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LIFE AFTER CHILDHOOD CANCER

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**Karolinska
Institutet**

Stockholm 2013

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Published by Karolinska Institutet. Printed by Universitetservice US-AB

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ISBN 978-91-7549-179-0

ABSTRACT

Young people who have undergone treatment for childhood cancer have a high risk of developing chronic health problems that could have a potential strong impact on their lives. How a childhood cancer experience affects the lives of young survivors has only been studied to a limited extent. The overall aim was therefore to investigate how adolescent and young adult survivors of childhood cancer perceive that their lives are affected by having had cancer. In study I, the aim was to gain a deeper understanding of how childhood cancer affects the lives of survivors by exploring adolescents' and young adults' views of what it is like living with this experience. In study II, the aim was 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.

The thesis includes two studies using cross-sectional designs, drawing on data from two different samples based on interview and survey data. In study I, 59 young survivors (12-22 years) were interviewed a median of five years after diagnosis (response rate 66%), and the interviews were analysed with qualitative content analysis. In study II, 246 long-term survivors (18-35 years) were interviewed a median of 16 years after diagnosis (response rate 64%), as well as 296 randomly selected controls (response rate 51%). Quality of life was assessed using the Schedule for the Evaluation of Individual Quality of Life- Direct Weighting (SEIQoL-DW) and self-reported health status was assessed using the Short Form Health Survey (SF-36).

The results from study I revealed that the young survivors of childhood cancer could be divided into three groups depending on how they perceived having had cancer affected their current lives: 'Feeling like anyone else' (the informants who described that the cancer experience had almost no influence on their current life) (49%), 'Feeling almost like others' (those who described some influence) (44%) and 'Feeling different' (those describing a great influence on current life) (7%). The results from study II showed that long-term survivors rated their overall quality of life and self-reported health status almost in parity with the comparison group. In both groups, family life, relations to other people, work and career, interests and leisure activities were the areas most frequently reported to influence quality of life. The long-term survivors only differed from the comparison group on one of eight SF-36 scales, which reflected problems with daily activities owing to physical health.

In conclusion, survivors appear to get along well after treatment for childhood cancer, although many informants described lives that were to some extent affected by having had cancer. To meet the needs of young survivors who perceive that the cancer experience to a large extent influences daily life and may find the health impairments hindering, follow-up care should be able to identify those having trouble in daily life and offer them support to strengthen their resources and ability achieve a good quality of life.

Keywords: childhood cancer, long-term survivors, adolescents, young adults, late-effects, sense of coherence

LIST OF PUBLICATIONS

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

- I Berg Doukkali, E., Winterling, J., Eriksson, L. E., Lampic C., Silván Hagström, A., Wettergren, L. Adolescents and Young' Adults Experiences of Childhood Cancer – Descriptions of Daily Life Five Years After Diagnosis. (Submitted)
- II Sundberg, K., Doukkali, E., Lampic, C., Eriksson, L. E., Arvidson, J., Wettergren, L. (2010). Long-Term Survivors of Childhood Cancer Report Quality of Life and Health Status in Pariety With a Comparison Group. *Pediatric Blood Cancer*, 55, 337-343.

CONTENTS

1	Preface.....	1
2	Background.....	2
2.1	Childhood cancer.....	2
2.1.1	Cancer treatment.....	2
2.1.2	Disruption in life continuity.....	3
2.1.3	Survivorship and follow-up.....	4
2.2	Health and quality of life.....	6
2.2.1	Measurement of quality of life and health.....	7
2.2.2	Health and quality of life among survivors.....	8
3	Aim.....	9
3.1	Rationale.....	9
3.2	Overall aim.....	9
4	Method.....	10
4.1	Design.....	10
4.2	Samples.....	10
4.2.1	Sample Study I.....	10
4.2.2	Sample Study II.....	11
4.3	Data collection.....	12
4.3.1	Data collection Study I.....	12
4.3.2	Data collection Study II.....	13
4.4	Procedures.....	14
4.4.1	Procedure Study I.....	14
4.4.2	Procedure Study II.....	14
4.5	Data analysis.....	14
4.5.1	Data analysis Study I.....	14
4.5.2	Data analyses Study II.....	15
5	Ethical considerations.....	17
6	Results.....	18
6.1	Results Study I.....	18
6.2	Results Study II.....	20
7	Discussion.....	23
7.1	Methodological Considerations.....	25
7.1.1	Study I.....	25
7.1.2	Study II.....	26
8	Conclusion.....	27
9	Implications.....	28
10	Summary in swedish – Sammanfattning.....	30
11	Acknowledgements – Tack till.....	31
12	References.....	33

LIST OF ABBREVIATIONS

ALL	Acute lymphocytic leukemia
AML	Acute myeloid leukemia
CNS	Central nervous system
CRC	Convention on the Rights of the Child
HRQOL	Health related quality of life
MCS	Mental component summary
PCS	Physical component summary
QOL	Quality of life
SCCR	Swedish Childhood Cancer Registry
SEIQoL-DW	Schedule for Evaluation of Individual Quality of Life-Direct Weighting
SF-36	Short Form-36 Health Survey
SOC	Sense of coherence
SPAR	Statens person och address register
SPSS	Statistical Package for Social Science
WHO	World Health Organization

1 PREFACE

I have had a deep engagement in nursing within the area of cancer care for many years. My master's thesis (2007) focused on distress among adults with inoperable lung cancer and a few years later I had the opportunity to also deepen my knowledge of the lives of those who have undergone treatment for childhood cancer. I had the opportunity to participate in the planning phase and data collection in a study conducted at the Division of Nursing (study II). During the years of my research education, I have gained deeper knowledge and understanding about how undergoing cancer during childhood can influence the lives of young survivors and long-term survivors.

2 BACKGROUND

2.1 CHILDHOOD CANCER

About 300 children and adolescents in Sweden are diagnosed annually with pediatric cancer (Gustafsson, Heyman, & Vernby, 2007). Nordic countries and Sweden represents a survival gold standard with survival rates of up to 80% (Gatta et al., 2003). The cause of childhood cancer is still largely unknown even though the impact of environmental factors and viruses has been discussed and hereditary disposition has been identified in only a few diagnoses (e.g., retinoblastoma (Kogner et al., 2008)). Childhood cancer tumors are known to be more aggressive and faster growing than most cancer among adults. The most common cancer diagnosed during childhood, in ages 0-4 years, is acute lymphatic leukemia (ALL), which represents 85% of all leukemia among children with cure rates greater than 80%. The largest group of solid tumors in childhood is tumors of the central nervous system (CNS), which represents one fourth of all cases and is evenly distributed across ages. Other types of common solid childhood cancer are: Ewing sarcoma, soft tissue sarcoma, retinoblastoma, hepatic tumors, Non-Hodgkin's lymphoma and osteosarcoma (Gustafsson et al., 2007; NOPHO, 2011).

If a malignancy is suspected, the child is referred to a pediatric oncology center to receive a definite diagnosis and start treatment. In Sweden, pediatric oncology care is organized into six regional cancer centers (Umeå, Uppsala, Stockholm, Linköping, Lund, Göteborg). The diagnosis is determined in accordance with the international classification of childhood cancer and the treatment given follows the international and Nordic treatment protocols (NOPHO, 2011). The length of the treatment varies according to diagnosis and treatment protocol from 3-4 months up to several years.

2.1.1 Cancer treatment

Treatment protocols often include use of multimodal therapy with a combination of modes (chemotherapy, radiation therapy and surgery, immune therapy, hormone therapy) (Kogner et al., 2008). Chemotherapy treatment is received by the majority of pediatric patients and often with multiple drugs as part of the therapeutic regimen. Cytotoxic drugs act by inducing cell death and the effect is most potent in cells that have rapid cell growth like cancer cells. The mechanism will also have an impact on healthy tissue since rapid cell growth may cause unwanted side-effects. Temporary side-effects commonly reported are fatigue, mucositis, nausea and vomiting, infections and neuropathies as well as changes in appearance (i.e., alopecia and weight alteration) (Miller, Jacob, & Hockenberry, 2011). Additionally treatment procedures are often painful and related to distress. Long-term effects from the cancer itself and/or treatment can include physical conditions (endocrine problems, cardiopulmonary dysfunction, gastrointestinal disorders, impact on vision, hearing loss, mobility problems, and fatigue), psychosocial problems (depression and anxiety), and neurocognitive difficulties (Bleyer, 2005; Oeffinger, Nathan, & Kremer, 2008).

The goal when using radiotherapy treatment is to decrease the size of the tumor and often involves a combination with chemotherapy and/or (before or after) surgery. Use of radiation therapy requires careful weighting of the risks for negative side-effects

depending on which part of the body is treated. Radiotherapy treatment can cause acute complications including nausea, vomiting, alopecia and fatigue. Long-term late complications include hearing loss, endocrine dysfunction with impact on growth and fertility, and neurocognitive effects and secondary neoplasm (Minturn & Fisher, 2013).

The purpose of surgery treatment is to radically eliminate the tumor and regional lymph nodes as well as limit the damage of surrounding tissues. Surgery also has prognostic value and is used to remove solid tumors or in combination with chemotherapy and radiation therapy. Despite not undergoing primary cancer treatment with surgery, many patients will undergo some sort of surgical intervention like biopsy or getting an intra venous device (Blaauwbroek, Groenier, Kamps, Meyboom-de Jong, & Postma, 2007; Kogner et al., 2008; Oeffinger et al., 2008)

Survival is dependent on diagnosis, the stage of the disease and age at diagnosis. Deriving from new knowledge there has been a drastic shift in treatment intentions from palliation to cure in the last five decades. The strategy of treatment is to cure the young patient with the intention to minimize the risk for developing late complications from disease and treatment (Jenney & Levitt, 2002). This shift has also resulted in a change whereby the prognosis of the once fatal childhood cancer diagnosis has evolved to be defined as a chronic illness (Eiser, Hill, & Vance, 2000). Therefore the great majority of the childhood cancer diagnoses are curable and will respond well to treatment and the main factor influencing survival is patients' access to treatment and use of up-to-date treatment protocols.

2.1.2 Disruption in life continuity

The cancer experience can be viewed as a disruption in the continuity in a young person's life and the normal development from childhood to adolescence and from adolescence to adulthood (Larouche & Chin-Peuckert, 2006) Even though most of the treatment is given within open care units, the child will spend a lot of time in the hospital, and the treatment-related side-effects will require that the child/adolescent have intense contact with the hospital (af Sandeberg, Wettergren, Björk, Arvidson, & Johansson, 2012). Both early complications such as nausea, pain and fatigue and treatment-related infections hinder the child/adolescent from participating in daily activities (Bleyer, 2005; Enskär & von Essen, 2007; Kestler & LoBiondo-Wood, 2012; Ruland, Hamilton, & Schjødt-Osmo, 2009). The cancer treatment will also have an impact on body appearance in form of with hair loss and weight changes, which can lead to withdrawal from social contacts and activities (Drew, 2007).

Undergoing cancer treatment and being bound to the hospital environment for an indefinite time means a break in the child's normal life activities and contacts with peers (Hinds, 2004). Daily life is scheduled around treatment and space for other activities is limited which can impact on the individual's ability to uphold continuity in social contacts and school attendance. This means that the child's opportunities to participate in normal activities such as maintaining contacts with friends, school attendance and leisure activities often are limited for some time (af Sandeberg, Johansson, Björk, & Wettergren, 2008). Research investigating adolescents stress the importance of having their friends and family accept them despite them having bodily changes and wanting to be treated as "the same old" person as before the cancer (Williamson, Harcourt, Halliwell, Frith, & Wallace, 2010; Woodgate, 2005).

School is an important activity for children and adolescents, and in Sweden school attendance during treatment is recommended if general health permits this. Results from a study with the same sample as the present thesis, showed that school attendance at the start of childhood cancer treatment was low, but increased significantly during the first five months following the start of initial cancer treatment (af Sandeberg et al., 2008). The most common reasons for absence from school were hospital visits and fatigue, and a positive relation was found between self-reported Health Related Quality of Life (HRQOL) and numbers of attended school days. In another Swedish study in which, adolescents undergoing cancer treatment were asked to list issues that worried them, concerns about being able to attend school were expressed as main issues (Hedström, Ljungman, & von Essen, 2005). These results are in concordance with a North American report which showed that adolescents who had high absence from school also rated higher on cancer related stress and showed a lower degree of adaptation to their changed life situation as a person with cancer (Hockenberry-Eaton, Manteuffel, & Bottomley, 1997). Modern information technology and the Internet are helpful tools during hospitalisation for maintaining contact with friends, communication and on-going participation in school activities (Hokkanen, Eriksson, Ahonen, & Salanterä, 2004).

These life changes and uncertain future associated with a may induce worries and depression related to not being able to continue life as usual (Woodgate & Degner, 2002). Research has shown that young persons diagnosed with childhood cancer often experience decreased self-confidence with feelings of loneliness and alienation about no longer belong to the social group (Moody, Meyer, Mancuso, Charlson, & Robbins, 2006). As the contact to the world outside the family narrows during the treatment period, the young person will be more dependent on the family for social relations, as well the communication between the adolescent and health care personnel often is mediated by the parents (Evan & Zeltzer, 2006). The dependency on the health care system means the adolescent to some extent loses control over her/his life situation. To avoid feelings of uncertainty the young person must be provided age-appropriate information about childhood cancer and treatment-related issues (Palmer, Mitchell, Thompson, & Sexton, 2007).

2.1.3 Survivorship and follow-up

When the treatment phase is finished the child/adolescent enters the post treatment phase, which will also bring challenges to the child/adolescent as he/she continue with his/her life and engages in all aspects of life as a childhood cancer survivor. There are different meanings about the term survivorship, for some the term applies immediately after diagnosis (Shepherd & Woodgate, 2010) while others argue that it applies to after completion t of treatment (Miedema, Hamilton, & Easley, 2007). The description that anyone who has been diagnosed with cancer is a survivor, from the time of diagnosis until life ends, is one that the survivorship organisations uses (Aziz & Rowland, 2003). According to the American National Cancer Institute survivorship is defined as, “the physical, psychosocial, and economic issue of cancer, from diagnosis until end of life. It includes issues related to the ability to get health care and follow-up treatment, late effects of treatment, second cancers, and quality of life” (American Cancer Society, 2007). According to Aziz and Rowland (2003), the survivorship experience can be described in relation to three phases: the acute phase that covers the time from diagnosis to completion of initial cancer treatment: the extended phase in which the

initial cancer treatment: ends and normal life is resumed; which step-by-step leads to the third phase of permanent survival with diminished risk of recurrence (Aziz & Rowland, 2003). The samples that follow in the present thesis, are in the third phase of permanent survival.

Compared to adult cancer survivors, childhood cancer survivors have their adult life ahead of them. Many of them will experience diminished health status from chronic conditions including limitation in activity and functional impairment (Ness et al., 2005). Studies have shown that up to 70% of childhood cancer survivors experience one long-term complication and that approximately 30% have one severe or life threatening late-complication (Oeffinger et al., 2006).

Although some studies on the long-term consequences of childhood cancer describes a favorable outcome despite the cancer experience, other studies that have shown that childhood cancer survivors perceive difficulties in finding a partner and starting a family (Gerhardt et al., 2007) and to succeeding studies and career (Gatta et al., 2003; Stam, Grootenhuys, & Last, 2005). As a result, they face increased risk for developing depression and anxiety compared with peers without cancer experience (Zebrack & Chesler, 2002).

Under this at risk scenario, the young survivors may have to manage on-going health problems that require long-term follow-up care by health professionals. Due to the fact that most childhood survivors are very young at the time of diagnosis and treatment and may have limited understanding and knowledge in relation to their cancer diagnosis and treatment, they may therefore be unable to recall details about their medical history. When childhood survivors were asked if past treatment could cause future health problems, 35% of participants responded affirmatively; 46% responded negatively; and 19% did not know (Kadan-Lottick et al., 2002).

Lifelong follow-up is recommended for all survivors. In Sweden, young survivors receive follow-up in the pediatric oncology clinic up to the age of 22 years. The most common situation in Europe and in the United States is that the survivor is referred back to the primary care physician without formal transition from the pediatric oncology setting (Bleyer, 2005; Landier, Wallace, & Hudson, 2006). Transition to adult services may be hampered by lack of adult physicians with special knowledge of late complications of childhood cancer. To meet the needs of the growing population of childhood survivors, it is essential to develop long-term follow-up delivered with a comprehensive risk-based approach in an age-appropriate environment that is supported by nurse-led services (Edgar, Borthwick, Duffin, Marciniak-Stepak, & Wallace, 2012). The ideal is risk-based survivor care through health monitoring and prevention based on the previous diagnosis, cancer treatment, lifestyle behaviors, and comorbid health conditions (Nunez, Mulrooney, Laverdiere, & Hudson, 2007). In the Swedish health care system the young long-term survivor receives followed-up at a pediatric oncology center to the age of 20 years, and then are referred to adult oncology departments for continuation of disease appropriate follow-up (Arvidsson, Söderhäll, Eksborg, Björk, & Kreuger, 2006). Some pediatric oncology centers have established ongoing cooperation with oncologists and work together to prepare the young survivors for a successful transition to adult oncology.

2.2 HEALTH AND QUALITY OF LIFE

Health is a concept that can be viewed from different perspectives. In the naturalism approach to health the biostatistical definition equates the focus absence of disease with health and has functional ability as a core concept. This approach to health contrasts with the holistic definition in which an individual is seen as a whole person and having good health is connected to the individual's ability to achieve vital goals and health is compatible with the presence of the disease (Nordenfelt, 2007). The holistic definition is also used within nursing science in which health is the purpose of caring with the overall aim to support and strengthen people's health processes. To be in a state of health means the person experiences well-being, feels good and is able to implement life projects, as well as experiences meaning and coherence in life. An individual's understanding of health does not exclude disease but the individual must be able to relate to disease or other kinds of health barriers to be able to experience health (Dahlberg & Segesten, 2010). When the health concept is related to long-term survivors of childhood, it is the holistic health definition and the nursing perspective that is used in this thesis.

Another description of health and well-being is provided by Antonovsky's (1987) salutogenic framework. It addresses the factors that account for health and well-being to explain how people manage well despite adverse health experiences. Antonovsky argued that disability should be addressed in terms of positive adaptation and resolution to stress and moves beyond the biostatistical definition of illness. In the salutogenic framework, the individual at a given time point can be seen as moving along a continuum between total health/and total ill health. According to Antonovsky, the interest should be on what constitutes health and not on the course of disease (e. g., how an individual can stay healthy in spite of stressful life events). Antonovsky's idea was that the movement could be explained by the personal propriety "generalised resistant resources" (GRRs) which facilitates the individual's ability to cope successfully with stressful life events. When Antonovsky studied what united the GRRs, it led to the concept sense of coherence (SOC) with the key features of; comprehensibility, manageability and meaningfulness. Sense of coherence helps the individual to make sense of his/her social world as rational, understandable, structured and predictable. Comprehensibility is the way the individual apprehends situations in life and understands why things happen. Manageability is connected to the individual's resources and ability to cope with a situation; the individual will perceive having sufficient resources to help to cope with problems he/she confronts. Meaningfulness can be related to the individual's wish to control and make sense of a situation; and the ability to find meaning, motivation and value to persist when confronting/dealing with disruptive conditions (Antonovsky, 1987). Critics have stated that the theory does not attend to the dynamic interrelationship between comprehensibility, manageability and meaningfulness and it is a rational model which minimizes emotions and affective behavior (Geyer, 1997). Despite this criticism, sense of coherence has shown that it can be successfully operationalised using the SOC scale, a self-reported questionnaire measuring a person's ability to respond to stressful situations.

The health definition suggested by World Health Organization (WHO, 1948) Health Declaration, states that health is "*a state of complete physical, mental and social well-being and not merely the absence of disease and infirmity*", focus on the physiological and social dimensions of well-being and can be seen as a combination of the two perspectives and pertains a dialectic relation between health and disease, where one

concept need the other. The WHO definition has a positive multidimensional direction and can be seen as a start point for defining the concept quality of life (QOL) (ref). Many studies use the aspect health related quality of life (HQOL) instead of QOL. The content of HQOL relates to function and well-being in relation to disease and treatment. Quality of life is a broader concept than health related quality of life (HRQOL) and according to Bullinger (2002) the term HRQoL refers to a changed perspective on medical outcomes in relation to health and subjective well-being in relation to treatment. HRQOL is a 'multidimensional construct which cover physical, emotional, mental, social, and behavioral components of well-being and function perceived by patients and proxies (Bullinger, Schmidt, Petersen, & Group, 2002).

QOL is a concept that can be seen from many perspectives and is difficult to define and measure. Calmans (1984) definition belongs to the goal-oriented approach to QOL and states that quality of life measures the gap or the difference between an individual's hopes and expectations at a particular time point and the individual's present experiences (Calman 1984). Another perspective of QOL is the needs-based approach described by McKenna and Doward (2004). The authors define needs-bases QOL by the fact that "life derives its quality from the ability and capability of the individual to satisfy certain human needs. QOL is good when most needs are fulfilled and poor when few needs are satisfied" (Mc Kenna & Dowland, 2004). WHO defines QOL as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (WHOQOL Group, 1993, p. 153). Quality of life as a holistic concept that goes beyond activities of daily living and disease groupings and directs attention to the more complete social, psycho social and spiritual being. Higher QOL can be experienced when the gap between the person's capacity and environmental constraints are reduced. Good quality of life reflects a balance between body, mind and spirit and poor quality similarly reflects the absence of balance (Albrecht & Devlieger, 1999). Illness and dissatisfaction with life can be seen as consequences of alterations in this balance.

2.2.1 Measurement of quality of life and health

The impact from treatment on patients' quality of life is an important measure in medical interventions and health care programs. Bullinger (1997) states that use of generic measures, which measure HRQOL across health conditions, might have the disadvantages that small changes may not be detected. But use of disease-specific measures hinders comparison between illnesses. Therefore a combined approach is appropriate (Bullinger, 1997). How individuals rate their QOL is dependent on which measurement is chosen and the predefined areas included in the instrument. To examine how the individual views their QOL, it is important that the individual himself/herself are asked to define areas of importance for his/her individual QOL. One way to do this is to use the Schedule for the Evaluation of Individual Quality of Life-Direct Weighting (SEIQoL-DW).

QOL is related to health status in that it is a reflection of the way patients' perceive and react to their health status as well as to other aspect of life and the importance of health

and physical function in relation to need fulfillment (Gill & Feinstein, 1994). In a meta-analysis Smith and Avis (1999) examined if QOL can be separated from health status and found that QOL and health status are distinct constructs. When patients rated their QOL they gave greater emphasis to mental health than to physical function but in contrast when they rated their health status, physical functioning was more important than mental health (Smith, Avis, & Assmann, 1999). One effective way to measure health status is by use of standardised questions whereby the responses are scored similar to that in Short Form-36 (SF-36). The SF-36 Health Survey is an instrument that can be considered sensitive to cross-cultural differences and variations. The surveys should address general health concepts as functioning and emotional well-being in relation to physical, social and role functioning, mental health, and general health perceptions (Ware & Sherbourne, 1992).

2.2.2 Health and Quality of life among survivors

When the literature is reviewed with a focus on how young survivors of childhood cancer describe how they live their lives and what impact having had cancer has had on their quality of life, the findings are inconsistent and provide a diverse description.

A literature review of 30 empirical studies focusing on quality of life found that most survivors reported being in good health and functioning well psychologically but reported problems obtaining work, lower rates of marriage and parenthood and worrying about their reproductive capacity (Langeveld, Stam, Grootenhuis, & Last, 2002). A later study, with a large Italian cohort of long-term survivors reported high scores for both overall HQOL and each health attribute assessed (Alessi et al., 2007). In assessing whether childhood survivors differ concerning self-reported QOL and self-esteem compared to healthy peers another study report that 10.9 percent of survivors in self-reports rate their overall health as fair or poor, compared with 4.9 percent of the siblings (Hudson et al., 2003).

Despite being more likely to repeat a grade and miss school survivors were similar to their peers on most educational and occupational outcomes (Gerhardt et al., 2007). In another study young childhood cancer survivors had achieved fewer milestones than their peers or achieve the milestones at an older age than their peers (Stam et al., 2005). Young adults survivors of childhood cancer on one hand feel that the cancer experience made them different from their peers, while on the other hand, the experience made them create long lasting and stronger relationships with family and particular friends (Enskär & Berterö, 2010).

3 AIM

3.1 RATIONALE

Long-term survivors of childhood cancer are a growing population in society and therefore increased knowledge about adolescent and young adult survivors own descriptions of long-term complications is essential to meet the needs of this new group within health care.

3.2 OVERALL AIM

The overall aim for this thesis was to investigate how adolescent and young adult survivors of childhood cancer perceive that their lives are affected by having had cancer.

The specific aims were as follows:

- The aim was to gain a deeper understanding of how childhood cancer affects the lives of survivors by exploring adolescents' and young adults' views of what it is like living with this experience (Study 1).
- 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).

4 METHOD

4.1 DESIGN

The thesis includes two studies which both use a cross-sectional design. Study I employs a qualitative approach and study II is a descriptive case-control study with a quantitative approach including qualitative data.

4.2 SAMPLES

4.2.1 Sample Study I

The sample in study I was based on a national cohort including all school-aged children (7-16 years) diagnosed with cancer and who started initial cancer treatment January 2004 through May 2006. The cohort initially was investigated with a focus on school attendance, HRQOL and infections on three occasions during initial cancer treatment (N=118). Excluded were patients treated exclusively with surgery as they were not treated at the pediatric Oncology Department as well as those who underwent stem cell transplantation as their possibility to attend regular school was very small; additionally, those with insufficient fluency in Swedish were excluded. Eligible survivors from the national cohort (N=90) a median of five years (63 months) were invited to participate in the study. Fifty nine adolescents and young adults consented to participation (response rate 66 %) (Figure 1.). The median age of the informants was 17 (range 12-22).

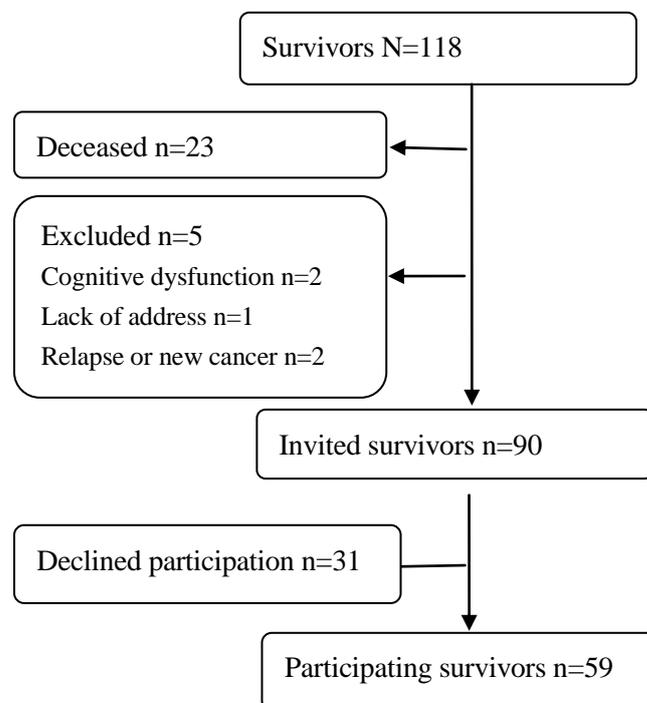


Figure 1. Overview of participants and non-participants in study I

4.2.2 Sample Study II

Study II included one group of young adult survivors of childhood cancer and a comparison group from the general population.

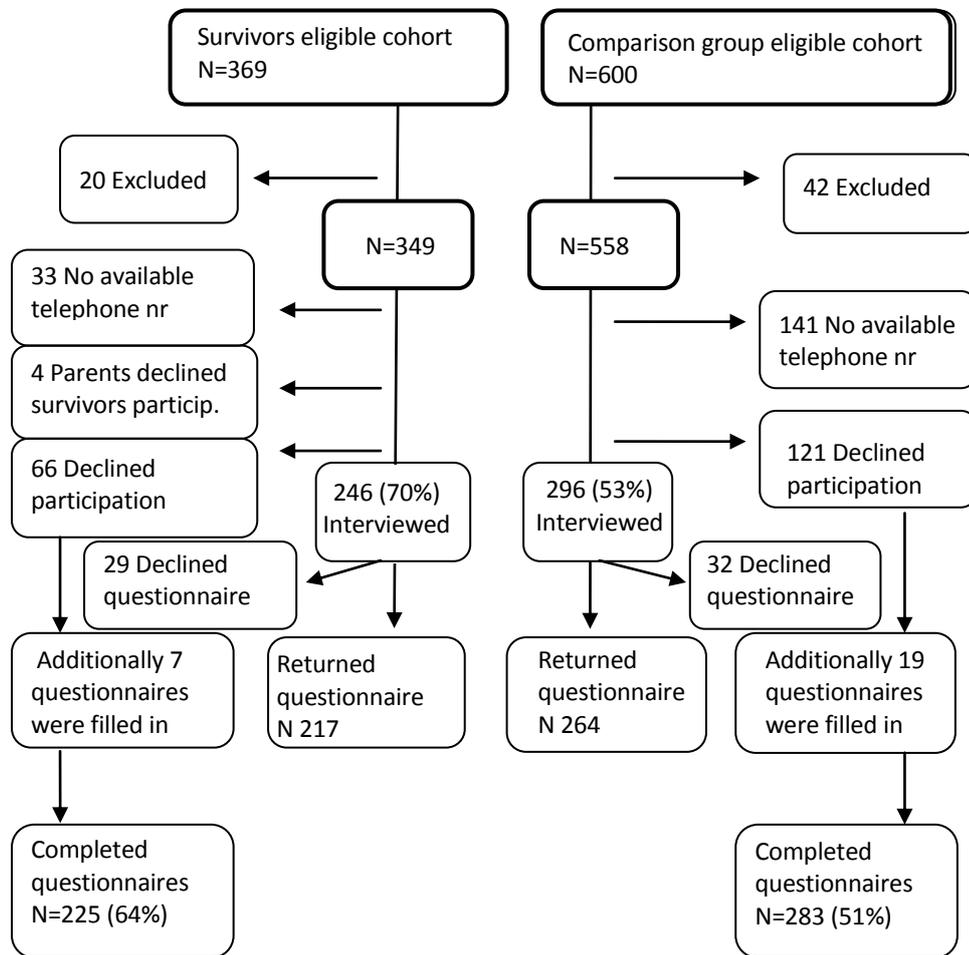


Figure 2. Participants and non-participants in study II

4.2.2.1 Long-term survivors

A cohort of 369 former patients diagnosed with childhood cancer and treated in Stockholm County during the period 1985-1999 was identified in the Swedish Childhood Cancer Registry. At the time of the study they were at least 18 years of age and at least five years beyond diagnosis. Please see figure 2 regarding participation and response rate.

4.2.2.2 Comparison group

The comparison group was randomly drawn from the Swedish population register (SPAR) among persons living in the greater Stockholm area. The sample was matched and stratified for age and gender as to resemble the participating long-term survivors; see figure 2 regarding participation and response rate.

4.3 DATA COLLECTION

The two studies included in this thesis are presented in Table 1.

Table 1. Overview of the studies, participants and methods

Study	Main content of the studies	Participants	Method of data collection/instruments	Method of data analysis
I	Descriptions of how a childhood cancer experience affects the lives of young survivors	59 adolescents and young adult childhood survivors	Telephone interviews/ semi-structured interview guide	Qualitative content analysis
II	Quality of life in relation to self-reported health status	246 long-term survivors 296 persons from the general population	Telephone interviews based on SEIQoL-DW; Standardized self-reported instrument Short Form 36 (SF-36)	Content analysis Chi-square test Student's t-test Hierarchical multiple regression

4.3.1 Data collection Study I

4.3.1.1 Telephone interviews

The data in the study were collected using telephone interviews. The interviews were based on a study specific interview guide with semi-structured questions; this is according to Patton a way to ensure that the interview is highly focused and that the interviewee gets asked the same questions (Patton, 2002). The open ended questions focused on the informants' current life situation in relation to having had childhood cancer and important areas in life (e.g., school situation/occupation, leisure activities and relation to friends). Examples of included interview questions were:

- If you think back to what your life was like while receiving treatment for cancer, what do you think of when you think back?
- How are things in school in school would you say?
- How is it now if you compare with your friends, do you think that something is different for you because you have had cancer? If you compare yourself with friends, do you think there is anything that is different for you due to having had cancer (in relation to your school situation/occupation, leisure activities, and friends)?

Throughout the development of the interview guide, the included questions/ areas were pilot tested among young adults with former childhood cancer experience, and based on

the results minor adjustments of the included questions were performed. During the interviews the semi-structured questions were followed-up on by probing questions aimed to encourage the informants to elucidate on their experiences or develop and clarify their descriptions. The interviews lasted a median of 19 minutes (range 13 to 60) minutes and were tape-recorded and transcribed verbatim.

4.3.2 Data collection Study II

4.3.2.1 Telephone interviews

Data was collected using semi-structured telephone interviews based on the interview-based instrument Schedule for the Evaluation of the Individual Quality of Life-Direct Weighting (SEIQoL-DW) (Hickey et al., 1996). During the interviews the respondents were asked the question: “If you think about your life as a whole, what are the most important areas --- both good and bad --- in your life presently that are crucial to your quality of life? The respondents were free to nominate as many areas as they wanted and were then asked to select five of the areas and rate their functioning or satisfaction with each area on a seven-point category scale. The verbal anchors of the category scale were “as bad as could possibly be” (scored 1) and “as good as could possibly be” (scored 7). Furthermore, the respondents were asked to also rate their global quality of life on the same category scale. The SEIQoL-DW instrument has been developed for assessment of individual quality of life by allowing the respondent to freely nominate the most important areas in life that are crucial for their quality of life. The SEIQoL-DW has been modified and translated to Swedish (Wettergren, Kettis-Lindblad, Sprangers, & Ring, 2009) and has been shown to be feasible and valid (Wettergren, Björkholm, & Langius-Eklöf, 2005). During the interviews with the comparison group the nominated areas/domains were written down and the telephone interviews with the long-term survivors were tape-recorded.

4.3.2.2 Standardised instrument

After completing the telephone interview respondents in both groups were sent a package of self-reported questionnaires including The Short Form-36 Health Survey (SF-36). SF-36 is a generic instrument for assessment of health status, which was constructed to capture two major dimensions of health, physical and mental health (Ware & Sherbourne, 1992). The questionnaire consists of 35 items divided into eight subscales, Physical functioning (PF), Role-physical (RP), Bodily pain (BP), General health (GH), Vitality (VT), Social functioning (SF), Role-emotional (RE) and Mental health (MH); Additionally one single item determines the perceived differences in health status over the past year. The response choices vary from two to six. For the dimensions physical and mental health respectively, based on the eight scales, two summary index scores, Physical Component Summary Score (PCS) and Mental Summary Score (MSS) are constructed. All scales influence the scores in the summary score although the PCS measures primarily the scales of PF, RP, BP and GH whereas MCS mainly contains VT, SF, RE and MH (Sullivan & Taft, 2002). The Swedish version of SF-36 has been found to be a valid and reliable instrument that has been used in a variety of populations including populations of long-term survivors of childhood cancer (Wettergren et al., 2005).

4.4 PROCEDURES

4.4.1 Procedure Study I

The data for study I was collected during the time period January to August 2009. The identified presumptive participants were sent a letter with information about the study and highlighted that the participation was voluntary and that they at any point could withdraw from the study. When the informant was aged 11-15 the letter was addressed to the parent/parents. For those aged 16-to 17, years the letter was sent directly to the informant but also contained a letter to the parents. For those aged 18-22, the letter was sent directly to the informant. Included with the letter was a form for informed consent that was obtained from all informants, and for those below the age 18 years consent was also acquired from parents. Some days after the letter was sent potential informants were contacted by telephone. If the young person agreed to participate, a suitable time was scheduled. In some cases according to the informants' choice the interview was conducted immediately. Potential participants difficult to reach were sent reminders. The telephone interviews were performed by three interviewers after training in the interview technique. After the interview, the informant was sent a cinema ticket as compensation for participating in the study.

4.4.2 Procedure Study II

The data collection for study II was conducted during the time period 2005-2007. The possible participants in both samples were approached by mail and were sent a letter with written information about the study, which stressed that participation was voluntary and confidential. One week after the letter was sent potential participants were contacted by telephone to request their participation. If the person agreed to participate, the interview according to their choice either could be conducted there and then or at a time suitable to the participant. The telephone interviews were performed according to procedures for administration of the SEIQoL-DW. The questions posed by the interviewers followed a strict order with probing questions to elucidate the answers. The tape-recorded interviews lasted a median of 10 minutes. Soon after the telephone interview the participant were sent a SF-36 questionnaire by post together with a cinema ticket as compensation for the participation. In case the questionnaire was not returned within two weeks' time a reminder was sent.

4.5 DATA ANALYSIS

4.5.1 Data analysis Study I

4.5.1.1 Qualitative content analysis

Qualitative content analysis is described in the literature as a suitable approach for drawing conclusions by systematic and descriptive identification of the evident content that is communicated in text on various abstraction levels (Patton, 2002). This qualitative content analysis approach was inspired by Graneheim's and Lundman's (2004) description of the content analysis process, and was chosen for the analysis of the verbatim transcripts derived from the telephone interviews (Graneheim &

Lundman, 2004). The method is described as useful for analysing content from interview data with a focus on people's experiences.

The analysis was carried out in the following steps. (1) All transcripts were read several times with the purpose to get an overall sense of the interview content. (2) The interviews were reread specifically to identify meaning units that described how childhood cancer affected the lives of the young survivors. (3) Each informant's total interview transcript with descriptions of impact on life was summarised. (4) When scrutinising the informants' statements the content analysis revealed three clear patterns. How the informants described how the experienced influence their current lives was divided into three separate groups. (5) Identified meaning units of each of the three groups were compared and four categories emerged revealing the differences between the three groups.

The analysis was carried out by the first author, in continuous dialogue with four of the co-authors. The analysis was an on-going process with repeated revisions and modifications until agreement was reached among the research group. Finally, to establish the credibility of the analysis we used triangulating analyst (Patton, 2002) whereby one member of the research group not previously involved in the analysis process read half of the interviews and categorised them according to the identified descriptions of the three groups (Table 3, page 28). When the new categorisation was compared with the original categorisation the agreement was 90%.

4.5.2 Data analyses Study II

4.5.2.1 SEIQoL-DW

The data collected through telephone interviews based on SEIQoL-DW were analyzed according to content analysis (Graneheim & Lundman, 2004). During the analysis process the data from the interviews with the two samples were analyzed separately. The analysis was carried out in the following steps. (1) The entire transcripts were read through several times. (2) To allow coding the interview transcripts, the written answers were divided into meaning units that reflected a content of QoL. (3) In the next step, units sharing the same or similar content were put together in sub-categories and categories. (4) The research group read the codes and preliminary sub-categories and discussed and defined boundaries and central characteristics of the categories. (5) During the process of categorisation, repeated meetings took place in which the sub-categories and categories were modified until a final agreement was reached. The analysis was validated by one member of the research group not previously involved in the categorisation process which resulted in 96% agreement for the survival group and 95% for the comparison group. The final categories derived from the content analysis showed that the most frequent mentioned areas were the same for the two groups and therefore the categories from the two samples were merged.

4.5.2.2 Statistical analysis

The statistical analysis to compare proportions of categorical variables between the two groups, long-term survivors and the comparison group was performed with Chi-square statistics. For comparison of means between groups the Student's *t*-test was used.

For statistical analysis of SF-36 each statement raw scores were coded, summed and transformed into a scale from 0 (worst possible health status) to 100 (best possible health status) following the SF-36 standard algorithms (Ware & Sherbourne, 1992). To compare SF-36 mean scores with Swedish normative data on young adults (Jörngården, Wettergen, & von Essen, 2006) a one sample *t*-test was used. By summing the ratings and dividing by the number of nominated areas (SEIQoL-DW) an overall individual QOL index was calculated. A hierarchical multiple regression analysis in two steps was performed to account for the variance in the scores of the dependent variable QOL (SEIQoL Index , overall quality of life index). In the first step, the confounders 'living alone', a living situation which is known to differ between long-term survivors and the general population, was included together with sex and group (long-term survivors vs. comparison group). In the second step the two SF-36 summary index score, physical (PCS) and mental (MCS) health were forced into the model. Impact of other confounding factors such as 'age at diagnosis', 'time since diagnosis', 'married/living with a partner', were tested in a first set of analyses but excluded from the final model as those predictors had negligible impact on the variance of quality of life. A significance level of $P < 0.05$ was applied in all analyses.

5 ETHICAL CONSIDERATIONS

Ethical approval for study I was obtained from the Regional Ethical Review board in Stockholm Dnr 03-662 (Supplement 04-208, 2009/1069-32, 2010/033-32). Study II was considered unproblematic from an ethical point of view and was not found to require ethical approval under the Act (2003:400). The Regional Ethical Review board in Uppsala gave an advisory statement about the study.

In research in which children are involved it is important to see the children as active participants in the research process in accordance with the Convention on the Rights of the Child (UNICEF Sverige, 2008). The CRC states that children have valuable knowledge to contribute and the right to be consulted in matters that involve them but that there must be a fair distribution between burdens and benefits of participation in research (Neil, 2005).

Prior to the interviews, information letters about the studies were sent to the potential participants. The letters included information about the studies with contact telephone numbers to the researchers in charge, purpose and benefits of the study, method and procedures for gathering the data, and any consequences for the individual as well as information that participation was voluntary and participants have the freedom to withdraw from research at any time. Included was also information about confidentiality and that the results would be presented in such a way that the participants' identity would be protected. Different letters were formulated to meet the parents, the young survivors (study I), the adult survivor (study II) and the comparison groups' (study II) level of comprehension. Approaching children who have had childhood cancer five years after diagnosis in interviews which the question areas focus on their previous cancer experiences, the participation might evoke distressing thoughts and feelings. Information informing that participation was voluntary and that they were free to withdraw from participations at any time without adverse consequences was given both in the information letter and in the introduction phase of the interview. At the end of the interview, a standard notice about where the participant could turn if he/she had questions after the interview was provided.

6 RESULTS

6.1 RESULTS STUDY I

The 59 participating survivors had been treated for ALL (n=20), skeletal and soft tissue sarcoma (n=13), tumors of the CNS (n=8), Hodgkin's lymphoma (n=6), non-Hodgkin's lymphoma (n=5), acute myeloid leukemia (n=3), and for other diagnoses (n=4).

What it is like to live with a childhood cancer experience

Study I describes how adolescents and young adult survivors of childhood cancer perceive that the cancer experience affects their current lives five years after diagnosis. The qualitative content analysis resulted in three clear patterns based on how the cancer experience influence on daily life was described. The three groups were: 'Feeling like anyone else', 'Feeling almost like others' and 'Feeling different'. In each group, the influence of the cancer experience was described in terms of four categories: thoughts about having had cancer, presence of complications in daily life, ability to handle complications and view of life (Table 3).

Feeling like anyone else

Forty nine percent of the informants were identified as belonging to the group "feeling like anyone else", since they reported that the cancer had almost no influence on their daily lives. Informants rarely thought about the cancer experience or perceived the thoughts as troublesome. Informants experienced minor or no complications and if they did it was often limitations related to physical performance, problems with fine motor skills, changes due to scars or having to handle on-going medication. If informants had any complications, these were easily handled and not perceived as hindering nor did take much attention in their daily life. Their view of life was rarely influenced and even though the cancer experience was a negative and undesirable event in life, the experience also had a positive effect in relation to changed priorities and values in life.

Feeling almost like others

Forty four percent of the informants were identified as belonging to the group, "feeling almost like others" since they perceived that the cancer experience influenced daily life to a small extent. They sometimes thought about having had cancer but the experience did not have a central role in their lives. Informants had complications but did not perceive this as hindering daily life. Described complications were both physical and mental complications and physical complications included excess weight, visual scars, short stature, and having prostheses or using a wheelchair for transportation. Mental complications included worries about the recurrence of cancer and concerns about changes in body appearance. They handled the physical complications and integrated them into normal life conditions, even though they had to do things differently compared to peers. Their view of life was often described as being influenced by the cancer experience. Informants reported feeling more mature and/or having a changed view about what they regarded as important in life.

Feeling different

Seven percent of the informants were identified as belonging to the group, “feeling different” since they perceived that the cancer experience affected and hindered them in daily life to a large extent. The informants thought about having had cancer frequently or all the time as there were things in their current lives that reminded them about the cancer and its on-going influence. They had complications that affected their daily lives very much. The descriptions of mental complications included worries and concerns about not yet having dealt with having had cancer. Cognitive complications (e.g., memory problems) were also reported as affecting school performance and/or impacting their ability to recall things in everyday life. The complications were described as hindering daily life and informants consciously tried to find strategies to handle the complications, but found it hard to find strategies that were adequate and helpful. Their view of life was influenced and the changes in view on life were both negative and positive. Negative consequences were: being marked for life, feelings of grief about having had cancer and having a vulnerable health condition. However, informants also reported that the cancer experience had a positive effect on their view of life, changing their values and priorities.

Table 2. Description of the characteristics of the four categories by the three identified groups

Identified Groups	Thoughts of Having Had Cancer	Presence of Complications in Daily Life	Ability to Handle Complications	View of Life
Feeling like anyone else	Only rarely think about having had cancer	Have minor or no complications from disease or treatment that influence daily life	If any complications, these are easily handled and not perceived as hindering daily life	View of life is rarely influenced
Feeling almost like others	Sometimes think about having had cancer	Have complications from disease and treatment that to a small extent influence daily life	Complications are not perceived as hindering daily life	View of life is often influenced
Feeling different	Often think about having had cancer	Have complications from disease and treatment that to a large extent influence daily life	Complications are perceived as hindering daily life	View of life is often influenced

6.2 RESULTS STUDY II

The 246 participating survivors had been treated for CNS tumors (n=59, 24%), leukemia (n=55, 23%), lymphoma (n=46, 19%) and 86 (34%) for other tumors. Other tumors included soft tissue sarcomas, malignant bone tumors, renal tumors, retinoblastoma, sympathetic nervous system, hepatic tumors, germ-cell tumors, other carcinomas and peripheral PNET.

Areas of importance for quality of life

The results from the telephone interviews with respondents from the two groups based on SEIQoL show that the nominated areas influencing quality of life were the same in both groups (Table 5.). Both groups nominated an average of four areas of importance (range 1-5).

Table 3. Differences in nominated areas being most important in life as measured with SEIQoL-DW in long term survivors (N=246) and in the control group (N=296)

Important areas	Long-term survivors	Control group	χ^2
	n (%)	n (%)	
Family life	189 (77)	198 (67)	6.2*
Relations to other people	167 (68)	179 (60)	5.2*
Work, career	113 (47)	145 (49)	
Interests, leisure activities	94 (38)	102 (34)	
Own health	64 (26)	88 (30)	7.3**
Relationship to a partner	60 (24)	66 (22)	
Studies, education	54 (22)	50 (17)	
Housing, living conditions	22 (9)	34 (11)	
Finances	17 (7)	40 (14)	5.4*
Satisfied with life and self	19 (8)	32 (11)	
Pets and animals	11 (4)	8 (3)	
Love	9 (4)	9 (3)	
Goals, new experiences	9 (4)	9 (3)	
Choice and independence	9 (4)	7 (2)	
Others health	7 (3)	23 (8)	5.6*
Available time	4 (2)	21 (7)	8.5**
Miscellaneous	8 (3)	5 (1)	

* p=<0.05

** p=<0.01

The categories 'Family life' and 'Relation to other people' were to a higher extent reported as important areas for quality of life by the survivor than by the comparison group. A smaller proportion of the survivors reported that the areas categorised as 'Own health' (survivors M 4.6, SD 1.5 vs. comparison group M 5.2, SD 1.3, P<0.01) and 'Finances' (survivors M 3.6, SD 1.0 vs. comparison group M 4.2, SD 1.0, P<0.01)

were of importance for quality of life, and they reported being less satisfied with those areas than the comparison group did.

When the overall quality of life mean score (SEIQoL Index) was calculated there were no significant differences between the scoring of long-term survivors (M=5.5, SD 0.82, range 2.3-7.0) and that of the comparison group (M=5.4, SD 0.80, range 3.0-7.0).

Reports of health status

The long-term survivors reported worse health status than the comparison group did on one of the eight multi-item scales that assess health concepts of the SF-36. In the survivor group the mean score for Role limitation-physical was significantly lower than in the comparison group (Table 6.).

Table 4. SF-36 reported in survivor group (n= 217) and control group (n=264)

	Survivors		Control	
	Mean	SD	Mean	SD
^a Physical function	92.7	14.6	95.0	11.6
Role limitation-physical	81.6	32.8	87.5*	25.4
Bodily pain	79.2	24.7	80.5	21.6
General health	72.2	22.2	75.8	18.2
Vitality	57.3	22.6	58.1	19.3
Social functioning	80.1	23.4	82.6	20.0
Role limitation-emotional	74.5	36.9	75.8	36.3
Mental health	70.0	19.3	71.4	17.5
^b Physical component scale	52.7	8.6	54.1	7.6
Mental component scale	42.3	12.0	42.8	11.5

* p-value is significant at the 0.05 level

^a Maximum score 100 equals to best possible

^b Score 50 and above equals to good functioning

Health status and socio-demographic characteristics (age, sex, and living alone) were weak predictors of overall quality of life (SEIQoL Index) as shown in the three hierarchical regression analyses (Table 7).

Table 5. Multiple regression analyses with SEIQoL Index as the dependent variable

	Survivors	Controls	Survivors and controls
Predictors	β	β	β
MCS	.397***	.328***	.363***
PCS	.110	.051	.009*
Age	.116	.210**	.177***
Sex	.253***	.073	.159***
Group			.052
Adj R ²	.223	.102	.157

β are standardized regression coefficients

* p=<0.05 ** p=<0.01

*** p=<0.000

In the first step of the regression, the influence from the socio-demographic variables on overall quality of life accounted for 6% of the variance. When the variable health status was added in the second step this gave a model in which 17% of the variance in overall quality of life was accounted for by the influence of the predictor variables.

7 DISCUSSION

The overall impression of the findings from the present thesis is that the majority of long-term survivors in both samples, despite the difference in time since diagnosis (median 5 and median 16 years), appear to get along well in life in spite of reported complications from diagnosis and treatment. This confirms findings from other studies reporting that long-term survivors experience having a favorable outcome despite the childhood cancer experience (Mattson, Ringner, Ljungman, & von Essen, 2007; Wicks & Mitchell, 2010). However, there is also a small group in the present material that perceives that complications are hindering in daily life five years after diagnosis. Consistent results were found in a study of adolescent long-term survivors, and those who experienced late effects also reported lower QOL compared with healthy controls (Bradley Eilertsen, Jozefiak, Rannestad, Indredavik, & Vik, 2012). Another study reported lower HRQL for both physical and psychosocial health in survivors compared to the general population (Speechley, Barrera, Shaw, Morrison, & Maunsell, 2006).

Even if a great majority of the participating survivors in both studies reported having complications from the childhood cancer, most of them said that the experienced complications were manageable and could be handled and therefore were not perceived as hindering in daily life. One way to explain this could be in relation to response shift, referring to the idea that individuals as a result of health alterations may undergo changes in internal standards, values or conceptualisation of QOL (Sprangers & Schwartz, 1999). According to Mallinson (2002), individuals' adaptation to physical limitations following illness and disability includes finding new ways to achieve their objectives, whereby the individual will recalibrate the judgment about the severity of the limitation (Mallinson, 2002). Another way to understand this adaptation process could be through the concept of normalisation. Results from one study showed that returning to a level of "normalcy" after completion of treatment was described as the predominant goal by young adult long-term survivors but that this "normal" was not necessarily the same normal as before diagnosis (Miedema et al., 2007). Normalisation is a process connected to reintegration to normal living with the purpose that the individual shall resume well-adjusted living following disease and are seen to contribute to quality of life. Important domains of recovery are mobility, self-care, daily activities, social activities, general coping skills, personal relationships and presentation of self to others (Wood-Dauphinee & Williams, 1987). Strategies to promote normalisation can involve a shift in thinking and to resume normal routines, previous activities and roles in life including returning to school, taking up relations with friends and leisure activities (Hilton, 1996). In this thesis, the salutogenic view of health can be used for understanding the different ways the informants handled the perceived influence from complications and to examine factors that account for health, well-being and satisfaction with life despite perceived complications and health impairments (Antonovsky, 1987). The theory of sense of coherence with the key features of comprehensibility, manageability and meaningfulness can help us to understand how individuals can perceive health and well-being despite experiencing complications from childhood cancer.

There was also a clear pattern within the material the informants' descriptions of the influence of complications in daily life differing among participants. Despite the fact that the great majority of participants in both samples included in the present thesis reported no or minor influences of complications, there were also long-term survivors

who reported having physical complications that to a large extent were perceived as affecting and hindering their daily life. Impact on daily activities was also reflected in that adult long-term survivors rated significantly lower in the SF-36 scale Role limitation-physical, which was the only scale on which survivors differed from the comparison group. A low rating in the SF-36 scale reflects problems performing work and other every day activities owing to physical health. This is in line with findings from other research that has used the same instrument in studies with long-term survivors (Pemberger et al., 2005).

Long-term complications following childhood cancer include limitations in physical performance that may have an impact on the individual's ability to participate in activities in daily life. When long-term survivors' ability to perform physical activities in daily life were studied in a cohort of survivors and compared with a group of siblings, the survivors were more likely to report physical limitations than did siblings (Ness et al., 2005).

It is unknown whether long-term survivors who report an impact on physical activity due to complications actually have more severe complications or if the complications are perceived as hard to handle. Seen from the perspective of the sense of coherence, this can be understood as key feature of manageability and the individuals' perception about his/her own capability to deal with demands and challenges in life (Antonovsky, 1987). The long-term survivors who reported having physical complications that they perceived to a large extent to be affecting and hindering their daily life may not perceive that they have sufficient resources to manage their complications and successfully integrate them into their daily life. To support those long-term survivors, follow-up services with a risk-based approach including health monitoring and prevention may facilitate the normalisation process and potential to integrate complications into daily life (Edgar et al., 2012; Hilton, 1996).

In both studies included in this thesis, the findings show that most of the long-term survivors are doing fine, but some are not. Even though adult long-term survivors and a matched sample from the general population rated their overall quality of life similarly, the survivors rated less satisfaction with their health than the comparison group (study II). According to the holistic health definition, to be in a state of health can be when the individual experiences well-being and is able to achieve life goals in spite of perceived complications from childhood cancer (Dahlberg & Segesten, 2010). The fact that the long-term survivors were less satisfied with their health may be an expression that they perceived that their health following childhood cancer was hindering in relation to achieving their life goals. The individuals' views of health, well-being and life satisfaction are often conflicting with their objective health status and health barriers (Drew, 2007).

There were descriptions from a majority of young long-term survivors about a changed view of life, with reports about a change in values that influence priorities and areas regarded as important in life as well as increased maturity following the childhood cancer experience. One described impact on the view on life was that the informants perceived having higher demands on relations to others. Enskär and Berterö (2010) found that the young adults survivors after childhood cancer on one hand felt that the cancer experience made them different from their peers but on the other hand the experience made them create long lasting and stronger relationships with family and particular friends (Enskär & Berterö, 2010). The long-term survivors prioritise "family life" and "relations to others" as areas of importance for QOL to a higher extent than

the comparison group. Also in earlier studies these areas been reported for quality of life among adult long-term survivors (Wettergren, Björkholm, Axdorph, Bowling, & Langius-Eklöf, 2003).

The positive impact on view of life after childhood cancer can be understood as a way to find meaning from the experience as a part of life. This is consistent with the SOC component 'meaningfulness' in which finding a meaning in things that happen in life may help the long-term survivor to get control and move on in life to regain health (Antonovsky, 1987; Flensburg-Madsen, Ventegodt, & Merrick, 2005). However, in the present study, there were also informants who reported negative consequences on their view of life, like being marked for life and feelings of grief about having had cancer. For some young long-term survivors this negative effect was still too strong for them to handle, and they had difficulties handling and making sense of their cancer experience in daily life.

7.1 METHODOLOGICAL CONSIDERATIONS

In this thesis the long-term survivors view have been investigated using different approaches, both qualitative and quantitative, which increases the possibility to investigating survivorship from different angles.

7.1.1 Study I

The trustworthiness of a qualitative study can be considered from the concepts of credibility, dependability and transferability (Graneheim & Lundman, 2004). The credibility of the findings in the present study relies on the selection of the sample, the data collection and the analysis procedures. The sample selection is considered to be strong, since all informants belonged to a national cohort that included variations in age, gender and residence in both urban and rural areas, and is representative of the most common childhood cancer diagnoses among school-aged children in Sweden.

Another strength is that this sample represents the view of the young survivors themselves, rather than reporting from parents or healthcare professionals. A response rate of 66% must be considered as acceptable, however the reasons for non-participation are unknown and it is therefore hard to determine whether non-participants experienced no, small or large effects from the childhood cancer experience. Telephone interviews were used for geographical reasons and because they were suitable for this age group of informants, who seemed to be at ease with sharing their experiences in the relative privacy of a telephone call. However, a limitation was that the younger informants more often gave short answers while the older informants gave more detailed descriptions. Our judgment is that qualitative content analysis was a suitable method for gaining a deeper understanding of adolescents and young adults' view of what it is like to live with this experience, and the inter-rater reliability of 90% ensures the credibility of the analysis.

The dependability of the findings in the present study is confirmed by the fact that data was collected in the same way from every informant. An interview guide with open-ended questions was used and the interviewers had special training in conducting

telephone interviews. During the analysis process, quality was ensured by continual discussions among the co-authors.

The transferability of the findings in the present study is dependent upon a good description of the study's context, selection and characteristics of the participants, data collection, the process of analysis and an in-depth presentation of the findings. I can be assumed that the results of the present study could be transferred to other groups of young people who are living with long-term health conditions.

Our pre-understanding in this thesis concerning the situation of young long-term cancer survivors was guided from earlier research findings on how having had childhood cancer may have a long-term impact on survivors daily lives five years post diagnosis (Drew, 2007). In study I, the research question pointed out that our interest was on how the childhood cancer experience influenced current life, and this could implicitly be seen that the area under examination was related to experience consequences on health following childhood cancer. Even though this did not directly focus on how the young long-term survivors perceived their health status, their answers often seemed to take stance from how they experienced that their current health were influenced in daily life.

7.1.2 Study II

The high non-response rate in the comparison group should be regarded as a risk for selection bias. In relation to official statistics for the general population in Stockholm County, participants in the comparison group seemed to be better educated and more "socially stable" than average. Thus, selection bias may account for some of the differences between the present survivor and comparison groups, which calls for caution when drawing conclusions. However, when comparing health status between the survivors and recent Swedish normative data in one age group (age 20-23) ²³ we found no discrepancies with our present results (data not shown). Another limitation of the study is the difficulty in determining the health status of the 30% of survivors who did not participate. Nevertheless, the fact that no clinical differences were detected between the participating and the nonparticipating survivors indicates that health status should be the same in both groups. In study II, the SEIQoL-DW was used to examine important areas in relation to quality of life.

8 CONCLUSION

Although the results from this thesis show an overall positive picture of the long-term survivors' current life situation, but it is also important to take into consideration how the daily life may be facilitated for those long-term survivors who perceive that their daily life is impacted from having had childhood cancer.

The results stress the importance of understanding that long-term survivors can perceive long-term complications from disease and treatment, which for a long time will have an impact on their daily life. The difference in time since diagnosis between the two studied samples illustrates the impact from disease and treatment from five up to sixteen years after cancer diagnosis, and gives a broad picture of how life as a long-term survivor is perceived. The results from study I, in which the young survivors provide their own descriptions of how having had cancer during childhood influences daily life, supplement and give more nuanced descriptions of the results presented in study II.

9 IMPLICATIONS

Contemporary knowledge in the society about the situation following childhood cancer may still be built on the image that childhood cancer is equal to a deadly disease, and not up-dated with knowledge of the high survival rates due to improved treatment modalities. To improve the situation for long-term survivors, it is important to highlight the fact that there can be a knowledge gap in society about the life span of this growing and “new” group of long-term survivors. Patient associations can be a way to take care of/ monitor the interest of long-term survivors and to spread knowledge in society of their current and future health care needs following the complication from childhood cancer. There are discussions in the literature concerning what is to be seen as suitable standard/model for follow-up for long-term survivors. There will be a stage in the young survivors’ life when they will be transferred from pediatric care to the adult oncology service, often considered to be more anonymous and with a different care culture. This can be experienced as a big change from having ongoing relations with the staff in the pediatric department, in a safe and well-known care culture and environment in which the young survivor has been taken care of for a long time period. There are alternative forms for follow-up whereby the young person is transferred to a special unit, specializing in long-term survivors and the focus can be more holistic and on different dimensions such as physical, mental, and psycho social. The individuals’ knowledge about risk for developing long-term conditions following the cancer diagnosis and treatment is one important area to highlight during follow-up. This includes increasing early detection by taking preventive actions. Coping styles can be learned and adjusted to be productive and supportive so that the individual influence from perceived complications in daily life can be minimized. This is an important target for follow-up to detect long-term survivors in need of individual support. Furthermore, it is important to transfer knowledge to the primary care settlement about preventive care to minimise the risk of chronic health conditions. Primary care will often be the first contact when long-term conditions occur, or in situations related to identified fertility problems. Knowledge should be acquired and precautions taken to identify those at risk for perceiving that the childhood cancer experience is hindering them in their daily life.

In line with previous reports, the findings in the present study show that most of the adolescents and young adults appeared to get along well in daily life, although many informants reported that life was to some extent affected by having had childhood cancer. However, a small group of survivors were troubled in daily life and would benefit from support: this would help them to mobilise the resources needed to manage their daily living situations. Follow-up care in which nurses and other health care professionals can identify those young survivors of childhood cancer that have trouble with daily life and offer them targeted support is needed (Friedman, Freyer, & Levitt, 2006). By using the salutogenic health model and structured dialogues in health care, it may be possible to identify difficulties in relation to having had cancer among young survivors. There are other studies (Griffiths, Ryan, & Foster, 2011) that have concluded that SOC theory can be used to understand how people reflect on how they cope with problems in their everyday lives. Sarenmalm et al. (2013) suggest that SOC could be useful as a tool for identifying individuals in need of support to cope with breast cancer (Sarenmalm, Browall, Persson, Fall-Dickson, & Gaston-Johansson, 2013). Strengthening existing resources and identification of new resources could help young survivors adapt to a changed life following childhood cancer. The chance to talk about

the experience with health professionals and peers who also have survived childhood cancer is a way to do this, since studies show that this increases the informants' knowledge about the disease and helps mobilise coping resources (Patterson, Millar, Desille, & Mc Donald, 2012). Further, the survivors may also benefit from improved help from health professionals in terms of their medical complications (such as pain and handling long-term medication regimens).

Clinicians must be aware that long-term survivors of childhood cancer may have different expectations and goals with respect to their current health status than clinicians do, and that specific questions may need to be asked to elicit important issues. Using a tool that identifies what is considered important in life in addition to regular follow-up may be helpful in establishing the individual's priorities and health promoting activities for the achievement of a good quality of life.

10 SUMMARY IN SWEDISH – SAMMANFATTNING

Unga personer som har genomgått behandling för cancer under barndomen löper stor risk att utveckla långvariga hälsoproblem efter genomgången behandling. Hur en cancererfarenhet inverkar på ungdomars och unga vuxnas liv i ett längre perspektiv har endast studerats i begränsad omfattning. Det övergripande syftet för med denna avhandling var därför att undersöka hur ungdomars och unga vuxnas liv är påverkade av att ha haft cancer i barndomen.

I de två studierna som ingår i avhandlingen användes en tvärsnittsdesign; data har insamlats med intervjuer och ett frågeformulär. I delstudie I var syftet att undersöka hur cancersjukdom i barndomen inverkar på ungdomar och unga vuxnas livssituation 5 år (63 månader) efter diagnos. Data insamlades genom telefonintervjuer med 59 unga personer (min 11 år-max 22 år) som ingick i en nationell kohort av skolbarn som diagnostiserats med cancer i barndomen under en tvåårsperiod; svarsfrekvens 66%.

Telefonintervjuerna utgick från en studiespecifik intervjuguide med frågor om nuvarande livssituation, skolsituation/arbetsituation, fritid och kamratrelationer. Intervjuerna bandinspelades och transkriberades för att sedan analyseras med kvalitativ innehållsanalys. I analysen identifierades tre grupper utgående från hur informanterna beskrev att deras dagliga liv var påverkat av cancererfarenheten: ”Känna sig som alla andra” där cancererfarenheten nästan inte alls beskrevs ha någon inverkan (49%), ”Känna sig nästan som andra” då cancererfarenheten beskrevs ha viss men begränsad inverkan (44%) och gruppen ”Känna sig annorlunda” (7%) där informanterna beskrev att cancererfarenheten hade stor inverkan på livet.

I delstudie II genomfördes telefonintervjuer med en kohort av 246 unga vuxna (min 18 år-max 37 år) (18-37 år) som hade diagnostiserats för cancer i barndomen en mediantid av 16 år (min år-max år) tidigare; svarsfrekvens 64%. En jämförelsegrupp ingick också och bestod av 296 unga vuxna slumpvis utvalda ur allmänheten i Stockholms län som identifierades via Statens personadressregister (SPAR); svarsfrekvens 51%. I strukturerade telefonintervjuer baserade på instrumentet The Schedule for Evaluation of Individual Quality of Life -Direct Weighting (SEQIoL-DW) tillfrågades alla deltagare om sin livskvalitet. Respondenterna uppmanades nominera viktiga områden i livet som de ansåg ha betydelse för sin egen livskvalitet och skatta sin tillfredsställelse med dessa områden på en sjugradig skala. Efter intervjun tillsändes respondenterna ett frågeformulär (Short Form-36) för att mäta hälsostatus. Resultatet visade att de unga vuxna med cancererfarenhet skattade sin övergripande livskvalitet likvärdig med jämförelsegruppen, inte heller de områden som angavs som viktiga i livet (familj, relationer till andra människor, arbete och karriär, intressen och fritid) skiljde sig mellan grupperna. Vid skattningen av hälsostatus skiljde sig grupperna i endast en av de åtta delskalorna, de unga vuxna med cancererfarenhet skattade mera problem med dagliga aktiviteter till följd av fysisk hälsa än jämförelsegruppen.

Resultaten från båda studierna visade att majoriteten av ungdomarna och de unga vuxna som hade behandlats för cancer i barndomen ansåg att cancererfarenheten hade liten inverkan på dagligt liv och rapporterade en god livskvalitet. Även om det övergripande resultatet indikerar att unga personer med cancererfarenhet har en hanterbar livssituation trots hälsohinder så är det viktigt att identifiera dem som beskriver problem i livet relaterat till den cancersjukdom de har behandlats för.

Sjukvården skall kunna erbjuda uppföljningsprogram med individualiserad långtidsuppföljning med fokus på individens egen upplevelse av hälsa där olika professioner inom vården samarbetar i syfte att stödja och stärka individens resurser och förmåga att hantera konsekvenser av cancererfarenheten i det dagliga livet.

11 TACK TILL...

Alla ungdomar och unga vuxna som deltagit i studierna och tagit sig tid att svara på alla frågor. Tack för att ni så generöst låtit mig ta del av de erfarenheter ni har med er i livet. Jag hoppas att min avhandling kan bidra med ny kunskap om hur det är att ha genomgått cancer i barndomen.

Min huvudhandledare Lena Wettergren som tog mig med på resan genom forskarutbildningen. Tack för att du alltid funnits där för mig och så generöst delat med dig av din kunskap. Ibland har det känts trögt och svårt att ta nästa steg men då har vi tillsammans provat nya vägar, och jag kom ju faktiskt fram till slut.

Mina bihandledare, Lars Eriksson och Claudia Lampic för vänligt stöd och uppmuntran och för att ni alltid haft tid att läsa mina manus och återkopplat med konstruktiv kritik. Våra samtal kring forskningen har fördjupat min förståelse för forskningsprocessens olika delar.

Alla mina medförfattare till publikationerna. Tack Jeanette Winterling för bra samarbete med stimulerande och kreativa Skype möten. Tack Anneli Silvéen Hagström för bra synpunkter. Min externa mentor Helena Hemmingsson.

Forskargruppen HIP-LC: Anna Jervaeus, Lise-Lott Rydström, Maria Wiklander, Jenny Nilsson, Maria Lindberg och Gabriela Armuand för gemenskap och stöd.

Mina ”forskarkompisar” Kay Sundberg, Kaisa Fritzell och Margareta af Sandeberg för att vi blivit så goda vänner och är så bra på att hitta på roliga upptåg.

Min vän i nöden Monir Mazaheri, vad hade jag tagit mig till i sista minuten utan dig?

Carol Tishelman och alla min ”gamla” forskarkompisar i ”Lungcancergruppen”, min introduktion till forskningens värld.

Eva-Lisa Lundgren och Eva Broberger, mina gamla lärare som gav mig chansen att bli lärare, jag är kvar än... Mina vänner i bokklubben och ”syjuntan” för trevlig samvaro och samtal om allt mellan himmel och jord.

Alla mina kollegor och vänner och på sektionen för omvårdnad, ni som bidrar till att skapa en bra och trevlig arbetsplats.

Min chef Eva Johansson för stöd och uppmuntran i slutfasen av arbetet med avhandlingen.

Institutionen NVS med Prefekt Kerstin Tham som gav mig möjlighet att inleda forskarutbildningen genom att tilldela mig strategiska medel för lärares kompetensutveckling.

Till Barncancerfonden som bidragit till finansieringen av mitt avhandlingsarbete.

Min ”lilla” familj, Magnus, Filippa och Mattis som stått ut med jag alltid haft mycket på gång. Nu kanske jag äntligen kan svara på frågan: vad håller du på med egentligen?

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