Adjustment in siblings of children with chronic health conditions: Associations with parent depression, anxiety, and stress

Marit Aall Henriksen Hughes

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Forfatter: Marit Aall Henriksen Hughes

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Abstract

Author: Marit Aall Henriksen Hughes
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Supervisor: Krister Westlye Fjermestad, Associate Professor, University of Oslo

Data: The data is from an ongoing exploratory trial of a sibling and parent group intervention for families of children with chronic health conditions (Frambu/Institute of Psychology, University of Oslo). The data is based on self-reports from healthy siblings and parents at baseline (T1). The author of this thesis did not take part in the planning and design of the study, but has taken part in the intervention as a group leader for the parent-only groups and joint sessions of parents and children.

The present thesis is a quantitative correlational study of 92 healthy siblings age 8 – 16, and parents of children with chronic health conditions (CHC) recruited at Frambu resource centre for rare disorders and collaborative associations. Informed by a substantial, but divergent, field of research, the thesis aimed to investigate the level of siblings’ adjustment and the association between adjustment and general socio-emotional difficulties. Further, the thesis aimed to investigate whether the levels of adjustment and socio-emotional difficulties were associated with siblings’ age and gender, and possible correlations with parents’ depression, anxiety, and stress. The healthy siblings were found to generally report satisfactory adjustment and low levels of socio-emotional difficulties. The parents reported generally low levels of depression, anxiety, and stress. Contrary to our expectations, there were no significant differences between mothers and fathers on depression and anxiety. However, there was a significant difference between mothers’ and fathers’ report of stress, with mothers reporting higher levels of stress. The only significant correlation between parents and children was between children’s level of adjustment and fathers’ depression. In sum the general picture is of well-functioning families, where both parents and children are well adjusted and are coping with their life-situation. Possible explanations may be high SES and highly educated parents, in particular mothers. Other possible explanations include gender equality in the household, economic support and a well-functioning health care service, all elements important for adjustment, coping and resilience. An implication of the findings of this study is that both parents, i.e. both mother and father, should be included in interventions to improve
siblings’ adjustment and socio-emotional functioning. Another important implication is that siblings as a group are well adjusted and coping well, further research is therefore needed to identify the individual siblings, parents, and families at risk of negative adjustment and socio-emotional difficulties.
Preface

I would like to thank the siblings and the parents participating in “the Sibling Project”. Thank you to the project and Torun Vatne for giving me access to the data, and for the opportunity to take part in the intervention as a group leader for the parent groups.

Thank you, Krister, the best supervisor I could dream of: Precise, thorough and fast as lightning. Thank you for your no-nonsense approach; that was exactly what I needed.

Thank you to Yngvild Haukeland for being so helpful and kind.

Thank you to my fellow students, for late nights in the reading room and lots of fun. Thank you Marte and Guro – you’ve become real friends during these past six years. And thank you for reading my thesis and giving useful feedback.

Thank you Mamma for always being there for me – from the very beginning.

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And last but not least, thank you to Leo, Edward & Ellinor, my beautiful children, the best trio of siblings I know.
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1 Introduction

The family has a central position in the life of an individual and represents humanity’s most fundamental and enduring social institution. Children are socialized within and through the family, and it is in the family that both young and old receive social, emotional and instrumental support. Thus, the quality of family relationships has as a profound effect on the quality of life of the individual, and how families function affect society at large (Koerner & Schrodt, 2014).

A vital part of family structure is the inter- and intra-generational relationships. Sibling relations are central and unique intra-generational relationships. They more often than not include shared and common life experiences, common biological background, and they often last a lifetime – longer than the relationship between parents and children. Sibling relationships have the potential to influence and shape a person throughout the life course (Davies, 2015). Sibling relationships are powerful components of socialization because they foster the development of important instrumental and affective relationship skills (Rodrigue, Geffken, & Morgan, 1993). What is learned from relating to siblings can potentially influence cognitive, affective, and social skills, as well as the development of a positive self-image. The importance of sibling relations in the context of child development has led researchers to investigate the psychological adaptation of siblings of children with chronic health conditions (CHC) or developmental disorders (DD) e.g. (Bluebond-Langner, 1996; Davies, 2015; Dyson, Edgar, & Crnic, 1989; Moyson & Roeyers, 2012; Rodrigue et al., 1993).

The sibling bond is no less enduring nor less influential when one sibling has a chronic health condition (CHC) (Rossiter & Sharpe, 2001). Having a sibling with CHC will impact on the psychological development and functioning of their typically developing siblings, as well as on the parents, and the entire family system (Rossiter & Sharpe, 2001). The present thesis is concerned with the adjustment of healthy siblings and the mental health of parents of children with CHC. The main aims of the thesis are to investigate the impact CHC in children have on the psychological adjustment and socio-emotional functioning of the healthy siblings and whether the siblings’ adjustment is associated with parental depression, anxiety, and stress.
Background

There has been a dramatic increase in the number of children and youth living with a chronic health condition (CHC) in the last four decades (Perrin, Bloom, & Gortmaker, 2007). This is mainly due longer life expectancy for children with genetic disorders, chronic illnesses and mental disabilities due to advances in medical treatment and improved general care, but also in better diagnostic tools following the advances in genetic testing. With the increased incidence and prevalence, the number of families with a chronically ill child has also increased. This increase happened simultaneously with a transfer of increasingly complex medical care to the home situation (Hatzmann, Maurice-Stam, Heymans, & Grootenhuis, 2009). The right to grow up in a family within a local community is today inscribed in international agreements as the United Nations Convention on the Rights of the Child (FN, 2003) and the United Nations Convention on the Rights of Persons with Disabilities (UN, 2006). All this naturally has led to better quality of life for children with CHC and their families, but also leads to greater caregiving demands on the family. The demands can be extensive and may lead to adverse psychosocial consequences of different degrees for parents (Cousino & Hazen, 2013; Folkman, Lazarus, Gruen, & Delongis, 1986; Hatzmann et al., 2009; Plumridge, Metcalfe, Coad, & Gill, 2011) and siblings (Alderfer et al., 2010; Fisman et al., 1996; Sharpe & Rossiter, 2002; Vermaes, van Susante, & van Bakel, 2012).

Around 10-15% of Norwegian children have a physical or mental disorder affecting their everyday functioning (Norwegian Directorate of Health, 2010). With the number of siblings and parents who are next of kin to these children, the number of individuals affected by chronic conditions and disabilities increases substantially.

Current systems of care for children with CHC depend on parents being able to adopt unexpected “informal caregiving careers”, roles that typically span the course of a child’s life (Raina et al., 2005). Parents provide long-term care that often requires extraordinary physical, emotional, social, and financial resources. In addition to being responsible for the physical and emotional care of their child, parents must co-ordinate their child’s numerous and multifaceted medical treatment, education and developmental interventions while balancing competing family needs, including the needs of their typically developing children (Murphy, Christian, Caplin, & Young, 2007; Silver, Westbrook, & Stein, 1998).
In recent years there has been an increased awareness within the health care system of children as next of kin. However, the focus has mainly been on children as next of kin to parents with physical or psychiatric illness, or substance abuse. There has been less focus on children as next of kin to chronically ill siblings, and how having a chronically ill child in the family affects both parents, the healthy siblings, and in effect the entire family system. In 2009 there was a revision of the law for health care personnel (HCP) in Norway by the Norwegian Directorate of Health (Helsedirektoratet, 2010). The law accentuates the rights of children as next of kin to parents with somatic or psychiatric illness or substance abuse (§ 10a). However, children as next of kin to children with a chronic condition (i.e., siblings) are not mentioned. This is a major limitation of the law and shows the importance of focusing on siblings as a group in clinical research and settings.

Research has shown that these siblings have an increased risk of developing both internalizing and externalizing difficulties in relation to siblings growing up in normal families (Alderfer et al., 2010; McKeever, 1983; O’brien, Duffy, & Nicholl, 2009; Sharpe & Rossiter, 2002; Vermaes et al., 2012).

1.1 Perspectives on stress and coping

When studying siblings of children with CHC it is important to address both positive and negative aspects of their psychological functioning. The presence of negative functioning does not imply the absence of positive functioning, and vice versa. Psychological functioning is here therefore understood both in terms of mental health problems and positive self attributes. An important line of distinction in the literature of siblings is the stress versus the coping paradigm. Are we exploring mechanisms of coping, or are we exploring how the situation of living with a CHC sibling affects the child negatively? Alternatively, one might pursue a perspective on the healthy siblings containing both the particular stress they live under and how they cope with it.

In a qualitative study of families where a child had Cystic Fibrosis, Bluebond-Langner (1996) found that the healthy siblings’ understanding of themselves and others was strongly linked to their parents’ responses. Further, the study shows that despite the burdens placed on all family members, the families appeared to function very much like other families. Bluebond-Langner (1996) concluded that understanding the impact of the disease on healthy siblings does not
come from searching for something wrong in the lives of the healthy siblings, nor does it come through the identification of some form of pathology or from the discovery of abnormal behavior caused by a trying situation. Rather, understanding involves appreciating how the family goes about preserving integrity, living life as it is, in the face of the intrusion of the illness (Bluebond-Langner, 1996).

The concept of salutogenesis was coined by Antonovsky (1979). Instead of asking what factors are causing pathogenesis, he asked what factors make people maintain and develop their health even under difficult external circumstances. According to Antonovsky, people who have sufficient and adequate generalized resistant resources at their disposal and learn how to use them, can gradually develop a strong sense of coherence. A strong sense of coherence is defined as a life orientation that helps people to perceive life as comprehensive, manageable, and meaningful, and is assumed to reduce the perceived strain of life. Salutogenesis is a system theory in which the coherence between the individual, the group and the environment plays a major role in the development of sense of coherence (Antonovsky, 1979, 1987). These perspectives of salutogenesis and how a family goes about living their life and preserving their integrity, in a situation more or less dominated by the CHC of a family member, is important to keep in mind when we explore the psychological health of the healthy sibling and the parents. According to Antonovsky’s theory it is more important to focus on peoples’ resources and capacity to create health than the classic focus on risks, ill health, and disease.

In this thesis I will let these two perspectives, stress and coping, both stay at the front; the siblings in this study are healthy and normally developing children, and the parents are healthy adults. However, their life situation is to different degrees trying and potentially stress evoking due to the chronic health condition of one of the children in the family.

1.2 Development of Resilience: A Model

The Norwegian child psychiatrist Hilchen Sommerschild termed the concept “mestring” (Sommerschild, 1998). The word in Norwegian contains the meaning of English “to master (something), to overcome, to handle, to cope”. Sommerschild describes her concept of “mestring” as wider, but containing, elements of Lazarus’ coping (Folkman et al., 1986), Rutter’s resilience (Rutter, 1981, 1985, 1993), family therapeutic approaches (De Shazer,
It is a concept evading translation, and as “mastery” gives somewhat different connotations in English, I have chosen to use related terms “resilience”, “coping” and “adjustment” in this thesis, bearing in mind that in doing so, I lose some of the qualities of the Norwegian word, and that for instance “resilience” in a clearer way writes itself into a traditions of research, whereas “mestring” stays closer to the colloquial language and the experience of handling the challenges that life throws at you.

As a child psychiatrist Sommerschild worked with families facing numerous challenges, and amongst them were families with disabled children. Some were coping well with their situation, and some were struggling. Sommerschild divided them in three groups: The families who appeared to cope with their situation, the potentially coping families, and the families who appeared enduringly weak in their coping. Rather than studying risk factors and possible negative trajectories, she was intrigued by the processes leading to coping. She set about to explore coping and the processes leading to it. One simple, temporary answer could be that coping is to “meet life’s challenges with resilience”. However, the question of what the processes behind coping and resilience are, remains unanswered.

Through her model, Sommerschild argues alongside Rutter (1981), that what the child first and foremost needs to develop resilience is belonging, in the sense of having at least one close confidant (See Figure 1, p. 6). Further, the child needs a family offering predictability and confirmation, as well as a wider social network of a community with shared values and social support. In the model, the individual’s need of competence is accentuated. Competence is here understood as knowledge and “know how”, but also a sense of “being of use” in relation to others, to be given responsibility and take responsibility. The two most important conditions for coping according to Sommerschild are therefore belonging and competence, as is shown in the model (See Figure 1). The model accentuates the individual’s position within a social network as well as the individual’s sense of self-worth as equally important factors for coping and resilience.
Conditions for resilience

<table>
<thead>
<tr>
<th>Belonging</th>
<th>Competence</th>
</tr>
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<tbody>
<tr>
<td><strong>The dyad:</strong></td>
<td><strong>Knowledge, “to know how”</strong></td>
</tr>
<tr>
<td>At least one close confidant</td>
<td>Be of use/be useful</td>
</tr>
<tr>
<td><strong>The family:</strong></td>
<td><strong>Be given and take responsibility</strong></td>
</tr>
<tr>
<td>Predictability, confirmation, belonging</td>
<td>Displaying love for the other</td>
</tr>
<tr>
<td><strong>The network:</strong></td>
<td><strong>Encounter and master adversity</strong></td>
</tr>
<tr>
<td>Shared values, social support</td>
<td></td>
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</tbody>
</table>

**Sense of self-worth**

**Resilience**

Figure 1. Conditions for resilience (Sommerschild, 1998).

The main elements of the model can, as already mentioned, be found in other theories and models of coping. Rutter identified three broad sets of factors leading to resilience that resembles the factors of Sommerschild’s model: (a) personality characteristics such as autonomy, self-esteem, and a positive social orientation; (b) family cohesion, warmth, and an absence of discord; and (c) the availability of external support systems that encourage and reinforce a child’s coping efforts (Rutter, 1993). Rutter (1985) defined protective factors as those factors that modify, ameliorate, or alter a person’s response to some environmental hazard that predisposes them to a maladaptive outcome. Risk and protective factors can be categorized into three broad groups: dispositional attributes of the child, attributes of the family environment, and of the extra-familial system (Garmezy, 1985).

Given what is known about the risk factors connected with being a healthy sibling of a child with CHC, it is important to focus on the factors that foster resilience in a child exposed to
this known risk factor. Sommerschild’s model of the conditions for resilience have therefore guided the research questions of this thesis, the ultimate goal being to develop interventions directed towards healthy siblings aiming to strengthen and foster coping and resilience.

1.3 Siblings of children with chronic health conditions

1.3.1 Challenges and risk factors

CHCs often involve important life events as well as daily challenges. Both will act as stressors for the affected child and the family. In meeting such stressors one can hypothesize that siblings are affected in multiple ways. Possible moderating factors are the nature of the illness (intrusive or non-intrusive), on siblings’ own personal coping strategies, their parent’s coping and mental health, the family environment, and the wider psycho-social context.

A number of studies have investigated the impact of CHCs on siblings’ psychosocial functioning. However, evidence regarding the psychosocial functioning of these siblings is mixed (Havermans, Croock, Vercruysse, Goethals, & Diest, 2015; Knecht, Hellmers, & Metzing, 2015; Kozlowska & Elliott, 2016; Macks & Reeve, 2007; Mulroy, Robertson, Aiberti, Leonard, & Bower, 2008; Stoneman, 2005). Tudor and Lerner (2015) suggested that the mixed results are due to small sample sizes, negative biases about sibling functioning, and too broadly defined study groups. Several studies have suggested that at least some siblings may possess an elevated risk for behavioral, social, or internalizing problems (Macks & Reeve, 2007; Tudor & Lerner, 2015; Vermaes et al., 2012). Thus, having a sibling with a CHC may represent a risk to the emotional health of some healthy siblings, including anxiety, depression, posttraumatic stress symptoms, poorer quality of life, behavioural difficulties, and/or peer problems (Alderfer et al., 2010; O’Brien et al., 2009; Rossiter & Sharpe, 2001; Sharpe & Rossiter, 2002; Vermaes et al., 2012).

A more recent meta-analytic review of 56 selected studies showed a small but significant negative effect ($d = -.10$) of CHCs on siblings, compared to siblings of healthy children. Siblings of children were found to have more internalizing problems ($d = .17$), more externalizing problems ($d = .08$), and less positive self-attributes ($d = -.09$) than controls (Vermaes et al., 2012). Siblings of children with highly intrusive and/or life-threatening CHC
appear especially at risk for internalizing and externalizing problems and less positive self-attributes. The siblings appeared less resilient than controls (effect size $d = -0.09$). Resilience was in this study defined as positive self-attributes. According to the findings, older siblings’ positive self-attributes were more negatively affected than younger siblings’ self-attributes. Overall, the review shows that siblings are at risk of poorer emotional health (Vermaes et al., 2012).

Vermaes and colleagues (2012) investigated six demographic factors: gender, age, birth-order, type of CHC, intrusiveness of treatment, and life-threatening potential. No significant moderation effects of gender, birth-order, and type of CHC were found. This is somewhat surprising given the literature on gender differences in the prevalence of internalizing (girls more than boys) and externalizing problems (boys more than girls) (Vermaes et al., 2012). These findings, however, are consistent with a previous meta-analysis (Sharpe & Rossiter, 2002). Other studies have shown that younger siblings, especially male, report greater negative adjustment than their parents (Hodapp, Glidden, & Kaiser, 2005).

Amongst the themes for future research on siblings, Vermaes et al. (2012) pointed to the mechanisms of risk and resilience through studying moderating effects of important intrapersonal factors (e.g., temperament and coping styles) and socio-ecological factors or interpersonal factors (e.g., social support, parental support). They further emphasized the role of child disclosure of positive and negative emotions, parental contingent sensitivity, parent-child dynamics, and the longitudinal development of positive self-attributes and internalizing problems (Vermaes et al., 2012).

As the field of risk research has developed, some investigators have cautioned against focusing on direct linear relationships between life stress and adjustment-related outcomes (e.g., Fisman et al., 1996). The emphasis should be on transactional mechanisms that affect outcome. A transactional orientation in the study of stress and coping may allow a better understanding of the variability in the healthy sibling’s functioning than the elucidation of single risk or protective factors (Fisman et al., 1996). Garmezy, Masten, and Tellegen (1984) suggested a curvilinear mechanism in which stressors can enhance competence in the presence of protective factors, provided the level of stress is not too high. When the level of environmental stress becomes overwhelming, those factors that were previously operative no longer serve to be protective. Rutter (1993) emphasizes the importance of both preceding and succeeding circumstances; and that resistance to stress has to be considered a graded
phenomenon, dependent on personality characteristics (e.g., autonomy, self-esteem, a positive social orientation, family cohesion, warmth, absence of discord; and the availability of external support systems that encourage and reinforce a child’s coping efforts).

Siblings’ emotional experiences are characterized by diverse and contradictory feelings (Haukeland, Fjermestad, Mossige, & Vatne, 2015). The factors explaining why siblings may be at risk for emotional difficulties are complex, and previous research has linked these difficulties to illness experiences, such as witnessing potentially traumatizing medical procedures without possibility of alleviating or affecting the situation (Alderfer et al., 2010; Vermaes et al., 2012) or changes in health status of the sick child (Hartling et al., 2014). Siblings’ experiences have also been linked to parent and family factors, such as differential parental treatment due to disproportionate care for the affected child and extra caregiving or domestic responsibilities. Insufficient parent-child communication and lack of illness knowledge have been found to partially explain adjustment problems and emotional distress among healthy siblings (Vatne, Helmen, Bahr, Kanavin, & Nyhus, 2015). Other risk factors are lack of knowledge or misunderstandings about the disease (Lobato & Kao, 2002), difficulties expressing emotions (Long et al., 2013), and being affected by parental distress reactions (Incledon et al., 2015; Mulroy et al., 2008).

1.3.2 Interpersonal factors and adjustment

Sommerschild’s concept of belonging might be informed by the framework provided by attachment theory (See Figure 1, p. 6). Based on John Bowlby’s attachment theory, attachment can be understood as a basic, and biologically based, form of behavior that leads to a child achieving a close relationship to one preferred individual, usually a parent or another close confidant who is conceived as stronger and wiser (Bowlby, 1998). The child gradually develops an internal working model of the self and others, and in particular of their caregivers. The attachment behavior, both of the child and of the parent, is biologically rooted and activated by environmental cues, such as the need for protection and care. In other words, attachment behavior is part of a genetically programmed survival function (Bell & Ainsworth, 1972). It can be argued that attachment theory informs the understanding of the inner mechanisms of social bonding and belonging, the way in which past experiences influence the establishment of relationships with other, and the role that attachment plays as a buffer against stress (Mikulincer & Shaver, 2007). Implicit in Sommersheild’s model is an
understanding of attachment (summed up as the individual’s relation to a significant other in the dyad and as belonging) as a precondition of sense of self-worth and resilience (see Figure 1, p. 6.)

Attachment theory holds that an individual’s attachment system is activated during stressful situations (Yahy & Sochos, 2013). In families of children with chronically ill children, both parents and healthy siblings are subject to potential stress, activating their attachment system. The importance of a healthy attachment system for children’s psychological well-being, and the intricate connections with parental stress highlights the importance of the entire family system as target for interventions.

1.3.3 Competence

Competence as understood through Sommerschild’s model is a wide concept, consisting of several attributes and behaviors of the individual, and possibilities given by the child’s environment. Important elements of competence in the model, is being useful and to be given and taking responsibility. This can together be understood as a form of agency. Another important part of competence is knowledge. Knowledge can be understood as a “knowing how” to do something, how to act, and how to understand something. It can also be understood as knowledge of communication; how to communicate and how to interact with significant others through communication. Shared knowledge through communication is again tied in with belonging (see Figure 1, p. 6.)

Agency

Havermans et al. (2015) studied siblings´ self-reported quality of life and the impact of four different paediatric illnesses (type 1 diabetes, cancer, congenital heart disease and cystic fibrosis) (n = 131). The initial hypothesis was that siblings of children with diabetes and cystic fibrosis would report lower quality of life, than siblings of children with congenital heart disease or. This hypothesis was based on the assumption that the lives of siblings of children with these two diagnoses were more disrupted by the illness (i.e. intrusive) due to the prescribed treatment regimen on a day-to-day basis (e.g., medicine intake, physiotherapy, nebulizing, injections, diet). However, contrary to the initial hypothesis, the results showed that the siblings of children with diabetes and cystic fibrosis reported less behavioral and internalizing problems than siblings of children with cancer and congenital heart disease. The
authors explained this by the possible role of what they called “(masked) stress, worry and life threat” associated with the cancer and congenital heart disease. Further, this finding was thought to be explained by the impact of information, beliefs and communication about an illness. These themes contribute to siblings’ representations of illness and coping. Siblings of children with diabetes and cystic fibrosis are confronted with the effects of the illness and its’ treatment on a daily basis, which may actually provide some sense of control. One might hypothesize that having concrete treatment procedures, although only to alleviate symptoms, potentially add to this sense of control. They are given a responsibility that they manage to handle. These siblings are believed to experience and understand what is going on, and they have learned what they can do to help in the daily care of their brother or sister. Self-efficacy in the form of a sense of control was shown to impact on how siblings perceive their lives and how they report quality of life in this study (Havermans et al., 2015). This ties in with what Sommerschild calls “to be of use” and “take responsibility” in her model (see Figure 1, p. 6). According to Sommerschild this gives nurture to the child’s self-efficacy and positive self-attributes, which in turn may contribute to resilience.

1.4 Parents of chronically ill children

The relationship between family variables and child adaptation is complex, but parenting factors seem to provide important protective functions for children in families that experience adverse stress. It is known that well-functioning, healthy families help children develop along a positive trajectory. Strong parental relationships, cohesive families, and low family conflict help the child to develop positive self-concept and a sense of self-worth (See Figure 1, p. 6), and protect against a range of externalizing and internalizing difficulties. Several studies have found that higher relationship satisfaction among parents of children with CHC was associated with better coping, adjustment, and higher levels of self-esteem in siblings (Friedman et al., 2004; Giallo & Gavidia-Payne, 2006; Raina et al., 2005). With satisfying partnerships, both parents and siblings appear to adjust better to having a developmentally disabled child in the family (Rodrigue et al., 1993). These aspects are in line with the conditions for resilience described in Sommerschild’s model as the need for belonging to a family; predictability, confirmation, shared values, and social support (see Figure 1, p. 6). However, these are needs not only in the child, but also in the adults of the family.
1.4.1 Challenges and risk factors

Families in which a child is diagnosed with a CHC have to face the diagnosis with the following symptoms, prognosis and treatment. They have to make adjustments in their caregiving, and in their general expectations to many aspects of family life. Like in sibling research, there is great variability in descriptions of the psychological functioning of parents of children with CHC (Pelchat, Lefebvre, & Perreault, 2003; Silver et al., 1998). The variation in study findings and range of functioning reported, suggest that factors other than the child’s health status also influence parental adaptation. A wide variety of risk and resilience factors, including characteristics of the child’s illness, coexisting life stressors, parental mental health, coping style, relationship satisfaction, and social support from the wider network and community, determine how parents cope with their situation.

**Stress/distress**

Parenting stress, or stress directly related to the role of being a parent, is important to understanding family dysfunction and psychopathology, and may have a variety of effects both on the parents and the children. There is substantial evidence suggesting that parents of children with disabilities often experience higher levels of stress than parents of typically developing children (Awijma, Masino, & Hodapp, 1997; Cousino & Hazen, 2013; Murphy et al., 2007; O’Connell Corcoran & Mallinckrodt, 2000; Rao & Beidel, 2009). Lindström, Åman, and Norberg (2010) found that there was an increased prevalence of burnout symptoms in parents of chronically ill children. Cousino and Hazen (2013) found in their meta-analysis of 13 studies and qualitative analysis of 96 studies, that parenting stress among caregivers of children with CHC. Further they found that these parents report significantly greater general parenting stress than caregivers of healthy children (effect size $d = 0.40$). Greater parenting stress was associated with poorer psychological adjustment in caregivers, and in the children with CHC. A number of studies have documented associations between parenting stress, adverse caregiving, and child psychological sequelae (Cousino & Hazen, 2013; Kozlowska & Elliott, 2016; Orsmond & Seltzer, 2009). Thus, it seems probable that parental mental health and family functioning mediate the effects of stressors on children’s mental health (Rutter, 1985).

Raina et al. (2005) investigated the direct and indirect associations between caregiver characteristics, sources of caregiver stressors, family functioning and informal social support.
on the well-being of caregivers of children with Cerebral Palsy \((n = 468)\). The study showed that psychological and physical health of caregivers was strongly influenced by caregiving demands. The perceived stress was the result of direct effects of child factors and general family functioning mediated by parent’s self-perception (Raina et al., 2005). A study examining family stress and sibling reactions in families of children with 5p- (cri du chat) syndrome aged 1 to 18 years who were living at home, found that the best predictor of familial stress was the child’s amount of maladaptive behavior, accounting for 12% to 38% of the variance across different stress measures (Awijma et al., 1997; Mazaheri et al., 2013).

Different aspects and consequences of the CHC have been investigated as sources of stress: *i.e.* illness severity (intrusiveness, progressiveness, mortality), functional limitations, cognitive impairment and maladaptive behavior. Although the role of illness severity in parental distress probably has been examined most frequently, most studies fail to demonstrate convincingly that medical indicators of severity relate to parents’ psychological status (Silver et al., 1998).

Silver et al. found comparatively higher distress only among parents of children whose conditions involved functional limitations, *i.e.*, restrictions or impairment in hearing, seeing, communicating, play, social roles and age-appropriate function, or activities of daily living. They may represent a particularly high-risk group among parents of children with CHC and confirm the results of other studies demonstrating that functional impairment in children relates to poorer parental adjustment (Silver et al., 1998).

**Parent gender and depression**

In a study of risk and protective factors affecting the adjustment of siblings of children with chronic disabilities, parents of children with pervasive developmental disorder (PDD) reported higher levels of parent distress and depression than parents of healthy children with parents of children (with Down Syndrome (DS) falling between these two groups) (Fisman et al., 1996). The distress factor (composed of parental stress and depression scores) was found to mediate the relationship between group membership (PDD, normal controls or DS) and parent reports of both internalizing and externalizing behavior problems in the healthy sibling (Fisman et al., 1996).
Much research on families with a child with CHC has focused upon the impact on parents, but the impact on mothers have been explored most frequently. Multiple studies show that mothers of children with developmental disorders are more likely to suffer from depression than mothers of typically developing children e.g. (Mulroy et al., 2008; Singer, 2006). Parental depression is known to be an important risk factor for difficulties in parenting, so factors that protect against depression in parents are likely to enhance parenting. In typically developing youth, maternal depression is a predictor of inconsistent and inattentive parenting, which is associated with both behavior problems and internalizing symptoms in youth (Tudor & Lerner, 2015).

Several studies have shown that being a mother of a child with CHC is associated with higher frequency of parenting stress and burnout symptoms e.g. (Lindström et al., 2010; Streisand et al., 2008). The same associations for fathers were less apparent. This might be a reflection of fathers expressing their distress in other ways, or a reflection of mothers being more involved in the day-to-day care of the child with CHC and taking greater responsibility for the well-being of the whole family. Across studies the response rate among fathers are lower than mothers, perhaps reflecting this difference in involvement and perceived responsibility. It seems safe to infer that less is known about the mental health of fathers of children with CHC than mothers, given the number of studies exclusively including mothers in addition to the lower response rate among fathers in studies that include both genders of caregivers. The reason for fathers’ withdrawal from participation in studies remains to be explored.

1.4.2 The Family System

The internal and external resources parents have available, including parenting competence, self-esteem, and social support, have the potential to influence the stress that parents experience and the level of distress they report (Silver et al., 1998). Quinton and Rutter (1984) argued that parenting cannot be seen as an attribute of individuals irrespective of their current circumstances. These circumstances can be understood as encompassing all the levels in Bronfenbrenner’s ecological model (Bronfenbrenner, 1977); it is the individual with temperament and personality (influenced by both biological and relational factors); the microsystem consisting of family, peers, and siblings; the exosystem with extended family, school, work environment, media, and neighbours; and the macrosystem with history, social conditions, laws, culture, and economic system. Using Bronfenbrenner’s insight of the
importance of ecological environments in influencing child development Stoneman (2005) made a model representation of the complexity and interactivity between the different systems in which the child and the parents are interconnected (see Figure 2). The individual child, the sibling relationship, and the parents enter into these systems and are affected by them in different ways. The model works as a reminder of how culture, with values and beliefs about disability, and sibling roles and expectations, works on the individual. The community affects the individual family through family support, services, and attitudes. The parenting is affecting the children, and the parenting is affected by the relationship between the parents, as well as child factors and characteristics of the CHC. In other words; the individual and the individual family enters into numerous complex and interrelated systems.

Figure 2. A model of interacting systems (Stoneman, 2005).
Such an ecological framework contributes to an understanding of a child’s development within the context of the systems of relationships that form his or her environment. It also serves as a good image of how the parents and the children in a family are included in the same systems and how these systems interact and affect each other. Thus one might think of both the children and the parents as belonging in Sommerschild’s sense to different systems; the dyad, the family, and the network, and that the quality of each one affects the individual. A safe attachment, secure relationships offering predictability, confirmation, shared values and social support being equally important for the child and for the parents.

Research has provided support for a contextual perspective on families living with CHCs, and the importance of viewing the individual child within the family, and the family within the wider societal context. Giallo and Gavidia-Payne (2006) studied families (n = 49) of children with intellectual, sensory, physical, or developmental disabilities residing at home and found that parent and family factors were stronger predictors of sibling adjustment difficulties than the sibling’s own experiences of stress and coping, highlighting the importance of familial and parental contributions to the sibling adjustment process. More specifically, socio-economic status, past attendance at a sibling support group, parent stress, family time and routines, family problem-solving and communication, and family hardiness predicted sibling adjustment difficulties. The findings highlight the relationship between parent emotional functioning and sibling adjustment, and the need to consider sibling adjustment within the context of the family (Giallo & Gavidia-Payne, 2006).

Other studies are in line with this; one such study found that family system variables are more powerful predictors of adaptation and adjustment than disease factors or illness severity. The synergy of the total family system is more important to resilience than specific, isolated family factors (Cohen, McDaniel, & Campell, 1999).

Stress and adversity of any sort in any of the systems in which the individual parent, individual child, or individual family partakes, can affect adjustment and coping. As an example, one study found that the self-concept of siblings of disabled children was lower in the presence of high parental stress (Dyson et al., 1989). And as a sense of self-worth according to Sommerschild’s model is not only the result of belonging and safe attachment to a significant other, but also a condition for resilience (see Figure 1, p. 6) it is important to strengthen the systems around the child to improve resilience in the individual child.
Parents’ perceived stress is therefore a factor not only affecting parents, but also the children in the family. Raina et al. (2005) found that perceived stress was the result of direct effects of child factors, but also mediated by parent’s self-perception and family function. The understanding of the wider contexts in which the individual child, parent and family enter into are therefore too important to ignore, and such an understanding should be the starting point of interventions aiming to improve the conditions for healthy siblings.

1.5 The current thesis

The current thesis analyzed material collected through the “Sibling Project” (Haukeland et al., 2015; Vatne, Fjernestad¹, Haukeland, & Mossige, In prep; Vatne et al., 2015) led by researchers from Frambu Resource Center for Rare Disorders (Frambu, 2016) and the Department of Psychology at the University of Oslo, Norway. By October 2016, 92 families have been recruited for this ongoing project. Baseline data from self-reports from 92 healthy siblings and their parents were analyzed in this thesis.

1.5.1 Research questions

Four main research questions are addressed in this thesis:

1. **What is the level of adjustment among siblings of children with CHC?** Adjustment was measured with the Negative Adjustment Scale (NAS) (Lobato & Kao, 2002) and is conceptualized as interpersonal and intrapersonal functioning, and fear of the disease. Adjustment in the current sample will be compared to results from a US study involving siblings of children with various CHC (Lobato & Kao, 2002). It is expected that the level of adjustment is comparable in the two samples as our sample also consisted of various CHC.

2. **Is the level of adjustment among siblings of children with CHC associated with siblings’ socio-emotional difficulties?** Socio-emotional difficulties was measured with the Strengths and Difficulties Questionnaire (SDQ) (Goodman, Ford, Richards, Gatward, & Meltzer, 2000), and is conceptualized as emotional symptoms, conduct problems, hyperactivity/inattention, and peer relationship problems. Because siblings have been found to be at increased risk of psychological difficulties (Vermaes et al., 2012), it was expected that their level of psychological difficulties would be significantly correlated with their adjustment.
3. Are the levels of sibling adjustment and psychological difficulties associated with siblings’ age and gender? Based on previous studies it was expected that girls would report more psychological difficulties (e.g., Goodman, 2001). Due to inconsistency reported across previous studies concerning age effects on siblings’ adjustment and psychological difficulties (Vermaes et al., 2012), these questions were explored openly with no a priori hypothesis.

4. Are siblings’ adjustment and psychological difficulties correlated with parents’ depression, anxiety, and stress scores? Parent symptoms were measured with the Depression Anxiety and Stress Scale (Lovibond & Lovibond, 1995). Based on a theoretically informed understanding of how the family affects the individual (Antonovsky, 1979; Bronfenbrenner, 1977; Rutter, 1985; Sommerschild, 1998) and empirical evidence (Cohen et al., 1999; Fisman et al., 1996; Giallo & Gavidia-Payne, 2006), it was expected that siblings’ adjustment and socio-emotional functioning would be significantly correlated with parents’ scores.

5. What is the level of parental depression, anxiety, and stress? Based on previous findings it was hypothesized that mothers would report higher symptoms than fathers.
2 Method

2.1 Samples

2.1.1 Child sample

The child sample comprised 92 children. The age range for the children was 8 to 16 years. The mean age was 11.4 years (SD = 2.1) and 54.7% were girls. All were biological siblings of a child with a CHC. In terms of sibling order, 56.5% were older and 34.8% were younger than the sibling with disorder. One (1.1%) was a twin. Sibling order information was missing for 7 participants (7.6%). The number of children in the families ranged from two to six, and 67.4% (n = 62) lived with both parents.

2.1.2 Parent sample

The parent sample comprised 83 fathers and 81 mothers. For 39.2% of the healthy siblings both parents participated in the study. For 34.8% of the siblings only the mother participated, and for 15.2% only the father participated. For 10.8% this information was missing. The mean age of the mothers was 40.8 years (SD = 4.7; range 31 to 53). The mean age of the fathers was 43.8 years (SD = 7.3; range 34 to 59).

The majority of mothers and fathers were married or cohabiting; 75.2% and 73.2% respectively. The number of mothers and fathers who were divorced or separated was 14%. In terms of parental educational level, 35.5% of mothers had higher education up to 4 years (i.e. had completed more than high school), and 38.7% had completed higher education more than 4 years. The equivalent numbers for fathers were 30.1% and 23.7% respectively.

49.5% of the parents reported that they their family economy was good or mainly good, 24% reported that it was neither good nor bad, 8.7% reported their economy to be bad or very variable over time.

2.1.3 Chronic health conditions

The sample represents a wide range of different diagnoses for the child with CHC in line with a non-categorical approach, assuming that different CHCs share certain characteristics that
affect daily functioning of the child with CHC, the parents and the siblings. Some of the
diagnoses have primarily cognitive impact (e.g., impaired intellectual level, memory and
attention functions, language functions). Others have mainly somatic impact (i.e. cardiac
problems, physical disability). Some of the diagnoses have an impact on both cognitive and
somatic functioning. The categorization shown in Table 1 gives an overview of the CHCs
included in the sample. However, it is important to note that there is considerable individual
variability within each diagnosis, and that the category “primarily cognitive in nature” does
not necessarily imply mental retardation. For some of these CHCs, it is not uncommon to
have cognitive impairment without mental retardation, in addition to primarily somatic
symptoms, as is the case for Duchenne muscular dystrophy (Anderson, Head, Rae, & Morley,

Another distinction in the data set is between rare disorders and more prevalent disorders.
Approximately half the sample (49.4%) comprises families where the child has more
prevalent CHCs, such as for instance Down syndrome, Cerebral Palsy and Autism spectrum
disorders. The other half of the sample (50.6%) comprises families where the children have
CHCs classified as rare disorders, such as for instance Fragile X syndrome, Smith-Magenis
syndrome, and Velocardiofacial syndrome (See Table 1 for an overview of diagnostic
categories). A CHC is defined as rare when it affects less than 1 in 2 000 citizens (Eurordis,
2014).

Table 1. Chronic health conditions represented in the sample (N = 92)

<table>
<thead>
<tr>
<th>Sibling diagnosis</th>
<th>n</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autism spectrum disorders</td>
<td>26</td>
</tr>
<tr>
<td>Rare disorders, primarily cognitive impact</td>
<td>25</td>
</tr>
<tr>
<td>Rare disorders, primarily somatic impact</td>
<td>13</td>
</tr>
<tr>
<td>Congenital heart disease</td>
<td>8</td>
</tr>
<tr>
<td>Down syndrome</td>
<td>7</td>
</tr>
<tr>
<td>Ultra-rare or unknown disorder</td>
<td>7</td>
</tr>
<tr>
<td>Neurodegenerative disease</td>
<td>4</td>
</tr>
<tr>
<td>Cerebral palsy</td>
<td>2</td>
</tr>
</tbody>
</table>
2.2 Procedures

The participants were recruited from Frambu (www.frambu.no) and the following collaborative associations: Ups & Downs Norway, the Cerebral Palsy Association Norway (CP-foreningen), the Association for children with congenital heart conditions (Foreningen for hjertesyke barn), NevSom – National Resource Centre for Neurodevelopmental Disorders (NevSom – Nasjonalt kompetansesenter for nevroutviklingsforstyrrelser og hypersomnier), and the Norwegian Association for Autism Spectrum Disorders (Autismeforeningen).

Recruitment started in 2014 and is ongoing. 155 families have been approached to date, of which 92 (60%) have consented to participation.

The target age group of siblings was 8 – 16 years. The aim was to include different types of CHCs with different challenges in accordance with a non-categorical approach; some primarily somatic, some primarily cognitive, some with aspects of both. Some of the diagnoses are progressive in nature. The diagnoses have different levels of intrusiveness in the lives of the families and pose different challenges on the siblings and the family system.

At baseline, parents completed a questionnaire on demographic and illness variables, and self-reports on mental and emotional variables, before the first group session. The healthy siblings filled out self-reports on emotional and communication variables.

The aim of the Sibling study is to measure changes in various sibling and parent measures before and after a parent-child intervention.

The intervention aims to improve parent and child communication about CHC, and the challenges siblings’ face, the ultimate aim being to increase psychological health in siblings through supportive group sessions, communication training, and psychological education. Since the current thesis is based on baseline data only, a short description of the intervention is to be found in the attachments.

2.2.1 Setting

The study took place at five-day family residential courses at Frambu, or at family gatherings organized by the collaborating associations. Courses at Frambu aim to provide parents with information about the disability and their social rights, and create an opportunity to meet other
families in similar life circumstances which is believed to be of importance for children with CHC, healthy siblings and parents. The children attend activities such as arts and crafts, swimming, outdoor activities, and school work (Vatne et al., 2015). The family gatherings organized by the different associations had mainly a social aim for the families to meet other families with similar challenges. Some of the gatherings were specifically aimed at healthy siblings and their parents.

### 2.3 Measures

#### 2.3.1 Negative Adjustment Scale (NAS)

The Negative Adjustment Scale (NAS) (Lobato & Kao, 2002) based on the Sibling Perception Questionnaire (SPQ) (Sahler & Carpenter, 1989) is a child self-report measure including 18 items designed to measure three dimensions of sibling adjustment to non-categorical CHCs: Interpersonal, relating to interpersonal interactions and relationships and how disease influence the relationship between the healthy sibling and significant others (e.g. “I wish my parents would spend less time with my brother/sister”). Intrapersonal, concerned with how siblings perceive the disease to affect them (e.g. “I wonder why my brother/sister got sick”). Fear of disability/disease (e.g. “I worry that I can catch cancer from my brother/sister”). The siblings are asked to respond according to how they generally (not time specific) experience their situation as a brother or sister to a child with CHC, and rated the items on a Likert scale from 1 (aldri/never) to 4 (mye/usually). Higher scores reflecting more negative sibling adjustment to CHC. Lobato and Kao (2002) reported an alpha coefficient for the composite Negative Adjustment scale as .79. In the current sample the inter-item reliability of the Negative Adjustment Scale (18 items) was $\alpha = .71$.

#### 2.3.2 The Strengths and Difficulties Questionnaire (SDQ)

The Strengths and Difficulties Questionnaire (SDQ) (Goodman, 2001; Goodman et al., 2000; www.sdqinfo.com, 2012) is a 25-item self-report measure designed to assess socio-emotional functioning in children aged 3 to 16 years. The child is asked to rate 25 items based on the last six months. The 25 attributes items, some positive and others negative, comprise 5 subscales; emotional difficulties, conduct problems, hyperactivity and inattention, peer difficulties, and pro-social behavior. Subscales 1 to 4 generate a total difficulties score (based on 20 items).
Respondents rate each item on a Likert scale from 0 to 2 (0 stemmer ikke/not true, 1 stemmer delvis/somewhat true, or 2 (stemmer helt/certainly true). Higher scores indicate more problems, except for the subscale pro-social behavior where higher scores indicate better adjustment. The SDQ has demonstrated sound psychometric properties for all subscales, and adequate test-retest reliability (Vostanis, 2006). The SDQ also has adequate concurrent validity and ability to distinguish between community and clinical samples (Goodman, 2001; Goodman & Scott, 1999) In the current sample the inter item reliability of the SDQ was $\alpha = .80$.

### 2.3.3 The Depression Anxiety Stress Scales–21 (DASS-21)

The Depression Anxiety Stress Scales–21 is a 21-item self-report questionnaire designed to measure the severity of a range of symptoms common to depression, anxiety, and stress (Lovibond & Lovibond, 1995). The psychometric properties of DASS has been shown to possess satisfactory psychometric properties and the factor structure was substantiated both by exploratory and confirmatory factor analysis (Lovibond & Lovibond, 1995). DASS has demonstrated satisfactory reliability, convergent and discriminant validity with other instruments (Brown, Chorpita, Korotitsch, & Barlow, 1997; Wergeland et al., 2015). Henry and Crawford (2005) reported the reliabilities for the DASS-21 in a non-clinical sample, broadly representative of the general adult UK population ($N = 1,794$) to be; $\alpha$ was .93 for the total scale, for the Depression scale $\alpha$ was .82, for the Anxiety scale $\alpha$ was .90, and for the Stress scale $\alpha$ was .93. Wergeland et al. (2016) reported excellent internal consistency of the DASS with $\alpha = .95$ based on a Norwegian non-clinical sample.

In completing the DASS, the individual is required to indicate the presence of a symptom over the previous week. Each item is scored from 0 (passer ikke i det hele tatt/did not apply to me at all over the last week) to 3 (passer best, eller mesteparten av tiden/applied to me very much or most of the time over the past week). The essential function of the DASS is to assess the severity of the core symptoms of Depression, Anxiety and Stress.

Internal consistency of the DASS in the current sample was excellent $\alpha = .95$.

The inter item reliability for the DASS total was $\alpha = .91$ for mothers, and $\alpha = .92$ for fathers. The inter item reliability for the subscale Depression was $\alpha = .87$ for mothers, and $\alpha = .88$ for fathers. The inter item reliability for the subscale Anxiety was $\alpha = .87$ for mothers and $\alpha = .61$.
for fathers. The inter item reliability for the subscale Stress was $\alpha = .86$ for mothers and $\alpha = .84$ for fathers.

### 2.4 Data analysis

The data was analyzed using SPSS version 22.0. Descriptive analyses and correlational analyses (Pearson’s $r$) were run. Effect size differences were calculated using the formula (Cohen’s $d = \frac{M_{group\ 1} - M_{group\ 2}}{pooled\ SD}$). Cohen’s (1992) criteria were used to define magnitude of effect sizes (0.10 to 0.29 = small, 0.30 to 0.49 = medium, and >0.50 = large).

### 2.5 Ethical issues

Those willing to participate provided written consent. For children aged $\geq 16$ years, written consent was obtained both from parents and the child, while children $< 16$ years old gave verbal assent and parents provided written consent. Reasons for nonparticipation were not registered to avoid unnecessary pressure on the families.

The project was approved by the local institutional review board, The Regional Committee for Medical and Health Research Ethics, South-East Norway (project no. 2013-2118).
3 Results

3.1 Negative Adjustment Scale (siblings)

The mean pr. item NAS score for the total sample was 2.1 (SD = 0.4, range 1.4 - 3.2, possible range 1.0 – 4.0) with no significant difference between boys and girls (p = .555). There was no significant correlation between the NAS score and child age (r = -.09, p = .45). In a previous study of siblings of children with CHC, the mean pr. item NAS score was 2.3 (SD = 0.4) (Lobato & Kao, 2002). The effect size difference between these samples was medium with $d = 0.49$.

3.2 Strengths and Difficulties Questionnaire (siblings)

The mean pr. item SDQ score for the total sample was 0.5 (SD = 0.3, range 0.1 – 1.3), possible range 0.0 – 2.0). The difference between boys´ scores (M = 0.5, SD = 02) and girls´ scores (M= 0.6, SD = 0.3) approached significance (p = .059). There was no significant correlation between the SDQ score and child age (r = -.12, p = .28).

3.3 DASS-21 (parents)

See Table 2 for DASS scores for parents. The possible range of the DASS (mean pr. item) was 0.00-3.00.

3.3.1 Considerations of normality

Inspection of the data plots including skewness and kurtosis values indicated that the children’s variables were normally distributed. However, all the scores on the parent total and the parent subscales were positively skewed due to several low scores. The DASS anxiety scales for mothers had one clear outlier; all analyses were repeated without this outlier, however, this did not change the overall pattern of results. Our sample is small and somewhat positively skewed, due to the low scores (the parents report generally low symptoms) (Field, 2009).
Table 2
Correlations between child-reported negative adjustment, strengths and difficulties, and parent reported depression, anxiety and stress for 92 healthy siblings and their parents.

<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>
<th>8.</th>
<th>9.</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. NAS C</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. SDQ C</td>
<td>.56**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. DASS M</td>
<td>-.08</td>
<td>-.13</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Dep M</td>
<td>.00</td>
<td>-.03</td>
<td>.87**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Anx M</td>
<td>-.14</td>
<td>-.10</td>
<td>.86**</td>
<td>.65**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Stress M</td>
<td>-.95</td>
<td>-.18</td>
<td>.91**</td>
<td>.67**</td>
<td>.66**</td>
<td>1</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7. DASS F</td>
<td>.22</td>
<td>.04</td>
<td>.12</td>
<td>.24*</td>
<td>.17</td>
<td>-.05</td>
<td>-1</td>
<td></td>
<td></td>
</tr>
<tr>
<td>8. Dep F</td>
<td>.25*</td>
<td>.05</td>
<td>.13</td>
<td>.23*</td>
<td>.16</td>
<td>-.03</td>
<td>.94**</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>9. Anx F</td>
<td>.19</td>
<td>.05</td>
<td>.00</td>
<td>.16</td>
<td>.03</td>
<td>-.13</td>
<td>.84**</td>
<td>.71**</td>
<td>1</td>
</tr>
<tr>
<td>10. Stress F</td>
<td>.17</td>
<td>.01</td>
<td>.16</td>
<td>.25*</td>
<td>.22</td>
<td>-.02</td>
<td>.93**</td>
<td>.79**</td>
<td>.69**</td>
</tr>
</tbody>
</table>

*Note. NAS = Negative Adjustment Scale. C = Child. SDQ = Strengths and Difficulties Questionnaire. DASS = Depression Anxiety Stress Scale. M = Mother. Dep = Depression. Anx = Anxiety. F = Father. * = correlation is significant at the p < .05 level. ** = correlation is significant at the p < .001 level.*
There were no significant differences between mothers and fathers on the DASS total or the depression and the anxiety subscales (all $p > .110$). However, there was a significant difference between mothers and fathers on the DASS stress subscale, with mothers reporting more stress ($M$ mothers $= 0.73$ (SD = 0.59); $M$ fathers $= 0.48$ (SD = 0.50); $t = -2.917$, $p < .05$).

See Table 3 for correlations between all child and parent-reported variables. The only significant correlation between parent scores and child scores, was between children’ NAS and fathers’ depression. Note that none of the DASS scales were significantly correlated with child age (all $r < .22$, all $p > .059$; data not shown).
4 Discussion

The aim of this thesis was to investigate healthy siblings’ adjustment and socio-emotional functioning, and whether, or to what extent, sibling adjustment is correlated to parents’ depression, anxiety and stress. The possible correlation between parent’s mental health and the healthy siblings’ adjustment is an understudied theme in sibling research, although a family system perspective implies that the presence of a sibling with a CHC will impact on the psychological development and functioning of their typically developing siblings.

4.1 What is the level of adjustment among siblings of children with CHC?

It was found that siblings reported only slightly heightened negative adjustment with a mean score pr. item (NAS) of 2.1 on a possible 1.0 to 4.0 mean pr. item scale. In Lobato and Kao (2002)’s American sample of 54 healthy siblings, the mean score pr. item (NAS) was higher than this at 2.3 (Lobato & Kao, 2002). The effect size difference between these samples was medium. This means that the children partaking in our study are not children high in negative adjustment, they are typically developing, healthy children.

One possible explanation for this finding may be that the stressors siblings experience generates resilient responses, and that the siblings adjust and cope despite major life events and daily hassles due to the CHC. Living with a sibling with CHC may reinforce the healthy siblings’ self-concept, self-esteem, and sense of competence. In support of such a resilience hypothesis, a few studies have found that CHCs enhance prosocial behaviors, independence, and competence in siblings e.g. (Tøssebro, Kermit, Wendelborg, & Kittelsaa, 2012). Despite extra care-giving tasks, stressed and overworked parents, and experiences that can be exerting, most sibling grow up without significant difficulties.

An IASSIDDD Position Paper from 2014 provides a concise authoritative summary of existing scientific knowledge regarding the situation, challenges and well-being of families that include a child with intellectual impairment and/or disabilities. The overall finding was that it appears that siblings of children with CHC are at minimal (if any) risk of psychosocial problems (IASSIDDD, 2014). Our findings seem to be in line with this. Most siblings seem to think of themselves as normal children, although many have had to take on more
responsibility towards their siblings with CHC and towards their parents. Tøssebro, Kermit, Wendelborg, and Kittelsaa (2012) found that healthy siblings experience a strong solidarity and loyalty towards their family. This solidarity might be understood as an element of what Sommerschild in her model identifies as belonging to a family and network. The siblings, through their competence and through their normality provide something useful to the family system. There is possibly also an element of “a sociology of acceptance”; that the unusual becomes everyday-life, a new kind of “normality” for the siblings.

Parent and family factors have been found to be stronger predictors of sibling adjustment difficulties than siblings’ own experiences of stress and coping (Giallo & Gavidia-Payne, 2006). The socio-economic status (SES) of the family is a known predictor of sibling adjustment, and as the families in our sample had high SES including a very high education level, this might affect both the level of adjustment and psychosocial difficulties reported by the children, and the level of depression, anxiety, and stress reported by the parents. Thus, the low negative adjustment scores identified in the current sample may in part be a reflection of the high SES in the present parent sample.

Given the contradictory findings in the sibling literature, one cannot dismiss the fact that many studies find that healthy siblings are at risk of psychosocial difficulties. One possible explanation for our findings might be the method of data collection. In a review of twenty-five published studies from 1972-1999, Rossiter and Sharpe (2001) found that the method of data collection was a moderator variable. Self-report produced the smallest negative effect size, direct observation the largest (Rossiter & Sharpe, 2001). Both NAS and SDQ are self-report measures, thus it is possible that this might have had an effect on the results. The use of self-report may have led to under-reporting, as well as the children under-reporting the strain of their interpersonal relationships with significant others. It is well known from the sibling literature that the healthy siblings often strive to protect their overburdened parents (Vermaes et al., 2012). It is also well known that children are very loyal to their parents and their families, and one possibility is therefore that they have under-reported to protect their parents. The siblings filled out the self-reports at home, and might therefore have been conscious of the possibility of the parents reading their answers.

The findings of the current study show that the siblings in our sample, on group level, have little negative adjustment. Given these findings it is therefore important to identify the children who might struggle and who might be negatively adjusted. even though they, as a
group, don’t seem to have much negative adjustment. Perhaps identifying the individual children/families at risk is of more clinical urgency, than studying these families as a group.

4.2 Is the level of adjustment among siblings of children with CHC associated with siblings’ psychological difficulties?

Sibling adjustment was found to be significantly associated with socio-emotional difficulties in the current study, and the correlation was large. Based on this correlational finding, it cannot be concluded if general symptoms (measured through SDQ: socio-emotional functioning; emotional difficulties, conduct problems, hyperactivity and inattention, peer difficulties, and pro-social behavior) influence adjustment, or vice versa. It can simply be inferred that higher negative adjustment covaries with more general symptoms, in either direction. This can be regarded as validation of the NAS-scale. As it is associated with the validated and well used measure SDQ, the NAS scale can be argued to measure a phenomenon that is sibling specific, yet related to general symptom burden. This indicates that the NAS-scale is sound and that the low scores in our sample are valid. Negative adjustment is a phenomenon that is likely to be associated with socio-emotional difficulties, although the association might be bi-directional. Longitudinal studies are needed to investigate the direction of the association.

4.3 Are the levels of sibling adjustment and socio-emotional difficulties associated with siblings’ age and gender?

Some previous studies have found that the age of the healthy sibling plays a role in how they experience their situation, with older children reporting higher symptoms of negative adjustment and socio-emotional difficulties (Lobato & Kao, 2005). In the present sample no significant correlation with age was found. This is in line with what Vermaes et al. (2012) found in their meta-analytic study, which reported that mean age did not significantly moderate internalizing or externalizing problems, although it did moderate self-attributes (Vermaes et al., 2012).
No significant gender difference was found in the current sample. This was somewhat surprising, as previous studies have shown gender differences among siblings, with girls reporting more symptoms, especially internalizing difficulties, compared to boys. However, the finding is in accordance with the two most recent meta-analyses (Sharpe & Rossiter, 2002; Vermaes et al., 2012). The difference in SDQ scores between boys and girls approached significance, with girls scoring higher than boys. With an increased sample size and more variation in the SDQ scores, previous findings of significant gender differences on the SDQ could possibly have been replicated in the current sample.

Possible explanations for the non-significant correlation with age and gender might be the generally low scores amongst the children in the sample. Low scores give a smaller potential for differences. The sample size might also contribute to the non-significant correlation with age and gender. If the associations of age and gender with adjustment are small, a larger sample would be needed to uncover significant differences.

Based on the findings there is no basis to state that girls or younger/older siblings are more vulnerable. This is an important implication given that our finding is ecologically valid. An implication of this is that neither age nor gender should be sole factors in determining which children should be screened for negative adjustment problems or socio-emotional difficulties, nor in who should be offered to partake in interventions. A general symptom and risk evaluation, including the family system surrounding the child, should be the basis for identifying the children at risk and in need of support and clinical interventions.

4.4 Are siblings’ adjustment and psychological difficulties correlated with parents’ depression, anxiety, and stress scores?

Contrary to the research hypothesis, siblings’ adjustment was not correlated with parent’s depression, anxiety, and stress scores, with the exception of fathers’ depression and children’s NAS score. The role of paternal depression for child adjustment is an interesting and potentially important finding. It is important to note that fathers’ depression was only significantly correlated with children’s NAS scores, and not with the children’s SDQ scores. This indicates that there is something specific about the sibling situation that is associated with fathers’ depression, and not the general functioning of the children. However, the
direction of the correlation is not known; do negatively adjusted children make the fathers depressed or does having a depressed father lead to negatively adjusted children?

A limitation of past research has been the almost exclusive focus on maternal parenting, ignoring the potentially influential role of fathers in influencing child adjustment. Support has been found for the importance of paternal functioning for child adjustment. Friedman et al. (2004) found a greater number of significant effects for father-reported predictors than for mother-reported predictors in a study of the implications of parent functioning for preadolescents’ adjustment (CHC = Spina Bifida; \( n = 68 \)). Father-reported symptomatology and parenting stress, as well as a combined mother and father report of couple-relationship satisfaction, were all found to be associated with various domains of child adjustment. These findings testify to the important role of fathers in child socialization (Friedman et al., 2004).

A possible factor making paternal depression relatively more influential on the siblings’ adjustment relative to maternal depression, is the different roles that mothers and fathers undertake in child rearing. Bögels, Bamelis, and Van Der Bruggen (2008) suggest that the different roles are partly biologically driven and that fathers are more explorative and challenging in their parental behavior, with mothers being more protective. In a meta-analysis of associations between maternal and paternal parenting behaviors, anxiety and its’ precursors in early childhood, Möller, Nikolić, Majdandžić, and Bögels (2016) found a small association between father’s anxiety and anxiety in the child. This association is according to the authors partially explained by paternal parenting behaviors, and that typically paternal parenting behavior (challenging parenting, autonomy granting) challenge their children both physically and socio-emotionally, and thus impact on the child’s level of anxiety. One might hypothesize that a depressed father, in a similar manner as an anxious father, might affect the child’s adjustment. In a study of parenting and children’s internalizing symptoms Sluis, Steensel, and Bögels (2015) found that paternal parenting, contrary to previous assumptions and weighting in research, is as important as maternal parenting with respect to children’s internalizing symptoms. The implication of the findings of these studies, including the findings of the father’s role in relation to the children’s negative adjustment in the present study, is that fathers should be included in interventions and child treatment.

Another element emphasized by Möller et al. (2016) is that fathers are specialized evolutionarily in opening children to the outside world, encouraging independence, and stimulating risk taking behavior. Mothers are specialized in caring, nurturing, and protective
behavior. It might be expected that a lack of challenging and autonomy granting behavior in fathers would have a stronger effect on children than that of the mothers in this area. As afore mentioned, the Möller et al. (2016) study is concerned with the effect of anxiety in fathers on children, but it is possible to imagine depression in fathers having similar effects and thus affecting the adjustment of the children.

One implication of our finding is that it is important to assess and be aware of fathers’ depression as it might have a relatively strong influence on the healthy siblings’ level of adjustment.

### 4.5 Parents’ depression, anxiety and stress

The parents in the current sample reported low levels of depression and anxiety, and some stress (mothers significantly more than fathers) as measured on the DASS-21 scale. DASS-21 requires the respondent to indicate symptom levels experienced in the past week. However, for this population of parents, whose life situation represents a chronic source of stress, one may question the relevance of asking about the stress experienced exclusively over the last week. One could imagine that a more relevant measure would be retrospectively at time of diagnosis and at crucial points during the course of illness, and at typically taxing everyday-life situations.

#### 4.5.1 Socio-economic status and education

Another possible reason for the low scores amongst the parents on depression, anxiety, and stress might be the level of education and SES. In our study 35.5% of mothers had ≤ 4 years of higher education, in the general population the equivalent percentage is 27.3%. The number of mothers who had ≥ 4 years of higher education in our study was 38.7%, in the general population the equivalent percentage is 8.3%. The equivalent numbers for fathers were in our study: ≤ 4 years of higher education 30.1%, in the general population 18.7%, ≥ 4 years of higher education 23.7%, in the general population 10 % (SSB, 2016). These numbers suggest that the nature of the study appeals to people with higher education, and to parents who perhaps already have an understanding of, and a will to meet, the needs of the healthy siblings. There was no available information about non-respondents and as a consequence it was not possible to examine selection-bias statistically. However, the respondents in this
study were highly educated and it is known that the levels of anxiety and depression are lower in highly educated people than in people with lower education. It is also suggested that anxiety and depression in parents of a child with CHC are influenced by demographic, social, as well as disease related factors (Oers et al., 2014). Based on this, one might suggest that levels of anxiety and depression possibly are higher in parents of chronically ill children who don’t participate in such a study. And that their children possibly would be more affected by this.

One may connect level of education to Sommerschild’s concept of competence; that education leads to knowledge and knowing how. The health care system might appear elusive, with a complicated bureaucracy and a technical language far removed from everyday language and everyday experience. Having gone through higher education means that one has become acquainted to this kind of language and possibly making the parents more prepared to maneuver the system, and ultimately give a sense of agency as well as ensuring that the family gets the help they need.

The non-significant differences on maternal and paternal depression and anxiety is somewhat surprising given what is known about gender effects on these variables e.g. (Oers et al., 2014; Pelchat et al., 2003). Possible reasons for this might be the level of education amongst the mothers in our sample. The mothers in our sample are highly educated, and on average they have higher education than the fathers in the sample. Another possible reason might be the support and the economical safety provided by the Norwegian health care system. Women who traditionally would stay at home to care for the child with CHC might due to the systemic support, be able to keep a job outside the family, which one might speculate give the mothers a sense of being useful, and keep an identity that encompass more than being mother and caretaker exclusively. The system also offers the possibility of being paid as a caretaker for the child with CHC, allowing for an identity of someone working, contributing and being useful from a societal point of view.

Despite its shortcomings, The Norwegian Health Care System ensures that having a child with CHC does not mean economic ruin. The families receive financial support, and medical services are free of charge. Possible implications of this are that the families may concentrate on the caregiving and the parenting of their children with and without CHC, which may have the biggest influence on mothers who traditionally have stayed at home with caregiving tasks.
Another possible explanation for the lack of gender effects in our sample may be the fact that gender equality in Norwegian families is quite high; both mothers and fathers are expected to take turns at child rearing, house work and ultimately at caring for a child with CHC. The gender equality is possibly higher in families where the parents are highly educated. To sum up, our result may reflect a reality for families with higher education, high gender equality in the couple, living in a society with a relatively well-functioning health care system (Frønes & Kjølsrød, 2010). These are parents who are coping well possibly due to internal aspects of the individuals (higher education), the organizing of the roles within the family (gender equality), a society with a relatively well-functioning support system (laws, economics, culture, support). In addition, support from society may also reflect the values and attitudes towards CHCs, which in turn affect the families living with CHCs.

4.6 Limitations and strengths

There are several limitations in the present thesis. A main limitation is the representativeness and generalizability due to the relatively small sample size. In addition, our sample represents a range of different disorders with different pathological processes and different prognoses, and the results may therefore not be generalizable to other diagnoses. The large variation in both manifestations and courses of these disorders indicates that information on the specific impact of certain disorder may have been missed. It is likely that the intrusiveness of the disorder is important, and that it influences how it affects the family system and the healthy sibling. Some studies have found that highly intrusive disorders have a more negative impact on healthy siblings (Sharpe & Rossiter, 2002; Vermaes et al., 2012). How siblings and family systems are affected by specific diagnoses is an important variable to investigate in future research.

To be able to identify potential group differences, larger and more homogenous age and diagnostic groups may be needed. However, the broad range of different disorders included in the sample may represent a strength as the variation means that the findings may be applicable for siblings and parents of children with various disorders.

Siblings from a broad age range (8 – 16 years) were included. This might represent a strength as findings may apply across a broad developmental level. However, the wide age span in the
sample means that there are big differences in developmental levels between the children. Given the sample size this might represent a limitation of the study.

A possible bias of this study was the recruitment from special settings. The families were recruited either through Frambu or through user associations for different diagnoses (See 2.2 Procedures). This might have led to a bias towards families who were already concerned with, and conscious of how the special situation of their family might affect their healthy children. This possible bias is further strengthened by the fact that not all families who were offered participation accepted. Perhaps the families who signed up differ on several factors (e.g. commitment, interests, SES, time management) compared to families who did not sign up. One might hypothesize that this is self-selected sample, consisting mainly of families where the parents are both aware of the needs of the healthy sibling, and who have the energy to participate. As reasons for non-participation was not investigated, one is left to speculate that the families who chose not to participate might be the ones who are possibly overworked and overwhelmed with their situation.

As discussed earlier, the use of self-reports may affect the results due to under-reporting. However, using self-reports from healthy sibling, mother, and father might on the other hand be considered a strength. Most of sibling research have been based on reports from parents, and mainly the mother, on behalf of the healthy sibling. Most studies lack information from the father. This has been considered a general weakness of the field. A strength of our study is therefore that it includes the different perspectives of the healthy sibling, the mother and the father through self-report.

4.7 Implications

4.7.1 Research implications

The field of sibling research is quite substantial. Early studies tended to find stronger negative effects on siblings, possibly due to moderation effects of methodological aspects of studies, but also due to educational, medical, and political and societal contexts. Smaller negative effects on siblings in more recent studies, might in part be due to change in these contexts around the families which have incorporated more advocacy, intervention, and habilitation
efforts aimed at optimal participation and inclusion of children with CHCs and their families in society, as well as improved medical care (Vermaes et al., 2012).

Most studies in the field have relied solely on self-report, often meaning parent report on child functioning, and many only included mothers as the sole source of data. An older study showed that parents tended to underestimate their well children’s worries and challenges (Menke, 1987), whereas newer studies indicate that mother were more alert to their well children’s concerns (Taylor, Fuggle, & Charman, 2001). Another common limitation in the field is the lack of comparison sample, and it is therefore remains unclear whether effects are qualitatively or quantitatively different (Drotar, 1997). Other limitations of the field of research is the heterogeneity of diagnoses, the general skewness in the samples towards mothers, and the method of reports; many studies of healthy siblings are based on parent-report, not self-reports from the siblings themselves.

A number of studies have investigated the impact of CHCs on siblings’ psychosocial functioning. However, the results appear difficult to synthesize; although the two most recent meta-analyses conclude that there is a significant small negative effect of CHC on siblings (Rossiter & Sharpe, 2001; Vermaes et al., 2012), other studies have found none or only small effects on the healthy siblings. Our findings are in lines with this; according to our data, the healthy siblings are coping well, which may be interpreted as a sign of improvement of support and understanding in the systems around the families, as well as resilience in the individual child.

The field of research on parents of children with CHCs is likewise divergent. Although most studies find that parents experience more stress in their parenting role than parents of normally developing children, the findings on anxiety and depression are more divergent. Our sample of parents show that the parents are coping well, but that they do experience stress to some degree. In our sample the mothers report more stress than the fathers, which is in line with most previous research.

Vermaes et al. (2012) concluded that future research on siblings of children with CHCs should focus on mechanisms of risk and resilience. Our findings confirm this: The children do not seem to experience their situation as more taxing and challenging than they can handle with support from their parents, and the parents don’t report high levels of symptoms of depression, anxiety or stress. Thus a natural inference from this is that the families in our
sample show resilience and coping; the parents have a generally low levels of depression, anxiety, and stress, and the healthy siblings don’t report negative adjustment nor socio-emotional difficulties. Further, it is suggested that future research should focus on the moderating effects on resilience of important intrapersonal factors (e.g., temperament and coping styles) and socio-ecological factors (e.g., social support, parental support) (Vermaes et al., 2012).

**Diagnosis: Categorical vs. non-categorical approach**

To what extent is the stress experienced by parents and healthy siblings tied to the characteristics of the actual diagnosis of the child with CHC? Chronic illness or disorder vary greatly regarding severity, prognosis and progression, impact on daily life, behavioral problems, intrusiveness of treatment (little versus highly intrusive), and life-threatening potential (low versus high mortality). Another variable is whether the diagnosis is common or rare. Lack of knowledge of rare disorder might pose a specific challenge for the families where the child has a rare CHC. Lack of knowledge may affect the competence element in Sommerschild’s model; the parents and the health care system has less knowledge of the disorder and therefore is less likely to understand it and know how to deal with the impact of the disorder. Another possible consequence of a rare diagnosis is the way the family is met by the health care system, and some studies indicate that distress in siblings lessen as a consequence of education about the CHC condition (Lobato & Kao, 2002).

The sample in the present study is heterogeneous, including a wide spectrum of different diagnoses, some being rare and some being more common. Some have severe somatic symptoms (e.g. congenital heart disease) some are progressive in nature (e.g. Duchenne muscular dystrophy), some have mental retardation to varying degrees (e.g. Cri du chat syndrome, Prader Willi syndrome), some often involve behavioral problems (e.g. Autism spectrum disorder), and some have a combination of these symptoms.

These conditions vary along a number of dimensions including etiology, stability and predictability, threat to life, complexity and demandingness, and the physical and social restrictions they impose; in short, they vary to what extent they may be considered a “intrusive” or “non-intrusive” condition, a distinction often seen in the literature (e.g. Vermaes et al., 2012). However, they are similar in the sense that there are no known cures for these disorders, they result in regular contact with a wide variety of health services and
possible hospitalization, they require treatment, they impose various limitations on everyday life, and require personal readjustment of the total family. This has led some researchers to argue for a non-categorical approach to chronic illness, arguing that the similarities are greater than the specific variations of the individual conditions (Stallard, Mastroymnopoulos, Lewis, & Lenton, 1997). However, there are indications in the research that siblings of children with life-threatening CHCs and CHCs requiring intrusive day-to-day treatment had significantly more internalizing and externalizing problems, and had significantly less positive self-attributes (Vermaes et al., 2012). There is a need in the field for more diagnosis specific research to disentangle the effects of different diagnoses.

4.7.2 Clinical implications

Our study highlights the importance of familial and parental contributions to the sibling adjustment process. This reinforces the finding that sibling adjustment is in part determined by interactions with other family members, emphasizing the need to consider sibling experiences within the family context (Giallo & Gavidia-Payne, 2006). Further research is needed to identify those siblings who are particularly at risk for mental health problems and second, research is needed to investigate the principles of what works in helping siblings to cope with the stressful events related to CHCs (Vermaes et al., 2012).

Implications of the current study are as follows: 1) The Negative Adjustment Scale (Lobato & Kao, 2002) was reliable with this mixed sample. 2) Parents and healthy siblings need to be understood and approached with the perspective that they are normal, healthy, and coping; and that as a group they display resilience. 3) Some individuals, both among the parents and among the children report severe difficulties in the domains that was measured. An important focus for clinical practice is to find ways of identifying these individuals, and helping them. 4) Fathers reporting high levels of depression may have a unique negative influence on their children, or negative adjustment in siblings may have a particular effect on depression levels among fathers. An important focus for clinical practice is how to identify these among all the parents and siblings who are coping well. 5) It is important that both fathers and mothers participate in family support groups/interventions. Our finding confirms that the inclusion of fathers both studies/research and in interventions aimed at healthy siblings is necessary and needed. One natural implication of the importance of the parents’ role for the siblings’ adjustment and coping is to incorporate both mothers and fathers into sibling group
interventions, which is also pointed out in several previous studies (Fisman et al., 1996; Lobato & Kao, 2002).

It is a challenge to clinicians to design and implement intervention programs that take into account the entire multifaceted and dynamic context in which any given family is situated. Thus keeping in mind how the individual child and the individual family are deeply situated in the relationships, the family, the community, and the cultural place, and how effects move across these different systems, is important.

**Interventions**

In a systematic review of interventions to support siblings of children with chronic illness or disability, Hartling et al. (2014) found that improvements were found for a variety of outcomes in studies evaluating a support group format including reduced anxiety and depression, increased self-esteem and self-concept; increased involvement with siblings; increased knowledge; and improved mood state, feelings and attitudes.

In a systematic review of intervention and support for siblings (6-12 years of age) of youth with developmental disabilities Tudor and Lerner (2015) found that more positive outcomes were consistently demonstrated for youth who participated in treatment versus youth who did not e.g. (Giallo & Gavidia-Payne, 2008). Further findings of the review was that treatment that included social and psychoeducational components, and that included parental involvement was evidenced as more effective e.g. (Lobato & Kao, 2005). Based on our findings, it seems safe to suggest that it is important that the father is included in interventions, as well as the mother, and that interventions targeting the entire family system are to be recommended.

“The Sibling Study” is an exploratory trial of a manual based group intervention for siblings of children with chronic illness or disability and their parents (Vatne et al., In prep). Main aims are to explore and improve psychological health, knowledge about diagnosis, and communication between well child and parents in families of children with CHCs (see Attachment). Previous research has emphasized the importance of involving parents in interventions aiming to increase psychosocial wellbeing among siblings of children with CHC. Lobato and Kao (2005) carried out an intervention which was designed to address sibling challenges that cut across types of diagnostic conditions, which involved both healthy
sibling and parents, and included sessions of collateral and integrated sibling-parent groups. They found that sibling knowledge of the CHC and sense of connectedness increased.

Incorporating a parental component in interventions with a group format; e.g. parallel sessions for parents, has been found to enhance and lead to more long-lasting effects of the intervention.
5 Conclusion

This thesis aimed at investigating the assumed correlations between healthy siblings’ adjustment and socio-emotional functioning, and the possible association with parents’ mental health (depression, anxiety and stress) in families living with a child with a CHC.

According to our findings parents report low levels of depression, anxiety, and slightly higher levels of stress. The mothers’ report significantly higher levels of stress than fathers, which is in line with previous research. Contrary to our expectation no correlation between negative adjustment and socio-emotional functioning, and sibling age or gender was found. However, the present thesis found a significant correlation between fathers’ depression and the children’s negative adjustment.

Based on the findings of the present thesis it may be concluded that the NAS (Lobato & Kao, 2002) has satisfactory psychometric properties and is a fit measure of negative adjustment on the population in the present study.

The general picture is one of well-functioning families, where both parents and children are well adjusted and are coping well. Possible reasons for the findings implying coping and resilience are highly educated parents, especially mothers. Other possible reasons include gender equality in the household, economic support and a well-functioning health care service supporting the families in their continuous strive towards a functioning everyday-life.

The most important message arising from the findings is perhaps that little support was found for hypotheses indicating that the healthy siblings constitute a particularly vulnerable and maladjusted group. However, this does not mean that siblings as a group does not need the attention of the health services, and some siblings are struggling and need support. In addition to working towards more effective ways of identifying the siblings at risk, and developing good interventions, it is important to see the siblings as part of the family, and therefore also focus on the mental health of the parents and the functioning of the family system.
References


Eurordis. (2014). What is a rare disease?


Frambu. (2016).


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**SPØRRESKJEMA OM SØSKENS TILPASNING (NAS)**

**Du er eksperten!** Det er ingen som vet bedre enn deg hvordan det er å ha en bror eller søster med spesielle behov. Vi vil gjerne at du skal lære oss mer om dette ved å sette en ring rundt ordet som sier noe om hva du føler.

1. Jeg glemmer at broren eller søsteren min har et problem.
   - aldri
   - litt
   - noen ganger
   - mye

2. Jeg føler meg trist på grunn av broren eller søsteren min sitt problem.
   - aldri
   - litt
   - noen ganger
   - mye

3. Jeg har for mye å gjøre hjemme på grunn av broren eller søsteren min.
   - aldri
   - litt
   - noen ganger
   - mye

4. Jeg er redd for broren eller søsteren min sin funksjonsnedsettelse eller sykdom.
   - aldri
   - litt
   - noen ganger
   - mye

5. Jeg tenker på broren eller søsteren min sin funksjonsnedsettelse eller sykdom.
   - aldri
   - litt
   - noen ganger
   - mye

6. Jeg forstår hvorfor foreldrene mine må bruke tid på broren eller søsteren min.
   - aldri
   - litt
   - noen ganger
   - mye

7. Jeg føler meg sint på grunn av broren eller søsteren min sin funksjonsnedsettelse eller sykdom.
   - aldri
   - litt
   - noen ganger
   - mye

8. Jeg skulle ønske jeg kjente noen som forstår hvordan jeg har det.
   - aldri
   - litt
   - noen ganger
   - mye

9. Jeg skulle ønske foreldrene mine kunne bruke mindre tid på broren/søsteren min.
   - aldri
   - litt
   - noen ganger
   - mye

10. Jeg skulle ønske det var noe jeg kunne gjøre med funksjonsnedsettelsen eller sykdommen til broren eller søsteren min.
    - aldri
    - litt
    - noen ganger
    - mye

*SNU - Spørreskjemaet fortsetter på baksiden*
   aldri    litt    noen ganger    mye

12. Jeg skulle ønske foreldrene mine kunne bruke mer tid sammen med meg.
   aldri    litt    noen ganger    mye

13. Jeg er bekymret for om jeg kan få sykdommen eller funksjonsnedsettelsen til broren eller søsteren min.
   aldri    litt    noen ganger    mye

14. Vennene mine er bekymret for at de kan få sykdommen eller funksjonsnedsettelsen til broren eller søsteren min.
   aldri    litt    noen ganger    mye

15. Foreldrene mine ignorerer meg.
   aldri    litt    noen ganger    mye

16. Andre er mer interessert i broren eller søsteren min enn i meg.
   aldri    litt    noen ganger    mye

17. Andre bryr seg om hvordan jeg har det.
   aldri    litt    noen ganger    mye

18. Problemet til broren eller søsteren min endrer hva vi kan gjøre som familie.
   aldri    litt    noen ganger    mye

Oversatt fra Negative Adjustment Scale (NAS; Lobato & Kao, 2002)
### Sterke og svake sider (SDQ-Nor)


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<td>Jeg blir ofte sint og har kort lunte</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Jeg er ofte for meg selv. Jeg gjør som regel ting alene</td>
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<tr>
<td>Jeg gjør som regel det jeg får beskjed om</td>
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<tr>
<td>Jeg bekymrer meg mye</td>
<td></td>
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<tr>
<td>Jeg stiller opp hvis noen er såret, lei seg eller føler seg dårlig</td>
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<tr>
<td>Jeg er stadig urolig eller i bevegelse</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Jeg har en eller flere gode venner</td>
<td></td>
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<tr>
<td>Jeg slåss mye. Jeg kan få andre til å gjøre det jeg vil</td>
<td></td>
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<tr>
<td>Jeg er ofte lei meg, nedfor eller på gråten</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Jeg blir som regel likt av andre på min alder</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Jeg blir lett distrahert, jeg synes det er vanskelig å konsentrere meg</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>Jeg blir nervøs i nye situasjoner. Jeg blir lett usikker</td>
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<td></td>
<td></td>
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<tr>
<td>Jeg er snill mot de som er yngre enn meg</td>
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</tr>
</tbody>
</table>
Jeg blir ofte beskyldt for å lyve eller jukse

Andre barn eller unge plager eller mobber meg

Jeg tilbyr meg ofte å hjelpe andre (foreldre, lærere, andre barn/unge)

Jeg tenker meg om før jeg handler (gjør noe)

Jeg tar ting som ikke er mine hjemme, på skolen eller andre steder

Jeg kommer bedre overens med voksne enn de på min egen alder

Jeg er redd for mye, jeg blir lett skremt

Jeg fullfører oppgaver. Jeg er god til å konsentrere meg

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Tusen takk for hjelpen
**DASS-21**

Mor/Far

Les gjennom utsagnene og sett en sirkel rundt 0, 1, 2 eller 3 for å vise i hvor stor grad du føler at utsagnet passer for deg *den siste uken*. Det er ingen svar som er riktige eller gale. Ikke bruk for mye tid på et enkelt utsagn.

**Vurderingsskalaen er som følgende:**
0 Passer ikke i det hele tatt
1 Passer til en viss grad, eller noe av tiden
2 Passer godt, eller en god del av tiden
3 Passer best, eller mesteparten av tiden

<p>| | | | | |</p>
<table>
<thead>
<tr>
<th></th>
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<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Jeg ble opprørt av trivielle ting</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>2</td>
<td>Jeg var oppmerksom på at jeg var tørr i munnen</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>Jeg klarte ikke å oppleve noen positive følelser</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>4</td>
<td>Jeg hadde pustevansker (f.eks. pustet alt for fort, andpusten uten fysisk anstrengelse)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>5</td>
<td>Jeg kom ikke i gang med noe</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>6</td>
<td>Jeg hadde en tendens til å overreakere</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>7</td>
<td>Jeg følte meg skjelven (f.eks. følte at bena kom til å gi etter under meg)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>8</td>
<td>Jeg hadde vansker med å slappe av</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>9</td>
<td>Jeg opplevde situasjoner som gjorde meg så engstelig at jeg var utrolig lettet når de var over</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>10</td>
<td>Jeg følte at jeg ikke hadde noe å se frem til</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>11</td>
<td>Jeg ble fort opprørt</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>12</td>
<td>Jeg følte at jeg brukte mye nervøs energi</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>13</td>
<td>Jeg var trist og deprimert</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>14</td>
<td>Jeg ble utålmodig hvis jeg ble forsinket på noen som helst måte (f.eks. heis, trafikklys, når jeg måtte vente)</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>15</td>
<td>Jeg følte jeg kom til å besvime</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>16</td>
<td>Jeg følte jeg hadde mistet interessen for nesten alt</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>17</td>
<td>Jeg følte meg verdiløs som person</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>18</td>
<td>Jeg tok meg lett nær av ting</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>19</td>
<td>Jeg svettet mye (f.eks. i hendene) uten at det var varmt</td>
<td>0</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td>Jeg følte meg redd uten å ha særlig grunn for det</td>
<td>0</td>
<td>1</td>
<td>2</td>
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<td>-----------------------------------------------------</td>
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</tr>
<tr>
<td></td>
<td>Jeg følte livet ikke var verdt å leve</td>
<td>0</td>
<td>1</td>
<td>2</td>
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</tbody>
</table>
Description of the group intervention of “The Sibling Project”

<table>
<thead>
<tr>
<th></th>
<th>Session 1</th>
<th>Session 2</th>
<th>Session 3</th>
<th>Session 4</th>
<th>Session 5</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Siblings</strong></td>
<td>Introduction (20 mins)</td>
<td>Knowledge module</td>
<td>Joint session Communication about diagnosis</td>
<td>Emotion module</td>
<td>Joint session Problem solving</td>
</tr>
<tr>
<td><strong>Parents</strong></td>
<td>Information</td>
<td>Knowledge module</td>
<td></td>
<td>Emotion module</td>
<td></td>
</tr>
</tbody>
</table>

**Intervention**

The intervention aims to improve parent and child communication about a) the disorder and b) the challenges siblings’ face and through this increase psychological health in siblings through supportive group sessions, communication training, and psycho education. The preliminary manual comprises of:

1) An introduction module involving information to parents and a child group session with "get to know each other” activities.

2) A knowledge module partly inspired by Lobato and Kao’s (2002) intervention. The module entails a group session with children to assess their need of knowledge, a group session with parents with education on how to explore children's thoughts and provide developmentally appropriate information, and a joint parent child session where families, under supervision of group leaders, discuss diagnosis related topics.

3) An emotion module inspired by tasks from the CBT intervention ”Friends for life” (Barrett, 2005). The module entails a) a group session with children focusing on their feelings, thoughts, and behaviors related to their siblings’ disorder, b) a parent session focusing on how to communicate in an emotionally supportive manner, and c) a joint family session where families, under supervision, work together with tasks from ”Friends for life” (Barrett, 2005) focused on solving a problem experienced by siblings. See table for a description of the intervention.

Children and parents will be placed in groups with 3-6 participants, about 25 groups in total. There will be three separate child sessions, three parent sessions, and two joint family sessions. Sessions 2-5 last 60 minutes each and will be conducted in three or five days dependent on setting. The groups will be facilitated by trained psychologists/advanced...
psychology students/special education staff who will receive regular supervision by the project group.