and the challenge presented from the sibling group.</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Session 5 Sibling-Parent Dialogue on Challenges (20-60 mins) * - sibling and parents joint</th>
</tr>
</thead>
<tbody>
<tr>
<td>A sibling-parent dialogue based on the challenges presented by the sibling</td>
</tr>
<tr>
<td>Individual feedback on communication from a group leader</td>
</tr>
</tbody>
</table>

*Note.* S=Sibling group. P=Parent group. Mins=Minutes *Optional duration for participants, min 20 minutes, max 60 minutes.*
The intervention has a knowledge-module inspired by the intervention by Lobato and Kao (2002) and an emotion-module inspired by the CBT (cognitive behavioural therapy) intervention “Friends for life” (Barret, 2005). The last four sessions were conducted over three to four days. The sessions were led by a trained psychologist, advanced psychology students and staff with special education who were under supervision of the project group. Each group had one group leader and one facilitator. The sessions were conducted at Frambu, or at the centres for the different user organizations.

7.4 Measures

7.4.1 The Parent-Child Communication Scale

The Parent-Child Communication Scale (PCCS; Krohn, Stern, Thornberry, & Jang, 1992; McCarty & McMahon, 2003) was used to measure quality of family communication. In the current thesis, the PCCS was used to measure the subjective reported sibling-rated communication in the family. The PCCS comprises 10 items for siblings, where siblings report how open their mothers and fathers are in their communication with them (see Appendix C). The PCCS is scored from 1 (almost never) to 5 (almost always). Seven of the ten items are reversed, with higher scores representing poorer communication. The sibling completed the PCCS at pre- and post intervention both for mother and for father.

Respectable to very good levels of reliability (DeVellis, 2012) has been demonstrated for the PCCS, with Cronbach’s alpha coefficients ranging from $\alpha = .70$ to $\alpha = .89$ (Larrañaga, Yubero, Ovejero, & Navarro, 2016; McCarty & McMahon, 2003; Offrey & Rinaldi, 2017). In this thesis, the inter-item reliability of the PCCS indicate a respectable to very good internal consistency (DeVellis, 2012). See Table 3 for reliability and frequency of answers of the PCCS.

Table 3

<table>
<thead>
<tr>
<th>PCCS Questionnaires</th>
<th>PRE</th>
<th>POST</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n</td>
<td>Cronbach’s Alpha</td>
</tr>
<tr>
<td>PCCS communication with mother</td>
<td>101</td>
<td>.79</td>
</tr>
<tr>
<td>PCCS communication with father</td>
<td>101</td>
<td>.81</td>
</tr>
</tbody>
</table>

*Note. PCCS=Parent-Child Communication Scale. n=number of participants*
7.4.2 The Sibling Knowledge Interview

The structured quantitative interview and scoring system the Sibling Knowledge Interview (SKI; Lobato & Kao, 2002) was used to measure siblings’ knowledge about their sister or brothers’ diagnosis. The SKI measures three different aspects of sibling disorder knowledge, that is, the name of the diagnosis, their ability to explain the diagnosis, and their understanding of the etiology of their siblings’ diagnosis. The interview was conducted at the intervention site, or over the phone. All the interviews were audio taped.

The interview generated two different variables; name and explanation. The SKI Name (SKI-N) variable was coded by assessing siblings’ ability to name their brother or sisters diagnosis. The scoring ranged from 1 to 3, were 1 represented incorrect or no answer, 2 represented a vague but correct naming of the disorder, and 3 represented that the sibling used the specific and correct term.

The SKI Explanation (SKI-E) variable was scored from 1 to 5, where 1 and 2 represented no answer or total misunderstanding of disorder, 3 were vague but correct, 4 were partial understanding and was defined as being able to identify the core symptoms. The score 5 represented an accurate understanding, defined as clearly identifying the core symptoms of the disorder. The interview included four follow up questions in addition to “Can you explain (the disorders name) to me?” where a score of 5 reflected that at least 3 of these were answered correctly (see Appendix D). In this thesis, only the explanation-variable was used, as this was found to be the most valid measure of sibling disorder knowledge.

The coding of the SKI was conducted by three advanced students of psychology, including the author of this thesis. The main features of all the diagnoses in the thesis were identified and written up as a checklist by the student coders. These checklists were approved by the psychologists experienced with the disorders, and used to guide the quality of the coding. The coding of pre-data followed the following procedure: The first five interviews were done by all three coders together to agree on the scoring. These scorings were double-checked and approved by the psychologists. The coders then coded the same ten interviews separately, to test inter-rater reliability. The reliability of the inter-rater reliability of the SKI was measured with intraclass correlations coefficients (ICC), based on a mean-rating (k=3), absolute agreement, mixed-effect model. The ICC estimates was ICC (name) = 1.00 and ICC (explanation) = .75, which represents a good to excellent interrater reliability (Cicchetti, 1994). After coding ten different interviews separately, twelve interviews were double coded to test the reliability of our coding, with ICCs (name) = 1.00 and ICC (explanation) = .87,
which represent excellent interrater reliability. Then the remaining interviews were coded separately, twelve each, which gave a total of 92 interviews.

The coding of post-data were coded by the same three coders with 10% of the interviews double coded for reliability. The ICC (name) = 1.00 and ICC (explanation) = .68 which represent good to excellent interrater reliability (Cicchetti, 1994). The SKI was completed by 92 siblings at pre, and 81 siblings at post.

7.5 Data Analysis

The data analyses were conducted with IBM SPSS 24.0 and included descriptive analyses, pre-post t-tests (paired sample), and Pearson’s \(r\)-correlation analysis.

Cohen’s \(d\) (Cohen, 1988) was used as the computed effect size of the paired sample t-tests in this thesis. The Cohen’s \(d\) for the paired sample t-tests was computed with the formula: Cohen’s \(d = (M_2 - M_1) / SD\). Also, the calculations corrected for dependence among means by adding the correlation of the two means so that Morris and DeShon’s (2002) equation 8 could be applied. The interpretations of the Cohen’s \(d\) values used in this thesis were; 0.20 as small, 0.50 medium, and 0.80 as a large effect size (Cohen, 1988).

The relationship between the different variables in this thesis were described through Pearson’s correlation coefficient, \(r\). Interpretations of the Pearson’s \(r\) values used in this thesis are; 0.10 as small, 0.30 medium and 0.50 as a large effect size (Cohen, 1992).

The PCCS scores represent the sibling-rated communication with mother and sibling-rated communication with father. The variables representing the PCCS scores are therefore named communication with mother and communication with father.

Both the PCCS and the SKI did not exist in Norwegian, and was translated by the project group. A back-translation to English was then performed by a native English speaker. The new English version was then compared to the original version, and approved by the authors of the measurements. The SKI scoring guide was only used by the coders, and was kept in English.

7.5.1 Thesis plan

Table 4 show an overview of the different variables in the paired sample t-tests in this thesis. The t-tests are performed to answer the first research question with the two first hypotheses concerning the assumed improvement in communication and knowledge from pre-
to post intervention.

The variables in Table 4, together with age of sibling and the computed change-variables of communication with mother, communication with father and SKI, were analysed with Person’s correlations. This analysis was performed to answer the second research question with the two hypotheses concerning the assumed relationship between communication and knowledge.

Table 4

*The set-up of the design of the paired T-tests in the thesis with the three variables communication with mother, communication with father and SKI explanation at pre-and post.*

<table>
<thead>
<tr>
<th>Variables</th>
<th>Pre</th>
<th>Post</th>
</tr>
</thead>
<tbody>
<tr>
<td>Communication with mother</td>
<td>PCCS child about mother</td>
<td>PCCS child about mother</td>
</tr>
<tr>
<td>Communication with father</td>
<td>PCCS child about father</td>
<td>PCCS child about father</td>
</tr>
<tr>
<td>The SKI explanation</td>
<td>SKI-E</td>
<td>SKI-E</td>
</tr>
</tbody>
</table>
8 Results

8.1 Descriptive statistics

8.1.1 Parent communication

The PCCS is the sibling’s ratings of communication with mother (communication with mother) and with father (communication with father) at pre- and post. The means, including standard errors of the mean, are presented in Figure 3. Lower values on the PCCS, reflect better rating of communication.

![Figure 3](image)

*Figure 3.* Mean values for PCCS for communication with mother and communication with father at pre- and post. Bars represent standard error of the mean.

8.1.2 Disorder knowledge

The SKI Explanation (SKI-E) variable measured disorder knowledge, and higher scores reflect more knowledge. See Figure 4 for the means and standard error of SKI scores at pre- and post.

The SKI-E variable was skewed at both pre- and post, with low scoring frequency for lower scores compared to higher scores (see Appendix A for details).
8.2 Analyses of pre-post change

8.2.1 Change in communication

A paired t-test was performed for both communication with mother and communication with father, to investigate if the communication with mother and communication with father had changed from pre- to post. The t-test showed that there was a significant improvement for communication with mother from pre- \( (M = 2.08, SD = 0.59) \) to post, \( (M = 1.82, SD = 0.57) \), \( t(71) = 4.01, p < .001, d = 0.47 \) (two tailed). However, communication with father only showed a tendency of the same relationship from pre- \( (M = 2.25, SD = 0.68) \) to post \( (M = 2.13, SD = 0.75) \), \( t(69) = 1.71, p = .091, d = 0.21 \) (two tailed). The effect sizes of the change in communication with mother was small (almost medium), and the change in communication with father was small.

The quality of communication, as rated by siblings, were also explored by investigating the frequency of the data, the cumulative percent of the distribution. Two levels were created, the scores 1-2.5 were top range, and 2.5 - 5 were low range. Communication with mother was rated in the top range by 74.3% (pre) and 82.4% (post) of the siblings. Communication with father was rated in the top range by 64.4% (pre) and 73.6% (post) of siblings. For frequency distribution of scores, see Appendix B.
Table 5

Means and standard deviations for the paired t-tests from pre- to post; communication with mother, communication with father, and SKI-E.

<table>
<thead>
<tr>
<th>Variables</th>
<th>PRE</th>
<th>POST</th>
<th>t</th>
<th>df</th>
<th>p</th>
<th>Cohens’ d</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M</td>
<td>SD</td>
<td>M</td>
<td>SD</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Communication with mother</td>
<td>2.08</td>
<td>0.59</td>
<td>1.82</td>
<td>0.57</td>
<td>4.01</td>
<td>.000</td>
</tr>
<tr>
<td>Communication with father</td>
<td>2.25</td>
<td>0.68</td>
<td>2.13</td>
<td>0.75</td>
<td>1.71</td>
<td>.091</td>
</tr>
<tr>
<td>SKI-E</td>
<td>3.70</td>
<td>0.95</td>
<td>3.97</td>
<td>0.83</td>
<td>-2.81</td>
<td>.007</td>
</tr>
</tbody>
</table>

*Note.* M = Mean, SD = Standard Deviation, df = degrees of freedom, SKI-E = Sibling Knowledge Interview – Explanation.

8.2.2 Change in disorder knowledge

A paired t-test was used to explore if level of knowledge in siblings had changed from pre- to post. The level of knowledge in siblings increased significantly from pre- to post (see Table 5). The effect size for change in knowledge was small.

8.3 Relationship between communication and knowledge

8.3.1 The relationship between communication and knowledge

See Table 6 for the Pearson’s Correlations between age, the communication variables, and the knowledge variables at pre- and post intervention. The four communication variables (i.e., with mother/father at pre/post) were significantly correlated (all $p < .001$) with large effect sizes. However, correlations between the four communication variables and knowledge were small and non-significant. The correlation between pre- and post measures of knowledge were large and significant ($p < .001$).

The correlations between SKI-E and age of sibling were medium both at pre- and post (see Table 6). Knowledge increased with higher age. Correlations between age and the communication variables, were small and non-significant.
Table 6

*Pearson Correlations between age of sibling, and the SKI-E, communication with mother, and communication with father at pre- and post.*

<table>
<thead>
<tr>
<th></th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
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<tbody>
<tr>
<td>1. AGE</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Pre SKI-E</td>
<td>.43**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. Post SKI-E</td>
<td>.35**</td>
<td>.57**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Pre communication with father (1)</td>
<td>.14</td>
<td>.18</td>
<td>.08</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Post communication with father (2)</td>
<td>.15</td>
<td>.11</td>
<td>.04</td>
<td>.64**</td>
<td>1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Pre communication with mother (3)</td>
<td>.00</td>
<td>-.04</td>
<td>-.15</td>
<td>.74**</td>
<td>.56**</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>7. Post communication with mother (4)</td>
<td>.19</td>
<td>.14</td>
<td>-.03</td>
<td>.52**</td>
<td>.66**</td>
<td>.56**</td>
<td>1</td>
</tr>
</tbody>
</table>

*Note.* SKI-E = Sibling Knowledge Interview – Explanation **. Correlation is significant at the 0.01 level (2-tailed).

### 8.3.2 The relationship between change in communication and knowledge.

Three change variables were computed; i.e., change communication with father, change communication with mother, and change SKI-E. The change variables were computed to investigate if the change in communication was related to the change in knowledge. The correlation between change communication with father and change SKI-E was small and non-significant (*p* = .609). However, the correlation between change communication with mother and change SKI-E was significant (*p* = .006), with a medium sized correlation. See Table 7.

The relationship was negatively correlated which indicates that the more the level of measured disorder knowledge (SKI-E) increases, the more the rating of communication increases, but only for mother.
Table 7

Pearson Correlations between the change variables and age of sibling.

<table>
<thead>
<tr>
<th></th>
<th>Change in communication with mother</th>
<th>Change in communication with father</th>
<th>Change SKI-E</th>
<th>AGE</th>
</tr>
</thead>
<tbody>
<tr>
<td>Change in communication with mother</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Change in communication with father</td>
<td>.49**</td>
<td>1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Change SKI-E</td>
<td>-0.35**</td>
<td>-.07</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>AGE</td>
<td>-0.18</td>
<td>.02</td>
<td>-0.11</td>
<td>1</td>
</tr>
</tbody>
</table>

*Note.* SKI-E = Sibling Knowledge Interview – Explanation **. Correlation is significant at the 0.01 level (2-tailed).

The improvements in communication were only significant for mother, and only the change in communication with mother were significantly related to change in knowledge. To investigate further the difference in siblings’ rating of mother and father, the sibling-evaluated communication with mother and communication with father was analysed using paired sample t-tests. A paired sample t-test was used because it was a within-subject comparison between the siblings rating of the mother and the siblings rating of the father. The t-test showed that the sibling-evaluated communication with mother \((M = 2.17, SD = 0.63)\) and father \((M = 2.33, SD = 0.67)\) were significantly different at pre-intervention, \(t(98) = 3.45, p = .001, d = 0.35\) (two tailed). The sibling-evaluated communication with mother \((M = 1.88, SD = 0.62)\) and father \((M = 2.15, SD = 0.75)\) were also significantly different at post intervention, \(t(69) = 3.93, p < .001, d = 0.48\) (two tailed). Siblings rated communication with mother significantly better than communication with father, at both pre- and post intervention. The effect sizes for the difference in sibling-evaluated communication with mother and father at pre-and post intervention was small (almost medium at post).

In summary, communication with mother was rated to be significantly better from pre- to post, but only a tendency for communication with father of the same relationship. The level of knowledge in siblings significantly increased from pre- to post. Change in communication with mother was significantly correlated to the change in sibling knowledge (SKI-E). The increase in quality of communication with mother was related to the increase in
level of measured disorder knowledge. Additionally, siblings rated communication with mother better than communication with father at both pre- and post intervention, and the overall quality of rated communication was good.
9 Discussion

9.1 Analysis of pre-post change

The first research question involved the pre-post change in sibling-rated communication with mother and communication with father, and change in the level of disorder knowledge in siblings.

9.1.1 Pre-post change in communication

The first hypothesis assumed that siblings’ subjective ratings of communication with their mother and father would improve from pre- to post intervention. This hypothesis was largely supported. There was a significant increase in communication from pre- to post intervention, but only for communication with mother.

Improvement in communication pre-post intervention has not been given much research focus in previous intervention studies for siblings. One study with siblings of children with cancer found no difference in communication (Sahler & Carpenter, 1989). The study investigated communication with a 4 item-domain in a questionnaire about siblings’ perceptions at pre- and post intervention. There were no parents who took part in the intervention, and no explicit focus on communication between siblings and parents. Another study found no difference in communication pre- to post intervention with a 10-item questionnaire to measure problem-solving communication. However, the authors emphasized further development of the intervention in the study, i.e., including activities that promoted greater discussion between siblings and parents (Giallo & Gavidia-Payne, 2008). It might be essential that sibling-parent communication is included in the intervention for increasing quality of communication in families.

Only communication with mother improved significantly from pre- to post intervention, and some of the explanation for this could be due to the fact that there were almost twice as many mothers (n=67) than fathers (n=34) participating in the intervention. The finding could strengthen the support for the intervention, but due to the skewness of the variable, this was not explored. Nevertheless, the assumption is that the intervention was the cause of the increase in sibling-rated communication, and with more fathers participating, it
could be expected that the siblings-rated communication would increase for that group as well.

9.1.2 Pre-post change in knowledge

The second hypothesis assumed that siblings’ level of disorder knowledge would improve from pre- to post intervention. This hypothesis was largely supported. There was a significant increase in the level of sibling disorder knowledge from pre- to post intervention. The finding in this thesis, that the disorder knowledge improves from pre- to post intervention, is in line with previous studies (Hartling et al., 2014; Tudor & Lerner, 2015). This means that interventions are a much-supported way of increasing sibling disorder knowledge.

This intervention emphasized disorder knowledge through open questions and discussing challenges that may arise from having a sister or brother with a CD, but it did not include explicit psychoeducation about the disorder. The finding of increase in knowledge in this thesis, could therefore indicate that the role of the parent is essential as the component who increases the level of disorder knowledge in siblings.

The general level of disorder knowledge in the current sample was slightly lower than the level of knowledge found in Lobato and Kao study (2002) using the SKI. The mean scores for the Lobato and Kao study were higher than the mean score in this thesis at both pre- ($M = 4.0, SD = 0.89$), and post ($M = 4.5, SD = 0.69$) (the mean scores in this thesis was ($M = 3.70, SD = 0.95$) at pre- and ($M = 3.97, SD = 0.83$) at post intervention). This might suggest that the siblings in the Lobato and Kao study had a higher level of knowledge than the siblings in this thesis, especially at post intervention. Alternatively, this could be due to the fact that the intervention in the Lobato and Kao study had a longer duration than the intervention in this thesis. The intervention in the Lobato and Kao study included 6 x 90 minute sessions over a period of 6 - 8 weeks. Also, the post-intervention data in the Lobato and Kao study was collected only 2 weeks after the last group session (compared to 3 months in this thesis), and the siblings might therefore remember more of the knowledge. To support this assumption, a smaller follow-up group in the Lobato and Kao study (n = 20) did a new interview after 3 months, and the new mean ($M = 4.2, SD = 0.88$) reduced the difference of the means between the two studies. On the other hand, it could also be that the difference between the studies point to differences in coding between the coders in the Lobato and Kao study and the coders in this thesis. Although the coders in this thesis showed good inter-rater
reliability, it was not measured against the coders in the Lobato and Kao study.

Testing effects, like the learning effects of taking a test more than once could be of importance to consider since the siblings completed the SKI more than once. If siblings got learning feedback from the interviewer there could be a testing effect in the SKI data, but this was not the case. The interviewer was instructed to only ask for what the sibling knew, and gave no information about the disorder. It could be a testing effect on the scores of knowledge if they got bored from doing the same interview twice, but the interview lasted only about five minutes. It is also considered unlikely that boredom could be influential on the measured change, since siblings got better from pre- to post.

The hypothesis assumes that the change in communication and knowledge are due to the intervention, but the possibility that the change might be a result of a confounding variable must be considered. According to the affect RTPB model, social/ family collaboration behaviour is also important for knowledge sharing. The intervention might increase this factor, and this again could be what influenced the change in knowledge. Also, the effect of unplanned events between pre- and post measurements is a possible threat to internal validity for the one-group pre-post design in this thesis. The difference in measurement from pre- to post could be due to the intervention, or to history. This threat might not be that influential, due to the short period of time between the pre- and post-test. Only three months makes it less probable, but it is not impossible that something happened to influence the data.

The increase in communication and knowledge in this thesis represents important support for the intervention in this study. The intervention is largely focused on communication and knowledge based on the Sommerschild (1998) model. According to this model, this will lead to increased self-worth and resilience. The measures of self-worth and resilience are beyond the scope of this thesis, but the first step, increasing the quality of communication and level of knowledge, is supported.

9.2 The relationship between communication and knowledge

The second research question explored the relationship between communication and knowledge, both at pre- and post, and the change in communication and knowledge from pre- to post. The pre-measures and post-measures for communication and disorder knowledge were collected at the same time, so there was no basis for analysing data searching for causal
9.2.1 The relationship between communication and knowledge

The third hypothesis was that there would be a relationship between the measures of communication and disorder knowledge at pre- and post. This hypothesis was not supported. The siblings rating of quality of communication at pre- or post did not relate to the siblings measured level of disorder knowledge. The different measures of communication correlated significantly with each other, as did the two measures of knowledge, but no relationship was found between communication and disorder knowledge.

The explanation for this finding could be that the way the communication was measured in this thesis did not manage to capture the type of communication that siblings have about diagnoses. The measure of communication in this thesis does not explicitly mention or measure communication about disorder, only general communication quality. Furthermore, this finding could be explained by the fact that sibling-rated communication with mother and father was generally rated quite high. The quality of communication was found to be good, and siblings reported good over-all communication with their parents. The good rating of communication could reflect that siblings in this group are genuinely experiencing good communication with their parents. Still, this finding was in contrast with research showing the communication to be poorer in families with a child with CD than in families with typically developing children (Murphy et al., 2017). However, this may be due to the fact that the studies in the review by Murphy et al. (2017) measured communication through objective observation, and this thesis measured communication through subjective questionnaires. The use of only questionnaires in studies of communication are criticized by researchers for only having data on the subjective perspective of communication (Murphy et al., 2017). There might be some complicating factors with subjective report, and the inclusion of a measure based on subjectivity can result in a biased result. The questionnaire used in this thesis was not retrospective in nature, so the challenges of retrospective report or limited recall was avoided, but the results can be biased by social desirability. Siblings were asked to rate their communication with their parents, and this could be a challenging topic to be honest about. It could be that siblings are reluctant to display discontent with parents in an unfamiliar context, especially since parents take part in the intervention. Still, only sibling’s subjective rating of communication is interesting in this thesis. It was more important to the thesis to
know if the siblings were happy with communication than if the communication was objectively good. The results show that the communication was rated to be good in general.

Finally, some of the difference in rated communication may be due to cultural differences. Most of the studies in the review by Murphy et al. (2017) was American. The socioeconomic status is an important factor in family functioning (Mulroy et al., 2008), and would therefore most likely be important for communication quality. Norway has a good welfare system which possibly provide families with a child with CD with more care, economic support, and advice than many other countries in the world. Cultural differences were investigated in a study comparing Latina and Anglo mothers, and found that culture played a part in how families communicated, and what they communicated about (Lauderdale-Littin & Blacher, 2017). Further, one of the differences between the groups was that Anglo mothers, less often than the Latina mothers, planned for the siblings to take a caregiver role in the future. This could indicate that siblings from different cultures are expected, by their parents, to take different levels of responsibility for the child with CD. This in turn, could vary the degree to which siblings perceive this as a burden or a stressor.

9.2.2 The relationship between the change in communication and knowledge

The fourth hypothesis was that the change in communication would be related to the change in disorder knowledge. This hypothesis was supported. The change in communication with mother correlated negatively and significantly with the change in knowledge. This indicates that the more the communication improved, the more knowledge improved from pre- to post, or the more knowledge improved, the more communication improved, but only communication with mother.

There are not any existing empirical intervention studies for siblings of children with CD that investigate the relationship between communication and knowledge. The empirical findings in this thesis support a relationship between communication and knowledge.

The communication with mother was rated significantly higher than communication with father at pre- and post intervention. Additionally, only the communication with mother changed significantly from pre- to post intervention and was significantly related to the change in communication. These findings are assumed to be due to the difference between the participation of mothers and fathers in the intervention, but the significant difference between communication with mother and communication with father pre-intervention would still need
an explanation. One perspective is that men and women talk differently. Women are more likely to use language to form and maintain connections with others, and men are more likely to use language to assert their independence and to achieve utilitarian goals (Philips, 1980; Tannen, 1994). Studies also show that mothers and fathers talk differently to their children (Gleason, 1987). A meta-analysis found that mothers talk more with their children, and use more supportive speech (Leaper, Anderson, & Sanders, 1998).

General high sibling-satisfaction with parents’ communication, and especially mothers’ communication, was found in a study by Jackson et al. (1998). Another study by Barnes and Olson (1985) found that adolescents reported having difficulty in communication with both parents, but indicated more positive interactions, i.e., more open communication with their mothers than with their fathers. Overall, the adolescents were more negative reporting on communication than their parents (Barnes & Olson, 1985).

The relationship between the increase in communication quality and disorder knowledge are in line with the affect RTPB model. An increase in quality of communication was linked to an increase in disorder knowledge. However, there was no relationship between the quality of communication and level of disorder knowledge at neither pre- nor post intervention. This could be due to general knowledge and disorder knowledge being different concepts and would therefore not respond to the quality of communication in the same way. Disorder knowledge about the diagnoses in this thesis are often very complex knowledge. According to the affect RTPB model, the more you believe what you know about what you share, and the more you believe you will be able to share, and the more likely it is that you will share the knowledge. The CDs in this thesis are complex, often rare, and we do not always know everything about the etiology or manifestation. This makes it harder for parents of children with CD to believe that they have a good understanding of their child’s disorder, and therefore it would influence if they share it. The other argument is linked to the fact that many of these diagnoses are genetic and parents can have reservations against sharing because of insecurity in how to address future implications for siblings. This would affect their belief about if they can share it with the sibling, which, according to the affect RTPB model, would affect the behaviour of sharing. The theoretical foundation in this thesis suggest a causal link between communication and knowledge, with communication leading to knowledge sharing. The empirical data in this thesis supported the link between communication and knowledge, but make no assumption about the direction of this relationship.
9.2.3 Age of sibling

Age was investigated as a possible influence on communication and knowledge, and correlated significantly with knowledge. Several studies have found a strong relationship between knowledge about illness and age (Glasberg, 2000; Myant & Williams, 2005). The study by Myant and Williams (2005) investigated children’s understanding of diagnoses with four age groups ranging from 4 to 12 years of age. Significant effects of age in the children’s explanations of illness were found, which became more sophisticated and accurate with increased age.

The theories of Piaget (1969) for the development of cognition in children (Piaget, 1969) represents a relevant framework for understanding children’s development of illness understanding (Perrin & Gerrity, 1981). It would therefore be expected that age and knowledge about the diagnosis would correlate, and in this thesis, it did. It was investigated if the significant relationship that was found between change in communication and knowledge could be explained by age, but this was not the case.

9.3 Methodological considerations

9.3.1 The SKI - Explanation variable

The SKI-E variable consisted of scores from 1 to 5, and the variable was used although it had an uneven distribution at both pre- and post (see Appendix A). A bigger sample size would be desirable for better variation in the scores.

Some of the explanation for the skewness of this variable could be in the scoring system of the SKI. The qualitative score of 1 is “don’t know” or “no response”, a score of 2 is “total misconception of disorder”, a score of 3 is “vague, but correct’. Therefore, if the sibling said anything about the diagnosis that was correct, they got a score of 3. “Total misunderstanding of diagnosis” may represent a valid value, but it does not necessarily represent a higher level of knowledge than “don’t know”. To get more variability in the scores it could be a solution to reassign new criteria for the three lower scores. A score of 1 could be “don’t know”, “no response”, or “completely misunderstanding of the disorder”. A score of 2 could be “very vague, one or two correct statements”. and a score of 3 could be “vague, but three or more correct statements”, The score of 4 as “partly understanding of the disorder” and score of 5 to be “accurate understanding of the disorder” could be kept.
9.3.2 Exclusion of the variable SKI - Name

The SKI - Name (SKI-N) of the interview was excluded from the analyses in this thesis. The reason was partly the variability in the name of the diagnosis. The names vary from terms like “Autism” to “Chromosome 5q deletion syndrome” and that raises a question if the variable measures knowledge, or if it also measures the complexity of the name of the diagnosis. The SKI name-variable was excluded in the analysis, and the SKI explanation-variable was kept. The SKI explanation was a measure of how much siblings knew about their brother or sisters’ diagnosis and therefore a better measure of sibling disorder knowledge for the hypotheses in this thesis.

9.3.3 Type of disorder

This thesis includes several different diagnoses. The challenges in the different diagnoses vary from profound cognitive challenges to normal cognitive functioning but with prominent somatic challenges. All the diagnoses in the thesis are chronic, but they differ in how rare they are, and the amount of information or knowledge that exists. As an example, it can be problematic to compare a sibling’s knowledge about Downs syndrome to a different diagnosis where the cause is not known. Differences between disorders were not investigated in this thesis, due to the large number of disorders included. Comparisons would therefore involve very small groups. Difference in outcomes across disorders represent a subject which deserves further scrutiny in future research (Hartling et al., 2014).

Reviews of intervention studies for siblings of children with CD note a wide array of different diagnoses across studies (Tudor & Lerner, 2015). Different diagnoses, from diabetes to autism spectrum disorders, are all categorized as CD. The review by Tudor and Lerner (2015) included 16 studies, and only one found differences between the diagnoses in the reported intervention outcomes. The explanations for this varied. Some studies only had one diagnosis in their sample, like autism spectrum disorder (Cooke & Semmens, 2010) and some did not differentiate between diagnoses. One study included five different diagnoses; cystic fibrosis, diabetes, spina bifida, cancer and developmental disabilities. The parents rated the severity of the condition and the ratings were validated by nurses. No statistically significant difference in severity between the diagnoses was found (Williams et al., 2003).

Other studies have found that type of diagnosis impact intervention outcomes. One study found that autism spectrum disorder and developmental disorder differed from attention
deficit hyperactivity disorder (ADHD), Asperger syndrome, and physical disorders in terms of reported disorder knowledge and siblings relationship from pre- to post intervention (Granat et al., 2012). The risk for overall adjustment problems was also found to vary with disorder type (Lavigne & Faier-Routman, 1992), and siblings of children with life-threatening and/or intrusive CD were at higher risk than siblings of children with a less serious diagnosis (Vermaes et al., 2011). The severity of the CD diagnosis has been linked to different impacts on siblings in studies (Limbers & Skipper, 2014). To support this, a study found symptom severity in children with autism spectrum disorder to be positively associated with adjustment difficulties in siblings (Meyer, Ingersoll, & Hambrick, 2011). Another study found a minimal risk of internalizing problems in siblings of children with high-functioning autism spectrum disorder (HFASD) (Rodgers et al., 2016).

Grouping different CDs to investigate differences between diagnoses, or common denominators, might be of importance. A common denominator across diagnoses might be the degree of challenging behaviour in the child with CD. Some research suggests that more challenging behaviour in the affected sibling results in more stress and more negative feeling in siblings and parents and therefore may lead to more adjustment problems (Lauderdale-Littin & Blacher, 2017; McCaffrey, 2016) as well as more depressive symptoms for the sibling (Lovell & Wetherell, 2016). It might seem as though severity of the CD or challenging behaviour of the affected child, might result in challenging consequences for the sibling and the family, that again can lead to the heightened risk of adjustment problems for siblings.

9.3.4 Past interventions, outcomes and methodology

Several reviews on intervention studies for siblings made comments on the variation in quality of the methods used, outcomes measured, and findings and emphasised the need for more systematic approaches (Hartling et al., 2014; Tudor & Lerner, 2015). Also, the review by Hartling et al. (2014) included 14 studies, and 9 of the studies were uncontrolled. The review pointed out that the consequence of lack of control groups and randomizing might be a biased result. More randomized controlled intervention studies are called for in future research. The review done by Tudor and Lerner (2015) also highlighted the need for studies with control groups, and the need for follow-up data collection. Further, no studies presented statistical power analysis and the studies’ sample sizes varied to a great degree. To pursue larger sample sizes in future studies was noted by both reviews for stronger statistical power. This thesis did not include the comparison to a group of families without a child with CD.
There was no control group who did not get the intervention or who got treatment as usual. Therefore, the result of the intervention might be biased. However, this is a pilot study and the data from this study will inform the next step; a randomized controlled intervention study.

This thesis was conducted with families who were contacted because of their link to Frambu or one of the user organizations mentioned. The sample was therefore not randomly selected and could thus be biased. The families who agreed to participate could have a higher level of knowledge of the diagnosis, or could have more motivation for optimizing family communication than the families who were not in contact with the organizations in question, or who did not want to participate. An alternative is that the families who agreed to participate had lower level of knowledge of the diagnosis, and more challenges in communication than the families who were not included in this thesis (Vatne et al., 2015). Additionally, the demographic profile of the sample consisted mostly of Caucasian, married couples which could reduce generalizability of the data because of limited demographic variation.

9.3.5 Statistical regression and instrumentation

Statistical regression are the effects of selecting samples based on extreme behaviour, that over time will tend to move towards the mean. The ratings of communication in this thesis are generally good and could represent a bias due to siblings’ reluctance to rate their parents poorly, but the ratings do not regress towards the mean at post. On the contrary, the ratings improved. So, in this thesis, this was likely not the case.

The change in communication and knowledge from pre- to post is assumed to be a result of the intervention, but the possibility that the change could come from other explanations are important to consider. Instrumentation, or the effects of changing the cut-off point or criteria of a measurement, is also a threat to the one-group design. In this thesis, there was a change in how the data of both the SKI and the PCCS was collected. At pre, the SKI was done in person at the intervention site, and the PCCS was also completed at the intervention site. At post, the SKI was done by phone, and the PCCS was sent by mail to the families. The families completed the PCCS at home, and returned the questionnaires by post. This difference in data collection could bias the data. Additionally, the number of participants who completed the SKI and the PCCS was lower at post-data than pre-data. Consequently, there could be some missing data due to the fact that the data collection at post demanded somewhat more effort from the participants than the data collection at pre.
9.4 Ethical considerations

This thesis has some ethical considerations that need to be addressed. The sample in the thesis consist of children, and REK approval (the regional ethical comity) has been applied for and granted. The participants signed informed consent and were voluntary participants. They were informed that they could withdraw from the study at any time and without an explanation. They would not lose any benefits from Frambu or the different user-organizations if they withdrew. They were informed that all information or data they contributed would be handled with confidentiality and that their anonymity would be protected. Only the relevant participants were addressed and recruited to this thesis.

This thesis strived to uphold the ethical consideration to do no harm, and have also, to the best of our knowledge, achieved to uphold this criterion. Siblings were asked to rate their parent’s communication, and this can be a distressing situation for a child. To assess their parent might be something the sibling has not thought of before or can be a vulnerable topic, especially if the sibling is not so happy with communication. The communication in the family was a topic in the interventions, and were an important focus. Still, there needs to be awareness that this can be a difficult thing to do for some siblings.

The interview can also be stressful in the sense that siblings are asked about their brother and sister with CD. They are asked about the diagnosis and what their brother or sister find difficult. This can be stressful for the sibling. It can be hard to talk about the challenges the child with CD meets in everyday life, and it can also invoke a sense of performance or test anxiety to be interviewed for disorder knowledge. The setting is laid as to minimize the risks for either of these, but it can be important to bear in mind.

9.5 Implications for practice

The data from this intervention thesis are used to inform the next step, a randomized controlled prevention trial (RCT). The Sibling Study will advance the pilot intervention to allow health providers to offer evidence-based health services to siblings, and examine risk factors as outcome predictors. The project is currently in stage 4, a randomized controlled prevention trial for siblings and parents of children with neurodevelopmental disorders. Funding has been applied from the NRC’s “Better health” program. Stage 1 was a qualitative study of sibling groups, Stage 2 was development of an intervention manual, and Stage 3 was an open trial of the manual with 98 participating families. This thesis is based on data from
stage 3, which will be used to inform stage 4.

The importance of these interventions is made evident by the research on risk factors and outcomes for siblings. The urgent timing of these interventions is due to the new law postulating that siblings of children with a CD have the same rights as children of parents with serious health challenges, requiring appropriate support to this group to be provided.

9.5.1 Knowledge as part of the intervention

There is an ongoing debate regarding whether increase in knowledge is linked to better psychosocial outcomes. Presented in this thesis are the findings that increased knowledge lead to better adjustment (Houtzager et al., 2001; Meyer & Vadasy, 1994). However, one study found that knowledge about a siblings’ autism spectrum disorder diagnosis was found not to be associated with scores on the Child Behaviour Check List (CBCL; Achenbach, 1991). However, others find that 40% of siblings in their study had CBCL scores in the clinical or almost clinical cut-off (Ross & Cuskelly, 2006). This emphasizes the need for attention to this group, but suggests that knowledge might not be a crucial factor in developing adjustment problems.

There is also a perspective that was beyond the scope of this thesis, namely siblings who have very much knowledge about the diagnosis of their brother or sister. It can be important to investigate if there are any negative consequences for siblings of having too much disorder knowledge, and if an intervention with more information could possibly have unwanted effects or consequences for these siblings. The literature calls attention to the fact that information must be adjusted to the developmental stage of the sibling (Plumridge et al., 2011) so it would be important to explore if there could be a negative effect for some siblings of knowing more than the age-appropriate level of disorder knowledge. Future research should examine the possible negative consequences for these siblings. Based on the findings in this thesis, the importance of including knowledge about diagnosis in the intervention is emphasized, but it would also be important to investigate how knowledge influences adjustment, and why.

9.6 Implications for future research

This thesis aimed to investigate if rating of communication and level of knowledge could be sibling factors that separate those who would benefit more from the intervention
from those who might not profit to the same degree. This thesis linked the benefit in evaluated communication to the change in sibling’s level of knowledge from pre- to post intervention. Various possible explanations for this phenomenon exist, but in this thesis the mechanisms are explained through a model of coping and resilience in siblings, and the affect RTPB model. This could indicate that siblings with higher knowledge and better quality of family communication would be more resilient and might not need an intervention at the same level as siblings with less knowledge and poorer quality of family communication. Of course, more research will be essential before making a conclusion of any kind in this area, and more research targeting the affect RTPB model will be necessary when going forward and investigating the relationship and causation between communication and knowledge.

9.6.1 Intervention for all? Being a sibling is not a diagnosis

Several studies revealed several risk factors and negative outcomes for siblings, but there are studies that report that living with a child with CD can also have positive impacts on siblings. Benefits of having a brother or sister with a CD were related to personality characteristics in siblings such as more tolerance and acceptance, and siblings were found by parents to be more mature and patient (Mulroy et al., 2008), as well as empathic and caring (Tøssebro, Kermit, Wendelborg, & Kittelsaa, 2012).

Focus has long been on helping parents cope with the complex diagnoses of a child with CD. It has been found that enough information can help with the managing of panic, fear and anxiety. Still, the same needs in siblings have not been recognized (Metcalf et al., 2008). Presently, focus is given to siblings at the same rate as parents, and it is essential that siblings who need help receive it. However, it is also important to remember that not all siblings develop adjustment problems and not all siblings need support in the form of an intervention. A priority in intervention studies will therefore have to be to find the sibling and family risk factors, to offer the intervention to the families and siblings who need it the most.

Sibling relationship has shown to be important for the development of psychological challenges in healthy children (Buist, Deković, & Prinzie, 2013) and being a sibling of a child with CD is linked to undesirable outcomes such as adjustment problems (Lavigne & Faier-Routman, 1992; Sharpe & Rossiter, 2002; Vermaes et al., 2011). The causality of the adjustment problems needs more inquiry. There might be a more complex explanation than their sibling status that results in the increased risk in sibling adjustment problems, and this needs more attention and exploring from researchers. For future research, sibling participant
characteristics must be targeted, and what specific sibling factors are the risk factors of adjustment problems must be examined (Tudor & Lerner, 2015).

9.6.2 Family factors

Family functioning in families with a child with CD can have an impact on the adjustment of siblings. As such, exploring family functioning factors should be an important focus. In addition to influences such as being a single parent, the family’s socioeconomic status (Meyer et al., 2011; Mulroy et al., 2008), parental stress (O’Brien et al., 2009; Vanegas & Abdelrahim, 2016), as well as parental coping strategies (Cipolletta et al., 2015), parental culture and type of diagnosis (Lauderdale-Littin & Blacher, 2017) might have impact on sibling adjustment. Identifying the factors affecting sibling adjustment can help greatly in combination with a focus on communication and knowledge. Stress, coping and challenging behaviours all need to be understood and dealt with by the families, and the intervention helping people deal with this can be more effective if more information about the biggest stressors or risk factors with having a child with CD are mapped out.

9.7 Conclusions

This thesis investigated the pre-post change in communication and disorder knowledge, and the relationship between communication and disorder knowledge. The results show increased communication quality, but only for mother, and increased disorder knowledge from pre- to post intervention. The results also support a relationship between communication and knowledge. The improvement in communication with mother, was related to the improvement in level of disorder knowledge. Additionally, communication with mother was always rated by the siblings to be significantly better than communication with father, both at pre- and post intervention.

The focus of this thesis was to investigate the intervention for siblings used in this study and start exploring which siblings could benefit more from this intervention. Although it is only a beginning, support is found for this intervention on the outcomes of communication and disorder knowledge. The findings might also suggest that the siblings with less disorder knowledge and poorer communication with mother, might benefit more from the intervention than siblings with higher levels of disorder knowledge and better communication with mother.
References


Attachments

APPENDIX A:

Figures of the descriptive statistics for the SKI explanation variable, illustrating the distribution of the different scores at pre-and post.

Figure A1. The distribution of the scores from 1-5 at pre intervention.

Figure A2. The distribution of the scores from 1-5 at post intervention.
APPENDIX B:

Figures of the descriptive statistics for the PCCS, illustrating the distribution at pre- and post of the different scores for communication with mother and communication with father.

Figure B1. Scatterplot of the scores from 1-5 at pre- and post intervention for communication with mother.

Figure B2. Scatterplot of the scores from 1-5 at pre- and post intervention for communication with father.
APPENDIX C:

The Parent-Child Communication Scale (PCCS), both for communication with mother and communication with father.

FORELDRE-BARN KOMMUNIKASJON (BARN)

Vennligst kryss av for hvilket av svaralternativene til høyre som passer best for deg når det gjelder disse spørsmålene om moren din (den du tenker på/ oppfatter som din mamma).

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Oversatt fra Parent-Child Communication Scale (PCCS) (Conduct Problems Research Group; CPRG)
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Oversatt fra Parent-Child Communication Scale (PCCS) (Conduct Problems Research Group; CPRG)
APPENDIX D:

The Sibling Knowledge Interview (SKI) and scoring guidelines

**INTERVJU OM SØSKENS KUNNSKAP**

(Oversatt fra “Siblings Knowledge Interview”; Lobato & Kao, 2002*)

Vi ønsker å lære mer om hvordan det er for barn når de hare n bror eller søster som er syk eller hare n funksjonsnedsettelse. Jeg vil starte med å stille deg noen spørsmål om hva som er problemet til din søster eller bror.


SØSKENKUNNSKAP

1. Kan du fortelle meg navnet på det problemet som søsteren eller broren din har?

_Hvis barnet ikke gir spesifikk diagnosenavn så spør etter hvilke ord familien bruker når de snakker om barnets problem – eks, Hva kaller moren/faren din problemet? Hva sier legen at det kalles?

_Hvis barnet bruker vage eller generelle begreper som ”hun er treg” eller ”han er spesiell”, spør om det er noen andre ord som foreldrene, leger eller lærere bruker for å beskrive barnets problem.

2. Hva er __________? Forklar __________? For meg. Hva vil det si å ha __________?

Bruk det ordet barnet bruker som navn på problemet selv om det er feil eller vagt. Fullfør intervjuet med å bruke barnets navn på problemet.

_Hvis barnets spontane svar ikke inkluderer de dimensjonene av problemet som er listet opp under s bruk de følgende oppfølgingsspørsmålene for å få frem en mer fullstendig forklaring

   a. Kroppsdel: Hvilke deler av kroppen er påvirket av __________? Hvis et barn har ___________, hvilke deler av kroppen fungerer ikke helt som de skal da?

   b. Manifestering: Hvordan kan man vite at noen har __________?
c. *Funksjon/Svekkelse:* Hva har barn med ________ problemer med å gjøre? Er det ting de ikke kan gjøre så bra? Er det ting de kan gjøre ganske bra?

e. *Behandling:* Hvordan behandler man ________? Hva kan familie, lege eller lærere gjøre for at ________ blir bedre?

**SØSKENS FORSTÅELSE AV ETIOLOGI**

3. Hvordan fikk søsteren/broren din ________? Hva var årsaken til det?

   Hvis barnet sier "Hun/han ble født med det", spør "Hvorfor ble han /hun født med det?".

4. Er det noe annet du kan fortelle meg om ________ så jeg kan forstå det bedre?
MERK: Hvis barnet ikke kan navnet på barnets sykdom/funksjonsnedsettelse eller bruker et opplagt feil (i motsetning til et vagt eller lite spesifikt) navn (f.eks. sa at barnet med cystisk fibrose har astma), så spør følgende:

5. Har du noen gang hørt ordet _________? (Fyll inn det diagnostisk mest nøyaktige begrepet på barnets problem, som beskrevet av foreldrene)

Hvis barnet svarer ”Nei” hopp til delen ”Kommunikasjon mellom søsken og familie”. Hvis barnet svarer ”Ja”, gå videre med spørsmål 6-8.

6. Hva er (diagnostisk begrep)? Forklar (diagnostisk begrep)? For meg. Hva vil det si å ha (diagnostisk begrep)?

Hvis barnets svar ikke spontant inneholder at dimensjonene under nevnes, så bruk følgende sonderinger for å få frem en mer helhetlig forklaring.

   a. Kroppsdelar: Hvilke deler av kroppen er påvirket av _________? Hvis et barn har _________, hvilke deler av kroppen fungerer ikke helt som de skal da?

   b. Manifestering: Hvordan kan man vite at noen har _________?

   c. Funksjon/Svekkelse: Hva har barn med _________ problemer med å gjøre? Er det ting de ikke kan gjøre så bra? Er det ting de kan gjøre ganske bra?

   e. Behandling: Hvordan behandler man _________? Hva kan familie, lege eller lærere gjøre for at _________ blir bedre?

7. Hvordan fikk søsteren/broren din (diagnose)? Hva var årsaken til det?

   Hvis barnet sier ”Hun/han ble født med det”, spør ”Hvorfor ble han /hun født med det?”.

8. Er det noe annet du kan fortelle meg om (diagnose) så jeg kan forstå det bedre?
SIBLING KNOWLEDGE INTERVIEW  
(Lobato & Kao, 2002)  

SCORING GUIDELINES  

I._sibling’s NAME of the child’s disorder  

CODE  

1  

Does not know, no response, uses an incorrect term  

2  

Vague, but correct term (e.g., “Slow” for mental retardation)  

3  

Specific, correct term  

– using the correct initials to designate the disorder is considered correct (e.g., “CP” for cerebral palsy, “PDD” for Pervasive Developmental Disorder)  

II. Accuracy of siblings’ EXPLANATION of the child’s disorder  

CODE  

1  

Does not know, no response  

2  

Complete misunderstanding of the disorder  

– All the information is incorrect or irrelevant to the child’s disorder  

– States wrong body parts, functions, etc.  

– Answers none of the follow-up questions correctly  

3  

Vague, but generally correct understanding of the disorder  

– Uses vague terms (e.g., sick, goes to doctors, slow) to describe the child’s disorder  

– The definition may lack mention of the essential symptoms* of the disorder  

– Correctly answers one of the four follow-up questions  

4  

Partial understanding of the disorder  

– Clearly identifies the essential or core symptoms* of the disorder  

– Correctly answers two of the four follow-up questions  

5  

Accurate understanding of the disorder  

– Sibling reveals understanding of all the aspects of the disorder  

– Clearly identifies the essential or core symptoms* of the disorder  

– Correctly answers three or four of the four follow-up questions  

* Descriptions of the core, essential manifestations or features of the disorder will be considered as a correct, essential symptom. The sibling does not necessarily have to identify the etiology of the disorder. For example, for Downs syndrome the correct core features include: trouble learning, trouble talking, something is wrong with his/her brain. The sibling does not have to identify the chromosomal abnormality.