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Influence of childhood cancer on adult life

Quality of life, health status, sexual function and
sense of coherence among long-term survivors

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“The goal of nursing research is to provide the evidence for equitable quality healthcare for all populations”

Afaf I Meleis, Stockholm, June 2010

ABSTRACT

Treatment related health problems are widely acknowledged in the growing population of long-term survivors of childhood cancer. The overall aim was to investigate the influence of childhood cancer on quality of life and health in long-term survivors to increase the understanding for their needs for support in adulthood. The four studies used a cross-sectional design and were based on interview data and self-completed questionnaires. The survivor sample consisted of 246 \geq 18 years old long-term survivors ($>$ 5 years beyond diagnosis), diagnosed at ages 0-18 during the period 1985-1999 in the greater Stockholm area. In Study I, consequences due to the cancer experience were investigated through telephone interviews based on the Swedish version of the Schedule for the Evaluation of Individual Quality of Life-Direct Weighting. In Study II-IV, a comparison group consisting of 296 persons randomly selected from the general population was included. Two standardized instruments were used assessing health status (Short Form Health Survey, SF-36), and coping capacity (Sense of Coherence, SOC), as well as a questionnaire evaluating sexual function and experience.

The survivors described and rated quality of life and health status similar to the comparison group. Nearly one third of the survivors reported no negative impact on life due to the cancer experience, and half of the survivors described a positive view of life and self. The most frequently reported negative consequences include a variety of physical impairments and activity limitations. The survivors differed from the comparison group on one of eight health scales reflecting problems with daily activities owing to physical health. Male survivors reported sexual dysfunctions more often than men in the comparison group did. The sexual dysfunctions were associated with being single and diagnosed with central nervous system tumour. There were differences regarding sexual experience but not regarding sexual function among the women. There was no significant difference in the mean SOC score between the survivors and the comparison group. The 20% of the survivors who reported a current need for support also reported a significantly lower mean SOC score. A low SOC score and having received combined cancer treatment were strong predictors of having a need for support.

The results reveal both positive and negative aspects of the disease and treatment. At the group level, the survivors reported few health problems and overall good quality of life. Sexual difficulties were not frequently reported by the survivors. The results indicate however, that childhood cancer had some impact on self-reported sexual function among men and in sexual experience among women. Overall, the survivors appear to have the resources needed to cope with stressful situations in life to the same degree as people in general. However, a lower SOC was associated with having a need for support. Furthermore, the findings show that the influence of childhood cancer varies between different subgroups of survivors, why it is important to identify problems and needs for each individual. Understanding how long-term survivors of childhood cancer perceive adult life is important to the practice of nursing because it will offer the possibility to provide guidance and support for patients and their families during and after treatment.

Keywords: Childhood cancer, long-term survivors, young adult, quality of life, health status, sexual function, sense of coherence

LIST OF PUBLICATIONS

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

- I. Sundberg, K., Lampic, C., Björk, O., Arvidson, J., Wettergren, L.
Positive and negative consequences of childhood cancer influencing the lives of young adults
European Journal of Oncology Nursing, 2009: 13, 164-170
- II. Sundberg, K., Doukkali, E., Lampic, C., Eriksson, LE., Arvidson, J., Wettergren, L.
Long-term survivors of childhood cancer report quality of life and health status in parity with a comparison group
Journal of Pediatric Blood & Cancer, 2010: 55, 337-343
- III. Sundberg, K., Lampic, C., Arvidson, J., Helström, L., Wettergren, L.
Sexual function and experience among long-term survivors of childhood cancer
European Journal of Cancer, 2010 Oct 27. (Epub ahead of print)
- IV. Sundberg, K., Lampic, C., Arvidson, J., Wettergren, L.
Sense of coherence and need for support among long-term survivors of childhood cancer. (*manuscript*)

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LIST OF ABBREVIATIONS

ALL	Acute Lymphoblastic leukaemia
AML	Acute myeloid leukaemia
CNS	Central nervous system
GRR	General resistance resources
HRQoL	Health related quality of life
KASAM	Känsla av sammanhang
MCS	Mental component summary
PCS	Physical component summary
PNET	Primitive neuroectodermal tumour
QoL	Quality of life
SCCR	Swedish Childhood Cancer Registry
SCT	Stem cell transplantation
SEIQoL-DW	Schedule for the Evaluation of Individual Quality of Life-Direct Weighting
SF-36	Short Form-36 Health Survey
SOC	Sense of coherence
SPAR	Statens person och adress register
SPSS	Statistical Package for Social Science
WHO	World Health Organization

1 BACKGROUND

Due to advances in medicine over the past five decades, childhood cancer survival rates have improved considerably with a continuously growing population of long-term survivors. Describing quality of life among survivors has become a focus in paediatric oncology research. In some regards, the extant literature shows an ambiguous overall picture. There are relatively few studies in which young adults have freely expressed their perceptions of their current lives. Moreover, coping capacity and sexual function are areas that have been investigated to a very limited extent.

Central concepts to the discipline of nursing are the *human being*, *caring/action/relation*, *health* and the *environment*. The focus of caring actions is to support health, prevent illness, alleviate suffering and protect dignity. In nursing and in nursing research it is essential to understand the subjective experience if we are to understand how the individual perceives her/his situation. For example, it is important to not only be aware of the occurrence of different symptoms, but also of how troublesome these symptoms are perceived to be. Exploration and identification of individual obstacles and resources in relation to different aspects in life are essential to maintaining health. Using solely pre-determined questionnaires increases the risk of not capturing aspects considered most relevant by the individual, as does focusing primarily on negative aspects of life due to the childhood cancer experience. Therefore, in the present thesis an individualized measure was also used to assess perception and evaluation of important life issues.

1.1 HEALTH AND QUALITY OF LIFE

There is as yet no consensus about what exactly constitutes quality of life (QoL) as it takes on different meanings to different people (Fayers 2007). Use of the term QoL in multiple disciplines has led to a diversity of definitions but most definitions incorporate ideas of satisfaction/dissatisfaction and happiness/unhappiness (Farquhar 1995). The question of how to gain quality in life so as to achieve "the good life" has been raised throughout modern history. Quality comes from the word *qualitas* meaning "of what kind/sort" and has become an essential feature, a special attribute of life. The term quality of life (QoL) is used in everyday language and in academia, and questions about its definition and methods of measuring it have been lively debated from different perspectives (McKenna and Doward 2004). One view is that QoL can be considered in terms of objective indicators such as socio economic status or housing conditions which are more related to quantity (of life) than quality of life (Meeberg 1993). However, the view that QoL is subjective and multidimensional, representing the perspective of the individual satisfaction with areas such as physical, mental, emotional and social functioning is common (Bowling 2001). In this thesis, the subjective view is based on an individual's reported perception and evaluation of her/his current life.

Many attempts to define QoL are based on the definition of health provided by the World Health Organization “Health is a state of complete physical, mental and social wellbeing and not merely the absence of disease and infirmity”(WHO 1947). WHO’s definition reflects the broad nature of the concept and has been recognized as emphasizing positive qualities of health but it has also received some critique for reflecting an ideal condition (Larson 1999). Although this definition takes a holistic perspective, it suggests that health means as a complete wellbeing and thus proposes a disease-free state or condition which reflects the prevailing biomedical view of health. Within the context of nursing WHO’s health definition has been well used, and has provided a foundation for other theoretical orientations (Simmons 1989). In nursing, health is a basic concept focusing on the individual’s perception of *having health and to be in a state of health* and what this means to the individual rather than the absence of disease and symptoms. This implies that people may be physically ill, but they may also consider themselves healthy if they can adjust and fulfil social roles and tasks. Similarly, people may be healthy in terms of being disease-free, but they may still *feel* ill and not function the way they would like to. Thus, perceptions of health can have an impact on how individuals perceive QoL.

Identifying which factors constitute a good life may reveal an understanding about which areas of life are to be overlooked in order to enhance QoL. The impact of health on QoL represents one area of influence although there are many more influences such as personality (motivation and values), relationships, financial status, environmental support and culture (Doward and McKenna 2004). Rather than being a description of an individual’s health status QoL is a reflection of the way in which individuals perceive and react to their health status (Gill and Feinstein 1994). There is general agreement that QoL can only be described in individual terms, and that the components constituting QoL are personal (Calman 1984; Fayers 2007). One definition that underpins the importance of the individual’s perception of certain aspects of life is the following: “a person’s QoL is what he or she tells himself it is” (Joyce 1988). This also implies that QoL can be determined by both positive and negative aspects.

The term health-related quality of life (HRQoL) is frequently used to distinguish between QoL in its more comprehensive sense and the quality of those aspects of a person’s life that impact directly upon health (Fayers 2007). Although it is not always clear how HRQoL definitions differ from the broader and more general concept of QoL, distinctions have been made. One distinction that seems useful and applicable to the present thesis is that QoL represents the broadest range of human experience whereas HRQoL focuses on those aspects of life that are influenced by symptoms and function (Doward and McKenna 2004).

Calman’s (1984) conceptual definition of QoL originally developed for cancer patients is relevant to all forms of illness and to health, which is why it also seems appropriate in the context of long-term survivors of childhood cancer. He suggested that QoL constitutes the difference or the “gap” between the hopes and expectations of the individual and that individual’s present experience. There are periods of good times and

bad times. The gap therefore defines the individual's QoL: the smaller the gap, the better the QoL. According to Calman, the gap may be narrowed by improving the function or modifying the ambitions and expectations. Good QoL can be said to be present when the hopes of an individual are matched by experience.

Based on this definition Calman (1984) proposes a model in which certain assumptions follow:

- i) QoL can only be estimated and described by the individual and depends on present lifestyle, past experience, hopes for the future, dreams and ambitions.
- ii) QoL must take into account many aspects of life as well as the impact of illness and treatment.
- iii) QoL must be related to individual aims and goals, age and experience.
- iv) Improvement of QoL is related to the ability to identify and achieve these goals.
- v) Action is required to improve QoL, i.e. to narrow the potential gap between hopes and experience, by making expectations more realistic or by encouraging the individual to develop and grow in other ways.
- vi) QoL changes with time, as each goal is achieved new ones are identified.
- vii) The aim is to try to help people reach the goals they have set for themselves which may be done by the individual alone or with the help of others.

Calman (1984) states that QoL changes with time and that it is modified by age and experience. This implies that there are several reasons for assessing QoL in health care such as to monitor the effects of care and treatment, to identify vulnerable periods during treatment and to gain better understanding for decision making in nursing practice.

1.2 MEASUREMENT OF HEALTH AND QUALITY OF LIFE

Given the broad concept, health can be measured in different ways. In a clinical assessment, medical checklists and prompted questionnaires for reporting disease and symptoms and function are common. Assessing health also means measuring of the quality of those aspects of a person's life that impact directly upon his/her health (Fayers 2007)). In the population of long-term survivors of childhood cancer, numerous studies have presented inconsistent findings on self-reported physical, psychological and social functioning as measured by QoL and HRQoL instruments (Apajasalo, Sintonen et al. 1996; Eiser and Eiser 2000; Langeveld, Stam et al. 2002; Zebrack and Chesler 2002; Boman and Bodegard 2004; Stam, Grootenhuis et al. 2006; Alessi, Dama et al. 2007; Blaauwbroek, Stant et al. 2007; McDougall and Tsonis 2009). The interchangeable use of the two concepts health and QoL causes ambiguity in defining and selecting available instruments in research. If a clear definition of the concept that

is to be measured is not provided there will be a lack of construct validity concerning the ability of the instrument to reflect the area of interest. The variety of instruments used for evaluating self-reported health is often referred to as QoL instruments although not claimed for by the authors (Fayers 2007). This also causes ambiguity as low ratings of physical health leads to the implicit assumption that persons with physical limitations cannot have a good QoL. Although commonly described as QoL scales, these instruments are better regarded as measures of health status as they commonly focus on physical and emotional functioning

Health status is a description and/or measurement of the health of an individual or population at a particular point in time in relation to identifiable standards or health indicators (WHO 1984). For example, a person may report having problems walking a long distance, cleaning the house or participating in sports due to health problems. Some instruments that assess health status can be used on a general population whereas others are intended for persons with specific diseases and can vary from one global question to several questions reflecting different aspects of health. In the present thesis, health was measured in terms of self-reported health status using Short Form-36 Health Survey (SF-36). The SF-36 is a commonly used instrument to measure health status and is often referred to as a HRQoL instrument. The advantage of using formal scientifically rigorous HRQoL measures is that they allow us to compare findings across studies. Most QoL and HRQoL instruments are typically standardized questionnaires based on a nomothetic approach which focuses on general laws concerning biology and human behaviour (Mitchell and Bradley 2004). Such instruments contain items that may not be relevant for all individuals whose QoL is being assessed (Hickey, Bury et al. 1996) and they provide little understanding of how the individual define or value the different components of his/her life (Bowling 1995). Moreover, predetermined tools often fail to consider positive elements that may contribute to QoL. Results from a few studies, in which survivors were directly asked to report possible positive aspects following the cancer experience, show that positive psychosocial outcomes in addition to negative consequences are common (Parry and Chesler 2005; Wettergren, Björkholm et al. 2005; Mattsson, Ringner et al. 2007).

Quality of life is difficult to measure due to its multidimensional nature. To properly assess and study QoL, it is essential to obtain data from the individual's own perspective (Fayers 2007). Individual measures not using predetermined variables are derived from an idiographic approach to assessing individual QoL (Mitchell and Bradley 2004). Such an instrument is the Schedule for the Evaluation of Individual Quality of Life (SEIQoL) which was the choice for the present thesis. The SEIQoL was developed on the premise that QoL is individual in nature and that a person judges QoL on the basis of how he/she is doing in a number of salient life areas (O'Boyle 1994). Listing "life areas" covering all aspects of life enables the identification of problems and priorities of particular importance to the individual (Flanagan 1982; Bowling 1995; Hickey, Bury et al. 1996).

1.3 STRESS AND COPING

Young people generally face a multitude of ongoing stressful concerns related to education and future vocation, peer and family relationships, environment and unemployment (Frydenberg 1994). Being a long term survivor of childhood cancer may influence life in many ways and maybe an additional source of stress that can lead to varying degrees of psychological distress (Hobbie, Stuber et al. 2000; Meeske, Ruccione et al. 2001; Recklitis, O'Leary et al. 2003; Sharp, Kinahan et al. 2007). Stress can be viewed according to the nature of the stimulus (Frydenberg 2008). It can be a matter of daily hassles such as quarrelling with a friend, an acute time-limited stress such as parents divorcing, chronic intermittent stress such as completing assignments for school or a chronic stressor such as an ongoing illness. Stress occurs "when there is an imbalance between the demands of the environment and the perceived resources of the individual" (Lazarus and Folkman 1984). Also, recent research has shown that the way life stressors influence mood states in humans is strongly linked to genetic differences in the serotonin neurotransmitter system (Caspi, Hariri et al. 2010). Such findings provide a biological explanation that may contribute to the interindividual variability seen in responses to life stressors.

The way in which stress is handled independently of what has caused the stress is called coping (Lazarus and Folkman 1984). Lazarus (1993) defined coping as the response to the "on-going cognitive and behavioural demands that are taxing or exceeding the resources of the person". Coping represents thoughts, feelings and actions that an individual uses to deal with problematic situations encountered in everyday life and in particular circumstances (Frydenberg 2008). In the stress and coping transactional model by Lazarus and Folkman (1984) it is proposed that coping changes over time and in accordance with the situation in which it occurs. Coping is essentially a dynamic interaction between persons and their environment and present situation. In research and practice, coping has acquired a variety of meanings that are often used interchangeably with concepts such as mastery, defence and adaptation (Frydenberg 2008). According to the transactional model, coping is preceded by appraisal which first determines whether the situation is a challenge or a threat and second establishes whether the individual feels he/she has the resources necessary to cope (Folkman 1997). Appraisal of how controllable a stressor may be is essential for the coping response. Greater control is associated with higher levels of problem-focused coping, such as information seeking and taking direct action to solve a problem. Less control is associated with higher levels of emotion-focused coping, such as escape or cognitive reframing. These responses also called coping styles or strategies are methods of coping that partly reflect personal values, beliefs and goals.

In nursing, attention has increasingly turned to the concept of sense of coherence (SOC) (Nilsson, Axelsson et al. 2001; Wettergren, Björkholm et al. 2004; Langius-Eklöf, Lidman et al. 2009; Barthelsson, Nordstrom et al. 2010). SOC is a theoretical model designed by American sociologist Aron Antonovsky to advance understanding of the relations between stressors, coping and health (Antonovsky 1987). After interviewing persons who had experienced severe hardship and traumatic situations and despite this

remained healthy he postulated that their ability to stay healthy was dependent on the way they viewed their life in general. SOC can be applied to explain an individual's resources for dealing with the stressors confronted in daily life (Antonovsky 1987).

SOC is defined by Antonovsky (1987, p 41) as "A global orientation that expresses the extent to which one has a pervasive, enduring though dynamic feeling of confidence that;

- (1) stimuli deriving from one's internal and external environments in the course of living are structured, predictable and explicable;
- (2) resources are available to meet the demands posed by these stimuli and;
- (3) these demands are challenges, worthy of investment and engagement."

The three components of SOC are called comprehensibility, manageability and meaningfulness. Antonovsky hypothesized that the more prominent these properties are, the higher a person's SOC will be and the more likely he or she will be to cope successfully with life stressor situations, thus leading to better health and QoL. According to Antonovsky, health can be seen as a movement along a continuum on an axis between total ill health (disease) and total health (ease) and an individual's sense of coherence is a determining factor for his/her position on that continuum. Key elements in the concept of SOC are the resources that are available to deal with the demands of everyday life and that provide the energy to combat various stressors. Antonovsky used the term general resistance resources (GRR) to refer to a number of resources found within people that are bound to their person and capacity but also to their immediate and distant environment. The GRR which are shaped by life experiences, are both of a genetic and a psychosocial character such as ego identity (strength), knowledge, intelligence, material (wealth), social support, cultural stability and religion/philosophy. SOC is a construct that is universally meaningful, and is assumed to cut across gender, social class, religion and culture (Antonovsky 1993). While the theory of SOC bears resemblance to other theories on the impact of strain on health e.g Bandura's (1977) concept of "self-efficacy" and Kobasa's (1979) concept of "hardiness", SOC was considered by Antonovsky, to be the broader of these concepts. SOC is more clearly concentrated on factors promoting health rather than factors causing particular diseases. Antonovsky believed that SOC is broader than specific coping mechanisms and that a higher SOC is required for successful coping and should be regarded as the ability to find and utilize resources, even when they are scarce (Antonovsky 1987). Antonovsky operationalized the SOC construct to form a scale on which one could measure the individual's adaption to life. The scale refers to individuals' enduring attitude towards life and the capacity to identify, use, and reuse general resistance resources in stressful situations to maintain and improve their health (Eriksson and Lindström 2006). Antonovsky described SOC as being high and low, but he did not define what could be seen as a normal SOC (Antonovsky 1987).

Antonovsky (1984) considered that SOC was developed during childhood, adolescence and early adulthood and stabilized after the age of 30 years thereafter showing only minor and temporary fluctuations in response to radical changes in life. He assume that

by the end of their third decade, people will have been exposed to a sufficiently long and consistent pattern of experiences that their SOC would have become a stable personality factor - although inevitably and constantly attacked by stressors - throughout life (Antonovsky 1987). However, in Antonovsky's later work, he argued that those with a high SOC are more likely to stay stable whereas those with a modest or weak score are likely to move to lower SOC-levels over time (Antonovsky 1996). More recent research has shown that SOC is not as stable as Antonovsky proposed. For people with an initial high SOC, the score tends to increase with age throughout the whole lifespan (Eriksson and Lindström 2005; Nilsson, Leppert et al. 2009). Moreover, in relation to stressful life events such as severe accident trauma and a loss of social support a decrease in the SOC score has been shown (Wolff and Ratner 1999; Schnyder U 2000; Nilsson, Holmgren et al. 2003; Snekkevik, Anke et al. 2003). SOC is strongly related to self-reported mental health and psychological wellbeing (Eriksson and Lindström 2006; Nilsson, Leppert et al. 2009). The relation between SOC and physical health is much weaker than Antonovsky postulated and more associated with perceived health than with medical outcomes (Eriksson and Lindström 2006). The suggestion that SOC is a resource that enhances aspects of QoL has been pointed out in numerous studies of disease specific groups of patients (Eriksson and Lindstrom 2007).

The relationship between chronic illness in childhood and the development of sense of coherence has only been studied to a limited extent (Moons and Norekval 2006). Theoretically it can be assumed that the experience of cancer in childhood may influence the development of SOC. The cancer experience may have brought with it some positive elements in life that can enhance the development of SOC or the experience may have induced high stressors that have a negative impact on SOC.

1.4 CHILDHOOD CANCER

Approximately 300 children before the age of 18 years are diagnosed annually with a paediatric cancer in Sweden (Gustafsson 2007). The cause of cancer in children and teenagers is largely unknown. Hereditary cancer in childhood is extremely unusual and mostly occurs among children with certain rare syndromes. Childhood cancer is most common in children before 5-6 years of age and among boys (ratio: male/female = 1.18). The distribution of diagnoses before 15 years is 31% for leukaemia, 28% for central nervous system (CNS) tumours and 41% for other solid tumours. The incidence of the different types of cancers among children varies with age. There is an incidence peak in ages 2-4 years for leukaemia, while brain tumours have a more even distribution across all ages. The most common diagnosis during teenage years is bone tumours which are very rare in younger children. Although the risk of relapse of childhood cancer is small it persists and is most common in CNS tumours and acute lymphoblastic leukaemia (ALL) which accounts for a majority of all leukaemia cases.

1.4.1 Treatment

Over the past five decades, there has been a change in treatment philosophy regarding childhood cancer from palliation to cure owing to the intensification and adjustment of

therapies (Gustafsson 2007). The Nordic countries have established a close collaboration and have treatment programmes in common. These programmes have become very complex and specifically designed for each diagnosis and diagnostic subgroups. Surgery is the oldest tool used in childhood cancer. Although it is less frequently used nowadays it remains vital and is often used in combination with chemotherapy and radiation. Surgery is rarely the first option today as it often results in severe body impairments and scars. Instead chemotherapy is generally chosen to shrink the tumour or at best cause it to disappear as well as to see what effect the treatment has had on the tumour which is helpful when estimating a prognosis. The greatly increased use of chemotherapy over the past three decades has enabled a steady reduction in dose and volume of radiotherapy and has been of major importance in improving survival rates. Radiotherapy is avoided as far as possible as it is always associated with a high risk of adverse effects such as impaired growth and cognition. Twenty years ago, cranial radiation was used as prophylaxis in the treatment of ALL. Thereafter a more restricted regime was introduced and only those with a suspected disseminated disease to the central nervous system now undergo cranial radiation. Treatment for tumours of the CNS often includes a combination of surgery, radiotherapy and chemotherapy (Rasco Baggott 2001). Surgery is always the first consideration for CNS tumours and whether the tumour is surgically accessible depends on its location and histology (type and grade). Sometimes treatment with chemotherapy or radiation is performed to shrink the tumour before the operation. Treatment for CNS tumours may damage normal tissue surrounding the tumour with a high risk of unwanted side effects (Anderson, Rennie et al. 2001). Leukaemia is treated with an intense intermittent chemotherapy regime often for two and a half years depending on the type of leukaemia (Rasco Baggott 2001). With the successful development and increased availability of donors, allogeneic bone marrow or peripheral blood stem cell transplantation (SCT) can be offered to children with especially very high risk and recurrent leukaemia disease (Reismuller, Attarbaschi et al. 2009). The group solid tumours include a number of different diagnoses and the treatment can be surgery, radiotherapy, or chemotherapy alone or often in a variety of combinations depending on the diagnosis.

1.4.2 Childhood cancer care

There are six paediatric cancer centres in Sweden treating children and adolescents, aged 0-18 years, who have been diagnosed with cancer (Gustafsson 2007). The treatment is always directed from one of the centres where a majority of the treatment is administered and sometimes part of the treatment can be given at a local hospital. The cancer diagnosis is often devastating and strikes the whole family. A comprehensive view of the child with cancer and the family is fundamental in paediatric oncology. The parents and often the siblings as well as the ill child receive the same information about the different phases of the medical care. The frequency and duration of hospital stays vary with diagnosis and treatment. Many children have their treatment when they are admitted to the ward but it is also common to be treated as an outpatient especially towards the end of the treatment period. A childhood cancer clinic is organized to take care of a large age range of patients from small children to teenagers up to the age of 18 years. There are no restricted visiting hours for the family

so that the children and parents can feel comfortable around the clock. Often the whole family becomes involved and the parents take a great responsibility in the daily food and hygiene routines. On admission children are given the opportunity to attend hospital school during their stay and in between treatments they attend to their regular schools. Psychosocial support is available for the ill child and the whole family but it is not a routine for all patients to see a psychologist or counsellor at least once before the discharge from hospital. Nor are there any routines for talking to the adolescent about psychosexual issues. Cancer treatments are usually lengthy regimens, with numerous side-effects and complications, such as pain from procedures and treatments, nausea, mouth sores, hairloss, change in appetite, fatigue, and susceptibility to infections and anaemia (Ljungman, Gordh et al. 1999; Hedström, Skolin et al. 2004). Taking medications and having to plan everything around hospital visits are aspects of distress reported among adolescents when receiving chemotherapy (Hedström, Skolin et al. 2004; Enskär and von Essen 2007). Other worries include losing friends and not being able to attend school (Enskär and von Essen 2008).

1.4.3 Survival

The survival of children with cancer in the Nordic countries has steadily improved for almost all diagnoses (Gustafsson 2007). The most dramatic improvements in survival occurred during the 1970s and 1980s especially among children with ALL. This has been accomplished through improved treatment programmes and continuous medical development in many fields. Survival rates vary greatly in Europe and in Eastern Europe the survival rates are still quite low whereas the Nordic countries represent a survival gold standard with an overall five-year survival at almost 80% in Sweden. (Gatta, Corazziari et al. 2003). It is well recognized that both type and intensity of treatment and patients' age at treatment are important factors for overall survival (Mertens, Yasui et al. 2001). The survival rate for ALL and Hodgkin's lymphoma at 5 years after diagnosis is at present 89% and the overall survival rate for CNS tumours at 10 years follow-up is 72%. Acute myeloid leukaemia (AML), bone tumours and neuroblastoma are diagnostic groups with the worst prognosis, all with a survival rate between 60-70% five years after diagnosis (Gustafsson 2007). There is no difference in survival between boys and girls.

The population of long-term survivors of childhood cancer is continuously growing and currently in Sweden there is one survivor in every 700 young adults between the ages of 25 and 35 years. What characterizes young adults in Sweden today differs from what characterized them in earlier periods (Trondman 2003). Generally speaking, there is a delay in finishing school and beginning to hold a full-time job, and taking in assuming other responsibilities of adulthood. The phase between childhood and adulthood tends to be more extended than in previous decades. Young teenagers want to be older than their age and often try to behave as though they were, but older teenagers more frequently want to stay young and to post-pone adult life with all its responsibilities. The last phase of becoming an adult is that of

independence and self-supporting, as well as an increased focus on gaining stability and making plans for family life (Trondman 2003).

In the field of cancer there has been a great deal of discussion about use of the term survivorship. “An individual is considered to be a cancer survivor from the time of diagnosis, through the balance of his or her life” (Lewis 2006). Survivorship can also be defined as living five years beyond diagnosis. This 5-year time frame has become an important reference point for survivors as the risk of recurrence is considered to be much lower after that period of time (Leigh 2004). Those individuals living five years or more *beyond* their primary cancer diagnosis have been defined as *long-term* cancer survivors (Mullan 1985) and this is the definition used for the purpose of the present thesis. Survivorship can be seen as an ongoing process for the remainder of the life of the survivor (Prouty, Ward-Smith et al. 2006). Family members, friends and caregivers are also impacted by the survivorship experience (NCI 2006).

Understanding how survivorship is applied in the context of young adult survivors of childhood cancer is important to the practice of nursing because it could offer a possibility to provide guidance and support for survivors and their families. Because survivorship is an ongoing process, consequences such as treatment side effects and the development of second cancers may occur for during the entire future life of the childhood cancer survivor. Providing appropriate follow-up care has been recognised as a major emerging challenge in paediatric oncology (Henderson, Friedman et al. 2010). Ideally, all patients who leave paediatric oncology should have a follow-up care plan that is carried out in adult-oriented pre-organized settings. Internationally, cancer centres differ in how they enable transition from paediatric oncology to adult medicine (Peter, Forke et al. 2009). Childhood cancer survivors seek and receive care from a wide variety of health care professionals including oncologists, paediatric specialists, surgeons, primary care physicians, gynaecologists, nurses, psychologists and social workers (Henderson, Friedman et al. 2010). In Sweden there is at present no nationwide organized follow-up programme for childhood cancer survivors. During the past few years, many of the paediatric centres have become more restrictive in seeing survivors 18 years or older and there is general agreement among clinicians about the urgent need for follow-up clinics. The Swedish national cancer strategy includes a proposal for the coming years to link the paediatric and adult cancer centres and to have organized follow-up clinics within the centres (Socialdepartementet 2009).

1.5 HEALTH AND QUALITY OF LIFE AMONG LONG-TERM SURVIVORS

1.5.1 Clinician reported health outcomes

Not all children with cancer are cured and some of the children who are cured are burdened with serious complications (Gustafsson 2007). These are often chronic medical health conditions that justify ongoing medical interventions and can occur during or soon after treatment (long-term effects) and also many years after successful

completion of treatment (late effects) (Landier and Bhatia 2008; Diller, Chow et al. 2009). Commonly reported health problems include endocrinological disorders, cardiopulmonary disease, neurological adverse outcomes, renal impairment, gastrointestinal dysfunction, musculoskeletal sequelae and subsequent malignancies (Stevens, Mahler et al. 1998; Hudson, Mertens et al. 2003; Robison, Green et al. 2005; Oeffinger, Mertens et al. 2006; Geenen, Cardous-Ubbink et al. 2007; Landier and Bhatia 2008). It has been demonstrated that long-term survivors of childhood cancer carry a high burden of morbidity with 62% reporting at least one chronic health condition and 25% reporting conditions considered to be severe or life-threatening (Oeffinger, Mertens et al. 2006; Geenen, Cardous-Ubbink et al. 2007). Almost half of the long-term survivors will have moderate to extreme limitations in activity and functional impairment (Hudson, Mertens et al. 2003; Ness, Bhatia et al. 2005). A cumulative incidence of health problems has been seen over time and can still increase 25 years after diagnosis (Oeffinger, Mertens et al. 2006), the absolute excess risk of premature death from a second cancer, cardiovascular or pulmonary disease is significantly elevated beyond 30 years after diagnosis (Mertens, Yasui et al. 2001). Long-term survival after diagnosis with a CNS tumour, Hodgkin's disease or bone tumours have been associated with a strong negative impact on health status (Hudson, Mertens et al. 2003; Oeffinger, Mertens et al. 2006; Geenen, Cardous-Ubbink et al. 2007) and limitations in performing physical activities (Ness, Mertens et al. 2005). Survivors who have received more intensive treatment including radiation to the head, chest, abdominal and pelvic areas, have been found to be at greater risk of adversely affected health (Hudson, Mertens et al. 2003; Oeffinger, Mertens et al. 2006; Geenen, Cardous-Ubbink et al. 2007). The risk of a second primary cancer among childhood cancer survivors is mainly due to the radiotherapy and chemotherapy directed to the first cancer in childhood (Olsen, Moller et al. 2009). That risk persists throughout life and the number who fall ill with a second cancer will increase because of the growing number of survivors as well as the increasing average age of the childhood cancer survivor population.

1.5.2 Self-reported Quality of Life

A growing body of literature has described aspects of QoL in terms of self-reported physical, social and psychological outcomes among young adult survivors of childhood cancer. However, the findings have been inconsistent and sometimes contradictory across studies (McDougall and Tsonis 2009). Survivors who report substantial physical late effects demonstrate more negative psychological and psychosocial outcomes (Zebrack and Chesler 2002). Other factors associated with negative QoL outcomes are certain cancer and treatment types, female gender, unmarried status, socioeconomic factors such as lower educational attainment and unemployment (Langeveld, Grootenhuis et al. 2004; Stam, Grootenhuis et al. 2005; Grant, Cranston et al. 2006; Zeltzer, Lu et al. 2008). In addition, those survivors who report doing well overall still express some concerns with respect to aspects of their physical, psychological and social well-being (Zebrack and Chesler 2002).

Research shows that survivors worry about illness recurrence and about the future (Zebrack and Chesler 2002) as well as that they have worries about reproductive capacity and future health problems their children might experience (Langeveld, Stam et al. 2002). Furthermore, negative impact on social activities and interpersonal relationships, marriage rates and parenthood as well as on life goals such as education and work has been reported (Langeveld, Stam et al. 2002; Stam, Grootenhuis et al. 2005; Zeltzer, Lu et al. 2008; Gurney, Krull et al. 2009). Physical reminders of cancer treatment - such as various physical dysfunctions (Ness, Hudson et al. 2009) stretch marks, scars and hair loss - can be sources of embarrassment and contribute to a perception of being 'different' in relation to peers (Pendley, Dahlquist et al. 1997; White 2000). Moreover, diagnosis and treatment prior to and during the onset of puberty may affect the appearance of secondary sexual characteristics, linear growth, fertility and psychosexual development (Armstrong, Chow et al. 2009). Such concerns regarding physical appearance may have negative implications for self-esteem and views on the physical self (Woodgate 2005).

Sexual health - defined by the World Health Organization (WHO) - as a state of physical, emotional, mental and social wellbeing related to sexuality, has been recognized as an integral part of overall health and QoL (WHO 2004). The possible influence of childhood cancer on the sexual health of young adult survivors in terms of infertility and negative reproductive effects has been well documented (Green, Kawashima et al. 2009; Reulen, Zeegers et al. 2009; Green, Kawashima et al. 2010). However, the question of which sexual difficulties may be present has been investigated to a much lesser extent. Only one study has published results on sexual function among young survivors of childhood cancer (Zebrack, Foley et al. 2009). The study results showed significant associations between sexual function and health status and that almost half of the survivors reported at least something of a problem in one or more of the areas measured. Some studies have reported a negative impact on psychosexual development and sexual experience after childhood cancer (Ropponen, Siimes et al. 1992; Puukko, Hirvonen et al. 1997; Stam, Grootenhuis et al. 2005; van Dijk, van Dulmen-den Broeder et al. 2008). Reports have shown that survivors were older than a comparison group when they started to date and first experienced sexual intimacy and sexual intercourse (Stam, Grootenhuis et al. 2005; van Dijk, van Dulmen-den Broeder et al. 2008) and when having an intimate or romantic relationship (Larouche and Chin-Peuckert 2006). While there is no consensus regarding which aspects to assess when studying sexual function, attributes often included are interest, desire, arousal, performance, and overall satisfaction (Arrington, Cofrancesco et al. 2004). Sexual function can be defined as the capacity and wish to perform a sexual activity including perceived sexual pleasure, and this is the definition chosen for this thesis.

Some studies have shown that survivors of childhood cancer are seriously psychologically troubled, exhibiting post traumatic-stress disorder, and post-traumatic stress symptoms above the prevalence rates of the general adult population, (Meeske, Ruccione et al. 2001; Langeveld, Grootenhuis et al. 2004; Bruce 2006; Rourke, Hobbie

et al. 2007). However, others give a more positive overall picture of long-term survivors reporting psychological well-being similar to or even better than that of control groups (Elkin, Phipps et al. 1997; Langeveld, Stam et al. 2002; Pemberger, Jagsch et al. 2005; Grant, Cranston et al. 2006; Stam, Grootenhuis et al. 2006; Zebrack, Mills et al. 2007; Zeltzer, Lu et al. 2008). Interestingly, recent research in adolescents with cancer has shown higher levels of depression six months after diagnosis and lower levels of anxiety and depression than a reference group up to 48 months after diagnosis (Larsson, Mattsson et al. 2010). Nevertheless, survivors of leukaemia, lymphoma, brain tumours, neuroblastoma and bone tumours demonstrate higher psychological distress than their siblings do (Ness, Gurney et al. 2008; Zeltzer, Lu et al. 2008). A particularly vulnerable group has been shown to be survivors of CNS tumours who report more psychological distress, fatigue, cognitive problems and diminished life satisfaction than survivors of any other cancer types (Mulhern, Merchant et al. 2004).

In conclusion, numerous large clinical cohort studies have reported a high burden of physical health problems among long-term survivors of childhood cancer. The overall picture shown by the prevailing research within the field of QoL is not as discouraging as one would expect, however, it is somewhat contradictory. Therefore it is important to continue to investigate areas of QoL that have previously been described and to investigate new areas that have hitherto not been described.

2 AIMS

The overall aim was to investigate the influence of childhood cancer on quality of life and health in long-term survivors to increase the understanding for their needs for support in adulthood.

This was approached with the following specific aims:

- To describe the positive and negative consequences of childhood cancer that young adult long-term survivors consider influences their present life, and to relate these consequences to diagnosis, treatment and gender (Study I).
- To describe quality of life in relation to self-reported health status and socio-demographic characteristics among long-term survivors of childhood cancer as compared to that among a sample from the general population (Study II).
- To compare sexual function, sexual experience and quality of partner relationship by gender in a cohort of long-term survivors of childhood cancer with a sample from the general population. An additional objective was to compare the findings for survivors diagnosed with CNS tumours with the findings for those with other diagnoses (Study III).
- To compare SOC among a group of long-term survivors of childhood cancer with SOC in a group selected from the general population. Further, to explore the need for current support among the survivors and the association between need for support and SOC (Study IV).

3 METHODS

3.1 DESIGN

The four studies presented in this thesis have a cross-sectional design. Study I is descriptive and explorative and include both qualitative and quantitative data. Study II has a quantitative approach including qualitative data, study III & IV have a quantitative approach. Study II, III & IV are descriptive case-control studies.

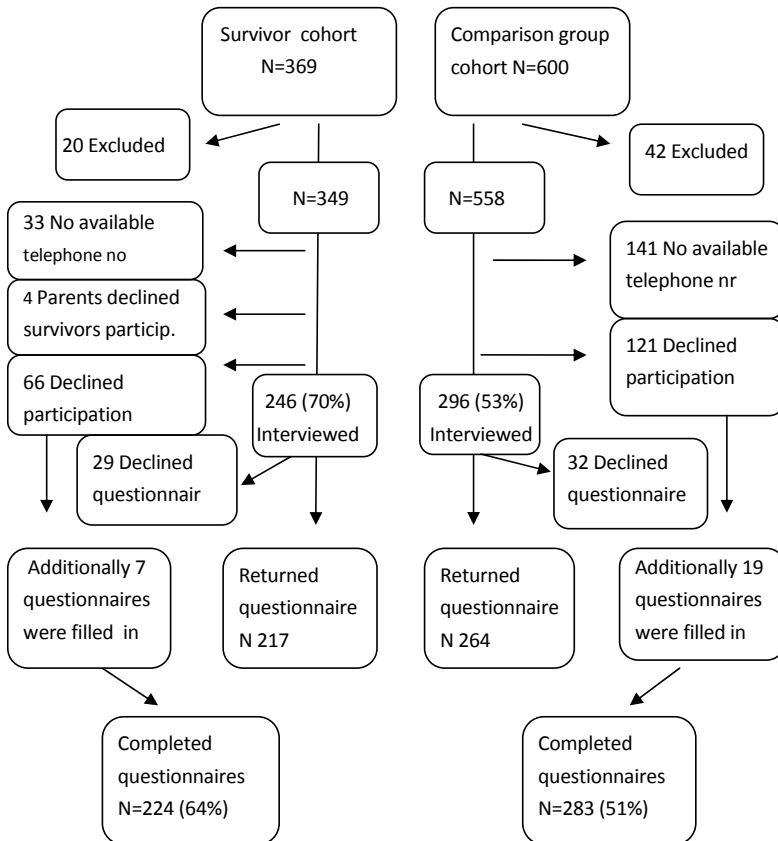


Figure 1. Participants and non-participants in the survivor group and the comparison group.

3.2 SAMPLES

3.2.1 Long-term survivors (I-IV)

Survivors of childhood cancer (N =369) were recruited from a regional cohort identified from the Swedish Childhood Cancer Registry (SCCR). They were all former patients diagnosed with cancer at ages 0-18 years during the period 1985-1999, at least

five years beyond their cancer diagnosis and at least 18 years of age at the time of the study. Excluded from the cohort were: those who had undergone bone marrow transplantation (n=14) because they were at the time included in another study, those who were experiencing a relapse or a second cancer in progress (n=2) or those who had a cognitive dysfunction (n=4). Of the remaining 349 survivors who were invited to take part in the study, 246 (70%) participated in an interview and 217 of the interviewees returned a self-reported questionnaire. In addition to this after being approached once more by a request letter, another 7 survivors who initially could not be contacted for an interview agreed on returning a self-reported questionnaire, resulting in a response rate of 64% for the questionnaires.

Table 1. Clinical and demographic characteristics of long-term survivors of childhood cancer, participants (n=246) and non-participants (n=103)

	Participants		Non-participants	
	n	%	n	%
Women	116	47	37	36
Men	130	53	66	64*
Diagnosis				
Leukaemia	55	23	21	20
Lymphoma	46	19	18	17
CNS ^a tumours	59	24	29	28
Soft tissue sarcomas	20	8	8	8
Malignant bone tumours	18	7	6	6
Renal tumors	13	5	6	6
Retinoblastoma	10	4	5	5
Other ^b	25	10	10	10
Treatment				
Chemotherapy solely	62	25	21	20
Combined therapy ^c	185	75	82	80
Relapsed disease	25	10	10	10
Age at diagnosis, mean (SD)	8.7	(5.0)	8.4	(5.2)
Time since diagnosis, mean (SD)	15.4	(4.0)	16.0	(4.0)
Age(years) at interview, mean (SD)	24.0	(4.1)	24.3	(4.6)

* p<0.05 tested for differences by χ^2 statistics

^a Central nervous system

^b Sympathetic nervous system, hepatic tumours, germ-cell tumours, other carcinomas, peripheral PNET

^c Surgery, radiation and a combination of surgery and radiation, all of them with or without chemotherapy

3.2.2 Comparison group (II-IV)

A random sample of 600 persons living in the greater Stockholm area was drawn from the Swedish population register (SPAR). The sample was stratified for age and gender so as to resemble the participating survivors. Forty-two persons were excluded because they did not speak Swedish (n=7), no longer lived in the Stockholm area (n=34) or had a cognitive dysfunction (n=1). Of the remaining 558 eligible participants who were invited to take part in the study, 296 persons (53%) agreed to be interviewed and 263 of the interviewees returned a self-reported questionnaire. Another 23 persons who initially could not be contacted for an interview agreed to return a self-reported questionnaire, resulting in a response rate of 51% for the questionnaires. In the comparison group, the mean age of the 296 participants was 25 years and did not differ from that of the non-participants. From the eligible sample, a higher proportion of females (62%) than males (52%) chose to participate ($p=0.03$). No statistically significant differences were found between the comparison group and the survivors with respect to age and gender.

Table 2. Socio-demographic characteristics of study participants; long-term survivors (n= 224), comparison group (n = 283)

	Survivors		Comparison group		p-value
	n	%	n	%	
Gender					0.571
women	115	51	144	50	
men	109	49	142	50	
Age					0.130
18-22	93	43	101	38	
23-29	103	45	132	44	
30-37	28	12	50	18	
Education level					0.001
junior compulsory	38	17	40	14	
senior high school	137	61	133	46	
postgraduate/university	49	22	113	40	
Occupation					0.000
student	92	41	62	22	
working	107	48	189	66	
unemployed	11	5	12	4	
sick leave	9	4	15	5	
parental leave	5	2	8	3	
Married/cohabitant/relationship	123	56	190	68	0.007
Single	101	44	93	32	
Months in current relationship, mean (SD)	40.6 (36.6)		51.8 (43.6)		0.019

Tested for differences by χ^2 statistics and Student's *t*-test

3.3 INSTRUMENTS

3.3.1 SEIQoL (study I & II)

The Schedule for the Evaluation of the Individual Quality of Life–Direct Weighting (SEIQoL-DW) is an instrument developed to assess individual QoL in the context of interviews without using predetermined variables (Hickey, Bury et al. 1996). The measure allows respondents to nominate the areas of life that are most important and to rate their level of current functioning or satisfaction with each area. A Swedish version of the SEIQoL-DW including both a generic part and a disease related part previously used in long-term cancer survivors (Wettergren, Björkholm et al. 2003) and shown to be feasible and valid (Wettergren, Kettis-Lindblad et al. 2009) was employed for this thesis. Each respondent was first asked the question “If you think about your life as a whole, what are the most important things - both good and bad - in your life at present that are crucial to your quality of life” (generic SEIQoL). It was possible to nominate as many areas as wanted. Subsequently, five of the nominated areas were selected by the participant and rated separately on a seven–point category scale with the verbal anchors “as bad as could possibly be” (scored 1) and “as good as could possibly be” (scored 7). An overall individual QoL score (SEIQoL index) was calculated by summing the ratings and dividing the numbers of nominated areas.

The Swedish version of the SEIQoL-DW is extended with a disease-related module to assess perceptions of the domains affected by the disease and treatment. The long-term survivors were asked the question: “If you think about the fact that you have been treated for childhood cancer, what in your life is influenced by this, both positively and negatively?” It was possible to nominate as many areas as desired. A selection of a maximum of five areas considered to have been influenced by the disease (positive and negative) was rated by the participants on a seven–point category scale depending on how troublesome or satisfying the consequence was perceived. The same calculation as mentioned above produced an overall disease index score.

3.3.2 SF-36 (Study II)

The Short Form-36 Health Survey (SF-36) is a generic instrument for assessing health status (Ware and Sherbourne 1992). The instrument groups 36 items into eight multi-item scales: Physical Function (PF), Role-Physical (RP), Bodily Pain (BP), General Health (GH), Vitality (VT), Social Functioning (SF), Role-Emotional (RE) and Mental Health (MH). Verbal response choices vary from two to six. Raw scores for each question are coded, summed and transformed into a scale from 0 (worst possible health state) to 100 (best possible health state), following standard scoring algorithms. Based on the eight scales, two summary index scores - Physical Component Summary (PCS) and Mental Component Summary (MCS) - are constructed for physical and mental health respectively. All scales influence the scores in the component summaries, although the PCS is primarily a measure of the PF, RP, BP and GH scales, whereas the MCS mainly comprises the VT, SF, RE and MH scales. Support for validity and reliability has been provided for the Swedish version of SF-36 (Sullivan 1998; Sullivan

2002), as well as when using the instrument in populations of long-term survivors of childhood cancer (Reulen, Zeegers et al. 2006).

3.3.3 Sexual function and experience (Study III)

A 30 items self-reported questionnaire was used to assess sexual function and sexual experience, as well as emotional quality in a partner relationship. Twenty-two items measures sexual functions/dysfunctions and sexual experience (19 items) and reflections regarding one's own sexuality (3 single items). This questionnaire has been validated in the epidemiological study "Sex in Sweden" thus providing population-based normative data (Lewin 1998; Öberg, Fugl-Meyer et al. 2004; Öberg and Sjögren Fugl-Meyer 2005). The items have also been found to be reliable when used in a group of men treated for testicular cancer (Eberhard, Stahl et al. 2009). Verbal response choices vary from four to six, defining to what extent a dysfunction had occurred during the last 12 months. Subsequently each dysfunction *per se* was reported in the same format if it was perceived as a problem. Only participants who had been sexually active with a partner during the past 12 months answered the items regarding sexual arousal and performance. The remaining eight items in the questionnaire exploring emotional quality of the relationship (Kreuter 1996) were answered by participants with a current partner relationship.

3.3.4 SOC (Study IV)

The SOC scale assesses an individual's capacity to respond to stressful situations and is a self-reported instrument, of which two versions are available. In its full version (The Orientation to Life Questionnaire) the sense of coherence scale consists of 29 items (Antonovsky 1987). In the present study we used the short version (13 items) which has been reported to have psychometric properties comparable to the full version (Schnyder U 2000; Feldt, Lintula et al. 2007). These 13 items comprise three components: comprehensibility (to which 5 items contribute), manageability (4 items) and meaningfulness (4 items). The respondents indicate agreement or disagreement with each item on a seven-point scale with two anchoring responses (e.g 1=never and 7=always). An example of an item is: "Has it happened that people whom you counted on have disappointed you?" After reversing the scores of the five items that are negatively worded, a total sum score, ranging from 13 to 91 is obtained. Higher scores reflect a higher SOC. The SOC scale has been shown to be applicable cross culturally (Eriksson and Lindström 2005) and has been translated into Swedish and tested showing acceptable results on both reliability and validity (Langius 1992; Nilsson 2000; Eriksson and Lindström 2005).

3.4 PROCEDURES

Data were collected through telephone interviews in Study I and II, and for Study II – IV postal questionnaires were dispatched. In Study I, only survivors of childhood cancer participated and data were collected during 2005. In study II-IV a control group was included and the data were collected during 2005-2007. In an invitation letter potential participants were informed about the study. It was highlighted in the letter that participation was voluntary and confidential. Within a week of dispatching the letters,

the research group contacted potential participants by telephone to request their participation. If they accepted, a suitable time for the interview was scheduled. Those who could not be contacted by telephone were sent an additional letter requesting them that they contact the research group if they wished to participate in the study. The interviewers were trained in the techniques used for administration of the extended SEIQoL-DW. During the telephone call the interviewer was supportive and asked neutral follow-up questions, for example, “What do you mean by..?” and “Can you tell me more about..?” in order to help the respondents elucidate their answers. The interviews lasted for 10-60 minutes and were tape recorded. A postal questionnaire including a package of self-reports was sent to the participants soon after the interview followed by a reminder letter if the questionnaire was not returned within two weeks.

3.4.1 Data analysis

The studies are based on one cohort of survivors and one cohort of controls, but depending on the primary measure used, the number of valid cases for analysis differs somewhat between the studies. Statistical calculations were performed using the Statistical Package for Social Sciences (SPSS), for Windows version 17.0. A statistical significance level of $p < 0.05$ was applied in all analyses.

3.4.2 Study I

The recorded interviews were transcribed verbatim and analysed according to content analysis (Graneheim and Lundman 2004). Content analysis is a method that can be used to draw valid conclusions about a manifest message in a communication through systematic identification of specified communication characteristics. Answers to open-ended questions are suitable for this technique. Words and sentences in the interviews were classified into mutually exclusive categories that reflect central messages in the interviews. Sentences classified in the same category are presumed to have similar meaning, either based on the precise meaning of the words or on words sharing similar connotation. An inductive reasoning was applied in all steps of the analysis whereas the data was taken from the specific observations to a more general level (Polit 2008). The analysis was carried out in the following steps: (1) The entire transcript was read through several times. (2) Words and sentences (meaning units) were identified reflecting a negative or positive consequence and codified. If a certain meaning unit was mentioned several times by the same respondent it was used only once. (3) The research group read the codes which were grouped into exclusive categories. (4) Repeated meetings within the research group took place to discuss the categories, define their boundaries and develop descriptions of the central characteristics of each category. (5) In this process, categories and sub-categories were modified until a final agreement was reached among the authors. The International Classification of Functioning, Disability and Health model (ICF) was used for inspiration (WHO 2001) when labelling some categories of the negative consequences thus using a deductive reasoning (Polit 2008).

For the quantitative data Student's *t*-test was used for comparison of means and Mann-Whitney test to test the differences in ranks of SEIQoL scores between groups. Chi-

square tests and Fisher's exact tests were used to compare proportions of categorical variables between groups.

Table 3. Example of categorization of statements reported by long-term survivors

Example of recording units	Central characteristics of the category	Subcategory	Category
"I had an operation when they removed one third of the muscle in my thigh which makes it weaker than the other leg"	Statements describing muscle, joint and skeletal dysfunctions related to movement.	Musculo-skeletal and movement-related functions	Bodily impairments and dysfunctions
"I am totally blind on my right eye and I can barely see anything with the left eye"	Statements describing dysfunction of the eye structure.	Impaired vision	
"I often have headaches and pain in my body due to problems in my arm, neck and back"	Statements describing various pain conditions.	Pain	

3.4.3 Study II

The tape-recordings and documentation from the interviews were transcribed verbatim and analysed according to manifest content analysis (Graneheim and Lundman 2004). The interview data from the two study groups were analysed separately by the first and the second author. There were repeated meetings within the research group before a final agreement was reached regarding the created categories thought to reflect the areas considered to be important in life. One of the authors not previously involved in the categorization process was asked to validate the analysis which resulted in 96% agreement for the survival group and 95% for the comparison group, thus indicating excellent agreement. Finally, the categories from the two groups were merged as they were judged to be equivalent.

Chi-square statistics were performed to compare proportions of categorical variables between groups. The Student's *t*-test was used for comparison of means between groups. A one sample *t*-test was used to compare SF-36 mean scores with Swedish normative data on young adults (Jörngården, Wettergen et al. 2006).

By two blocks, a hierarchical multiple regression analysis was employed to account for the variance in the scores of the dependent variable SEIQoL Index (overall quality of life). In the first block the living situation 'living alone' - which is known to differ between long-term survivors and the general population - was included, together with age, sex and group (long-term survivor vs. comparison group). In the second block, physical (PCS) and mental (MCS) health were added into the model.

3.4.3.1 Additional analysis

A multiple linear regression analysis was performed to determine the influence of the following predictor variables on the RP score (dependent variable) found to statistically differ between the survivors and the comparison group: group (0= comparison group, 1=survivors), age, sex (0= women, 1=men), marital/partner status (0=single, 1=married/partner) education (1= junior high/senior high, 2= post graduate/university) and occupation (3 variables: 0=no student 1= student;0=not working 1=working;0=no other 1= other).

3.4.4 Study III

Data were prepared by dichotomizing the verbal responses to facilitate the analysis and the interpretation of the data. The item measuring sexual desire had four response alternatives and was dichotomized into *never/rarely* and *sometimes/often*. Items with six response choices were cut off in the middle, for example experience of sexual interest during the past 12 months was divided into *frequent periods of low sexual interest* (all the time/ nearly all the time/quite often) and *rare periods of low sexual interest* (seldom/ almost never/never). The item measuring sexual satisfaction had five response alternatives and was dichotomized into *low sexual satisfaction* (very dissatisfying/ dissatisfying/ neither satisfying nor dissatisfying) and *high sexual satisfaction* (satisfying/very satisfying), the items regarding perceived problems were dichotomized in the same way. The differences were tested within gender, comparing the survivor group with the comparison group. Student's *t*-test was used for comparison of means between groups. Chi-square tests and Fisher's exact tests were used to compare proportions of categorical variables between groups.

3.4.4.1 Additional analyses

To investigate possible influence of the observed socio-economic differences (Table 2) between the long-term survivors and the comparison group logistic regression analyses were performed. Each item found to significantly differ between the long-term survivors and the comparison group in Chi-square analyses in Study III was entered as a dependent variable in a regression model (in males: sexually attractive, sexual interest, sexual satisfaction and orgasmic difficulty, in females: sexual experience). The independent variables used were group, marital/partnership status, diagnosis, level of education and occupation. In addition to the demographic variables found to differ between the two groups, age was used to examine the possible influence of age on sexual reporting. Sex was not controlled for in the analyses as men and women were analysed separately in different models. The

independent variables were coded as follows: group (0=comparison group, 1= survivors), education (1=junior high/senior high, 2=post graduate/university), occupation (1=student, 2=working, 3=other).

Logistic regression analyses were also performed with those variables (sexual satisfaction, sexual arousal problems, and sexual experience) found to significantly differ between male survivors diagnosed with CNS tumour and other diagnosis as dependent variables. Independent variables used were: age, time since diagnosis, diagnosis (CNS tumours vs other diagnoses), treatment modality (chemotherapy only vs all other modalities), marital status, education, occupation.

3.4.5 Study IV

The SOC score was regarded as an interval scale, and as the data were approximately normally distributed parametric methods were used. The Cronbach's alpha coefficient was used to assess internal consistency in the SOC scale and was 0.87 in the survivor group and 0.85 in the comparison group. Student's *t*-test was used for comparison of means between groups. Chi-square tests were used to compare proportions of categorical variables between groups Cohen's effect size (ES) was calculated to evaluate potential differences in means values of the SOC. According to Cohen $d=0.20-0.50$ indicates a "small", $d=0.51-0.80$ a "medium" and $> d=0.8$ a "large" effect size (Cohen 1988).

A multiple linear regression analysis was performed to determine the influence of the following predictor variables on the SOC score (dependent variable): group (0= comparison group, 1=survivors), age, sex (0= women, 1=men), marital/partner status (0=single, 1=married/partner) education (1= junior high/senior high, 2= post graduate/university) and occupation (3 variables: 0=no student 1= student;0=not working 1=working;0=no other 1=other).

Answers to the question regarding support were categorized into "yes", "sometimes" and "no" and the responses to the follow-up question regarding what kind of support were grouped into six categories. A logistic regression analysis was performed with need for support as the DV and age at interview, time since diagnosis, sex, diagnosis, treatment modality, marital status, education, occupation and the SOC mean score as the predictor variables. Age at diagnosis had a high correlation with both time since diagnosis and age at the time of study and was accordingly not used in the model. The reason for the chosen dichotomization regarding treatment (chemotherapy only vs all other modalities) was the well-known fact that chemotherapy alone more commonly results in fewer sequelae than a combination of other treatments. The diagnostic groups included in the model were leukaemia, CNS tumour and other solid tumours whereas the latter was kept as an indicator in the model.

Table 4. Outcomes/ variables, instruments and analyses used in Studies I-IV and Thesis.

<u>Study</u>	<u>Outcomes /Variables</u>	<u>Instrument</u>	<u>Analyses</u>
I	Consequences of childhood cancer	SEIQoL-DW (disease related)	Content analysis Student's <i>t</i> -test Mann-Whitney test Chi-square test Fisher's exact test
II	Important areas in life	SEIQoL-DW(generic)	Content analysis Student's <i>t</i> -test Chi-square test
	Self-reported health status	SF- 36	Student's <i>t</i> -test One sample <i>t</i> -test Hierarchical multiple regression
III	Desire Interest Arousal Performance Overall sexual satisfaction Sex with partner past 12 months Number of sexual partners in total Feeling sexually attractive Need to talk about one's own sexuality Turned to someone for advice regarding own sexuality	Sexual function and Sexual experience	Chi-square test Fisher's exact test Student's <i>t</i> -test
	8 items	Emotional quality of the relationship	
IV	Capacity to cope	13 item SOC scale	Student's <i>t</i> -test Cohen's effect size Cronbach's alpha Linear regression
	Need for support	Interview question	Content analysis Chi-square test Logistic regression
Thesis	Daily activity/physical health Selected Sexual function items Selected Sexual experience items	RP scale (SF- 36)	Linear regression Logistic regression Logistic regression

4 ETHICAL CONSIDERATION

The study was considered unproblematic from an ethical point of view by the Regional Ethical Review Board in Uppsala.

In any discipline that involves research with human beings, researchers must address a range of ethical issues. Based on the Declaration of Helsinki, codes of ethics have been developed for research (Polit 2008). The first ethical code to take into consideration for this thesis is that of beneficence. Human research should be intended to produce benefits for participants themselves or for other individuals or society as a whole. An invitation letter was sent to the potential participants in the survivor group informing them about the study and that they shortly would be contacted for an interview. The letter also informed about the benefits of the study, which were that we would learn more about quality of life among those who have been treated with childhood cancer, allowing us to better plan and provide support and follow-up care. The invitation letter to the comparison group also stated that their participation in the study was important, as it would enable a full understanding of the results from the survivor group.

Another consideration is maintaining respect for human dignity, which includes the right to full disclosure and the right to self-determination. In the letter, the nature of the study was fully described and information was given that participation was voluntary, including the right to disrupt the interview any time without having to provide a reason. The third ethical principle is that of justice. In this case, the potential participants in the survivor group were informed that any ongoing contact with health care would not be affected if they should choose to decline or disrupt participation in the study. Furthermore, by enclosing telephone numbers to the research personnel, the participants could receive clarification when desired. Finally, it was highlighted in the letter that participants have the right to expect that any data they provide would be kept in the strictest confidence and would not be presented in a way that would allow identification of individuals.

5 RESULTS

5.1 CONSEQUENCES OF CHILDHOOD CANCER (STUDY I)

The survivors were asked “Is your life in any way negatively and/or positively affected by having been treated for cancer as a child?” Fifteen percent of them answered that they did not consider their life to have been influenced by having had cancer. These responders were more likely to be men, diagnosed with leukaemia before the age of six and currently medication free. The remaining 85% reported a median of two consequences. Sixty-eight percent reported at least one negative consequence, 54% reported at least one positive consequence, and 36% reported at least one negative and one positive consequence. The consequences were grouped into categories and the negative consequences are shown in Table 5.

5.1.1 Negative consequences

Bodily impairments and dysfunctions were reported by 34% of the survivors. This was the largest of the negative categories and includes a variety of physical shortcomings due to the cancer experience. The survivors described having problems with mobility due to muscle, joint and skeletal dysfunctions. Also, various pain conditions were described related to the reported dysfunctions. Other physical impairments reported were related to vision, hearing, the mouth and stomach. The survivors could also be troubled by impaired balance, lack of physical fitness and feeling tired. Cognitive dysfunctions were described as difficulties with learning, reading, comprehending or having memory problems.

Limitations in activity and participation were reported by 28% of the survivors. A variety of social problems were described, such as difficulties with relationships to friends and with romantic and intimate relationships. Furthermore, there were statements describing difficulties in performing desired activities, restrictions in daily life situations, work and education. Other limitations in life could be due to constant hospital check-ups or treatments as well as to the drawbacks of being continuously on medication.

Psychological impact reported by 22% included statements about being in a depressed mood, having worries about the disease coming back or other future concerns. Also the survivors described being distressed by the memories of being ill and having a fear of hospital visits and hospital procedures. Other psychological impacts were described as lack of self-confidence or feelings of being different from peers.

Altered body appearance was reported by 15% of the survivors and the descriptions included concerns about personal appearances such as disturbing scars, poor hair quality, being short or overweight as well as impaired growth. Feeling embarrassed in front of other people about having a prosthesis was another concern.

Table 5. Negative consequences of childhood cancer nominated by long-time survivors (N=246)

Categories	Subcategories
Bodily impairments and dysfunctions	Musculo-skeletal and movement-related functions, Impaired vision, Effect on hearing, Oral and dental problems, Gastric and intestinal dysfunction, Pain, Cognitive dysfunction, Fatigue and lack of fitness, Easily infected, Impaired balance, Reproductive and sexual dysfunction, Epilepsy and cramps, Tingling and numbing sensations, Other disease related dysfunctions
Limitations in activity and participation	Physical limitations in performing desired activities, Interaction with others, Work and educational limitations, Tied up to medical routines, Difficult to meet a partner, Dependent of others
Psychological impact	Worry about the disease coming back or other future concerns, Lack of self-confidence, Distressing memories, Hospital anxiety, Depressed mood, Anxiety, Other impact on mood
Altered body appearance	Disturbing scars, Quality of hair, Overweight, Impaired growth, Other concerns about personal looks

5.1.2 Positive consequences

The survivors who reported positive consequences (54%) considered the cancer experience to have affected their perspectives in life. The statements described a fuller appreciation of life, changed values and a more positive view of life, making different priorities and not taking anything for granted. The cancer experience had also given them a more positive self- perception including being calm and mature, mentally stronger, having a good self-confidence and greater emotional openness and compassion for others. Other positive consequences described by the survivors included having better relationships with friends and family as well as a desire to help others. Moreover, the cancer had led to an increased interest in personal health and medicine.

5.1.3 Consequences reported in subgroups

Women, to a greater extent than men, reported that their cancer experience had a negative psychological impact on life and led to an altered body appearance. Women to a greater extent than men also reported positive interaction with others due to the cancer experience. There were some statistically significant differences between diagnoses regarding the reported consequences. Survivors diagnosed with CNS tumours reported bodily impairments and limitations in performing activities to a

greater extent than did those with other diagnoses. Moreover, those diagnosed with leukaemia reported a positive self-perception and interaction with others to a greater extent than did survivors with other diagnoses. Those treated with combined therapy (surgery, radiation and a combination of surgery and radiation with or without chemotherapy) to a greater extent reported bodily impairments and dysfunctions as well as limitations in activity and participation. A reverse pattern was seen regarding positive interaction with others which was reported by a smaller percentage of those treated with combined therapy compared to those treated solely with chemotherapy.

5.2 IMPORTANT AREAS IN LIFE (STUDY II)

Through the interviews, the survivors and the comparison group were asked to nominate important areas crucial to QoL. The most frequently reported areas categorized as - 'Family life', 'Relations to other people', 'Work, career' and 'Interests, leisure activities' - were the same for the survivors and the comparison group (Table 6). In both groups, participants nominated an average of four important areas each (range 1-5). The survivors to a higher extent than the comparison group reported that 'Family life' and 'Relations to other people' were important for QoL. Even though the survivors reported being significantly less satisfied with 'Own health' and 'Finances', those categories were reported to a lower extent among the survivors than among the comparison group. All nominated important areas were rated and no difference was found in overall QoL mean score (SEIQoL Index) between the groups.

Table 6 . Frequencies and mean rating scores of areas nominated as most important in life among long- term survivors (N=246) and a comparison group (N=296)

Areas	Survivors		Comparison group	
	n ^a (%)	mean ^b (SD)	n ^a (%)	mean ^b (SD)
Family life	189 (77 [*])	6.2 (1.0)	198 (67)	6.1 (0.9)
Relations to other people	167 (68 [*])	5.6 (1.1)	179 (60)	5.6 (1.1)
Work, career	113 (47)	4.7 (1.5)	145 (49)	5.1 (1.1)
Interests, leisure activities	94 (38)	5.2 (1.1)	102 (34)	5.0 (1.2)
Own health	64 (26 ^{**})	4.6 ^{**} (1.5)	88 (30)	5.2 (1.3)
Relationship to a partner	60 (24)	6.1 (1.2)	66 (22)	5.8 (1.5)
Studies, education	54 (22)	5.4 (1.3)	50 (17)	5.0 (1.2)
Housing, living conditions	22 (9)	5.4 (1.4)	34 (11)	5.1 (1.4)
Finances	17 (7 [*])	3.6 ^{**} (1.0)	40 (14)	4.2 (1.0)
Satisfied with life and self	19 (8)	5.2 (1.1)	32 (11)	4.8 (1.1)
Pets and animals	11 (4)	6.1 (1.3)	8 (3)	6.5 (0.9)
Love	9 (4)	4.2 (1.5)	9 (3)	5.4 (1.5)
Goals, new experiences	9 (4)	6.2 (1.1)	9 (3)	5.2 (1.1)
Choice and independence	9 (4)	5.2 (0.7)	7 (2)	5.1 (1.6)
Others health	7 (3 [*])	4.9 (1.5)	23 (8)	5.4 (1.3)
Available time	4 (2 ^{**})	4.5 (1.0)	21 (7)	4.7 (0.8)
Miscellaneous	8 (3)	5.5 (1.7)	5 (1)	5.0 (2.1)

^a tested for differences by χ^2 statistics

^b tested for differences by Student's *t*-test

* $p < 0.05$, ** $p < 0.01$

5.3 SELF-REPORTED HEALTH (STUDY II)

The long-term survivors reported health status similar to that of the comparison group and worse in only one of the eight functional scales of the SF-36, the mean score for RP (social role limitations caused by physical factors) which was statistically significant lower in the survivor group. A multiple regression analysis with the RP scale as the dependent variable showed that sex was a statistically significant predictor of health status. Being male sex was associated with less limitations due to physical conditions. The other predictors in the model (age, marital status, education level and occupation) were not significant.

In the regression analysis including the predictors survivor group and comparison group, the influence of self-reported physical (PCS) and mental health status (MCS) and socio-demographic characteristics (age, sex and living alone) on the dependent variable overall quality of life (SEIQoL Index) was tested. The results showed that these were all weak predictors of the ratings of overall QoL. In the final model only 17% were accounted for by the predictors of the variance in overall QoL.

5.4 ASPECT OF SEXUAL HEALTH (STUDY III)

There were no statistically significant differences regarding self-reported sexual function between the female survivors and the females in the comparison group. Male survivors more frequently reported periods of low sexual interest, low sexual satisfaction and orgasmic difficulties and less frequently reported feeling sexually attractive than the males in the comparison group. It was also twice as common among male survivors as among males in the comparison group to report frequent periods of low sexual interest and it was twice as common for male survivors to report this as a problem.

In contrast to female survivors, male survivors reported a statistically significant lower total number of sexual partners than the comparison group did. The female survivors differed in two ways from females in the comparison group regarding sexual experience. They were less likely to have had sex with a partner during the past 12 months and they reported being married, cohabitant or having a close relationship to a lower degree than did females in the comparison group. For men there was no significant difference in this respect. Neither women nor men in the survivor group differed from the comparison group with respect to mean time in months in a current relationship. Furthermore, there were no significant differences reported within genders regarding emotional quality in a partner relationship. Seventy percent of the female survivors and half of the male survivors had turned to someone for advice regarding their own sexuality which was similar to the numbers in the comparison group. The men most commonly had turned to their partner or to a physician whereas the women had most commonly turned to their partner or to a friend.

In multivariate logistic regression analyses with the outcomes of sexual function and experience that significantly differed between the long-term survivors and the

comparison group, neither of the two socio-demographic variables education level and occupation were shown to be statistically significant. Group (comparison group vs survivors) and marital status were the only predictors that were statistically significant. It was about twice as likely to feel less sexually attractive (male survivors $p=0.03$, OR 0.579) and three times more likely to have low sexual interest for male survivors ($p=0.011$, OR 0.356) than for men in the comparison group. It was three times as likely for male survivors ($p=0.016$, OR 0.439) and over six times as likely for singles ($p<0.0001$, OR 6.659) to have low sexual satisfaction, over four times as likely for male survivors ($p=0.035$, OR 4.673) and over six times as likely for singles ($p=0.009$, OR 6.435) to have orgasmic difficulties. Finally, in the model being sexually active only being single ($p<0.0001$, OR 0.013) was a significant predictor.

The impact of diagnoses on sexual function was in Chi-square analyses shown only among the male survivors. Men diagnosed with a CNS tumour statistically significant more frequently reported sexual arousal problems and overall low sexual satisfaction. They were less likely to have had sex with a partner during the past 12 months, and they reported a lower total number of sexual partners than did males with other diagnoses. Logistic regression analyses were performed with these variables of sexual function as dependent variables. The independent variables included were: age, time since diagnosis, disease, treatment modality, marital status, education, occupation. The models showed that it was four times as likely for those diagnosed with a CNS tumour ($p=0.041$, OR 0.254) and 15 as likely for those being single ($p<0.0001$, OR 15.019) to have low sexual satisfaction. Moreover it was nine times as likely for those diagnosed with a CNS tumour ($p=0.022$, OR 9.343) to have arousal problems and three times less likely for those diagnosed with a CNS tumour ($p=0.046$, OR 3.238) to be sexually active.

5.5 NEED FOR SUPPORT (STUDY IV)

The survivors were also posed the question: “Do you need any support today due to the cancer experience and if so what kind of support?” Twenty percent of the survivors reported a current need for support and 12% reported that they sometimes experience a need for support. Almost two thirds of those who reported a current need for support were females, and 70% had their cancer diagnosis at the age of six or older. Among the variety of different kinds of support that were mentioned, “psychosocial support” was reported by a majority and this refers to wanting to talk to someone i.e., a counsellor, a psychologist or to have peer support. Almost one third mentioned “medical support” which refers to wanting to see a physician about medical issues. Other kinds of support that were mentioned were physiotherapy, financial, educational and support in daily activities.

5.6 CAPACITY TO RESPOND TO STRESSFUL SITUATIONS (STUDY IV)

The capacity to respond to stressful situations was assessed by the SOC scale. Overall, the sense of coherence (SOC) did not differ, between the survivor and comparison groups. SOC was higher among men than among women in both groups. A linear

regression analysis with SOC as the dependent variable and group, age, sex partnership status, level of education and occupation showed that female sex and being single were significant predictors of a low SOC score. The survivors who reported a current need for support had a significantly lower mean SOC score than that of the remaining survivors. The logistic regression model with the predictors age, sex, diagnosis, treatment, partnership status, level of education and occupation accounted for between 24.0% and 36.8% of the variance in need for support status. The model showed that a low SOC score and combined treatment reliably predicted need for support. Those with combined treatment were over three times as likely to report a need for support. For every 10 units decrease on the SOC scale it was three times as likely to report a need for support. None of the other predictors were significant.

6 DISCUSSION

The results from the four studies will be discussed in this thesis on the basis of the descriptions given by the survivors themselves. In the interviews, the survivors were asked to give their views on life and their perceptions of the impact of childhood cancer on health and quality of life. The following is an attempt to give the reader an overall picture of the influence of childhood cancer on adult life as seen from a multidimensional perspective.

6.1 SELF-REPORTED HEALTH

The negative consequences due to childhood cancer reported in the interviews (SEIQoL) by the long-term survivors mainly concerned physical dysfunctions and activity limitations, which is in concordance with previous research (Hudson, Mertens et al. 2003; Ness, Bhatia et al. 2005; Hjern, Lindblad et al. 2007; Reulen, Winter et al. 2007). In the present study, survivors of CNS tumours and those having received a combined treatment reported these shortcomings to a greater extent than did survivors with other diagnoses and treatment modalities, as has been reported by others (Hudson, Mertens et al. 2003; Alessi, Dama et al. 2007; Boman, Hoven et al. 2009). The physical shortcomings are reflected in the SF-36 scale Role-Physical, which illustrates problems with daily activities owing to physical health, which in the present study was reported to be significantly worse among the survivors than in the comparison group. This was the only one of the eight SF-36 health scales in which the survivors differed from the comparison group. This finding largely agrees with the results of previous studies on long-term survivors using the same instrument (Langeveld, Stam et al. 2002; Zebrack, Zeltzer et al. 2002; PEMBERGER, Jagsch et al. 2005; Zeltzer, Lu et al. 2008). That as many as one third of the survivors reported bodily impairments (SEIQoL) indicates that physical health could be a larger problem than what is revealed by the SF-36. This may indicate an “under-reporting” of problems when health was assessed using the SF-36 health scale, which could be due to the instrument’s lack of sensitivity as regards distinguishing illness experiences (Sneeuw, Aaronson et al. 1997; Sprangers and Schwartz 1999).

The present results of self-reported health status are also in contrast to the overall view given by the body of literature describing clinician reported chronic health conditions among a majority of survivors of childhood cancer (Hudson, Mertens et al. 2003; Robison, Green et al. 2005; Oeffinger, Mertens et al. 2006). Several studies have shown that a patient’s own evaluation may differ considerably from that made by a clinician, suggesting that patients report less distress and dissatisfaction than what may be expected (Sprangers and Aaronson 1992; Sneeuw, Aaronson et al. 1997). Besides the different ways of measuring health, there could be additional explanations for the discrepancies found between the health of the survivors and the above-mentioned clinical studies. In the large cohort studies, the survivors were diagnosed with cancer at an earlier time period than the present survivor sample, which was diagnosed between 1985 and 1999. Over the past twenty years, therapeutic modalities have changed with a

possible reduced risk of adverse health. Research has shown that physical health status and late effects have been reported to be substantially worse among those diagnosed more than 20 years ago than among those diagnosed more recently (Blaauwbroek, Stant et al. 2007; Reulen, Winter et al. 2007). Also, the cumulative incidence of health problems has been seen to stretch even longer than 25 years after diagnosis (Oeffinger, Mertens et al. 2006).

Although the survivors did not frequently report (SEIQoL) that health was important to QoL, they did say they were less satisfied with their own health. This finding could indicate that the survivors considered poorer health less important, which would support the assumption that the individual may adapt to a loss of health by changing her/his perception of the importance of some aspects affected by such a loss (Lampic, Thurfjell et al. 2002). According to Calman's model of QoL, the differences between the hopes and expectations of an individual (the "gap"), which decreases QoL, may be narrowed by either improving the function or modifying one's ambitions and expectations (Calman 1984). Thus, in individuals with acquired health problems, minimization of such problems and enhancement of other aspects of life are required to improve the individual's QoL. For example, giving up something that has become problematic such as skiing or other leisure activities and focusing on something that is possible to achieve could be one way of narrowing the "gap" and thus improving QoL.

6.2 BODY APPEARANCE AND SEXUAL CONCERNS

The present finding of dissatisfaction with body appearance, such as disturbing scars and other changes to the body, was another negative attribute shown to be important to the survivors in the present study. Previous research on long-term survivors of childhood cancer has shown that endocrine disorders from disease and treatment during childhood have the potential to lead to short stature and undeveloped secondary sexual characteristics (Armstrong, Chow et al. 2009). These and other physical impairments, dysfunctions or dissatisfaction with the body may influence how sexually attractive a person perceives her-/himself to be, which could affect self-esteem and the likelihood of initiating sexual relationships. Despite this, sexual difficulties were not frequently reported by the young adult survivors in the present study.

Research in the general population has shown that women and men report different kinds of sexual dysfunctions and different frequency rates for these dysfunctions (Eplov, Giraldo et al. 2007; Beutel, Stobel-Richter et al. 2008; Christensen, Gronbaek et al. 2010). This is why we have chosen to study sexuality in childhood cancer survivors within gender. Women in the survivor and comparison group reported a similar frequency of sexual dysfunctions to those reported among Swedish women in general (Öberg, Fugl-Meyer et al. 2004). The differences between the survivor and comparison groups found among women and not among men concerned sexual experience. The female survivors were less likely to report having a partner and having sex. This was associated with being single, which is in good accordance with the relation between sexual activity and partnership status in the general population (Beutel, Stobel-Richter et al. 2008). The present results indicate that self-reported sexual function may be influenced by childhood cancer, particularly for men diagnosed with CNS tumours.

Being single and having had a CNS tumour diagnosis predicted less sexual experience and low sexual satisfaction among men. The findings are in line with previous reports of survivors of CNS tumours (Reimers, Mortensen et al. 2009), and these circumstances have been suggested to be a consequence of the impaired memory, physical shortcomings and emotional distress found in CNS survivors (Janson, Leisenring et al. 2009). Periods of low sexual interest were reported more frequently in the male survivor group than in the comparison group. Recent research has shown that adult male survivors treated for cancer in childhood are at risk for hypogonadism and androgen deficiency (Romerius, Stahl et al. 2009). This is known to influence to what extent a man wishes to have sex, and it has been argued that replacement therapy is under-used in male survivors of childhood cancer, as there is a lack of accurate assays and population-based normative ranges for testosterone concentrations (Bhasin, Enzlin et al. 2007). Hormone replacement therapy is common among both women and men who have survived childhood cancer, a circumstance that could influence the way they report their sexual function and experience. It is beyond the scope of this thesis to explain the causality of the present findings. Also, we have not approached the issue of gender differences in sexual function and experience, and further investigations are needed to gain a deeper understanding of how troublesome sexual difficulties are perceived by both men and women.

6.3 PSYCHOLOGICAL IMPACT AND MENTAL HEALTH

In the interviews, one fifth of the survivors reported a negative psychological impact on life due to the cancer experience. This was described as - living with uncertainty and fears of recurrence of the old cancer or of contracting another cancer, concerns about the impact the disease or treatment may have on their future health, fertility and on the health of their existing or future children – numerous worries that have previously been demonstrated by others (Langeveld, Stam et al. 2002; Zebrack and Chesler 2002; Gurney, Krull et al. 2009). Women more often than men reported a negative psychological impact on life as well as poorer self-reported mental health. Analyses of gender differences in the general population have shown that females report more psychological distress and worries across the life span (Macintyre, Hunt et al. 1996). According to Macintyre et al. (1996), one common explanation for higher rates of psychological distress symptoms among females may be that they are more sensitive to various discomforts and more willing to report symptoms of distress than men are. Mental health, measured by SF-36, was low in comparison to population norms (Sullivan, Karlsson et al. 1995) among the survivors, but just as low in the comparison group. This is in line with recent research suggesting a general decline in mental health for young adults over the past decades (Jörngården, Wettergen et al. 2006). Reports indicate that risk factors for psychological distress observed in the general population (female gender, not involved in a committed relationship, lower household income, and lower educational attainment and unemployment) are the same for survivors of childhood cancer (Zebrack, Zeltzer et al. 2002; Zeltzer, Recklitis et al. 2009). The present findings show that the survivors had a considerably lower frequency of employment, were less likely to have graduated from higher education as well as to have married or to be living with a partner than were members of the comparison

group. This is supported by previous research (Weigers, Chesler et al. 1998; Langeveld, Stam et al. 2002; Langeveld, Grootenhuis et al. 2004) and suggests a delay in life goals, indicating that important social and occupational aspects of life are affected by childhood cancer.

6.4 LIFE VALUE PERCEPTIONS

In the present thesis, a multidimensional evaluation of the lives of long-term survivors of childhood cancer is presented, where personal values in life were one of the aspects. The most important areas reported to influence QoL were very similar in the survivor group and the comparison group and in line with previous findings from survivors of adult cancer (Wettergren, Björkholm et al. 2003) and from the general population (Bowling 1995). This indicates that, despite differences in childhood health experience, people share similar values concerning what is important in life. Previous research among cancer patients has suggested that life value perceptions play a role in psychological adaptation (Lampic, Thurfjell et al. 2002). It can be assumed that individuals may limit their perceptions of what is important to those values to which they have access (i.e., narrowing the “gap”) in order to restore or maintain psychological wellbeing.

The present findings show that the survivors more often than the comparison group prioritized their relationships to family and friends, something also shown by others (Gray, Doan et al. 1992; Karian, Jankowski et al. 1998). According to attachment theory, during illness children seek security and become attached to adults and peers who are sensitive and responsive to their needs (Ainsworth M 1978). This intensified bonding generates trust and could lead to a heightened need for and valuing of relationships later in life. Moreover, it is possible that overcoming a life threatening illness makes an individual more alert to life and leads to a desire to invest in meaningful relationships.

The survivors also reported a deeper appreciation of life, changed values and a positive view of life and self. Being a survivor had accelerated their personal development and they felt mentally stronger and more mature than their peers. These findings are in accordance with other findings reporting gratifying outcomes after childhood cancer and could be seen as an expression of positive adaptation (Gray, Doan et al. 1992; Karian, Jankowski et al. 1998; Parry and Chesler 2005; Mattsson, Ringner et al. 2007). Being able to rise above one’s problems through personal growth and development can be understood within the concept of thriving (O’Leary 1998). It is believed that a key element of thriving is the ability to make positive meaning out of a traumatic experience, for example by establishing new life goals. One young man in the present study said that walking a long distance was a problem because of his amputated leg. Notwithstanding, meeting a large number of peers with similar problems, and enjoying new personal relationships, also had made him a stronger person. Calman (1984) proposes that the individual attaches positive value and meaning to expectations in life in order to narrow the “gap”. Thus, when things are going well, the individual may be able to cope with illness and stress and to achieve a good QoL.

6.5 CAPACITY TO COPE AND NEED FOR SUPPORT

The present findings suggest that, overall, long-term survivors have the resources needed to deal with life stressors despite having experienced cancer in childhood. Previous research has shown that people with a high sense of coherence (SOC) seem to be more resilient under stress, which can positively influence adaptation to stressful life situations and subsequently an individual's perception of QoL (Eriksson and Lindström 2006). According to Antonovsky (1987), it is important to strengthen existing general resistance resources and create new ones. Calman (1984) argues that action could be taken by, for example, health care providers to improve QoL by encouraging the individual to develop and grow in different ways. The aim is to try to help people reach the goals they have set for themselves.

In the present study, it was found that survivors who reported a current need for support also had a lower score on the SOC scale than did those reporting no need for support. This is in line with previous research showing a relationship between SOC and the presence of emotional as well as social support (Wolff and Ratner 1999; Nilsson 2000; Nilsson, Holmgren et al. 2003). The regression analysis showed a threefold risk for every ten unit decrease on the SOC scale of having the need for support. Combined treatment was also associated with the need for support. This "heavier" treatment modality includes surgery and radiation often in combination, with or without chemotherapy. An increased need for support could be reflected in the consequences of a more severe cancer disease and the known fact that heavier treatment modalities have the potential to cause serious adverse health outcomes in survivors of childhood cancer (Diller, Chow et al. 2009).

Psychosocial and medical support were the most common requests among the 20% of survivors who reported a current need for support. In a recently published study of health information needs among childhood cancer survivors, personalized information on late effects of treatment and information on self-care were highly prioritized (Knijnenburg, Kremer et al. 2010). In another study investigating childhood cancer survivors' knowledge of their disease and effects of treatment, only 84% could list their diagnosis, although all of the subjects stated that they knew their disease (Bashore 2004). The majority of participants in that study knew they had received chemotherapy and radiation, but only 30% were aware of what their risk of developing late effects could entail. In a Swedish study of the use of health services, 21% of long-term survivors stated that they had questions about their cancer experience, but did not know whom to consult or had failed to establish a relationship with a physician (Arvidson, Söderhall et al. 2006). In another recent study on survivors with an average of four years since diagnosis, opportunities to meet other survivors and the encouragement to pursue social activities were among the supportive care needs given the highest priority (Zebrack, Mills et al. 2007). According to Calman (1984), the individual's own list of problems and priorities should be assessed so action can be taken to improve QoL. In Sweden today, no organized support programmes in which survivors can address areas of concern and uncertainty are offered in follow-up care. In the Swedish national cancer

strategy proposal, it is recommended that sufficient competence and resources for follow-ups should be guaranteed (Socialdepartementet 2009).

6.6 QUALITY OF LIFE

Self-reported health status was not shown to have a major impact on overall ratings of QoL, suggesting that health and quality of life should be evaluated as different constructs. Clinical data may show various health impairments, but reveal less about what impact they have on the individual. According to Calman (1984), identification of problems and priorities makes it possible to develop realistic goals and to use these to reduce the “gap” and thus improve QoL. In agreement with Calman, it is not considered possible to make value judgements about individuals’ QoL; it is their own perceptions that matter. If health care professionals are to provide interventions and support for survivors of childhood cancer, they need to consider what the individual means by QoL. Research has shown that health care providers in cancer care can underestimate patients’ coping resources and QoL (Mårtensson, Carlsson et al. 2008; Pearcy, Waldron et al. 2008). Survivors of childhood cancer may have expectations and goals and may be affected by adversities with respect to their current health status that clinicians are not aware of. Asking about the needs that could be satisfied by a function rather than asking about that function *per se* may be one way to help. Thus, taking the individual’s perspective into consideration would seem to be crucial when providing care and support for survivors of childhood cancer.

6.7 METHODOLOGICAL CONSIDERATIONS

A major strength of the four studies presented here is the unique representative cohort of survivors included as participants. This survivor sample was recruited from one of the six childhood cancer centres and consists of the entire regional cohort during the time period 1986-1999, with the exception of the twenty patients who were excluded. All diagnostic groups were included, and the distribution of cancer diagnoses in the sample corresponds fairly well to the annual incidence of and survival from childhood malignancies in Sweden (Gustafsson 2007). The interview response rate was 70%, which can be considered fair. The return rate of questionnaires was 64%, which was lower than desired. It is not possible to know the health and QoL of those who chose not to participate, which is why the results must be interpreted with some caution. Feeling well and thinking they had nothing to report or not wanting to talk about their cancer experience could be some reasons for declining participation. Nevertheless, the similarities of the participating and the non-participating survivors regarding age and clinical characteristics decrease the risk of response bias.

Several steps were taken in an attempt to increase the response rate. The potential participants who could not be contacted by telephone were sent an additional invitation letter asking them to contact the research group. Also, a reminder was sent by post if the package of questionnaires had not been returned within two weeks of dispatch. If there was still no return after an additional two weeks, a telephone call was made to

check whether the respondent had any hesitations about filling in the questionnaire or whether he/she wished to receive an additional questionnaire.

To what degree the slightly larger non-responding proportion of males than females in the survivor group influenced the results is difficult to say with any certainty. It is a common finding that females more often respond to health questionnaires and has previously been demonstrated among long term survivors of childhood cancer (Larcombe, Mott et al. 2002). Women generally experience poorer health than men do (Macintyre, Hunt et al. 1996) and report higher levels of depression, distress and a variety of chronic illness (McDonough and Walters 2001). Furthermore, there may be a risk of selection bias in the survivor group due to the design of the study. Only those diagnosed between 1985 and 1999 and at least 5 years beyond diagnosis were included in the study. Accordingly, survivors 25 years of age or more at interview were 6-17 years old when they fell ill and, consequently, were diagnosed less often with childhood malignancies typically occurring in small children. This could influence physical health and function and lead to different dispositions in life. However, physical health was reported at similar levels in those diagnosed before the age of six years and in those diagnosed after the age of six, which suggests similar late effects and chronic health problems independent of any possible difference in diagnosis prevalence distribution.

The high non-response rate in the comparison group should be regarded as a risk for selection bias, which may limit the significance of the differences found between the two samples. Two areas of socio-demographics, education level and occupation, were found to differ between the comparison group and official statistics for the general population in Stockholm County. Participants in the comparison group had completed senior high school as the lowest level of education (junior high school/senior high school) to a higher degree than the general population and had a higher level of employment than average. It has been shown that being better educated and having higher socioeconomic status are associated with better health (Adler, Singh-Manoux et al. 2008). Nevertheless, neither of the two socio-demographic variables, education level and occupation, were shown to be statistically significant predictors of the outcomes that differed significantly between the survivors and the comparison group. Furthermore, we found that the health status of the comparison group was in parity with that found in the same ages in a recently collected Swedish normative data (Jörngården, Wettergen et al. 2006). Sexual function reported in the comparison group was also similar to that reported in the population-based normative data of a Swedish survey from 1996 (Lewin 1998).

To overcome time-consuming disadvantages for both researcher and study participants and to minimize the risk of high attrition, telephone interviews were used in the present study. It can be difficult for the interviewer to know of or control for any environmental factors distracting the attention of the participant during the interview (Musselwhite, Cuff et al. 2007). Changes in body language and other visual cues are lost when telephone interviews are used. However, the absence of a face-to-face contact during

the interview permits more anonymity and may result in a more relaxed interview. Another advantage of the telephone interview was the opportunity for the participants to directly receive an answer to any questions regarding the study aim or procedures. Questions have been raised about the quality of data obtained via telephone versus face-to face interviews, but there is little evidence to be found regarding their respective merits or shortcomings (Novick 2008). To avoid loss or distortion of any data, the telephone interviews were tape-recorded when interviewing the survivor group. Before interviewing the comparison group, a pilot study was performed. Twenty volunteers were recruited to interviews; ten were tape recorded and for the other ten only notes were taken. We could conclude that the information in both the subsequent analyses could be considered equivalent, which is why the comparison group interviews were not tape recorded. Finding valid phone numbers to some members of the identified comparison group proved to be a challenge. Through the SPAR registry, we had access to names and addresses, but only a valid phone number in 75% of the cases. Without a personal identity number it was difficult to track people down. Another problem was the common use of mobile phones and the frequent use of a pay card among young persons, which makes the phone number difficult to trace through a telephone directory. Despite these difficulties, the telephone mode of conducting interviews seems to be reasonable when studying young people, as it is a convenient and quick method and probably promotes participation (Musselwhite, Cuff et al. 2007).

Due to the cross-sectional design, the data provided in this present thesis can mostly tell us about the situation at a particular point in time. A prospective approach would provide more information about causes and changes and more conclusions could be drawn. The use of quantitative as well as qualitative data in the present thesis can be seen as an advantage, as this combined approach allows us to look at the research question from different angles (Clark 2008). The extended Swedish version of the SEIQoL -DW seems to be an appropriate instrument for assessing QoL in long-term survivors of childhood cancer. As previously reported, the questions used in the disease-related part of the SEIQoL were successful in capturing the consequences - both negative and positive - of the cancer experience (Wettergren, Björkholm et al. 2005). The present results indicate that a more detailed description of the survivors' health problems was given in the SEIQoL interview than reported in the standardized questionnaire (SF-36).

The advantage of choosing the SF-36 is that a generic instrument is suitable for comparing the survivors' results with those found in the comparison group. Another advantage with a well-used instrument is allows us to compare results across studies. One possible disadvantage, however, is if the items on the functional scales lack the sensitivity or specificity to capture the actual health status of the survivors (Westerman, Hak et al. 2008). It is difficult to say whether this explains the discrepancy between health in the present survivor group compared to studies of verified medical health problems. It is possible that a more disease-specific instrument would be better when assessing the actual health status of childhood cancer survivors. It has also been suggested that there is a risk of response bias when assessing health status using a

standardized instrument in long-term survivors of childhood cancer (O'Leary, Diller et al. 2007). If an item is found to be unimportant or not applicable to the person's situation, it may be ignored or the function might be guessed in activities that cannot be performed.

The questions used in the present study to assess sexual function have been validated in several studies (Lewin 1996; Öberg, Fugl-Meyer et al. 2004; Öberg and Sjögren Fugl-Meyer 2005; Eberhard, Stahl et al. 2009). However, it is not a standardized instrument, which may limit the possibility of comparing results across studies. The quality of the data, being on nominal and ordinal levels, limits the statistical analyses. It is difficult to choose a comprehensive instrument for assessing sexual health. Many instruments are focused on assessing sexual function and are disease-specific, and thus the selection of valid generic instruments is strongly limited (Daker-White 2002). The choice to use the present set of questions was made because the questions were considered relevant with respect to content validity in line with other measures of sexual function (Daker-White 2002).

Various aspects of the trustworthiness of research findings based on qualitative data deal with how appropriate the method of data collection is, how well categories cover the data, and how to judge the similarities and differences within and between categories (Graneheim and Lundman 2004). From the analysis of qualitative data, the researcher wants the most empirically meaningful information without too much loss of reliability. The majority of the interviews with the survivors were conducted by one person in the research team (myself), and for the comparison group, another person in the team was also involved. Both persons were trained in the techniques used for administration of the extended SEIQoL-DW. The content analysis was performed by the same persons; one of them analysed the data from the survivor group and the other the data from the comparison group. Regular meetings with the rest of the research team took place to discuss the categories until a final agreement was reached in the team. Additionally, the analyses were verified by a third party not primarily involved, and here a high percentage of achievement agreement was shown, also referred to as inter-rater reliability (Barbour 2001).

Due to the psychometric properties shown in the SEIQoL-DW (Wettergren, Kettis-Lindblad et al. 2009) and to the approximately normally distributed ratings of SEIQoL data in the present study, the data was analysed using parametric tests. It can be discussed, however, whether the SEIQoL scores can be considered to be on an interval scale. Therefore the Mann-Whitney test was also performed to test for differences in SEIQoL scores between groups and results showed statistical differences of the same magnitude as were shown when using the Student's *t*-test.

The objective of the study was not only to investigate the whole group of survivors, but also smaller subgroups of survivors, the aim being to gain a more comprehensive understanding of the data. Small subgroups affect the power of the analysis, however, which is why the number of statistical tests was limited in each study. A large number

of statistical tests increases the risk of obtaining significant results by chance, a type I error. One way of dealing with this problem is by using a more stringent p-value. On the other hand, there is a risk for type II errors, by wrongly accepting a false null hypothesis, when conducting extensive subgroup analyses. Therefore, the p-value was set at <0.05 in all analyses. To evaluate the clinical significance of the mean differences between groups of the SOC scores, Cohen's effect size (ES) was calculated.

7 SUMMARY AND CONCLUSIONS

The findings of the present thesis show that overall, young adult survivors of childhood cancer a median of 16 years after diagnosis reported health and QoL in a manner similar to that of age and sex matched peers. Nearly one third of the survivors reported no negative influences of childhood cancer, and a sizeable proportion also expressed gratitude and satisfaction with life. The negative consequences reported were mainly described as physical impairments and limitations in participating in activities. These problems with daily activities owing to physical health were also shown in the detected statistically significant difference between the survivor and comparison group in one of eight self-reported health status scales. Sexual difficulties were not frequently reported by the young adult survivors. Moreover, at group level the survivors appear to have the resources needed to cope with stressful situations in life to a similar degree as people in general do. We can conclude that our results do not indicate a need for an overall effort of special care for long-term survivors of childhood cancer. However, it should be pointed out that the present study does not cover the early years following the diagnosis or the period more than three decades after diagnosis when the needs may be different.

Cancer disease and treatment generally seems to have more impact on sexual function of male survivors than on that of female survivors and in particular on that of males diagnosed with CNS tumours. In female survivors, more impact was seen in relation to sexual experience. Therefore, we recommend assessment of sexual function and inquiry about sexual experience to be included in regular follow-ups after childhood cancer so that potential problems can be detected and addressed.

Among the one fifth of survivors who expressed a current need for support, psychosocial and medical support was most commonly reported. The survivors who have received combined treatment were identified as a group expressing need for support to a higher extent than the remaining survivors. The present findings reinforce the general agreement among clinicians about the need for organized follow-up clinics. In such clinics, survivors with special needs can be identified and could be offered besides medical advice, psychosocial support in terms of peer support as well as individual counselling.

8 IMPLICATIONS

Having knowledge of how long-term survivors of childhood cancer perceive adult life is important to the practice of nursing, as such knowledge will increase nurses' understanding of how to provide guidance and support for patients and their families during and after treatment. The influence of childhood cancer has the potential to lead to a variety of shortcomings unique for each individual, which in line with others, has been shown in this present thesis to vary between different subgroups of survivors. Therefore it is of the utmost importance to identify the problems and needs of each individual. The short-term consequences of disease and treatment are often identified directly, but the late effects, which have the potential to accumulate over time, also need to be taken into account. In addition to follow-up, considering the individual's perspective and asking him/her what is important in life may be helpful in establishing his/her priorities for achieving good quality of life. In the context of nursing practice, discussing an individual's resources for and impediments to health and quality of life in terms of comprehensibility, manageability and meaningfulness may be a structured way of better meeting the need for support in the follow-up of long-term survivors of childhood cancer.

Nurses in paediatric care could in practice talk about issues of sexuality and intimate relationships with adolescents during the treatment period and inquire about problems when seeing young adult survivors in follow-ups after childhood cancer. These are sensitive personal issues that can be difficult to discuss and require special communication and counselling skills. By discussing these matters, those who have or are at risk of having problems with their psychosexual development could be identified and sexual concerns could be detected and addressed.

Adverse effects of disease and treatment are well-known facts for paediatric oncology physicians and nurses, and in follow-up visits they often see the survivors with the highest burden of morbidity and those who have the greatest need for health care. With the knowledge that survivors often view quality of life in a positive way despite experiencing negative consequences, nurses and health care providers can be supportive when caring for worried children and families. It can be a comfort and encouraging hearing from healthcare professionals that after the disease and treatment periods are over, most survivors experience life in largely the same way their peers do.

9 FUTURE PERSPECTIVES

These studies contribute to the knowledge about perceived health and quality of life in young adult childhood cancer long-term survivors. Future research could build on this work by conducting longitudinal follow-ups of health and functional status as well as individual perceptions of the impact of childhood cancer. To gain a more profound understanding of how long-term survivors perceive their life situation from different perspectives, more in-depth qualitative studies are needed.

Future research, preferably longitudinal, is recommended to investigate how sexual dysfunctions may be related to different aspects of health, such as physical, emotional and social wellbeing. It would also be valuable, in the context interviews or focus groups to explore young adult survivors' own perceptions of their sexual lives and what consequences potential sexual dysfunctions may have. Furthermore, future studies could approach potential gender issues in this field.

According to the theory, SOC has not stabilized until the age of 30 years. The mean age of the study participants was 24 years which is slightly younger. It would therefore be interesting to investigate the stability of SOC in adulthood in a follow-up study. An increased need for support was found among certain subgroups of the survivors in this present thesis which highlights the call for research focusing on the interventions aimed at best address these needs.

10 SUMMARY IN SWEDISH

Behandlingsrelaterade hälsoproblem är välkända i den växande populationen av vuxna personer som behandlats för cancer i barndomen. Det övergripande syftet med denna avhandling var att undersöka hur genomgången behandling för cancer i barndomen påverkar livskvalitet och hälsa hos unga vuxna. Detta för att ge en ökad förståelse för gruppens behov av stöd i vuxenlivet.

I de fyra ingående studierna användes en tvärsnittsstudiedesign med intervjuer och självadministrerade enkäter. De som i barndomen diagnostiserats för cancer (> 5 år efter diagnos) bestod av 246 personer som var ≥ 18 år vid tiden för undersökningen och fick diagnosen i åldrarna 0-18 under perioden 1985-1999 i Storstockholm. I studie I undersöktes konsekvenser till följd av cancerupplevelsen genom telefonintervjuer baserade på den svenska versionen av Schedule for the Evaluation of Individual Quality of Life-Direct Weighting. I studie II-IV ingick en jämförelsegrupp bestående av 296 personer matchade för ålder och kön, slumpvis utvalda ur den allmänna befolkningen. Deltagarna undersöktes med två standardiserade instrument som bedömer hälsostatus (Short Form Health Survey-36), förmåga att hantera stress (Känsla av Sammanhang, KASAM) samt ett frågeformulär för utvärdering av sexuell funktion och erfarenhet.

De unga vuxna som haft cancer i barndomen beskrev och skattade sin livskvalitet och sin hälsostatus likartat med jämförelsegruppen. Nästan en tredjedel av de som genomgått cancer rapporterade inga negativa konsekvenser i livet på grund av cancerupplevelsen och hälften av dem beskrev också en positiv syn på livet och sig själv. De vanligast rapporterade negativa konsekvenserna innefattade olika fysiska problem och aktivitetsbegränsningar. De som haft cancer skilde sig från jämförelsegruppen i en av åtta SF-36-skalar, vilken avspeglar problem med dagliga aktiviteter beroende av fysisk hälsa. Män som haft cancer angav oftare sexuella dysfunktioner än män i jämförelsegruppen. Att vara singel och att ha blivit diagnosticerad med hjärntumör utgjorde en större risk för sexuella dysfunktioner. Hos kvinnor förelåg en statistiskt signifikant skillnad i fråga om sexuell erfarenhet, men inte vad avser sexuell funktion. Det fanns ingen signifikant skillnad i KASAM mellan de som genomgått cancer i barndomen och jämförelsegruppen. Tjugo procent av de som haft cancer rapporterade ett aktuellt behov av stöd och rapporterade en signifikant lägre KASAM-poäng. En låg KASAM-poäng och att ha fått kombinerad cancerbehandling var starka prediktorer för att rapportera ett behov av stöd.

Resultaten visar både positiva och negativa aspekter av sjukdomstiden och den genomförda behandlingen. På gruppnivå rapporterade de som överlevt en cancersjukdom i barndomen få hälsoproblem och övergripande god livskvalitet. Sexuella svårigheter rapporterades sällan av dessa unga vuxna som haft cancer. Dock tyder resultaten på att den genomgångna cancersjukdomen har en viss inverkan på självrapporterad sexuell funktion hos män och vad gäller sexuella erfarenheter hos kvinnor. Sammantaget verkar de som haft cancer ha de resurser som behövs för att

hantera stress i livet i samma grad som människor i allmänhet, även om en lägre känsla av sammanhang var förknippad med ett större behov av stöd.

Resultaten visar att påverkan av barncancer varierar mellan olika grupper och individer varför det är viktigt att identifiera varje individs behov av stöd. Att förstå hur de som genomgått cancer i barndomen uppfattar vuxenlivet är viktigt för utövande av omvårdnad eftersom det kommer att erbjuda möjlighet att ge vägledning och stöd till patienter och deras familjer under behandling och uppföljning.

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