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# **PARENTAL REACTIONS IN CHILDHOOD CANCER: DISTRESS, RISK AND RESILIENCE**

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## Abstract

Parents of children with cancer encounter an exceptionally difficult life experience. Acute and long-lasting distress adds to the initial traumatic experience of being told that a family member suffers from a potentially fatal illness. This thesis investigates the determinants of short and long-term psychological effects in parents in two Nordic sites with different routines for care and follow-up. A general aim was increased knowledge and understanding of parental reactions and psychosocial follow-up needs of families. Study I examined the significance of individual strengths for parental resilience against distress by studying the relationships between Sense of Coherence (SOC) and illness-specific and generic distress symptoms. We also studied whether parental *gender, level of education, and use of professional psychological support* influenced that relationship. Data were collected from 190 parents of 126 childhood cancer (CC) survivors, and 208 parents from the general population. SOC was assessed using the SOC-13 questionnaire. Distress outcomes were measured using the multi-dimensional Parental Psychosocial Distress in Cancer (PPD-C) questionnaire, which covers *uncertainty, loss of control, self-esteem, anxiety, disease-related fear, loneliness, sleep disturbances, depression, and psychological and physical distress*. Non-pathological psychiatric symptoms were screened for using the General Health Questionnaire (GHQ-12).

Study II, included 328 parents of 211 CC patients at one of two sites, either in Sweden or Iceland, and 208 control parents. We studied occurrence of cancer-related parental reactions, and compared distress symptoms in the two national samples. A goal was to identify of nation-, site- and organisation-related determinants of parental experiences. Outcomes were evaluated against non-clinical norm data.

Study I provided support for the hypothesis about a protective influence of SOC, and that SOC is associated with resilience also in this population. Influence of SOC was strongest regarding general psychiatric symptoms, physical and psychological stress symptoms, anxiety and depression, although low SOC was associated with more severe distress general. Influence of SOC was stronger among mothers.

Study II showed that both in the Swedish and the Icelandic groups, distress exceeded the level of a non-clinical norm group. In adjusted analyses comparing parent outcomes Icelandic parents scored significantly higher in distress on 5 of 11 PPD-C subscales. Significant differences, primarily concerning illness-specific distress, and uncertainty due to unmet informational needs, were found between parents at the two sites.

The findings in this thesis indicate that parental resilience to cancer-related distress varies with identifiable strength factors. This suggests that a strengths-oriented approach facilitates understanding of the kind of factors that govern parental adjustment to childhood cancer. Addressing resilience instead of pathology helps identifying parents at risk and in need of intensified psychosocial support.

Distress and needs for illness-related information was more prominent among parents at the smaller Icelandic site. This finding indicates a need for routines to meet parental uncertainty, adapted to conditions prevailing at a low-influx centre with limited resources for specialised psychosocial follow-up. Differences between study groups indicate that compensatory measures in local/national and/or site-related arrangements for care, surveillance and information are possible means by which parental psychosocial services can be improved. Differences in this study in parental outcomes may partly be due to the larger centre having more favourable preconditions for providing a multifaceted caring context, including parental fellowship where informal mutual support and shared information contribute to resilience against illness-related distress.



## LIST OF PUBLICATIONS

- I. Gudmundsdóttir, E., Schirren, M., Boman, K. K. (2011). Psychological resilience and long-term distress in Swedish and Icelandic parents' adjustment to childhood cancer. *Acta Oncol*, 50(3), 373-380.
  
- II. Gudmundsdóttir, E., Boman, K.K. Psychological outcomes in Swedish and Icelandic parents following a child's cancer - in the light of site-related differences and organisation. (*manuscript*).

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## **LIST OF ABBREVIATIONS**

ALL	Acute lymphoblastic leukaemia
AML	Acute myeloid leukaemia
ANOVA	Analysis of variance
CC	Childhood cancer
CNS	Central nervous system
GHQ-12	General Health Questionnaire
NHL	Non-Hodgkin's lymphoma
PPD-C	Parental Psychosocial Distress in Cancer
SOC	Sense of coherence
SOC-13	Sense of Coherence Scale
SPSS	Statistical Package for Social Sciences





## INTRODUCTION

Although a rare disease, childhood cancer (CC) in the industrialised countries is the fourth most common cause of death of children under the age of 20, after accidental death, birth defects, and deliberate harm (Centers for Disease Control and Prevention (CDC), 2007).

A child's cancer diagnosis is often followed by a lengthy and invasive treatment, which places most families in a life-situation characterized by extraordinary distress. The child's disease constitutes a parental stressor that existentially threatens the conception of life as predictable and safe, and that for most parents is followed by the immediate fear of losing their child. The change in everyday life requires coping with a suddenly altered life-situation characterised by strain, escalated situational stress, and heightened parenting demands. (Boman, Viksten, Kogner, & Samuelsson, 2004; Dixon-Woods, Findlay, Young, Cox, & Heney, 2001; Giammona & Malek, 2002; Kazak, 1998; Santacroce, 2002; Van Dongen Melman, et al., 1995).

During the past 2-3 decades, several studies have investigated parental reactions to a child's cancer, leading to increased knowledge concerning the psychosocial effects of the illness. The research focus has varied, but in addition to investigating a variety of distress indicators and their severity, earlier studies have also addressed the influence on parental distress of factors such as parental gender, ethnicity, number of children, and parental education, or aspects related to illness-treatment, age of child at diagnosis, time passed since diagnosis, and type of cancer (Boman & Bodegard, 2000; Boman & Lindahl, 2002; Steele, Dreyer, & Phipps, 2004; Vrijmoet-Wiersma, Egeler, Koopman, Norberg, & Grootenhuis, 2009; Wijnberg-Williams, Kamps, Klip, & Hoekstra-Weebers, 2006).

Focusing on the possible determinants of parental distress due to a child's cancer diagnosis may provide better and more specific understanding about parental psychological reactions. This, in turn, can facilitate the improvement of support services for parents, minimizing negative psychological distress and strengthening resilience in the difficult life-situation following a child's cancer.

## MEDICAL FEATURES AND INCIDENCE OF CHILDHOOD CANCER

The incidence of paediatric cancer patients the Nordic countries are approximately equivalent. Reports on incidences for 1985-2004 of eleven main diagnostic groups of childhood cancers in the Nordic countries shows that the most frequent diagnosis is acute lymphoblastic leukaemia (ALL, ~30%), followed by central nervous system tumours (CNS, ~27%), lymphomas/histiocytosis (~12%), and bone tumours (~10%) (NOPHO-Annual-Report, 2010).

Incidences of AML, ALL, and CNS tumours have been stable in the Nordic countries over the past two decades, with the incidence of childhood CNS tumours being among the highest in the world (Hjalgrim, et al., 2003; Schmidt, et al., 2010).

Childhood malignancies vary regarding a number of factors: e.g., peak age at diagnosis and occurrence in boys and girls (NOPHO-Annual-Report, 2010). Cancer in children is most frequent at the age of 5-6 years, and boys are at greater risk of developing cancer than girls (Gustafsson, Heyman, & Vernby, 2007).

The distribution of types of childhood malignancies in Nordic countries are presented in Figure 1.

## **Surviving childhood cancer**

Advances in research into childhood cancer have led to improvements in diagnostics and more efficient treatment. The result has been a significant increase in survival rates during the last 50 years, from a period when paediatric cancer was seen as an inevitably lethal disease to the present where around 80% of children living in developed countries become long-term survivors (defined as having finished treatment and being disease-free for at least 5 years) (Cantrell & Conte, 2009; Gustafsson, et al., 2007; Steliarova-Foucher, et al., 2004). The greatest improvements in survival rates took place between the 1970s and 1980s, most prominently among children with ALL and Non-Hodgkin's Lymphoma (NHL) (Gustafsson, et al., 2007).

The survival rates for various childhood cancers in the Nordic countries over the past 55 years (1950-2005) are presented in Figure 2.

### **Late effects**

In industrialized countries, survivors of childhood cancer are a rapidly growing group. Knowledge about late effects after treatment in childhood has also increased in recent years. Despite improvements in survival rates, there are potentially negative consequences of having been treated for cancer in childhood in terms of survivors' physical, psychological, and social wellbeing (Dieluweit, et al., 2010; Hovén, 2010; Lof, Winiarski, Giesecke, Ljungman, & Forinder, 2009; Lund, Schmiegelow, Rechnitzer, & Johansen, 2011; Schmidt, et al., 2010; Seitz, et al., 2010).

Studies have shown that survivors have a persistently high risk for a secondary cancer throughout the remainder of their lives. Findings in a study by Olsen et al. showed that patients treated with intensive, multiple-agent chemotherapy (1975-2005) had a relatively high incidence of age-specific secondary cancer (Olsen, et al., 2009).

This thesis involves two national sites treating childhood malignancies where the number of children annually diagnosed with cancer differs greatly. In Sweden, with a population of ~9.4 million, approximately 300 children are diagnosed each year (Gustafsson, et al., 2007). The annual influx rate at the Icelandic site is 12-14 children from a population of ~320 thousand (NOPHO-Annual-Report, 2010).

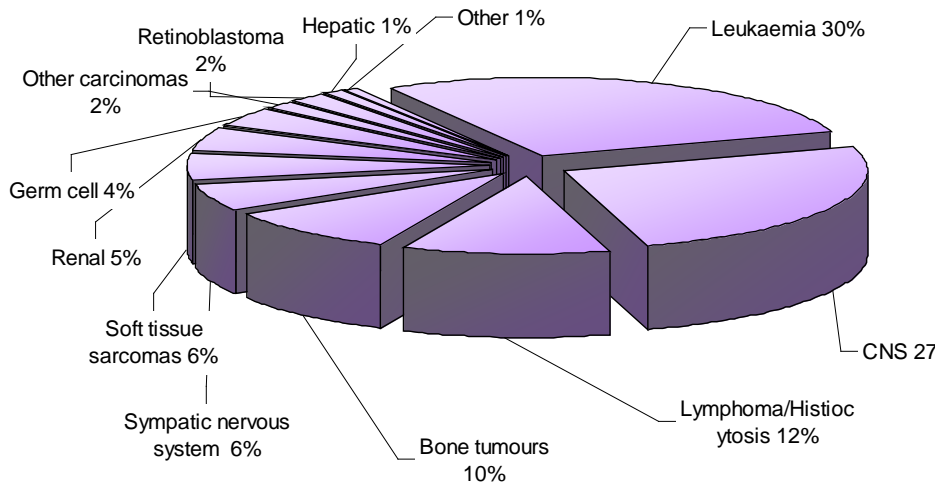


Figure 1 Distribution of different childhood malignancies in the Nordic countries.

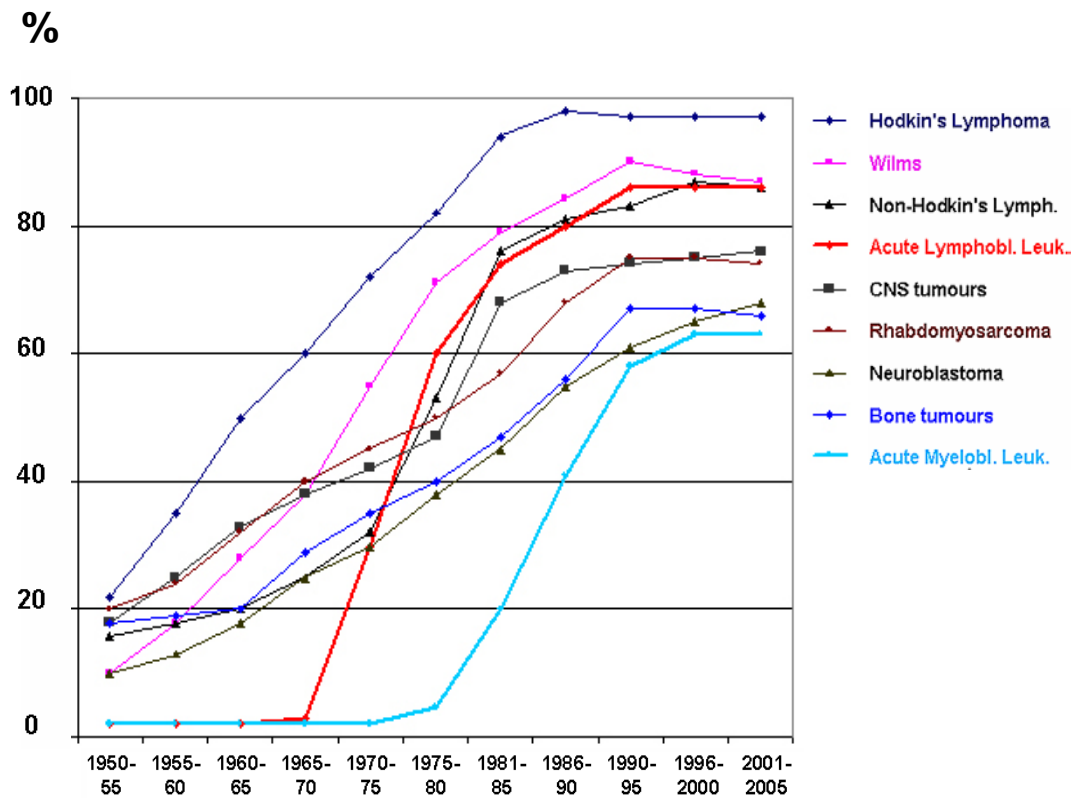


Figure 2 Survival rates for various childhood malignancies in the Nordic countries over the past 55 years (1950-2005). Source: Göran Gustafsson, Swedish Childhood Cancer Register, 2010.

# **DEFINITION OF CORE CONCEPTS**

## **SENSE OF COHERENCE**

Our focus on strengths is based on a theoretical construct named the salutogenic model, originally formulated by Aaron Antonovsky following his observations of holocaust survivors (Antonovsky, 1987). Antonovsky and his colleagues defined sense of coherence (SOC) as representing salutogenic (origin of health) resources that enable individuals to treat life experiences as comprehensible, manageable and meaningful (Antonovsky & Sagy, 1986). Thus, the salutogenic approach defines strong SOC as constituting a flexible, adaptive disposition, allowing individuals to cope successfully with adverse experiences (Surtees, Wainwright, & Khaw, 2006).

This approach, therefore, deals with the following question: What it is that enables some people but not others, to adapt to stress and manage crisis without being harmed in the process - or even, to be strengthened? (Antonovsky, 1987).

## **RESILIENCE**

The disposition of some individuals to cope better with adverse psychological reactions despite experiencing risk is referred to as resilience. The concept thus implies the relative resistance to risk, or the overcoming of stress or adversity. In viewing resilience as individual, the focus is on personal differences rather than on seeing it as a general attribute (Gudmundsdottir, Schirren, & Boman, 2011; Hoge, Austin, & Pollack, 2007; Rutter, 2006).

As such, resilience differs from both social competence and traditional concepts of risk and self-protection in the sense that it focuses on individual variations in response to comparable experiences (Hart, Wilson, & Hittner, 2006). Resilience includes both psychological and biological characteristics intrinsic to the individual; characteristics that might be modifiable and that confer protection against the development of psychopathology in the face of stress (Hoge, et al., 2007).

## **CARE AND ORGANISATION**

The aim of Study II was to improve the understanding of possible determinants influencing parental psychological reactions to childhood cancer. It therefore focused not only on the occurrence of distress, or the influence of demographical or cancer-related factors, but particularly on differences in healthcare services, such as size and type of medical centre, and routines for psychosocial follow up for families of childhood cancer patients.

## PARENTAL PSYCHOSOCIAL CONSEQUENCES

Cancer diagnosis and treatment in children has been shown to cause high levels of stress in parents. For some, the strain can become so overwhelming that it may threaten their ability to function as parents, both for the sick child and for other family members, resulting in difficulty sustaining the family through the illness. Studies have shown that the psychosocial situation of parents is strongly affected, both during treatment and follow-up, and even decades after the treatment is completed (Barakat, et al., 1997; Boman, Lindahl, & Björk, 2003; Boman, et al., 2004; Dixon-Woods, et al., 2001; Giammona & Malek, 2002; Kazak, 1998; Lindahl Norberg & Boman, 2008; Mu, Ma, Hwang, & Chao, 2002; Santacroce, 2002; Van Dongen Melman, et al., 1995); K Boman, et al., 2003; Van Dongen-Melman, 1995).

High levels of distress are often caused by uncertainty about final treatment success, adding to the initial traumatic experience of being told that a child in the family suffers from cancer. This strained life situation, characterized by on-going disease and treatment-related distress, makes parents vulnerable to acute and lasting psychological adverse consequences (Barakat, et al., 1997; Boman, et al., 2004; Dixon-Woods, et al., 2001; Giammona & Malek, 2002; Kazak, 1998; Mu, et al., 2002; Santacroce, 2002; Van Dongen Melman, et al., 1995).

Earlier studies have been inconsistent in their findings when it comes to parental distress associated with a child's cancer. This may be due to different factors, study designs suffering from shortcomings. Study groups may have been small, making it difficult to draw definitive conclusions on the basis of the trends observed (Best, Streisand, Catania, & Kazak, 2001; Kazak & Meadows, 1989; Lahteenmaki, Salmi, & Salo, 1996). Several studies have restricted their focus to only one or two dimensions of psychological reactions, and assessment instruments have often failed to cover illness-specific reactions associated with the unique problems, relevant for the population. Additionally, many studies have addressed mothers only (Greenberg, Kazak, & Meadows, 1989; Steele, et al., 2004), although mothers and fathers may have different experiences (Gudmundsdottir, et al., 2011; Yeh, 2002), which is important to acknowledge when aiming at a complete evaluation of parental reactions to a child's illness.

In recent years, there has been a growing understanding regarding the necessity of dealing with the psychosocial needs, not only those of the sick child, but of the entire family. This realization has resulted in the knowledge that psychological services are a critical component of complete cancer treatment (Pai, Drotar, Zebracki, Moore, & Youngstrom, 2006).

Mapping and analysing previously unexplored determinants of psychological adverse reactions in parents of children with cancer is of great help in designing and developing functional and tailored follow-up procedures.

## PREVIOUS RESEARCH

The way parents respond psychologically to their child's cancer has been investigated in a growing number of studies world-wide, beginning about 3 decades ago and expanding substantially thereafter (Best, et al., 2001; Boman, et al., 2003; Boman, et al., 2004; Hovén, Anclair, Samuelsson, Kogner, & Boman, 2008; Kazak & Meadows, 1989; Koocher, 1986; Peck, 1979; Vrijmoet-Wiersma, et al., 2009).

Studies have focused on various distress indicators, such as anxiety (Dahlquist, et al., 1993; Grootenhuis & Last, 1997; Van Dongen Melman, et al., 1995), depression (Grootenhuis & Last, 1997; Van Dongen Melman, et al., 1995), psychosomatic problems (Sloper, 2000), symptoms of stress (Lahteenmaki, et al., 1996), including traumatic and posttraumatic stress (Kazak, et al., 1998; Lindahl Norberg & Boman, 2008; Lindahl Norberg, Lindblad, & Boman, 2005; Manne, Duhamel, & Redd, 2000; Stuber, Christakis, Houskamp, & Kazak, 1996), marital distress (Dahlquist, et al., 1993; Fife, Norton, & Groom, 1987), and quality of life (Goldbeck, 2001).

The existential trauma caused by a child's illness affects the whole family, parents in particular, regardless of the individual's psychological resources. Nevertheless, as there appear to be more and less adaptive ways of coping with this existential threat (Forinder, 2004; Lindahl Norberg, Lindblad, & Boman, 2005), individual strengths and resources may influence the vulnerability for psychological distress (Kazak, et al., 2003). Findings exist from previous studies of patients and families coping with the threat of illness in general (Buysse, et al., 2008), and, in particular, cancer, or cancer treatment (Forinder, 2004; Mullen, Anderson, & Blaese, 1993).

Studies of parental reactions have rarely adopted a resources-oriented perspective, exploring the distress buffering effect of individual resistance factors. The dominant approach has been concerned with weaknesses, or the formation of psychological or psychiatric symptoms within a framework of psychopathology. However, when the focus has been on resources in studying parental reactions to other paediatric illnesses or medical conditions, outcomes have indicated the potential of sense of coherence for boosting coping and resilience against distress. Margalit et al., studying parents of children with learning difficulties, mental retardation and behaviour disorders, found a positive relationship between low SOC and avoidant non-adaptive behaviour (Margalit, Raviv, & Ankonina, 1992). A more recent study of Icelandic and American parents of paediatric asthma patients showed that sense of coherence moderated the effect of family demands on the situational adaptation of parents (Svavarsdottir, Rayens, & McCubbin, 2005). Also, SOC has been identified as a significant resource in reducing stress among mothers of children with hearing impairments (Hintermair, 2004), Down's syndrome (Hedov, Anneren, & Wikblad, 2002), and developmental disabilities (Olsson & Hwang, 2002). Nevertheless, prior studies have, as far as we know, not addressed the relationship between sense of coherence and a variety of symptoms of disease-related distress among parents of children with life-threatening cancer.

Previous studies on parental consequences following childhood cancer have typically involved samples from single countries. As far as we know, research on site-specific determinants and differences in organisational care, possibly influencing the outcome of parental stress due to a child's cancer, has not been carried out in a comparative approach including two countries. Collecting data from two Nordic sites makes it possible to study both national and site-specific potential determinants of psychological

parent distress. In order to improve our understanding of the determinants of parental psychological reactions, the second study in this thesis addressed the occurrence of parental distress at two quite different types of sites in two Nordic countries, to investigate whether essential differences related to centre type may predict parental outcomes.

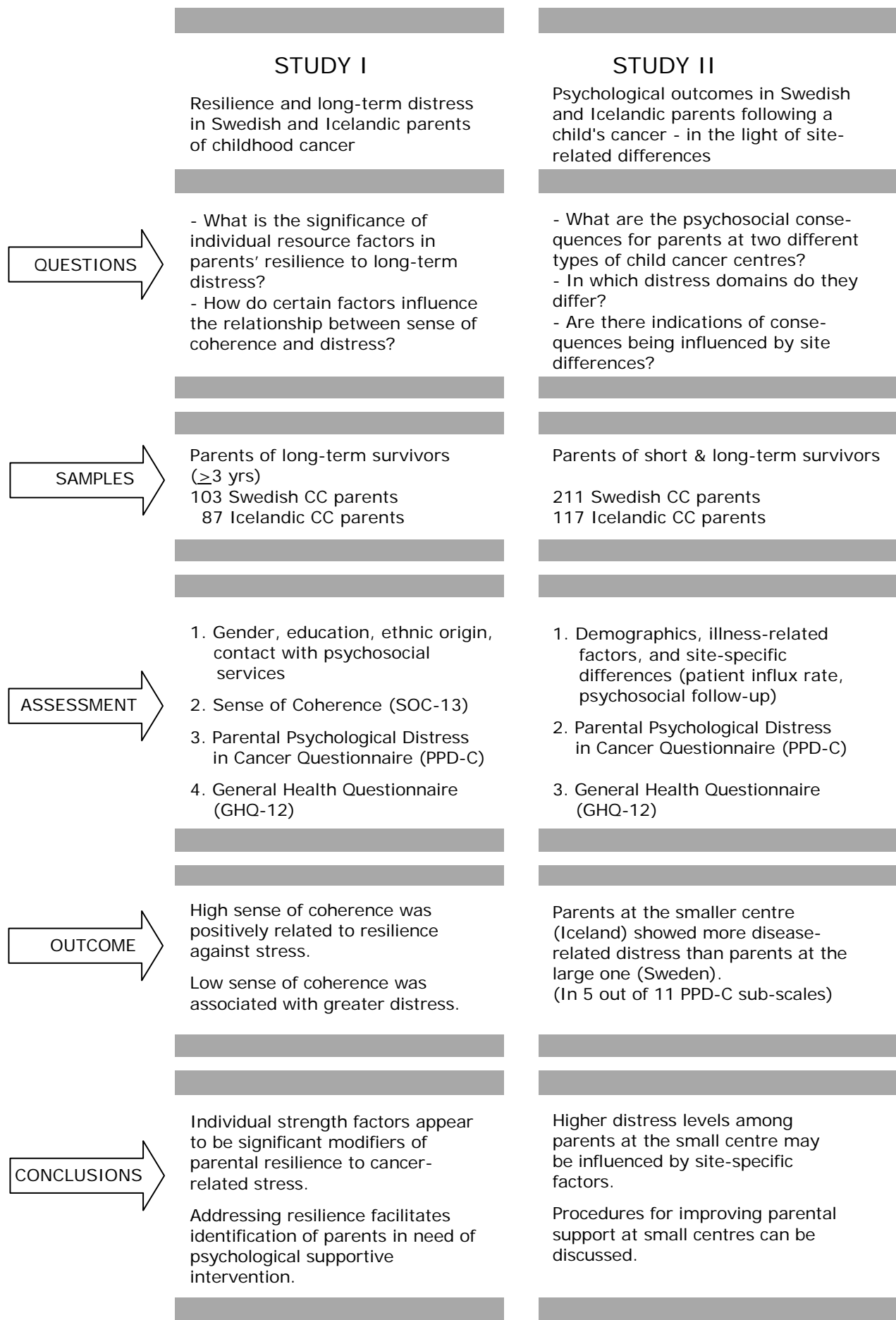


Figure 3 – Overview of thesis studies



# THE EMPIRICAL INVESTIGATIONS

## GENERAL AIM

The main objective of the present thesis was to examine, in a Nordic sample, the psychological reactions of parents of children diagnosed with cancer, and, at the same time, investigate the determinants of parental reactions. The two studies assessed short and long-term effects, and risk and resilience factors influencing parental reactions to having a child with cancer, and discussed the implications for possible psychosocial interventions and family care in the light of differences between treatment centres. The studies addressed parental distress resilience in general, and how differences related to type of treatment centre and organisation of care may predict parental distress outcomes – novel areas of study in this population. An ultimate goal is, by increasing understanding of the determinants of parental reactions and needs, to facilitate the development of individualized care and clinical follow-up for families of children with cancer. Investigations also studied the extent to which demographic factors (parental gender, ethnicity, number of children, and parental education), or aspects related to illness-treatment (age of child at diagnosis, time passed since diagnosis, type of cancer, utilisation of professional psychological support services), might possibly influence parental distress.

An overview of study aims, samples, methods, and main findings by study is presented in Figure 3.

## SPECIFIC AIMS

### *Study I*

The aim of the first study was to examine the resilience-boosting significance of individual strength factors (as defined by sense of coherence) on parental distress related to childhood cancer. The specific focus was on the relationships between parental sense of coherence (SOC) and illness-specific and generic distress symptoms. We also wanted to study whether parental (a) *gender*, (b) *level of education*, and (c) *use of professional psychological support* influenced the relationship between sense of coherence and distress.

### *Study II*

The aim of the second study was to determine the incidence of disease-related distress symptoms in parents treated at two quite different types of sites, and from two Nordic countries. We wanted to compare the two groups regarding distress outcomes to see whether findings could be understood in the light of national, site-specific, and/or organisational determinants; i.e., whether parental reactions in parenting a child diagnosed with cancer might be influenced by such determinants. The study thus addressed the question of how differences in type and size of center, and/or national and local solutions regarding care may predict parents' distress outcomes.

# METHODS

## PARTICIPANTS AND PROCEDURES

### *Study I*

Data were collected from 398 parents of children and adolescents, of whom 190 had children that had been diagnosed with cancer (clinical group), while 208 were reference parents in the community (non-clinical group). The clinical group comprised parents of 126 children who were either in treatment or had finished treatment at one of the two participating sites.

Children of parents in the clinical group had been treated at Astrid Lindgren Children's Hospital in Stockholm, Sweden (ALCH, 103 parents of 64 children), or at the Children's Hospital at Landspítali-University Hospital in Reykjavik Iceland (CH-LUHI, 87 parents of 62 children), who were diagnosed with any kind of malignancy or Langerhan's cell histiocytosis.

Only parents whose child had survived their illness were approached for this study. A known fatal diagnosis, palliative treatment phase, or insufficient knowledge of Swedish or Icelandic to complete questionnaires, were criteria for exclusion. A criterion for inclusion was that a minimum of 36 months had passed from the child's diagnosis in order to comply with the aim of investigating long-term distress and resilience.

Of the 347 eligible parents in the clinical group receiving invitations, 197 returned the questionnaires, an overall response rate of 57%. Seven respondents were excluded prior to analyses due to inadequate data and/or internal data decline. Data analyses were thus based on the remaining 190 parents.

Data regarding sense of coherence (SOC) were collected from a non-clinical reference group of parents, to evaluate the feasibility of the SOC scale being used for the intended purpose; we wanted to examine the degree to which the SOC scale might reflect situational distress in this population to ensure reliable interpretation of the scores (Svavarsdottir, et al., 2005; Wolff & Ratner, 1999). The non-clinical reference parents were selected from families who a) lived in the catchment area of ALCH, and b) had at least one child corresponding in age to that of the children in the clinical group. In a letter of invitation, both parents were asked to participate by completing individual questionnaires. The response rate in the non-clinical group was 56%.

Parents in the clinical and non-clinical groups received an invitation letter which included information about the project, and informed consent was obtained from all participants prior to inclusion. Mothers and fathers were instructed to complete their own questionnaires independently, without consulting the other parent.

### *Study II*

Data were collected from 328 parents of 211 childhood cancer (CC) patients. Children of parents in the clinical group had been treated at ALCH (217 parents of 130 children), or at the CH-LUHI (118 parents of 81 children) in Reykjavik, Iceland. Parents of children diagnosed with any kind of malignancy or Langerhan's cell histiocytosis, between the years 1986-2007, were eligible for participation.

As in study I, only parents whose child had survived their illness were approached. Other illness-related and language criteria were the same as in study I, that is; parents of children with a known fatal outcome of illness, those in palliative treatment phase, or insufficient knowledge of Swedish or Icelandic to complete questionnaires, were excluded.

Of the 529 eligible parents (Iceland N=232 – Sweden N=297) in the study group who received the questionnaire booklet, 335 (Iceland N=118, ~51% – Sweden N=217, ~73%) returned the questionnaires. Overall response rate of the two Nordic sites were 63%.

Due to a lack of identification data and/or internal data decline, 7 respondents were excluded prior to analyses. Data analyses were thus based on the remaining 328 parents.

Invitation took place while parents attended the in-patient or out-patient unit for treatment or follow-up, or by contacting them by mail in cases where regular follow-up at the hospital was no longer on-going. Time elapsed from time of diagnosis to study assessments ranged from 1 week to 20 years, mean elapsed time being 5.0 years. The national groups were similar regarding the range of time from diagnosis to follow-up. Data collected from a non-clinical population of comparison parents (same non-clinical sample as in study I) were used as a reference when evaluating clinical parents' distress in relation to norm data.

## **ASSESSMENT**

### *Study I*

For assessing comprehensibility, manageability and meaningfulness in the clinical study group, Swedish and Icelandic versions of the 13 item Sense of Coherence-Scale (SOC-13 scale) (Antonovsky, 1993) were used. SOC has been recognised as a health promoting resource, which strengthens resilience and advances a positive subjective state of health (Eriksson & Lindstrom, 2006; Hart, et al., 2006; Surtees, et al., 2006). The SOC-13 is a standardised scale that has been found to be cross-culturally applicable, and versions have been developed for use in both Sweden and Iceland (Eriksson & Lindstrom, 2005). Summary scores were calculated for parents individually. In prior studies using SOC-13, internal reliability has ranged from 0.74 to 0.91, and the instrument has demonstrated high content, face and constructs validity, as well as temporal stability (Antonovsky, 1993; Feldt, et al., 2007).

The Parental Psychosocial Distress in Cancer (PPD-C) is a standardized self-report distress measure originally developed by van Dongen-Melman et al., (Van Dongen Melman, et al., 1995), primarily to study illness-specific distress symptoms characteristic for parents of children who survived cancer (Van Dongen Melman, et al., 1995). The conceptual framework for the assessment model is based on theory, literature, and in-depth interviews with parents of childhood cancer patients.

The PPD-C consists of 11 subscales: *uncertainty*, *loss of control* (regarding personal functioning, parenting the patient, the sibling(s)), *self-esteem*, *anxiety*, *disease-related fear*, *loneliness*, *sleep disturbances*, *depression*, and *psychological and physical distress*. The response format of the 125 items asks parents to respond according to 2-, 3-, or 4-point Likert scales. The in-depth interviews with parents, which were part of the construction of the original questionnaire, ensure construct validity of assessment (Van Dongen Melman, et al., 1995). In this study, estimation of the reliability of the SOC questionnaires by Cronbach's alpha resulted in 0.88 for the clinical groups and 0.86 for non-clinical groups, 0.86 for the clinical groups and 0.82 for the non-clinical

groups regarding GHQ, and an alpha ranging from 0.70 to 0.95 for the sub-scales of PPD-C.

The General Health Questionnaire 12-item version (GHQ-12) was used, a reliable and sensitive tool for screening for non-pathological psychiatric symptoms (Van Dongen Melman, et al., 1995), commonly used world-wide. Items relate to the mastering of daily problems, self-esteem, stress, depression and anxiety. Cronbach's alpha in the Study I group was 0.86.

For both Study I and Study II, data regarding illness and treatment-related background information, parents' utilisation of professional psychological support, the family structure, educational level, ethnic background, and home language were also collected.

### *Study II*

Illness-specific distress was assessed using the Swedish and Icelandic versions of the 11-dimensional PPD-C, described above under Study I. Cronbach's alpha ranged in Study II from 0.68 to 0.95.

General, non-pathological psychiatric distress symptoms were assessed using the 12-item General Health Questionnaire (GHQ-12). Cronbach's alpha for GHQ in the clinical group was 0.86.

Descriptive, comparative and inferential analyses were completed by comparing outcomes with non-clinical norm data, whenever possible, and findings were discussed in the light of treatment centre characteristics.

## **STATISTICAL ANALYSES**

### *Study I*

Parents in the clinical and non-clinical groups were compared regarding background variables using non-parametric statistical procedures for categorical and ordinal variables, and parametric procedures for continuous variables.

Analysis related to the primary focus on the relationship between SOC and distress was carried out by conducting linear regression analyses, adjusted for potential confounders. In these analyses, distress symptom outcomes were inserted as the dependent variable in separate analyses, with the individual SOC-sum score inserted as the predictor.

With two-way ANOVA in a combined main and interaction effects model, we also examined whether the relationship between parental SOC and distress was influenced by potentially modifying factors.

In case a significant interaction was found in analyses involving parent gender, these analyses were re-conducted adjusted for the possible dependency between respondent pairs caused by, in some cases, both parents of the same child providing distress data.

### *Study II*

In this study, main outcomes were compared between parents at the Swedish and Icelandic study sites, and descriptive statistics for groups were presented regarding different background variables (e.g., education, utilized professional psychological support, ethnicity, and number of children in the family) and illness-related factors (diagnosis, child's age at diagnosis, time since diagnosis). Comparisons between the site groups regarding these variables were carried out using Mann Whitney U-Test.

Study sites were compared concerning distress outcomes of the PPD-C and GHQ-12, using t-test for independent groups. Distress outcomes were compared with normative data using independent t-test.

### **ADJUSTED ANALYSES**

The two clinical study groups were compared regarding distress outcomes. In order to control these analyses for the effect of possible confounders, multi-factorial ANOVAs were carried out with inserting confounders as covariates.

In this multi-factorial ANOVA, only factors that differed significantly between the two sites were used as covariates in the regression. Nationality (study centre) was inserted as a fixed factor, and potential confounders were inserted together as covariates.

# SUMMARY OF RESULTS

## STUDY I

### Basic outcomes

Comparison of background factors in the clinical study group showed that parents' educational levels were similar. On the other hand, the proportion of parents with an immigrant background was lower in the Icelandic group, and the mean number of children per family was higher. The utilisation of professional psychological support when confronted with the child's cancer differed between mothers and fathers in both groups, with mothers using such support more frequently.

### Sense of coherence and distress

Outcomes indicated that SOC had a modifying effect on generic distress symptoms, physical and psychological stress symptoms, anxiety and depression. The relationship between SOC and distress was inverse, with low SOC scores being associated with more severe distress in all-dimensions of the PPD-C and GHQ.

The influence of SOC varied with parents' gender, showing a stronger modifying influence among mothers.

Associations were found for all the dependent distress variables, showing a negative correlation coefficient (Pearson's  $r$ ) from moderate ( $r$  -0.17) for the subscale (*uncertainty* - PPD-C) to strong ( $r$  -0.79) for GHQ.

Outcomes showed a significant interaction effect between gender (independent background variable), SOC, and distress, with regard to *self-esteem* ( $p < 0.05$ ) and *disease-related fear* ( $p < 0.01$ ).

Subsequent analyses of the interaction effect of SOC and gender, adjusted for a potential dependency between parent couples, demonstrated that the effect was reduced to statistically non-significant for self-esteem ( $p = 0.077$ ) but strengthened for disease-related fear ( $p = 0.005$ ). Results indicated that the negative relationship between SOC and the distress variables were stronger for mothers than fathers. This tendency, although less strong, for this interaction effect between gender and SOC was seen regarding other distress variables as well.

Concerning the interaction effect between other background variables, SOC, and distress, a significant effect of education and SOC ( $p < 0.05$ ) on the *psychological and physiological distress* variable (subscale of PPD-C) was apparent. Parents with higher education showed more distress compared to parents with lower levels of education.

Comparison of SOC outcomes between the clinical and non-clinical groups demonstrated a non-significant tendency for higher SOC in parents belonging to the non-clinical group ( $t = -1.817$ ,  $p = 0.07$ ).

# RESULTS

## STUDY II

### Basic outcomes

Comparisons of various background factors between the Icelandic and Swedish sites showed that educational status was similar in both groups, as well as parental gender. On the other hand, the mean number of children was higher among Icelandic parents ( $t=-4.35$ ,  $p<0.001$ ) and the proportion of parents with immigrant background was lower at the Icelandic site (Mann Whitney,  $p<0.001$ ).

The two sites differed in utilising professional psychological support: 54.1% of Icelandic parents having used some kind of such support, compared to 34.8% of Swedish parents ( $t= -2.50$ ,  $p<0.05$ ).

Regarding illness-related factors, there were significant differences between the national sites concerning the child's age when diagnosed ( $t= -3.00$ ,  $p<0.005$ ), and time from diagnosis to assessment ( $t= -6.90$ ,  $p<0.001$ ).

### Main distress outcomes

Comparing the two national sites, there were significant differences concerning illness-specific distress, Icelandic parents (at the smaller site) scoring higher. The majority of the subscales where the Icelandic study group scored higher concerned disease-related symptoms.

In the entire clinical group of Swedish and Icelandic parents, distress among parents generally exceeded the level of our reference group data collected from the non-clinical group.

Results from t-tests regarding all studied dimensions of distress (PPD-C) showed that Icelandic parents scored significantly higher on 5 of the 11 subscales (*uncertainty* ( $p<0.001$ ); *loss of control regarding parenting the sick child* ( $p<0.05$ ); *disease-related fear* ( $p<0.001$ ); *sleep disturbances* ( $p<0.005$ ); and K: *psychological and physical distress* ( $p<0.005$ )).

For the remaining 6 subscales, the outcomes were similar when comparing the two sites, showing no significant difference between the clinical study groups.

Outcomes of the GHQ-12 concerning non-pathological psychiatric symptoms were similar for the two national groups.

### ADJUSTED ANALYSES

The two clinical study groups were compared regarding distress outcomes using t-test for independent groups. In order to control for possible confounders, multi-factorial ANOVAs with confounder factors inserted as covariates were carried out in those cases where confounder factors differed significantly between the two national sites. In these multi-factorial ANOVAs, nationality was inserted as a fixed factor and potential confounders were inserted together as covariates.

The results showed that in all adjusted analyses, the differences between national sites were still significant for all concerned distress sub-outcomes; in fact, the differences

became even stronger: *information-related uncertainty* related to unmet informational needs  $p<0.0001$ ; *loss of control /patient*  $p<0.0001$ ; *disease-related fear*  $p<0.001$  ; *sleep-disturbances*  $p<0.0001$ ; *psychological & physiological distress*  $p<0.0001$ .

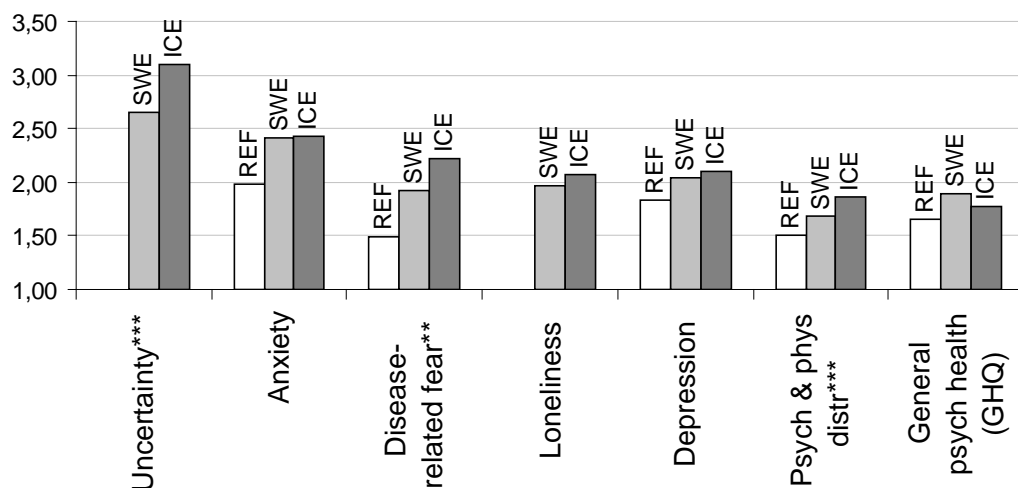


Figure 4. Group-wise outcomes for 4-point distress symptom scales (scale range=1-4). REF=reference parents, non-clinical; SWE & ICE= Swedish and Icelandic childhood cancer parents, respectively. \*\*\*= $p<0.0001$ , \*\*= $p<0.001$ , \*= $p<0.01$  in Sweden - Iceland clinical groups comparisons. Note 1: Uncertainty and Loneliness scales are illness-specific, and not applicable for the non-clinical reference group.

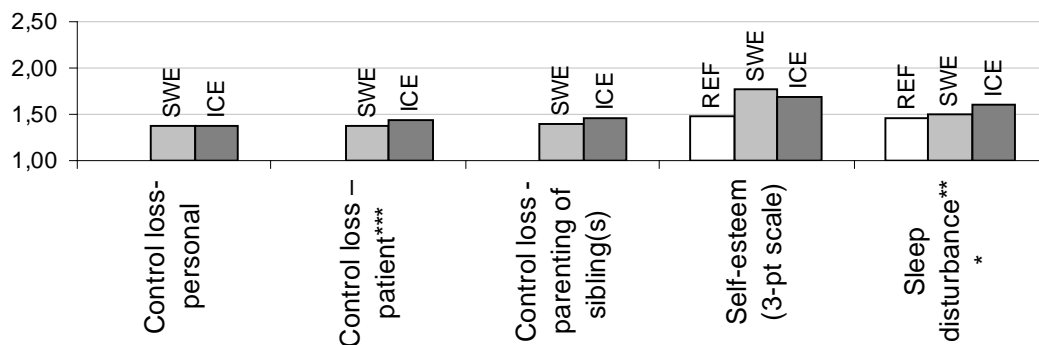


Figure 5. Group-wise outcomes for 3 & 2-point distress symptom scales. REF=reference parents, non-clinical; SWE & ICE= Swedish and Icelandic childhood cancer parents, respectively. \*\*\*= $p<0.0001$ , \*\*= $p<0.001$ , \*= $p<0.01$  in Sweden - Iceland clinical groups comparisons. Note 1: Control loss sub-scales are illness-specific, and not applicable for the non-clinical reference group.



## GENERAL DISCUSSION

The general aim of the studies in this thesis was to increase knowledge concerning short and long-term consequences of having a child with cancer, and possible factors associated with those consequences. This includes the investigation of parental distress in general, as well as specific risks and strength factors, including variations in organisation of care due to treatment centre type.

Novel areas of focus were parental resilience to distress, and a design that enabled a comparison between parents from two different types of treatment centre, investigating whether variations in parental distress might be understandable in the light of factors related to centre characteristics. The aim was to increase our understanding of the determinants of parental reactions and needs. This facilitates the development of the care and follow-up routines for families, paying attention to both individual risk and resilience factors, and to ways in which limitations related to treatment centre and organisational characteristics could be compensated.

A majority of earlier studies of parental reactions to childhood cancer have typically focused on the incidence and severity of distress, e.g., psychological and psychiatric reactive symptoms among parents of children with cancer. Findings indicate that these parents experience extraordinary strain which, in turn, can increase their vulnerability for developing various serious psychological symptoms (e.g., depression, anxiety, sleep-disturbances, and symptoms of post-traumatic stress) (Boman, et al., 2004; Kazak, et al., 2004; Lindahl Norberg & Boman, 2008).

As earlier studies have shown (Boman, et al., 2004; Hovén, et al., 2008; Van Dongen Melman, et al., 1995), and the outcomes of study II appear to confirm, the negative psychological symptoms have been found to persist years after diagnosis and successful treatment.

### STUDY I

As far as is known, few studies have investigated the effect of resilience-related individual characteristics among parents of children with cancer, although studies concerning children suffering from other medical conditions (Hedov, et al., 2002; Olsson & Hwang, 2002; Svavarsdottir & Rayens, 2005) have shown that SOC is a positive and helpful factor for parents in these stress-related circumstances.

Following Antonovsky's theory on sense of coherence (Antonovsky, 1987; Antonovsky & Sagy, 1986), and inspired by earlier studies investigating the effect of sense of coherence (SOC) on parents experiencing long-term distress due to illness-related situations (Hedov, et al., 2002; McCubbin, Balling, Possin, Friedrich, & Bryne, 2002; Olsson & Hwang, 2002; Sivberg, 2002; Svavarsdottir, McCubbin, & Kane, 2000), our hypothesis was that high SOC would strengthen parents' resilience against distress when faced with their child's cancer.

Overall, the outcomes of Study I showed that levels of sense of coherence were associated with higher resilience, here operationalised as lower levels of reported distress symptoms. Sense of coherence played a protective role in relation to both generic and illness-specific distress. On the other hand, lower levels of SOC were significantly associated with parents' showing more severe distress in all dimensions of the Parental Psychosocial Distress in Cancer (PPD-C) questionnaire and the General Health Questionnaire (GHQ).

These results supported the primary study question whether individual resistance factors, operationalised as sense of coherence, had any significance for parents' experienced levels of distress. This indicated that SOC can act as a protective psychological resource strengthening resilience among parents of children diagnosed with cancer.

The resilience-strengthening impact of SOC varied with parental gender, where levels of SOC had a stronger modifying effect among mothers than fathers. Also, regarding parents' level of education, the protective effect of SOC had a stronger influence on distress in parents with a lower level of education compared to those with a higher level.

Findings support the feasibility of the SOC-13 scale as a measure of individual strengths facilitating coping with the illness situation, as indicated by the fact that higher sense of coherence seems to be associated with protection against long-term distress in parents of childhood cancer patients.

Parents with a higher sense of coherence appear better prepared to manage the extraordinarily stressful conditions associated with the child's illness. Those showing stronger SOC-associated resilience seemed less vulnerable to cancer-related stress. This was seen regarding both general psychiatric symptoms as assessed by the GHQ, as well as illness-specific parental distress and fear.

These outcomes indicate that identifiable strength factors in individuals do play an important role concerning parents of children with cancer, in the same manner as has been shown in earlier studies on parents of children with other illnesses (Hedov, et al., 2002; McCubbin, et al., 2002; Olsson & Hwang, 2002; Sivberg, 2002; Svavarsdottir, et al., 2000).

Although our findings indicated support for the hypothesis concerning the resilience-strengthening effect of SOC, it is important to point out that being involved in long-lasting extensively stressful situations might moderate the resilience-boosting effect of SOC. When designing the study we were aware that studying parents shortly after the child's cancer diagnosis might influence the levels of parental SOC. Antonovsky, the founder of the theory of sense of coherence, points out that situations involving intense stress may influence parental coping and be associated with a decrease in SOC (Antonovsky, 1987). To reduce the possible bias of such a potential decrease of SOC due to intense stress, in Study I we stipulated 36 months as the minimum time from child's diagnosis to assessment for parental inclusion. The period directly after diagnosis and the initial treatment period were not covered, since this is a period known to be characterised by the most intense stress (Boman, et al., 2003; Levi, Marsick, Drotar, & Kodish, 2000; Poder, et al., 2007; Vrijmoet-Wiersma, et al., 2009).

The focus on medium to long-term parental distress instead of acute stress following diagnosis meant that the study group was composed of parents whose children had already completed 2–2.5 years of cancer treatment. Among these parents, SOC was at a level similar to those in the non-clinical reference-group, indicating relative stability of SOC across situational circumstances, at least when these are not characterised by acute and intense stress. As the findings indicate, distress levels in the post-treatment phase are less apt to affect parental "base-level" sense of coherence.

The clinical implications of the outcomes are that using the SOC scale, in order to counteract psychological vulnerability, screening for strengths can facilitate the detection of parents at risk, predicting the severity of stressful reactions, which in turn

can improve the identification of parents in need of intensified professional psychological support and psychosocial follow-up, when facing childhood cancer. The integration of a resources perspective would promote parents' resilience with regard to their child's illness.

## STUDY II

Earlier studies addressing parental reactions following childhood cancer have rarely, in a comparative approach, included samples from more than single nation samples. In our study we collected data from two national sites, one in Sweden and one in Iceland, quite different in size and frame of organisation. This enabled us to compare and study parental distress outcomes, and to investigate both national and site-specific potential determinants of parental psychological distress accompanying childhood cancer.

Regarding a focal question of the study, systematic differences in psychosocial outcomes were found between the parents at the two studied sites, with Icelandic parents presenting stronger disease-related distress symptoms than Swedish parents. This finding was to some extent unexpected, especially since a longer period of time had passed since diagnosis for the parents at the Icelandic site - although the two samples were within the same *range* of elapsed time from diagnosis to follow-up. These results were noteworthy, since earlier studies have usually shown disease-related distress to generally decrease with time. In contrast to our findings, other studies have sometimes indicated that parents of children with cancer report levels of distress symptoms similar to parents of healthy children (Jurbergs, Long, Ticona, & Phipps, 2007).

Another unexpected finding was that Icelandic parents had utilised more professional psychological support services than Swedish parents; while, at the same time, showing stronger distress symptoms, not less. Explaining this outcome is difficult; one reason may be that the Icelandic group experienced more disease-related distress, and consequently sought more professional help. Conversely, the outcome could be explained by the fact that the difference in the mean number of contacts is very small from a clinical perspective (Icelandic parents had on average received professional help 2.7 times, and Swedish parents 1.6 times). This may not be of clinical significance when it comes to reducing the severe distress symptoms seen in the childhood cancer parent population.

Another aim was to investigate whether and how parental psychological outcomes might be influenced by certain modifying factors, including parental gender, ethnicity, number of children in the family, and parental education, as well as factors related to the child's illness, such as age at diagnosis, type of cancer, and time passed since diagnosis. In studying two treatment sites that showed considerable differences (e.g., regarding size, patient influx rate, psychosocial services), we also were especially interested in whether differences in distress in study groups might be attributable to differences in type of medical centre, and centre-specific routines for psychosocial services and follow ups.

The significance of differences in parental distress between the two sites remained strong even when adjusted for modifying factors (parental factors, patient factors, education, and utilisation of professional psychological support). Icelandic parents scored significantly higher on 5 of PPD-C's 11 subscales, indicating that there may be site-specific factors influencing distress outcomes.

One of these could be the population difference in the two nations, affecting the influx rate of childhood cancer patients at each site. Sweden has ~9.4 million inhabitants, large enough to allow for a number of specialised childhood cancer centres treating ~300 newly diagnosed patients annually, while Iceland with a population of ~320 thousand has only one small treatment centre (with no specialized ward when it comes to in-patient treatment) responsible for treating 12-14 newly diagnosed paediatric cancer patients annually.

The limited influx of CC patients is likely to result in less peer support for parents and families during the child's treatment and thereafter. The parental fellowship with mutual support and sharing of experiences naturally occurring at a large centre probably functions as a modifier of parental uncertainty, providing a variety of informal illness and treatment-related information. However, the lack of such inter-parental support at a small low-influx centre could be compensated for, by, e.g., establishing regular meetings in an intensified and structured co-operation between the hospital and the local parental organisations. The establishment of volunteer-based groups, including parents with longer time elapsed since diagnosis and treatment could be a part of such routines, implemented in collaboration between the parent organisation(s) at the smaller treatment site as part of a structured follow-up.

## **REPRESENTATIVITY AND GENERALISIBILITY**

The relatively low response rate in the studied clinical groups (57% in Study I and 63% in Study II) must be acknowledged when considering the generalisibility of the findings for the whole population of parents of children with cancer. In particular, the lower response rate at the Icelandic site (Iceland ~51% – Sweden ~73%, Study II) raises the question of whether the study sample is representative of the Icelandic study population, affecting in turn generalisation of the Icelandic outcomes. On the other hand, the Icelandic group represented an entire national cohort, which to some extent, may compensate for the negative effect of the lower participation rate in the Icelandic group.

The threat to generalisibility is related to the possible reasons for attrition, and to the extent to which non-responders may differ from responders. There is a variety of possible explanations for the lower response rate in the Icelandic group. The Icelandic nation is small, with only 320 000 inhabitants. This can result in concerns about anonymity (fear of being identified/recognized) when participating in studies like this where sensitive data is collected about illness and individual psychological reactions. The situation is thus quite different from the one in Sweden, where data collection involved a large city population, the risk of similar identification hardly constituting a problem at all. The relatively small group of all families of children diagnosed with cancer also constitutes the only "national target group" for similar investigations in Iceland, a fact that may result in the group becoming the subject of a variety of similar and simultaneously on-going research studies. If so, being frequently contacted for research study purposes could make parents more reluctant to participate. Although attrition always constitutes an un-wanted weakness complicating the interpretation of results, the two types of hypothetical explanations for the attrition referred to above, although they are likely to be influential, need not seriously threaten the reliability and generalisibility of the findings.

Furthermore, the studies focused on parents of children who had *survived* cancer, with time from diagnoses to assessment ranging from less than one month to 20 years, the mean value being 8.7 years (study I) and 5.0 years (study II) for the Swedish and the

Icelandic groups respectively, and many of the children had completed their treatment at the time of assessment of parents. Therefore, findings naturally do not apply to subgroups such as those with a newly diagnosed child, nor the considerable group of those who lost their child to the illness.

## **ASSESSMENT VALIDITY AND RELIABILITY**

Regarding assessment validity and reliability, there are limitations that need to be recognized. While both the SOC and GHQ (Study I) questionnaires are well-established and frequently used in both Sweden and Iceland, the PPD-C questionnaire (Study I and II) was used here for the first time in an Icelandic context. Due to the lack of prior Icelandic experience from use of the PPD-C, we cannot fully know whether the instrument is equally applicable in both countries. Compared to the situation in Sweden, data is lacking from an Icelandic non-clinical norm sample. This fact limits the applicability of the use of the reference group used here (Swedish parents) in some of the analyses, i.e., those comparing the Icelandic clinical population with the Swedish non-clinical sample. Even though the two Nordic nations can be expected to be culturally similar, the absence of Icelandic non-clinical reference data constitutes a bias. It should be kept in mind, however, that in Study I, comparisons with reference data were not part of the study questions, and in Study II, comparisons between study groups were of primary interest.

Another methodological consideration in study I is that sense of coherence and parental distress were assessed at the same point in time. Even though the main outcomes provide interesting targets for future follow-up studies, we cannot make definite conclusions about suggested causal relationships between main variables in study I, since SOC was not assessed in parents prior to the child's cancer.

## CONCLUSIONS

- Findings indicate that a favourable sense of coherence moderates the experience of distress in parents of children diagnosed with cancer.
- Parents with high sense of coherence appeared more resilient, which makes them better prepared in managing the demanding situation related to their child's cancer.
- High sense of coherence is a strength factor, associated with parents being less negatively affected and displaying fewer symptoms of both general and disease-related distress.
- Screening for individual strengths can be useful in identifying parents at risk for developing high-level cancer-related distress.
- Comparison of parents attending the large childhood cancer centre (Sweden) and parents attending the small low-influx centre (Iceland) show higher levels of mainly disease-related distress in parents from the small site.
- Heightened concerns about survival and late-effects, more prominent in the Icelandic small centre group, indicate a need for information routines adapted to meet parental worries and uncertainties that may be attributable to preconditions of small centres with low patient-influx rate.
- Site characteristics, including influx rate of patients, local routines of care, surveillance and information are viable explanations for differences found between parents attending different types of treatment centres, and constitute targets for forthcoming study.
- A better understanding of organisation- and care-related determinants of parental reactions can facilitate improvement of psychosocial family services, and provide guidance for optimally organising resources to adequately meet parental needs despite size and type of treatment centre.

## **FUTURE PERSPECTIVES**

Forthcoming studies would benefit from investigating additional resilience-related determinants at an early stage. Repeated assessments over time in a longitudinal approach would add to our understanding of how strengths and distress are causally related, and whether parents with identifiable resources recover faster, or more completely. A prospective approach initiated closer in time to the child's diagnosis could address the question of how individual strengths are related to parental reactions to acute, chronic or ongoing stress.

Studies should address in more depth the specific organisation-related determinants that come across in this study as plausible explanations for the differences in distress found between the two studied groups. A more inclusive approach, including the study of other factors related to national, cultural or social factors, and/or differences in e.g., national solutions within the social welfare system, would be an interesting focus for forthcoming studies, to investigate the reasons for the perhaps unexpected differences in parental distress in our two Nordic parent groups.

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