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# **BEING YOUNG WITH A CANCER EXPERIENCE**

**Health-related quality of life with  
special focus on independence  
and sexuality**

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*“It’s difficult to make a long cancer story short...”*  
Young woman treated for Hodgkin’s lymphoma



## ABSTRACT

The increased survival rates for persons with a cancer experience from childhood imply that health care professionals working in different areas of the health care sector will encounter young persons with a cancer experience. It is therefore of importance to investigate perceptions of health and life situation including health-related quality of life (HRQoL) among young persons with such experience. Previous research in the area shows a somewhat varying picture and independence and sexuality have not been studied to a large extent, still aspects relevant for young people. The overall aim for this thesis was to investigate self-reported HRQoL with special focus on independence and sexuality among young persons treated for cancer during childhood.

Study I is a methodological study, studies II-IV primarily have a cross-sectional design while study III also include longitudinal data. Two samples of survivors after childhood cancer were included, for studies I-III a national Swedish cohort consisting of persons 4-6 years after diagnosis (n=63, aged 12-22) and for study IV, a sample of adolescents and young adults ( $\geq 5$  years after diagnosis) (n=133, aged 16-25). An age-matched comparison group was included in studies I-III (n=257). In study I, a Rasch-analysis was performed to assess the psychometric properties of the instrument KIDSCREEN-27, studies II-III had a quantitative approach and were based on semi-structured interviews and self-reported questionnaires and study IV on written online focus group discussions which were analysed with qualitative inductive content analysis.

The psychometric evaluation of KIDSCREEN-27 (study I) indicated that the instrument is acceptable to use among young persons with a cancer experience. Further, the overall results from study II and III revealed that the majority of survivors report satisfactory levels of HRQoL, similar or higher than a comparison group. A small group reported lower levels of HRQoL which corresponded to findings from the semi-structured interviews (study II). The survivors rated their independence as higher 5 years after diagnosis compared to the time at diagnosis and higher compared to the comparison group (study III). The analysis of the written online focus groups (study IV) resulted in one main category, *Sex as a given part of life*, with four generic categories: *Sex considered to be good*, *Feeling insecure and falling behind*, *Relating sex to a stable relationship* and *Physical concerns*. The majority had positive experiences of sex without any experiences of sexual problems. However, thoughts and worries included limited sexual experiences, feeling insecure, being behind peers and physical concerns such as vaginal dryness and erectile dysfunction. Experiences and concerns were sometimes related to the cancer experience but most often not.

The findings of the thesis illustrate that the majority of those with a cancer experience from childhood get along well in terms of self-reported HRQoL focusing on independence and sexuality. However, a small group seem to be in need of extra support, displayed by lower ratings of HRQoL and sexual matters such as worrying thoughts and physical concerns. Registered nurses are suggested to take an active role in the care of young persons with a cancer experience. By tailoring the follow-up care in concordance to the needs and desires from these persons, the health and life situation for this group can be improved. Continued research, in a longer time perspective, is suggested in the areas of sexuality and sexual function.

*Key words:* childhood cancer survivors, self-reports, health-related quality of life, independence, sexuality



## LIST OF PUBLICATIONS

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

- I. Jervaeus A, Kottorp A, Wettergren L (2013).  
Psychometric properties of KIDSCREEN-27 among childhood cancer survivors and age matched peers: a Rasch analysis.  
*Health and Quality of Life Outcomes*, 11(1), 96.
- II. Jervaeus A, Lampic, C, Johansson E, Malmros J, Wettergren L (2014).  
Clinical significance in self-rated HRQoL among survivors after childhood cancer- demonstrated by anchor-based thresholds.  
*Acta Oncologica*; 53(4), 486-492.
- III. Jervaeus A, af Sandeberg M, Johansson E, Wettergren L (2014).  
Survivors of childhood cancer report high levels of independence five years after diagnosis.  
*Journal of Pediatric Oncology Nursing [Epub ahead of print]; Mar 27.*
- IV. Jervaeus A, Nilsson, J, Eriksson, L.E., Lampic C, Widmark C, Wettergren L.  
Sex is something natural, important and taken for granted in life- findings from online focus group discussions with childhood cancer survivors.  
*manuscript*

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## LIST OF ABBREVIATIONS

ALL	Acute Lymphoblastic Leukaemia
CNS	Central Nervous System
DIF	Differential Item Functioning
DCGM-37	DISABKIDS Chronic Generic Module
ES	Effect Size
HRQoL	Health-related Quality of Life
MnSq	Mean Square
NHL	Non Hodgkin´s Lymphoma
NOPHO	Nordic Society of Pediatric Haematology and Oncology
PCA	Principal Component Analysis
PCM	Partial Credit Model
SD	Standard Deviation
SF-36	Short-Form 36
SPAR	Swedish Population Register [Statens Person- och Adressregister]
SPSS	Statistical Package for the Social Sciences
STI	Sexual Transmitted Infection
VIF	Variance Inflation Factor
WHO	World Health Organization
QoL	Quality of Life

# 1 INTRODUCTION

I have been strongly engaged with the area of cancer care since I became a registered nurse in 2003. I have worked with adult cancer patients in different contexts, including a ward, a day-care unit and as a research nurse. The period, during in which I worked in this last capacity, interested me in research. Therefore, after completing my master's education, I applied to the Doctoral School of Health Care Sciences, to projects related to cancer care, and luckily was accepted. It has been truly interesting and indeed a privilege to gain deeper knowledge and understanding during this educational period regarding persons with an experience of cancer during childhood and their situation, especially because I have no experience working with this patient group. I hope this thesis can contribute to the existing research, to students at different levels within care sciences and medicine, to health care professionals in general and registered nurses and nursing staff in particular, as well as to cancer patients and persons with a cancer experience.

## 2 BACKGROUND

### 2.1 BEING DIAGNOSED WITH CANCER AS A CHILD

When a child is diagnosed with cancer, the life situation of that child as well as that of the entire family including parents, grandparents and siblings changes dramatically (Neu, Matthews, & King, 2014). Frequent and sometimes long hospital visits for both the child and the parents might become a reality, and there they encounter many new people and a totally new environment. The hospital visits often include troublesome treatment periods and diagnostic procedures for the child. In addition the parent/parents might need to take parental leave from work so as to be with the child, and this may cause economic strain for the whole family.

Interviews with newly diagnosed children (aged 1-6 years) and their parents (Darcy, Knutsson, Huus, & Enskär, 2014), have revealed feelings of strangeness for both the child and the parent, of a loss of power and of isolation. The children expressed wanting their parents to be protectors, seeing to their comfort and safety, and wanted the health care professionals to treat them with respect, e.g. helping them feel included and involved in their care; this was especially true of nurses (Darcy, et al., 2014). In a study that also included older children and adolescents (aged 4-19 years), similar important aspects were described, including a familiar environment and support from parents (Gibson, Aldiss, Horstman, Kumpunen, & Richardson, 2010). Further, children worry about symptoms to become permanent and older children (6-12 years) feel unsatisfied when parents lead the communication with health care staff, suggesting that young people, having achieved increased autonomy (13-19 years) should take the lead in communicating directly with the health care professionals (Gibson, et al., 2010).

Distressful events related to diagnosis and treatment has been investigated through interviews with newly diagnosed children and adolescents (0-19 years), parents and nurses (Hedström, Haglund, Skolin, & von Essen, 2003). Among those aged 13 and up, the most frequently mentioned aspect related to physical distress was nausea and the most mentioned aspect related to emotional distress was changed appearance (Hedström, et al., 2003). In interviews with adolescents treated for different cancer diagnoses, the negative experiences that were described included fear of alienation, changed appearance, fear of dying, and physical concerns. However, positive experiences were also described such as good relationships with health care professionals and being well cared for (Hedström, Skolin, & von Essen, 2004).

Parents are the ones giving the most important support to the child, but at the same time they are struggling with their own worries and anxiety. In interviews, parents of newly diagnosed children described feelings of powerlessness, inadequacy, and constant worry and as living with shock and trauma (Darcy, et al., 2014) but they also attempted foster hope by accepting reality, having control, reorganising their hopes and thinking positively (Bally et al., 2013). Furthermore, the situation for the siblings can be difficult characterised by feelings of worry, anticipatory grief, concerns about the loss of normal life for the sick sibling and being forgotten and unimportant in the family (Jenholt Nölbris, Enskär, & Hellström, 2013).

The cancer treatments for children and for adults differ in that children have many more years to live, compared to an adult diagnosed at an older age. Children are growing, and normal development, including puberty and fertility concerns, must be taken into account when designing treatment (Childhood Cancer Foundation, 2009). Children and adolescents with cancer should be encouraged to continue school (af Sandeberg, Wettergren, Björk, Arvidson, & Johansson, 2013) and every-day-life activities, including meeting friends, separating from parents, and developing relationships with boyfriends and girlfriends (Childhood Cancer Foundation, 2009; Epelman, 2013; Hedström, et al., 2004). Research has shown that school attendance during initial cancer treatment does not seem to be associated with getting an infection that requires treatment (af Sandeberg, et al., 2013). Health care professionals must take all these aspects into account and organise their work in concordance to the individual patient so that normal routines can continue despite the cancer diagnosis (Childhood Cancer Foundation, 2009).

In Sweden, children and adolescents (aged 0-18 years) are diagnosed and mainly treated at one of the six Paediatric Oncology Centres in Umeå, Uppsala, Stockholm, Linköping, Gothenburg and Lund. Nowadays, care takes place more and more at outpatient clinics why time spent at hospital is generally less than before (af Sandeberg, 2011). However, children and adolescents still spend a great deal of time at hospital for intensive treatment periods, including care for treatment complications, and also because certain procedures and examinations must be performed.

When cancer treatment is over, the reality is to re-enter everyday life, which might be troublesome (Hauken, Larsen, & Holsen, 2013). Young persons, 24-35 years of age who had finished treatment within the latest five years, described in interviews that they were unprepared to re-enter everyday life and that meeting reality was hard. Their descriptions included lack of information about late effects, how to handle those and limited understanding from their surrounding networks, as well as from health care professionals conducting follow-ups (Hauken, et al., 2013). Meeting reality can also include the transition from the paediatric care setting to an adult care setting, something which can be experienced very differently among individuals and include many perspectives and dimensions (McCann, Kearney, & Wengström, 2013).

Cancer survivorship can be diverted into three phases being the acute (from diagnosis to end of initial treatment), the extended (from end of initial treatment including remission, follow-ups and more treatment if needed) and the permanent (from extended survival when probability of relapse is sufficiently low) (Aziz & Rowland, 2003). Focus in this thesis is primarily on persons being five years or more beyond their primary cancer diagnosis, encompassing the concept of permanent survival. Survivorship research focuses on the health and life of the person with a cancer experience and the aim is to build a knowledge base to be able to design the best follow-up care for persons with a cancer experience (Aziz & Rowland, 2003).

## **2.2 CHILDHOOD CANCER**

### **2.2.1 Incidence**

The cause of childhood cancer is mainly unknown, although genetic factors exist, such as for hereditary retinoblastoma. Internationally, the incidence of childhood cancer is reported to be highest in the Nordic countries, probably because these countries employ a precise registration of all cases. The careful registration and follow-up of the treatment given have led to that the Nordic countries have one of the best treatment results from an international perspective (Gustafsson, Kogner, & Heyman, 2013; Kogner et al., 2008). According to the Nordic Society of Paediatric Haematology and Oncology (NOPHO) report (2011), the largest diagnostic groups among children with cancer in the Nordic countries (cases and incidence during the period 2006-2010) are leukaemia, tumours of the central nervous system (CNS), lymphomas, soft-tissue sarcomas, renal tumours, sympathetic nervous system tumours and malignant bone tumours. Leukaemia and tumours of the CNS have remained the two largest diagnostic groups since the mid 1980s in the Nordic countries (Nordic Society of Pediatric and Haematology and Oncology, 2011).

In Sweden, the diagnostic distribution among those diagnosed from 1984 to 2010 follows the Nordic pattern; that is, 70% of diagnoses consist of leukaemia, tumours of the CNS and lymphomas (Gustafsson, et al., 2013). During this period, 7065 children under 15 years old (the majority are diagnosed before age 15), were diagnosed, corresponding to an annual incidence of 16.0 / 100 000 children, or 300 children per year. The incidence did not increase significantly during this period. Regarding age distribution, acute lymphoblastic leukaemia (ALL) has an incidence peak at ages 2-4 years, while tumours of the CNS have a more even distribution. Neuroblastoma is most common among those under one year of age, germ cells tumours mostly occur during the first years of life and among teenagers and bone tumours mostly affect adolescents. Childhood cancer (all malignancies) is more common among boys (ratio 1.17:1) and has an age peak around 5-6 at diagnosis. The prognosis is similar for boys and girls and for different age groups, except for those aged 10 and over, for whom the prognosis is worse (Gustafsson, et al., 2013).

### **2.2.2 Treatment**

An increased improvement has been seen regarding the outcome for all childhood cancer diagnoses (Gustafsson, et al., 2013). The most important change happened gradually in the 1970s and 1980s when treatments were intensified with combinations of radiotherapy, surgery and chemotherapy, according to new treatment protocols (Gustafsson, et al., 2013). In addition, the use of immune-based therapy can nowadays be beneficial in treating certain types of leukaemia in childhood (Shah, Dave, & Wayne, 2013); for instance, monoclonal antibodies have been evaluated for use in paediatric non-Hodgkin lymphoma (NHL) patients (Meinhardt et al., 2010). Although most types of childhood cancer can be treated and cured today with chemotherapy, often in combination with surgery and/or radiotherapy (Kogner, et al., 2008), cancer remains the most common cause of death due to disease, for children over one year old (Gustafsson, et al., 2013). Close collaboration both nationally and internationally

regarding development and follow-up on treatment protocols has contributed to positive development for children diagnosed with cancer (Kogner, et al., 2008).

Surgery is the oldest treatment type and solid tumours are often treated surgically, usually preceded by or in combination with other treatments such as chemotherapy and radiotherapy (Sjödahl, 2008). Chemotherapy impedes and eliminates both normal and cancer cells leading to positive effects on the cancer itself, but at the same time it causes side effects which are troublesome for the patient including nausea and vomiting, effects on muscles and nerves, alopecia, skin problems, effects on mouth and throat, diarrhoea, obstipation and other side effects affecting various organ systems both short- and long term (Hansson, Henriksson, & Peterson, 2008). Radiotherapy aims a certain amount of radiation towards the tumour in order to achieve cure or symptom relief (Zackrisson & Turesson, 2008). Both normal and malignant cells are damaged and destroyed, meaning that side effects are related to the radiation on the normal cells in the radiation field (Spreadborough & Read, 2000). Cranial radiation for children is problematic because development – in particular the myelination - of the CNS is not completed and cognitive ability is thus more negatively affected by radiation than that of adults (Bergenheim & Henriksson, 2008).

Hospitalised children and adolescents (with different cancer diagnoses and admitted to hospital for treatment, fever or for other reasons), who were asked about their most prevalent physical symptoms reported nausea, fatigue, lower appetite, pain, and a feeling of drowsiness. Furthermore the most bothersome, distressful, or severe symptoms for them were, nausea and pain, “less hair than usual” and vomiting (Miller, Jacob, & Hockenberry, 2011).

### **2.2.3 Survival**

The overall survival probability by ten years for all childhood malignancies is about 80% today and the survival rate has improved since 1990. According to the report (Gustafsson, et al., 2013), during the last 15 years survival seems to have reached a plateau, and new strategies are needed to move beyond this. The pattern for tumours of the CNS and neuroblastoma appears to be a bit different; improvements have been marked during the recent decades owing to better management, diagnostic measures, and improved and intensified treatment (Gustafsson, et al., 2013).

### **2.2.4 Late effects**

Given the increased number of persons in society, who have survived their cancer during childhood or adolescence, there are also a large proportion of those, around 75% according to the literature, who will experience late effects that is, effects mainly of treatment-related toxicity or complications from the cancer itself (Haddy & Haddy, 2010). Late effects can be assessed or diagnosed through clinical examination, blood tests, radiological examinations and other tests such as neurologic examinations and psychiatric tests (Haddy & Haddy, 2010). The occurrence of the majority of late effects increases with age, and often they become clinically apparent decades after treatment (Oeffinger & Hudson, 2004). With regard to different treatment types, late effects can affect a variety of organs and systems. Selected late effects of radiotherapy are for cranial radiation: neurocognitive impairments, growth hormone deficiencies, seizures

and strokes. For radiation to the chest or mantle region: secondary cancers such as breast and lung cancer, pulmonary disease and cardiac disease. For abdominal and pelvic radiotherapy: chronic enteritis, gastrointestinal malignancy, renal insufficiency and gonadal dysfunction (ovarian failure and testicular failure). Selected late effects from chemotherapy include hypogonadism, infertility, early menopause (alkylating agents), secondary malignancies, pulmonary fibrosis, bladder disease, hearing loss, tinnitus, neurocognitive impairments and cardiomyopathy (Oeffinger & Hudson, 2004). Using clinical criteria, the prevalence of certain late effects after treatment (chemotherapy, radiation) has been shown to be highest for pulmonary, auditory, endocrine, reproductive, and neurocognitive function in a sample of adult survivors diagnosed from 0-24 years of age and being 10 to 47 years after diagnosis (Hudson et al., 2013).

It is known that persons who have been treated for cancer during childhood report more health problems than controls and that these problems can affect their health-related quality of life (HRQoL). As previously described from focus groups with young persons, who had completed cancer treatment at least two years before the study, limited knowledge of late effects was something creating anxiety (D'Agostino & Edelstein, 2013). In another study, health problems (here equivalent to late effects) related to growth, hearing ability, thyroid, kidney, liver, immunologic, heart and fertility were reported almost four times more by young adults treated for cancer during childhood, range 3-22 years after treatment, than by controls, survivors of tumours of CNS were not included (Schwartz et al., 2010). In addition, scarring, disfigurement, and hair loss have been reported significantly more often by childhood cancer survivors, being at least five years post-diagnosis, than by sibling controls, and also negatively affect HRQoL (Kinahan et al., 2012).

Along with the increased risk of late effects, persons treated for cancer during childhood or adolescence have an excessive mortality risk (Oeffinger & Hudson, 2004; Reulen et al., 2010). One study, including children diagnosed before the aged of 15 and being at least five years beyond diagnosis, showed that the risk was persistent beyond 25 years after diagnosis and was caused by secondary malignancies and circulatory and pulmonary diseases (Reulen, et al., 2010). Therefore, future challenges include the preparation and implementation of new methods for treatment, with the possibility of curing more children but at the same time reducing the late effects caused by treatment. Follow-up studies focusing on quality of life among adults who have survived childhood cancer will be important since results from these studies will add important information to future treatment protocols (Gustafsson, et al., 2013). For instance, de-escalation of chemotherapy to patients in low-risk groups is being considered in some treatment protocols.

### **2.3 THE CONCEPT OF HEALTH**

The concept of health is complex and multidimensional and has varying meanings for different people and depending on the situation. It reflects the individual's present and total life situation, it is an integrated part of life and health is also the most central concept in health care as a whole. In care sciences, including the discipline of nursing,



knowledge from the discipline of medicine regarding health and disease interplay with the existential experience of health from each individual. Both parts are of importance for a person's experience of health; the biological involving bodily functions and systems and the existential regarding who you are, how you understand yourself and the interplay with the rest of the world and other people. To be in a state of health can be described as encompassing the experience of well-being, feeling well and being able to fulfil smaller and larger life projects. The concept of health does not exclude the event of a disease, still the individual must be capable of dealing with such events, if health, in terms of well-being and being able to, are to be present (Dahlberg & Segesten, 2010).

### **2.3.1 Health-related quality of life**

Health-related quality of life (HRQoL) can be defined as the aspects of quality of life that relate to an individual's health (Wilson & Cleary, 1995) that is, to assess to what extent disease and treatment influence important domains of functioning and well-being such as physical, psychological and social (Revicki et al., 2000). HRQoL can therefore be considered a component of the more general concept quality of life (QoL), which also can include aspects such as economic matters and political freedom (The European DISABKIDS Group, 2006). As previously shown, from in-depth interviews with healthy Norwegian adolescents (14-15 years), their view on QoL was separated from health. According to them, QoL has to do with positive life cycles including satisfaction with oneself, and the importance of good friendship and family relations (Helseth & Misvaer, 2010). These adolescents' description can be related to an evaluation done by Moons et al. (2006), although that study did not explicitly focus on young people, where they concluded that the best definition of QoL is framed in terms of satisfaction with life.

Both QoL and HRQoL are important as research areas in public health, and medical research and as criteria for outcome and evaluation (The KIDSCREEN Group Europe, 2006). QoL and HRQoL are also often used as outcome variables in nursing research, for instance describing life for persons with chronic conditions. Moreover, for nurses working in the clinical setting assessments of patients (Fawcett, 2005), or, in this case young persons with a cancer experience, are essential to meeting their needs and providing support when necessary. However, QoL and HRQoL are not well-defined by consensus and there is variation regarding which domains that are to be included (Doward & McKenna, 2004; Moons, et al., 2006; The KIDSCREEN Group Europe, 2006).

Going through puberty, the adolescent period and then entering into adulthood are associated with achieving and creating a new independence as well as with developing one's identity (Wrangsjö & Winberg Salomonsson, 2007). With regard to persons with an experience of cancer during childhood, Nightingale et al. (2011) performed a systematic review focusing on qualitative studies (based on interviews and/or focus groups with survivors) in which they suggested a revised HRQoL framework by adding three new domains; body appearance, resilience (including self-confidence/self-efficacy and independence) and fertility/sexual function to the core domains of physical, psychological, social and spiritual character. Eiser (2007) describe the essential

domains as similar, with the additions of body image and autonomy, considered especially important to young adults (Eiser, 2007). Concerns related to body image and independence have been mentioned by adolescents as affecting their QoL, in previously performed interviews and focus groups (Edwards, Huebner, Connell, & Patrick, 2002). The analysis resulted in four dimensions to which the “sense of self” was central, including belief in oneself (e.g., overcoming obstacles and reaching goals) and being oneself (e.g., feeling good about oneself, pleased with one’s looks, comfortable with one’s sexual feelings) (Edwards, et al., 2002). Given this background, the definition of HRQoL applied in this thesis includes the essential aspects such as physical, emotional and social along with independence and sexuality, with a special focus on the two latter.

HRQoL is in this thesis measured with two instruments, KIDSCREEN-27 and The DISABKIDS Chronic Generic Module (DCGM-37), and through online focus group discussions. Regarding KIDSCREEN, the KIDSCREEN Group Europe developed three instruments based on a literature review, a Delphi study with experts and focus groups in which children, adolescents and their families could express their opinions about what constitutes HRQoL. The developmental process resulted in three instruments and the dimensions included in KIDSCREEN-27 (*Physical Well-being, Psychological Well-being, Autonomy & Parent Relations, Social Support & Peers and School Environment*) are considered to be a description of HRQoL (The KIDSCREEN Group Europe, 2006). The developmental steps were similar for the DISABKIDS instruments, since these two European projects closely cooperated, and the process resulted in two chronic generic instruments (one longer and one shorter); one dimension (Independence) from the long version has been used in this thesis (The European DISABKIDS Group, 2006).

#### 2.3.1.1 *Health-related quality of life in survivors of childhood cancer*

Research results from a number of studies, including different diagnoses, show in general that the majority of persons, having survived their cancer at least four years, rate their HRQoL or QoL as similar to or higher than that of persons without a cancer experience (Bradley Eilertsen, Jozefiak, Rannestad, Indredavik, & Vik, 2012; Essig et al., 2012; Harila, Salo, Lanning, Vilkkumaa, & Harila-Saari, 2010; Langeveld, Grootenhuis, Voute, de Haan, & van den Bos, 2004; Mört, Salanterä, Matomäki, Salmi, & Lähteenmäki, 2011a, 2011b; Sundberg et al., 2010; J. van Dijk et al., 2007; Zebrack & Landier, 2011). Despite these similar results, it is noteworthy that inconsistencies exist across studies. These inconsistencies are explained by factors such as varying study focus and designs, a lack of distinction between the conceptualisation and measurement of QoL and HRQoL and a lack of qualitative input from those with a cancer experience during childhood (McDougall & Tsonis, 2009). Further limitations, as shown in a systematic review aiming at guiding future paediatric oncology research, are small sample sizes, heterogeneous samples, lack of longitudinal approaches and different measures used (Klassen, Anthony, Khan, Sung, & Klassen, 2011).

Most often the physical dimension displays the largest differences when survivors are compared to peers, while other dimensions such as psychological well-being and social well-being do not show such great divergence (McDougall & Tsonis, 2009). Factors

that influence HRQoL have been documented and two studies showed, among those having survived at least five years, all health problems and an increased prevalence of symptoms (in those studies these are not labelled late effects) were connected to lower self-reports of HRQoL, measured with Short-Form 36 (SF-36) (Huang et al., 2013; Rueegg et al., 2013). The health problems (e.g. overweight, vision impairment, hearing, memory, digestive, musculoskeletal/ neurological concerns) had the greatest impact on physical function and general health, and survivors who reported memory, musculoskeletal, or neurological problems were those most affected (Rueegg, et al., 2013). Further, a higher prevalence of symptoms (cardiac, pulmonary, motor/movement problems, pain, sensation impairments, learning/memory problems, anxiety, depression, and somatisation) (Huang, et al., 2013) and more severe late effects (Blaauwbroek et al., 2007) was connected to a longer time since diagnosis (up to 30 years) (Blaauwbroek, et al., 2007; Huang, et al., 2013). However, there seems to be a discrepancy between late-effects that are self-reported and those documented in medical records (Casagrande et al., 2013). Casagrande and co-workers showed that self-reported late effects among survivors of childhood cancer, at least 12 years post-diagnosis, were significantly higher (9 of 14 categories) than those reported by health care professionals reflecting that a more systematic follow-up might be needed regarding late effects among long-term survivors (Casagrande, et al., 2013). However, lack of thoroughness in the medical records might account for the discrepancy and differences in the perception of the severity of late effects between survivors and health care professionals.

Generally, factors such as older age at diagnosis, longer time since diagnosis and certain diagnoses and treatment types contribute to lower QoL or HRQoL (McDougall & Tsonis, 2009). However, examples exist where the results of QoL, in this particular study among survivors of Hodgkin's lymphoma, were not connected to time since treatment, age at diagnosis or therapy burden (Calaminus et al., 2014). Persons with a diagnosis of tumours of the CNS, osteosarcoma, Wilms tumour, neuroblastoma, (survivors of Wilms tumour and neuroblastoma here received more intensive treatment) (Bradley Eilertsen, et al., 2012; Mört, et al., 2011a, 2011b) have reported lower HRQoL. In addition, those treated with stem cell transplantation (SCT), which is a very intensive treatment program (Mört, et al., 2011a) have been shown to report lower HRQoL and females generally report lower QoL or HRQoL (Barrera, Teall, Barr, Silva, & Greenberg, 2012; McDougall & Tsonis, 2009; Mört, et al., 2011a).

### **2.3.2 Independence**

The lived experience of health influences a person's identity and self-esteem and the ability to live the life that is desired (Dahlberg & Segesten, 2010). For young person's growing up that can include the ability to make independent decisions about the future as well as separating from parents (Zebrack & Isaacson, 2012). A disease can interfere with a person's wish for an integrated identity and self-esteem, still it is known that such an event encompasses a potential for increased awareness of who you are and a re-definition of life goals (Dahlberg & Segesten, 2010).

Adolescence is a period of developmental transition (Im, 2010) whereas aspects related to independence and identity becomes essential (Erling & Hwang, 2001; Wrangsjö &

Winberg Salomonsson, 2007). Researchers commonly divide the adolescent period into early adolescence (ages 10-13), middle adolescence (ages 14-17) and late adolescence (ages 18-early 20s) (Smetana, Campione-Barr, & Metzger, 2006). Three different areas of independence are usually discussed: emotional independence in close relations especially to parents; the ability to make decisions and to complete those decisions; and the development of values concerning morality, politics and religion. Independence development during the adolescent years is sometimes referred to as the second individuation and entails increased responsibility for oneself and one's actions (Erling & Hwang, 2001). Adulthood's beginning is less clearly marked, than the onset of the adolescent period when more dramatic changes occur owing to pubertal development, but it has been defined in terms of marriage, parenthood, finishing education, and entering the work force. It has been suggested, given that entering adulthood occurs later in today's society, that the years between ages 18 and 25 should be labelled emerging adulthood (Smetana, et al., 2006).

A cancer experience during childhood hampers a young person's course of life, and thereby his or her development towards independence because hospital visits affect, for instance, the person's school situation and relationships to peers. To the best of our knowledge, research regarding self-rated independence, (e.g. emotional independence and the ability to make decisions), is limited among persons that have survived childhood cancer. Among identified reports, one presented results in which retinoblastoma survivors, a mean of 11 years since diagnosis, rated their autonomy higher than peers did (measured with KIDSCREEN-52 and including questions such as being able to choose what to do during free time) (J. van Dijk, et al., 2007). However, a different report showed that those with a cancer experience from childhood (leukaemia, solid tumours, brain tumours) were less autonomous (measured by questions regarding employment during secondary school and being on holiday without adults) than peers (Stam, Grootenhuis, & Last, 2005). Another aspect of independence, studied by Koch and co-workers and Stam and co-workers, is leaving the parental home as a young adult and to live on one's own. In those two studies, no significant differences were found when comparing survivors of childhood cancer, the majority being five years beyond diagnosis, and peers (Koch et al., 2006; Stam, et al., 2005) except for survivors of CNS tumours who left home to a significantly lower extent (Koch, et al., 2006).

The adolescent and emerging adulthood periods are stages in life when relationships other than those with one's parents become important, (e.g., partner relationships). The period involves a separation from parents and increased independence, as already mentioned, as well as other physical, emotional, social and cognitive changes (Epelman, 2013). Research has shown that partner relationships generally become normative and stable during middle adolescence, influencing psychosocial development and during this period interactions with romantic partners are more frequent than those with parents, siblings or friends (Smetana, et al., 2006).

Research results regarding engagement in partner relationships, marriage and parenthood among persons with a cancer experience during childhood vary somewhat. Still, the majority of studies, including different diagnoses all but one study also include persons with tumours of the CNS, report a more negative outcome for survivors, when

stated being off treatment or had survived three years and beyond, than for peers (Gurney et al., 2009; Jóhannsdóttir et al., 2010; Pivetta et al., 2011; Stam, et al., 2005; Thompson, Marsland, Marshal, & Tersak, 2009; Wengenroth et al., 2013). However, some findings, these studies did not include tumours of the CNS, do not show differences between survivors, mean time since diagnosis 7 years, and peers in terms of having a relationship or dating (Gerhardt, Vannatta, Valerius, Correll, & Noll, 2007) or in terms of satisfaction with, conflict in, and duration of relationships (Thompson, et al., 2009). Still, those with a cancer experience have been shown to have fewer relationships and to experience greater discomfort at the end of relationships (Thompson, et al., 2009).

### **2.3.3 Sexuality**

Having a satisfactory sexual life is crucial for people's well-being and health (Dahlberg & Segesten, 2010; National Institute of Public Health, 2012) and sexuality is an important part of every individual's desire and opportunities for intimacy (Bolte & Zebrack, 2008).

During adolescence, the sexual perspective becomes more visible, including thoughts and questions about sexuality, combined with pubertal development (Epelman, 2013; Helmius, 2010) and the individual's social context.

The following is a working definition of sexuality, developed by international experts and highlighting that sexuality is dependent on social and cultural factors and further, this working definition has been used in cancer research (Rasmusson, Plantin, & Elmerstig, 2013).

*Sexuality is a central aspect of being human throughout life and encompasses sex, gender identities and roles, sexual orientation, eroticism, pleasure, intimacy and reproduction. Sexuality is experienced and expressed in thoughts, fantasies, desires, beliefs, attitudes, values, behaviours, practices, roles and relationships. While sexuality can include all of these dimensions, not all of them are always experienced or expressed. Sexuality is influenced by the interaction of biological, psychological, social, economic, political, cultural, ethical, legal, historical, religious and spiritual factors.*

(World Health Organization, 2006) (page 5).

Adolescents suffering from chronic health conditions such as cerebral palsy, cystic fibrosis, type 1 diabetes, epilepsy, and spina bifida may experience problems that affect their pubertal development, sexual maturation and sexual function (Rew, 2007). This may place them behind peers and affect their development during the adolescence. For young persons with a cancer experience, too, sexuality and intimacy are crucial aspects of QoL but influenced by the cancer disease and treatment where some treatments and side effects might be permanent affecting sexuality and sexual behaviour (Bolte & Zebrack, 2008). The subjects of sex and sexuality are often

avoided by health care professionals, or are not addressed in the follow-up care, and perhaps are not brought up by survivors either (Bolte & Zebrack, 2008; Park et al., 2009). Nevertheless, sexuality is a subject where health care professionals can play an important role when meeting persons with a cancer experience, since it is part of our professional role to focus on factors that maintain health (National Institute of Public Health, 2012). One study that particularly analysed conversations between adolescents and physicians found that the adolescents themselves never brought up sexual issues (Alexander et al., 2014).

Going through cancer treatment during childhood may have an impact on fertility (Epelman, 2013; Jacobs & Pucci, 2013) and further, lower sexual confidence and the feeling that one is not attractive enough might be effects of a changed bodily appearance and treatment may affect sex drive and sexual function (Langeveld & Arbuckle, 2008). Radiation exposure to the ovarian-, testis-, and cranial- regions like certain types of chemotherapy (alkylating agents), can lead to delayed puberty or its absence affecting, for instance, libido, sexual maturation, and erectile function (Armstrong, Chow, & Sklar, 2009; Haddy & Haddy, 2010). Therefore, redefining intimacy and sexual behaviour (finding a “new normal”) might be needed for adolescent and young adult cancer survivors (Bolte & Zebrack, 2008). A literature review showed that sexual development, sexual function and sex life may be potential worries for those that have survived cancer during childhood along with uncertainty regarding fertility status (Mattsson, Lindgren, & von Essen, 2008). In addition, survivors, being five years or more beyond diagnosis, who had received chemotherapy, radiotherapy, and surgery were older at the time of their first sexual experience, than comparisons were (Stam, et al., 2005).

### 2.3.3.1 *Sexual function and dysfunction*

Sexual dysfunction has been defined as a lack of sexual interest, decreased arousal, erection- and ejaculation difficulties, orgasm dysfunction, dyspareunia and vaginismus (Fugl-Meyer & Fugl-Meyer, 2010). Regarding sexual dysfunction in the general population, a Danish study (Christensen et al., 2011) showed that men and women report similar figures (11% of both men and women reported at least one dysfunction). As the authors concluded, this finding contrasts with other reports, in which women in general report more sexual dysfunctions than men but it may reflect the fact that more women reported not being sexually active and were therefore excluded (Christensen, et al., 2011).

Sexual function and dysfunction among persons with a cancer experience from childhood is an understudied area and studies that take a qualitative approach are especially lacking. Among the studies found, participants being at least 2 years post-diagnosis, aged 17 and above, 20-29 % of them had experienced sexual dysfunction (e.g., no interest in sex and arousal problems) (Bober et al., 2013) and a limited sex life because of their disease, (they reported not feeling certain about their bodies, difficulties revealing emotions, scars and potential infertility) (E. M. van Dijk et al., 2008). Comparisons of those who experiencing sexual dysfunction and those who did not revealed that older survivors experienced sexual dysfunction to a significantly higher extent (Bober, et al., 2013) and older survivors and those diagnosed at an older

age had higher symptom scores related to sexual dysfunction (Zebrack, Foley, Wittmann, & Leonard, 2010). Sexual function in these studies did not show any significant differences with regard to type of diagnosis (Bober, et al., 2013; Zebrack, et al., 2010) or exposure to treatments with a presumable effect on sexual function (Bober, et al., 2013). However, Zebrack et al. (2010) commented on the fact that male survivors of tumours of the CNS were more likely than male survivors of haematological cancers and solid tumours to leave an item blank or to choose the response category “not applicable” in that study. Furthermore, another study showed that male survivors of tumours of the CNS were the most vulnerable diagnostic group with regard to sexual dysfunction (Sundberg, Lampic, Arvidson, Helström, & Wettergren, 2011).

Differences between women and men, with regard to sexual desire, have been studied to some extent, and large population studies show that men report stronger sexual desire than women do (Beutel, Stobel-Richter, & Braehler, 2008; Eplov, Giraldi, Davidsen, Garde, & Kamper-Jorgensen, 2007) but also that this desire decreases with age for both sexes (Eplov, et al., 2007). However, sexual function and sexuality may be perceived differently for men and for women, possibly influenced by stereotypes, norms, emotional and sociocultural factors, in today’s society and culture (Beutel, et al., 2008; Rasmusson, et al., 2013; Thaler-DeMers, 2001).

Among persons with a childhood cancer experience, women report having more sexual problems than men do (Bober, et al., 2013; Zebrack, et al., 2010). Zebrack et al. (2010) showed that 32% of men and 52% of women reported “a little of a problem” related to sexual function, and the overall mean symptom score, related to sexual function, among women was more than twice that of men. In addition Bober et al. (2013) have reported that women were almost twice as likely to be sexual dysfunction cases. Because of the possible different perceptions of sexual function among women and men, it is preferable to compare the results for those who have had cancer to corresponding findings among peers and to conduct analyses by sex, something which has not been done in the previously mentioned studies (Bober, et al., 2013; Zebrack, et al., 2010). A few studies have taken this approach, yielding somewhat differing results with regard to women and men, respectively. Puukko et al. (1997) included assessments of self-reported data, interviews and projective psychological tests and found that young female survivors of leukaemia, the majority were more than five years beyond treatment, reported more negative attitudes regarding sexual pleasure and their sexual identity was considered less feminine and more infantile, than that of female controls. However, a more recent study showed that male survivors had more problems with sexual function than male comparisons did, while no such differences were seen among female survivors and female comparisons (Sundberg, et al., 2011).

## **3 AIM**

### **3.1 RATIONALE**

Health care professionals involved in care of persons with an experience of cancer during of childhood require extended knowledge and understanding in several areas to be able to meet their needs and deliver appropriate care. This is essential and relevant in light of the description of health and the aim for survivorship research. On the one hand, from a medical point of view regarding different cancer diagnoses, treatments and occurrence of late effects. On the other hand from an existential point of view, how these young person´s look upon themselves towards achieving an increased independence and identity development together with the interplay with other people and the rest of the world.

Advances in medicine have led to increased survival rates for persons with a cancer experience from childhood implying that health care professionals working in different areas of the health care sector will encounter them. It is therefore of importance to investigate these persons perceptions of their health and their life situation, including health-related quality of life (HRQoL). The existing body of literature, within the field, show a somewhat varying picture and certain research areas, such as independence and sexuality have not been studied to a large extent, areas of importance for adolescents and young adults. Within care sciences and the discipline of nursing, central concepts are human being, nursing action/relation, health and environment. The nursing discipline originates from the lived experience by each human being why self-reports become crucial and where this thesis has its point of departure. It is of importance to use relevant and validated instruments to be able to identify needs and to fully grasp the impact and complexity of childhood cancer. In addition, there appears to be a lack of especially qualitative studies regarding sexuality, which is a crucial aspect taking into account when tailoring the follow-up care in concordance with the needs from this growing population.

### **3.2 AIMS**

The overall aim for this thesis was to investigate self-reported HRQoL with special focus on independence and sexuality among young persons treated for cancer during childhood.

The specific aims were as follows:

- Study I To evaluate the psychometric properties of the five dimensions in KIDCSREEN-27 for use in survivors of childhood cancer.
- Study II To investigate HRQoL in childhood cancer survivors with a standardised instrument and establish thresholds for clinically significant differences by using qualitative interviews as anchors. An additional aim was to investigate survivors´ HRQoL in relation to an age-matched comparison group without cancer experience.
- Study III To investigate independence by comparing self-rated independence in childhood cancer survivors five years after diagnosis with ratings during initial cancer treatment and in a comparison group. Further, to determine whether and how certain clinical and demographic variables had any impact on self-rated independence.
- Study IV To investigate how childhood cancer survivors think and how they reason about sexuality and sexual experiences.



## **4 METHODS**

### **4.1 DESIGNS**

A summary of the four studies is displayed in Table 1.

The four studies in this thesis have different methodological approaches. Study I is a methodological study and the three following (studies II-IV) have a cross-sectional design. Study III also includes longitudinal data, i.e. several data collection occasions with the same individuals over a certain period of time.

Study I, II and III include follow-up data from a national Swedish cohort study, from diagnosis up to 4-6 years after diagnosis, with totally four data collection occasions (Figure 1). Results from the time closer to diagnosis are partially presented in this thesis (study III) while the majority of those results are presented elsewhere (af Sandeberg, 2011). Study IV includes data from another data collection. The focus in this thesis has been on self-reports of HRQoL, with different methodological approaches; both semi-structured interviews, questionnaires and written online focus group discussions, from young survivors of childhood cancer as well as peers.

The importance of using valid and sensitive measurements, relevant for the population under study, was achieved by performing a Rasch analysis (study I) before further evaluations were performed. A combination of quantitative and qualitative methods were used (study II), self-reports of independence were compared between time of diagnosis and 4-6 years post-diagnosis (study III) and comparisons to peers were made (studies I-III). Lastly, exploring a relatively new area, i.e. sexuality among survivors of childhood cancer, was achieved by written online focus group discussions using a descriptive approach (study IV), relevant when little is known of the subject under study.

Table 1. Outcomes, number of participants, measures and analyses in the four studies

Study	Study outcomes	Participants	Measures	Analyses
I	Psychometric properties of KIDSCREEN-27	Survivors, n=63 aged 12-22 4-6 post-diagnosis Comparison group, n=257 aged 11-23	KIDSCREEN-27 Medical records	Chi-square statistics Partial Credit Model (PCM)
II	Clinical significance HRQoL		KIDSCREEN-27 Interview data Medical records	Chi-square statistics Multiple regression models Effect sizes
III	Independence		The DCGM-37 dimension Independence Medical records	Chi-square statistics Independent-samples t-test Paired-samples t-test Multiple regression models Effect sizes
IV	Thoughts and reasoning around sexuality and sexual activities	Survivors, n=133 aged 16-25 ≥5 years post-diagnosis	Written online focus group discussions Register data (age, sex, diagnosis)	Chi-square statistics Inductive content analysis

Sample, Data collection, procedure and data analysis are first presented for studies I-III and thereafter for Study IV.

## 4.2 SAMPLE STUDIES I-III

### 4.2.1 Survivors

All school children (aged 7-16) in Sweden that were diagnosed with cancer and started treatment (chemotherapy and/or radiation therapy) between January 2004 and May 2006 were eligible (N=145) for inclusion in a national cohort study, 87 males and 58 females (ratio 1.5:1). The aim from the beginning was to investigate the school situation and HRQoL among children during and after cancer treatment (af Sandeberg, 2011). Those treated with stem cell transplantation (they are not recommended to

attend school during first six months after treatment) or solely with surgery and those with language difficulties were excluded. For the initial studies, data was collected on three occasions (1 month; 2.5 months; 5 months after treatment start, respectively) when school attendance (all three occasions) and HRQoL (the second and the third occasion) were assessed. Regarding school attendance, n=126 participated at least once and these data are presented elsewhere (af Sandeberg, 2011).

Relevant for this thesis are the two assessments of HRQoL (with Disabkids Chronic Generic Module, DCGM-37), at 2.5 and 5 months after treatment start, where n=118 participated at least once. As noticed, there is a discrepancy regarding how many that participated in the assessments regarding school attendance and HRQoL; eight individuals chose to terminate their participation after the first assessment of school attendance (af Sandeberg, 2011).

At the present follow-up, four to six years after diagnosis (median 63 months, range 50-74), 23 of those (n=118) had died and three were excluded due to cognitive dysfunction (n=2) or unidentifiable address (n=1). This resulted in 92 survivors eligible for inclusion in the present follow-up. Among the 92 survivors, 63 accepted participation (68%) in the follow-up, aged 12-22, median 17 years, 26 females and 37 males (Figure 1).

Regarding participation: 61 participated in both the semi-structured interview and answered the questionnaire by telephone while two chose to answer only the questionnaire and at home instead of telephone participation.

#### **4.2.2 Comparison group**

Age-matched participants were randomly selected from the Swedish population register (SPAR), 50% females and 50%. Exclusion criteria were: language difficulties (n=5), cognitive dysfunction (n=3), prior cancer experience (n=1) and being abroad (n=10). Five were excluded due to unidentifiable address. From the remaining 476 eligible participants; 257 (54%) agreed participation, aged 11-23, median 16 years. Two hundred and nineteen declined participation; actively (n=171) or passively (n=48), e.g. did not respond to letters (4 did not send back the questionnaire and 44 were unable to reach or did not answer) (Figure 1).

Regarding participation; 65 participated in both the semi-structured interview and answered the questionnaire by telephone, 165 answered the telephone administered questionnaire and 27 chose to answer the questionnaire by hand in their home, instead of participating in a telephone interview.

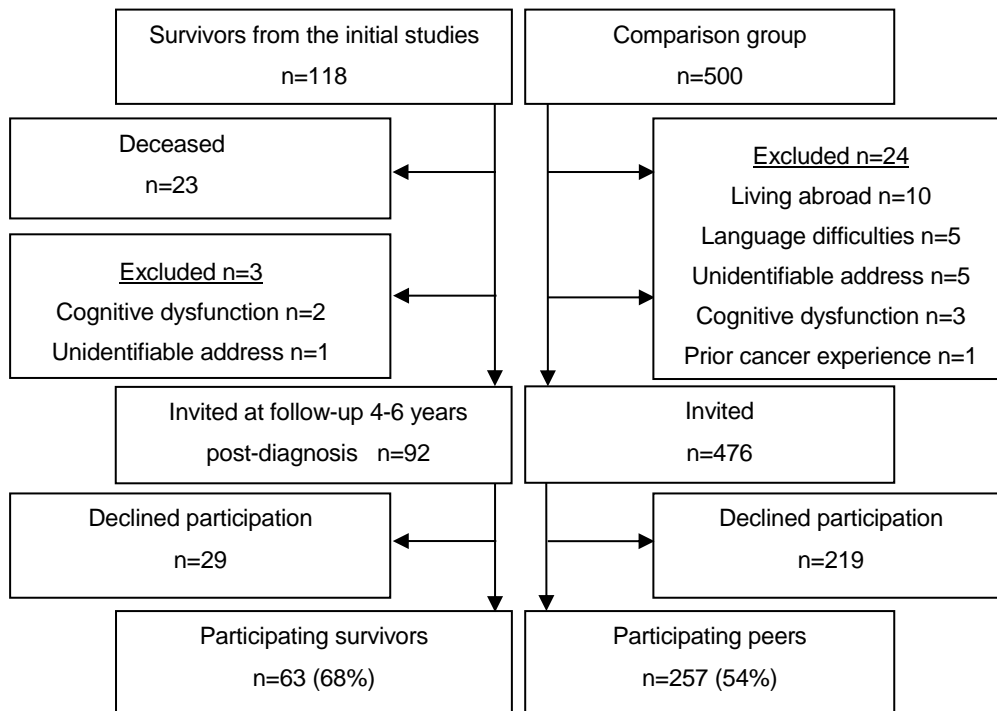


Figure 1. Overview of the inclusion of survivors from the initial studies and the comparison group , with permission from the author, Margareta af Sandeberg (af Sandeberg, 2011).

### 4.3 DATA COLLECTION STUDIES I-III

For the follow-up 4–6 years after diagnosis data were collected through telephone interviews (studies I– III) during 2010 and 2011 including both open-ended and structured questions.

#### 4.3.1 Measures

##### 4.3.1.1 KIDSCREEN-27 (study I, II)

KIDSCREEN-27 was designed to assess generic HRQoL among both chronically ill and healthy children and adolescents aged 8–18 years. It was developed within a European collaboration with a simultaneous approach in each of the 13 European countries using a cross-cultural harmonisation and development. The instruments were developed through literature reviews, expert consultation and focus groups in all countries to identify items and aspects of HRQoL relevant in all countries. Thereafter; item reduction and selection, translation, pilot studies and finally a survey study was performed. There are three KIDSCREEN instruments (KIDSCREEN-52; KIDSCREEN-27; KIDSCREEN-10). KIDSCREEN-27 consists of 27 items in five dimensions (*Physical Well-being; Psychological Well-being; Autonomy & Parent Relations; Social Support & Peers and School Environment*). All items refer to the previous week and follow a 5-point Likert scale ranging from 1 to 5 (poor to excellent; not at all to extremely; never to always). Both a self-report and a proxy version are available and in this thesis, the self-report version was used (The KIDSCREEN Group Europe, 2006). KIDSCREEN-27 has shown to be a valid instrument when tested psychometrically in a large representative European sample (Ravens-Sieberer et al.,

2007; Robitail et al., 2007; The KIDSCREEN Group Europe, 2006). The raw scores obtained from the participants are scored and analysed using standard scoring algorithms. A syntax is used and the different steps result in T-values with scale mean values of 50, standard deviation of 10 and were higher values indicate higher HRQoL (The KIDSCREEN Group Europe, 2006).

#### 4.3.1.2 *DISABKIDS Chronic Generic Module (DCGM-37) (study III)*

The DISABKIDS project is a European collaboration and cooperates with the KIDSCREEN project (see above). The developmental process involved literature review, focus groups, item development and translation, pilot study, field test and implementation. The DISABKIDS instruments are useful in different national and cultural contexts and focus on seven chronic conditions; asthma, juvenile rheumatic arthritis, epilepsy, cerebral palsy, diabetes mellitus, atopic dermatitis and cystic fibrosis. Apart from the disease specific instruments, there are two chronic generic DISABKIDS instruments and the longer version was used in this thesis (DCGM-37). The instrument assesses HRQoL in children and adolescents aged 8-16 years with different chronic conditions. It consists of six dimensions; Independence, Emotion, Social inclusion, Social exclusion, Physical limitation and Treatment (The European DISABKIDS Group, 2006). In this thesis the Independence dimension was used. Raw scores from participants are coded, summed and transformed into a scale from 0 (lowest possible Independence) to 100 (best possible Independence) using standard scoring algorithms for the instrument (af Sandeberg, 2011). The entire instrument DCGM-37 have been used and psychometrically tested (feasibility, data quality, reliability, construct and criterion-based validity) in the same sample of childhood cancer survivors, while under treatment, with overall acceptable results (af Sandeberg, Johansson, Hagell, & Wettergren, 2010).

#### 4.3.1.3 *Semi-structured interviews (study II)*

Qualitative data, obtained from individual interviews (n=61) with the cohort of survivors were used. The interview guide covered areas of present life situation, school situation and/or occupational situation, leisure and relationships to friends. The interviews were audio recorded, transcribed verbatim and analysed using content analysis, findings reported elsewhere (Doukkali et al., 2013). The analysis resulted in the identification of three groups “Feeling like anyone else”; “Feeling almost like others” and “Feeling different” based on how the cancer experience influenced daily lives for the survivors. Thereafter, meaning units (words or sentences that contain similar aspects) from the three groups were compared and four categories were created; thoughts regarding the cancer experience, complications in daily life, ability to handle complications and view of life, revealing the differences between the three groups. The survivors in the group “Feeling like anyone else” described that they seldom thought about the cancer experience, they had minor or no complications, and if any complications, those were handled easily and survivors in this group expressed hardly any influence on view of life. For the group “Feeling almost like others” descriptions included that they sometimes thought about the experience, they had complications that to a small degree influenced, without hampering, daily life and they often mentioned that the experience had influence over their view on life. Survivors in the group “Feeling different” expressed that they often thought about the experience, had complications largely influencing and hampering daily life and described that the experience often influenced view on life (Doukkali, et al., 2013).

#### 4.3.1.4 *Study-specific questions and medical records (studies I-III)*

Selected socio-demographic and clinical characteristics such as sex, age, diagnosis, treatment modality, event, living situation and occupation were obtained from the study specific questionnaires and medical records.

## **4.4 PROCEDURES STUDIES I-III**

### **4.4.1 Survivors**

An information letter was sent to eligible participants. Information about confidentiality and the possibility to withdraw from the study were stressed in the letter and immediately before the interview. For those aged 11-15 years, the letter was addressed to the parent/parents, for those aged 16-17 to the possible participant and to the parent/parents and for those aged 18 years and above, directly to the participant. Within two weeks potential participants, or their parents, were contacted by telephone and for those who gave consent to participate, a suitable time for the interview was agreed upon. For those difficult to reach, written reminders were sent. For those <18 years written informed consent was obtained from both participants and parents and for those  $\geq 18$  years from participants. The interviews, including the semi-structured part (see 4.3.1.3) and the structured questions (study-specific, KIDSCREEN-27 and the Independence dimension in DCGM-37), were tape-recorded and answers to the structured part were also written down. All participants received a cinema ticket as a form of compensation.

### **4.4.2 Comparison group**

The procedure for the comparison group was similar to the group of survivors, except for the consent procedure. For those aged <18 years, written informed consent was obtained from participants and parents and for those  $\geq 18$  years, oral consent was tape-recorded before the interview. All participants received a cinema ticket as a form of compensation.

## **4.5 DATA ANALYSES STUDIES I-III**

Statistical calculations were performed with the IBM<sup>®</sup> SPSS<sup>®</sup> Statistics, versions: 20-22. A significance level of  $p < 0.05$  was applied in all analyses, if nothing else is stated. Following Cohen's classification standards, effect size of  $d = 0.20$  was considered small;  $d = 0.50$  as moderate and  $d = 0.8$  as large (Cohen, 1988).

### **4.5.1 Study I**

The Rasch approach offers a method for analysis by simultaneously generating measures for persons according to their ability, and items according to their difficulty (Bond & Fox, 2007) based on ordinal data. The approach is increasingly used in health sciences research with the intention of both develop and examine the measurements that are being used (Hagquist, Bruce, & Gustavsson, 2009). The construct of KIDSCREEN's five dimensions was evaluated (using the raw scores) by the partial credit model (PCM), a Rasch model developed for polytomous data (Bond & Fox, 2007). The Rasch analysis software program WINSTEPS<sup>®</sup> Rasch Measurement, version 3.72.2 and 3.73 (Linacre), was used to perform the analysis.

The rating scales were examined by analysing the category structure, expressed by the observed average and outfit Mean Square (MnSq) values and the guidelines by (Linacre, 2004) were followed. The guidelines recommend that, e.g., (a) all rating scale categories and thresholds are supposed to advance monotonically and (b) the rating scale category outfit MnSq value should be below 2.0.

Internal scale validity and person-response validity was investigated by item and person goodness of fit statistics. Calculated statistics were represented by MnSq residuals and standardized z-values, indicating to what extent responses from KIDSCREEN-27 match expected responses in the Rasch model, for items and persons. In addition, floor and ceiling effects were examined.

Unidimensionality was investigated by monitoring the variance explained for each KIDSCREEN-27 dimension by the use of a principal component analysis (PCA) of residuals.

Uniform differential item functioning (DIF) was examined to investigate the stability of item difficulty comparing childhood cancer survivors to a comparison group.

#### **4.5.2 Study II**

When combining quantitative and qualitative approaches while studying HRQoL among survivors of childhood cancer, stronger evidence can be provided with regard to the HRQoL in this group. In addition, according to Wyrwich et al. (2005), reporting of p-values does not elucidate if a certain finding is clinical significant, i.e. statistical significance is not per se similar to a meaningful difference in the investigated variable (Wyrwich, et al., 2005). One way of investigating clinical significance is by using a distribution-based method, e.g. effect size calculations, together with an appropriate anchor (Wyrwich, et al., 2005), to be able to highlight clinical importance of possible mean value difference. Therefore, self-reported HRQoL (KIDSCREEN-27) from the survivors was illustrated by the results from the qualitative analysis of the interview data (reported elsewhere) (Doukkali, et al., 2013) to enable establishing of thresholds for clinically significant differences. Calculations of mean values, standard deviations (SD) and effect sizes (ES) were performed and compared to anchors (the survivors expression of influence on daily life) from the qualitative interview data and the previously identified subgroups (“Feeling like anyone else”, “Feeling almost like others”, “Feeling different”).

To explore possible influences on HRQoL, the following independent variables sex (female vs. male), age (11-16 vs. 17-23 years) and group (survivors vs. comparison group) were included in the multiple regression analyses, four regression models one for each KIDSCREEN-dimension. Additional variables (main occupation, living situation and siblings) did not display significance in univariate regression analyses and were therefore not included in the final models.

The three subgroups were further evaluated with regard to age and the group “Feeling different” with regard to diagnostic representation.

Additional regression analyses with regard to the group of survivors were performed. In univariate regressions none of the variable diagnosis (tumours of the CNS vs. other diagnoses; solid tumours vs. other diagnoses), event (event: secondary cancer/relapse, vs. no event), main occupation (studies/work vs. unemployed/sick leave), living

situation (with parents vs. not with parents), age at diagnosis, or siblings (yes vs. no) were significant. Solid tumours, in both study II and III, refer to skeletal and soft tissue sarcoma, Hodgkin's lymphoma and other diagnoses (sertoli leydig tumour and germ cell tumour).

The independent variables displaying significant results in the univariate analyses, sex, age and initial cancer treatment (combined treatment vs. chemotherapy only) were included in the final multiple regression analyses, one for each KIDSCREEN-dimension, i.e. four regression models. One participant, from the group of survivors, was excluded in the *Physical Well-being* and two in the *Social Support & Peers* dimension due to high Cook's distances (0.238; 0.296; 0.202), cut-off at 0.20, and therefore having a negative influence on the result, after recommendations from a statistical consultant.

### 4.5.3 Study III

A correlation matrix was performed to show the relationships between the dependent variable and the independent variables treatment type (combined and chemotherapy solely) and type of diagnosis.

Differences in means were analysed with independent-samples t-test (between groups: males vs. females and survivors vs. comparison group) and paired-samples t-test (survivors during treatment and a median time of 5 years post-diagnosis). Cohen's effect size was used to investigate the clinical importance of mean value differences.

To explore potential predictors of self-rated independence, a multiple regression analysis was performed. The following independent variables were entered; age at present follow-up, sex, (male vs. female,) diagnosis (solid tumours vs. all other malignancies) and (tumours of the CNS vs. all other malignancies) and type of initial cancer treatment (combined treatment vs. chemotherapy solely).

## 4.6 SAMPLE STUDY IV

For study IV data was collected through written online focus group discussions during spring 2013.

### 4.6.1 Survivors

Young survivors after childhood cancer (n=400) diagnosed with selected solid tumours and tumours of the CNS, were identified through the Swedish Childhood Cancer Registry. The diagnoses were chosen with regard to that the treatment given can have an impact on fertility, and were: Hodgkin's lymphoma, Ewing/Ewing-like sarcoma, osteosarcoma, rhabdomyosarcoma, neuroblastoma and tumour of the CNS. The inclusion criteria were that survivors should be at least five years beyond date of diagnosis and the age range 16-24, at day of inclusion. The total sample in the register regarding solid tumours filling the inclusion criteria were selected (N=280). Survivors treated for tumours of the CNS were randomly selected (n=120) from a sample of n=529.

Twenty-eight persons were excluded due reasons such as; cognitive impairment (n=7), emigrated (n=7), being abroad (n=7), missing personal identification number (n=1), deceased (n=1) and other reasons (n=1). Additionally, four persons were excluded due



to unidentifiable addresses. From the remaining 372 eligible participants, 133 (36%) accepted participation, aged 16-25, median 21 years, 67 females and 66 males. A total of 238 declined participation, either actively (n=154) or passively (n=84) e.g. did not respond to letters. For one online discussion, there was only one participant and therefore not included in the analysis.

## **4.7 DATA COLLECTION STUDY IV**

### **4.7.1 Measures**

#### *4.7.1.1 Written online focus group discussions (study IV)*

The study has a qualitative design and data was collected through synchronously written online focus group discussions, in a chat format. The method has shown to be advantageous when collecting data concerning possible sensitive topics (Elmir, Schmied, Jackson, & Wilkes, 2011). A previous study also revealed that online group discussions, performed asynchronously, were a useful methodology in paediatric oncology settings, the study included both patients, parents and survivors (aged 10-19) (Tates et al., 2009). Well performed focus groups discussions have the possibility to move beyond individual interviews due to the illustrative aspects of different perspectives, views, beliefs and opinions in the interactions among participants (Willis, Green, Daly, Williamson, & Bandyopadhyay, 2009). Qualitative description was used as described by Sandelowski (2000, 2010) with the theoretical foundation of naturalism and with the intention to stay as close to the data as possible (Sandelowski, 2010). According to Sandelowski (1994) the goal of qualitative and nursing research is to get at close as possible to the experience by the human being. The qualitative description can therefore be considered relevant when one wants to know answers to questions such as “What are the concerns of people about an event?” “What are people’s responses toward an event (thoughts, feelings, attitudes)?” and focus groups can be a relevant method (Sandelowski, 2000).

An existing chat platform was used, developed together with an internet consultancy company (Carmona AB) and used in previous research regarding sensitive topics (Blomberg et al., 2011). The platform made it possible for the participants to access the online discussions from any computer. A discussion guide was developed after discussion among the moderators aiming to cover the topics: thoughts and experiences around dating and partner relations, sexuality (e.g. sexual experiences and feelings concerning these experiences or associated with having no experiences, thoughts about having sex and sexual problems related to diagnosis and/or treatment). Thoughts about fertility and having biological children in the future and the experiences of the care provided from the health care sector were also included in the discussion guide. The platform was evaluated and tested before data collection started, i.e. some family members received log-in details and performed a discussion together with some of the moderators. The guide was flexible during the discussions to allow for new topics to be brought up by the participants and continuously developed among the moderators during the data collection process. Follow-up questions were added based on the interaction and discussion in each performed online focus group discussion. The focus for study IV was on sexuality, sexual experiences and possible sexual problems. The results regarding thoughts about fertility and having biological children will be presented elsewhere.

#### 4.7.1.2 *Data from The Swedish Childhood Cancer Registry (study IV)*

Demographic and clinical characteristics such as sex, age, diagnosis and time since diagnosis were obtained from the registry.

### **4.8 PROCEDURE STUDY IV**

An information letter was sent to eligible participants with detailed information about the study. Potential participants were contacted by telephone within two weeks after the information letter was sent. Written informed consent was obtained from all informants that agreed to participate. For those who agreed to participate, a suitable time was agreed upon for the online discussion together with other participants of approximately the same age. Number of participants in each online focus group discussion varied from two to five, as well as diagnoses and sex distribution. Each participant received log in details before the online discussion and they were informed that they could use their first name or any pseudonym, if so desired. Each online discussion lasted approximately 90 minutes (median 87 minutes, range: 65-130 minutes) and one to two moderators were engaged in each discussion. Moderators had different professional backgrounds (registered nurses with different specialties, one psychologist and one midwife) and clinical experiences, e.g. counselling in cancer care. After each online group discussion the participants had the opportunity to evaluate the discussion as well as continue the discussion online with the participants from the same discussion, in a separate chat forum. Forty online group discussions were performed but one discussion, for which there was only one participant, was excluded from the analysis, i.e. 39 online discussions were used for the analysis. All participants received a cinema ticket as a form of compensation.

### **4.9 DATA ANALYSIS STUDY IV**

Participants vs. non participants were compared with regard to diagnostic- and sex distribution.

For the analysis, qualitative inductive content analysis was used (Patton, 2002) and the inductive approach is recommended when previous knowledge about a phenomenon exists to a limited extent (Elo & Kyngäs, 2008). The analysis steps, described by Elo & Kyngäs (2008) were followed and the steps include: preparation, organising and reporting (Elo & Kyngäs, 2008). NVivo 10 (QSR International), a software program, was used to facilitate the analysis process and all transcripts generated from the discussions were directly transferred into NVivo. The preparation phase was done before the analysis process started, i.e. deciding the unit of analysis. The organising phase started with that the transcripts were read a number of times and notes and headings were written down by using the coding function in NVivo (open coding) to describe the content as fully as possible (done by the first author). Following that, notes and headings were freely grouped into preliminary categories (done by the first author). The organising phase also include the abstraction process (Elo & Kyngäs, 2008): to generate a description of the research topic by creating sub-categories, generic categories and a main category/categories. This was done by collapsing, keeping or changing the preliminary categories into sub-categories, generic categories and a main category/categories, trying to enhance the categories to be mutual exclusive during the process. Sub-categories with similar content were grouped together under the same categories and categories under the same main category (Elo & Kyngäs, 2008; Juvani,

Isola, & Kyngäs, 2005). Verbatim quotes were selected in order to illustrate the findings without distorting or misrepresenting the meaning. All authors (same as the moderators) were engaged during data collection and data analysis and development of sub-categories and generic categories were discussed until consensus was reached.

## 5 ETHICAL CONSIDERATIONS

Ethical approval including amendments for studies (I-III) have been obtained from the Regional Ethical Review Boards in Umeå, Uppsala, Stockholm, Linköping, Göteborg and Lund (Dnr: 03-662, and amendments: 04-208T, 2009/1069-32, 2010/0033-32, 2010/1520-32, 2010/2115-32/4) and for study IV from the Regional Ethical Review Board in Stockholm (Dnr: 2012/1600-31/1 and amendment: 2013/410-32).

Research involving human beings must follow certain ethical principles and an international set of standards is the Declaration of Helsinki, recently revised in October 2013 ("World Medical Association Declaration of Helsinki: ethical principles for medical research involving human subjects," 2013). Three crucial ethical principles are included in most guidelines: beneficence, respect for human dignity and justice and what actions that are taken to adhere to those (Polit & Beck, 2012).

Approaching children and young persons can be sensitive due to their status as a potential vulnerable group (Polit & Beck, 2012) and to encounter them about their cancer experience several years earlier can cause negative thoughts and feelings. Discussing and posing questions regarding sensitive issues, i.e. sexuality can further cause feelings of inconvenience. All participants received written and oral information about the study and informed consent was obtained from those who agreed to participate and from parents, when applicable (*respect for human dignity and the right to full disclosure*) (Polit & Beck, 2012). For both data collection occasions, the potential risk of causing distress among participants was considered against the benefit of improving the future care of young persons with a cancer experience (*beneficence including risk/benefit assessment*) (Polit & Beck, 2012). For the data collection for studies I-III it was stressed to the participants that they could contact the researcher for advice regarding where to turn for help, if needed. For the data collection for study IV, useful links and additional information was provided in connection to the written online group discussions and it was stressed in the information letter that participants could contact the researchers if needed.

However, discussing a previous cancer experience can be beneficial and especially together with others with the same experience which was especially noted during the online focus group discussions, were many participants stressed the benefit of interacting with others with the same experience and also to discuss relationships, sexuality and fertility. To all participants, information about confidentiality was given, their possibility to withdraw from the study at any time and the fact that the decision regarding participation would not affect their care in any way (*respect for human dignity and right to self-determination*) (Polit & Beck, 2012). For study IV, efforts were taken so individual participants were not possible to identify in the manuscript and being a man or a woman was only stated if participants revealed it themselves during the online focus group discussions (*justice including confidentiality*) (Polit & Beck, 2012).

## 6 RESULTS

Results from study I are presented separately, results from studies II and III together, when applicable, and thereafter findings from study IV are presented.

Demographic and clinical characteristics for participants in studies I-III are presented in Table 2.

Table 2. Demographic characteristics of participating survivors and comparison group (studies I-III)

	Survivors n=63	Comparison group n=257	p <sup>a</sup>	Total sample n=320
Age, median (range)	17 (12-22)	16 (11-23)	0.050	
Age ≤16, n (%)	27 (43)	148 (58)		175 (55)
Age ≥17, n (%)	36 (57)	109 (42)		145 (45)
Sex, n (%)			0.092	
Female	26 (41)	139 (54)		165 (52)
Male	37 (59)	118 (46)		155 (48)
Living situation, n (%)			0.364 <sup>b</sup>	
With parent/parents	56 (89)	214 (83)		270 (84)
Alone	4 (6)	18 (7)		22 (7)
With partner	1 (2)	14 (6)		15 (5)
Other constellation <sup>e</sup>	2 (3)	11 (4)		13 (4)
Occupation, n (%)			0.802 <sup>c</sup>	
Education	52 (82)	206 (80)		258 (81)
Work	6 (10)	41 (16)	0.274 <sup>d</sup>	47 (15)
Unemployed	3 (5)	10 (4)		13 (4)
Sick leave	2 (3)	0		2 (<1)
Diagnoses, n (%)				
Acute lymphoblastic leukaemia	21 (33.0)			
Skeletal and soft tissue sarcoma	15 (24.0)			
Tumours of the CNS	10 (16.0)			
Hodgkin's lymphoma	6 (9.5)			
Non-Hodgkin lymphoma	6 (9.5)			
Acute myeloid leukaemia	3 (5.0)			
Other diagnoses <sup>f</sup>	2 (3.0)			
Event, n (%) <sup>g</sup>	12 (19.0)			

<sup>a</sup> Tested for differences in proportions by Chi-square test. Degree of freedom: 1.

<sup>b</sup> Tested between those living with one or two parents vs. those reporting other living arrangements.

<sup>c</sup> Tested between those in education vs. those not in education.

<sup>d</sup> Tested between those working vs. those not working.

<sup>e</sup> Other constellation: includes living with friends, other relatives or combined living, e.g parents/student apartment.

<sup>f</sup> Sertoli leydig cell tumour and germ cell tumour.

<sup>g</sup> Relapses (n=11) and Secondary malignancy neoplasm (n=1).

### 6.1 STUDY I

Aim: Evaluate the psychometric properties of the five dimensions in KIDSCREEN-27 for use in survivors of childhood cancer.

Ratings scales/category function overall displayed acceptable results with a few exceptions. Regarding internal scale validity all items except four showed MnSq values within the range (0.6-1.40), indicating to what extent the responses match the expected responses in the Rasch model. The assessment of unidimensionality revealed that four of the five dimensions had acceptable results (>50%) while the dimension *Autonomy & Parent Relations* displayed a lower value (39.8%) and based on this result, that dimension was excluded in study II. Person response validity showed acceptable results within four of five dimensions, indicating reasonable good person response validity.

The last assessment performed also included the comparison group, and regarded DIF. No uniform DIF was detected between survivors and the comparison group, indicating no difference in item difficulty across those two samples. Overall the psychometric assessment performed with a Rasch approach revealed a satisfactory result.

## **6.2 STUDY II AND STUDY III**

Aim study II: To investigate HRQoL in childhood cancer survivors with a standardised instrument and establish thresholds for clinically significant differences by using qualitative interviews as anchors. An additional aim was to investigate survivors' HRQoL in relation to an age-matched comparison group without cancer experience.

Aim study III: To investigate independence by comparing self-rated independence in childhood cancer survivors five years after diagnosis with ratings during initial cancer treatment and in a comparison group. Furthermore, to determine whether and how, certain clinical and demographic variables had any impact on self-rated independence.

To summarise study II and III, the results reveal that the majority of those treated for cancer during childhood reported "Feeling like anyone else" or "Feeling almost like others", based on the analysis of the findings in the telephone interviews (Doukkali, et al., 2013), and survivors rated their independence higher, compared to the comparison group, five years after diagnosis. However, a minority reported "Feeling different" and they also rated their HRQoL as lower compared to the two other groups. Older age at interview (17-23), both in the group of survivors and when analysed together with the comparison group, was associated to lower ratings of HRQoL.

Mean values and standard deviations (SDs) of the KIDSCREEN-27 for survivors and the comparison group are presented in Table 3 together with effect size calculations for the identified subgroups (study II).

Table 3. Descriptive statistics of KIDSCREEN-27 (survivors, comparison group) and survivors presented by subgroups including effect sizes (ES)

KIDSCREEN-27 <sup>a</sup>	Survivors n=63 Mean (SD)	Comparison group n=257 Mean (SD)				
Physical Well-being	48.0 (8.6)	46.9 (8.9)				
Psychological Well-being	53.3 (8.5)	51.5 (9.1)				
Social Support & Peers <sup>b</sup>	51.4 (9.1)	51.3 (8.7)				
School Environment <sup>c</sup>	54.9 (9.0)	52.3 (7.7)				
<b>Survivors divided by subgroups</b>						
	Feeling like anyone else; n=29; 47% Mean (SD)	Feeling almost like others; n=26; 43% Mean (SD)	Feeling different; n=6; 10% Mean (SD)	ES <sup>d</sup>	ES <sup>e</sup>	ES <sup>f</sup>
Age median (range)						
At interview	14 (12-21)	17.50 (12-21)	20 (12-22)			
At diagnosis	10 (7-15)	13 (8-15)	14.50 (7-16)			
Physical Well-being	51.3 (5.8)	47.6 (8.1)	36.4 (9.6)	1.88	1.26	0.53
Psychological Well-being	54.7 (5.5)	54.7 (7.4)	39.6 (8.7)	2.07	1.87	n/a <sup>g</sup>
Social Support & Peers	52.7 (5.6)	52.7 (8.2)	44.1 (8.4)	1.20	1.04	n/a <sup>g</sup>
School Environment <sup>h</sup>	53.7 (9.3)	57.6 (7.3)	45.0 (5.5)	1.14	1.95	0.47

<sup>a</sup> Possible range for each dimension: Physical Well-being 12.1-73.2; Psychological Well-being 4.5-73.5; Social Support & Peers 11.2-66.3; School Environment 16.3-71.0

<sup>b</sup> Comparison group: n=253 due to missing values.

<sup>c</sup> Survivors: n=49; comparison group: n=188 due to not all attending school the previous week.

<sup>d</sup> Effect size: Calculated between "Feeling like anyone else" and "Feeling different".

<sup>e</sup> Effect size: Calculated between "Feeling almost like others" and "Feeling different".

<sup>f</sup> Effect size: Calculated between "Feeling like anyone else" and "Feeling almost like others".

<sup>g</sup> n/a: not applicable.

<sup>h</sup> Different n, by group, due to not attending school in the previous week: n=24; n=20; n=3.

As shown in Table 3 the magnitude of difference was large (indicating clinical significant differences) for all KIDSCREEN-27 dimensions (*Physical Well-being*; *Psychological Well-being*; *Social Support & Peers*; *School Environment*), when comparisons were made between the KIDSCREEN-27 scores for the two groups "Feeling like anyone else" and "Feeling almost like others" to the group "Feeling different". The magnitude of difference was indicated by effect sizes between the groups, as shown in Table 3. There was no clear pattern regarding the diagnostic distribution in the group "Feeling different"; osteosarcoma (n=1), ALL (n=1), CNS (n=2), NHL (n=2). However, they had a higher median age, both at diagnosis and at interview.

The four multiple regression models regarding KIDSCREEN-27 performed in study II, when analysing survivors and the comparison group together, revealed that those with a cancer experience rated the dimension *School Environment* (questions including being happy at school, getting along well, ability to pay attention and ability to get along well with teachers) significantly higher than did the comparison group. Further, the models showed that female sex (in *Physical Well-being* and *Psychological Well-being*) was significantly associated with a lower HRQoL and that older age (17-23 years), at the time of interview, was associated with lower self-rated HRQoL in all KIDSCREEN-dimensions.

Additional results (Table 4) regarding the four multiple regression models, using KIDSCREEN-27 for those with a cancer experience only, showed that females rated their HRQoL significantly lower than males for the dimension *Psychological Well-being*. The variable age, showed the same pattern for *Psychological Well-being*, where older age at interview (17-22 years) contributed to lower ratings of HRQoL. Further, the treatment variable had a significant association to the dimension *Physical Well-being* where combined treatment was associated with lower ratings of HRQoL (Table 4).

Table 4. Multiple regression analysis by KIDSCREEN dimensions (survivors, n=63)

Independent variables	Physical Well-being <sup>a</sup>	Psychological Well-being	Social Support & Peers <sup>b</sup>	School Environment <sup>c</sup>
	Std β	Std β	Std β	Std β
Sex <sup>d</sup>	0.084	0.321*	-0.222	-0.038
Age at interview <sup>e</sup>	-0.227	-0.247*	-0.098	-0.227
Treatment <sup>f</sup>	0.316*	0.068	-0.014	-0.196
R <sup>2</sup>	17.4%	18.0%	6.0%	9.1%
F	4.064*	4.311**	1.209	1.494

P-value: \* <0.05; \*\* <0.01

<sup>a</sup> n=62, one excluded participant due to high Cook's distance (0.238)

<sup>b</sup> n=61, two excluded participants due to high Cook's distance (0.296; 0.202)

<sup>c</sup> Lower number (N=49) due to not all attending school the previous week.

<sup>d</sup> Female=1; Male=2.

<sup>e</sup> Age: 12-16 years=1; 17-22 years=2.

<sup>f</sup> Combined treatment=1; Chemotherapy solely=2.

A correlation matrix for the dependent variable (Independence) and the independent variables diagnosis, in two groups, and treatment are shown in Table 5. No correlation was above 0.70, chosen as cut-off, and there was no indication for multicollinearity (measured with Tolerance and Variance Inflation Factor, VIF) (Pallant, 2010).

Table 5. Correlation matrix for the dependent variable (Independence) and the independent variables diagnosis and treatment

	1	2	3	4
1. Independence (DCGM-37)	1.00			
2. Treatment <sup>a</sup>	-0.12	1.00		
3. Diagnosis <sup>b</sup>	-0.03	0.40	1.00	
4. Diagnosis <sup>c</sup>	0.06	0.57	-0.33	1.00

<sup>a</sup> Combined treatment (1) vs. chemotherapy solely (0).

<sup>b</sup> Tumours of the CNS (1) vs. all other malignancies (0).

<sup>c</sup> Solid tumours (1) vs. all other malignancies (0).

As shown in Table 6, the majority of those in the group solid tumours and those diagnosed with tumours of the CNS received combined treatment, while those diagnosed with leukaemia mainly received chemotherapy only.



Table 6. Type of treatment with regard to diagnosis

	Combined treatment	Chemotherapy solely
Solid tumours		
Skeletal and soft tissue sarcoma <sup>a</sup>	15	0
Hodgkin's lymphoma	4	2
Other diagnoses <sup>b</sup>	2	0
Tumours of the CNS	10	0
Acute lymphoblastic leukaemia	2	19
Acute myeloid leukaemia	0	3
Non-Hodgkin lymphoma	1	5

<sup>a</sup> Ewing sarcoma, osteo sarcoma, rhabdomyosarcoma, soft tissue sarcoma and synovial sarcoma.

<sup>b</sup> Sertoli leydig cell tumour and germ cell tumour.

Results from study III showed that survivors rated the Independence dimension, measured by the DISABKIDS Chronic generic Module (DCGM-37), significantly higher five years after diagnosis, compared to the time of treatment. Furthermore, those with a cancer experience showed a higher level of independence than the comparison group. No significant differences were seen between female and male survivors, five years post-diagnosis. The result of the regression analysis performed showed that none of the independent variables (age, sex, diagnosis in two groups and treatment in two groups) significantly predicted the ratings of independence five years after diagnosis.

### 6.3 STUDY IV

**Aim:** To investigate how childhood cancer survivors' think and how they reason about sexuality and sexual experiences.

Demographic and clinical characteristics are presented in Table 7. Regarding marital status and sexual experience; around 36% of the participants reported being in a relationship, a few more females than males. Almost 50% reported being single at the time of the online discussion. The majority (77%) stated that they had sexual experience, again more females than males.

Table 7. Demographic characteristics of participants

	Females n=67	Males n=66	Total sample n=133
Age, median (range) <sup>a</sup>	21 (16-25)	22 (16-25)	21 (16-25)
Time since diagnosis, years, median (range) <sup>b</sup>	12 (5-23)	13 (5-24)	12 (5-24)
Diagnosis, n (%)			
Hodgkin's lymphoma	13 (19)	19 (29)	32 (24)
Tumours of the CNS	16 (24)	14 (21)	30 (23)
Rhabdomyosarcoma	7 (10.5)	13 (20)	20 (15)
Neuroblastoma	11 (17)	8 (12)	19 (14)
Osteosarcoma	13 (19)	5 (7.5)	18 (13.5)
Ewing/Ewing like sarcoma	7 (10.5)	7 (10.5)	14 (10.5)

<sup>a</sup> Age at participation.

<sup>b</sup> Time since diagnosis at time of study participation.

Participants and non-participants were compared with regard to sex and diagnosis (Table 8). A significant difference was seen in the diagnostic distribution among participants and non-participants; a higher proportion of those diagnosed with tumours of the CNS were among non-participants compared to the other diagnoses.

Table 8. Sex and diagnostic distribution between participants vs. non-participants

	Participants n=133	Non-participants <sup>a</sup> n=239	p <sup>b</sup>
Sex, n (%)			0.313
Females	67 (50)	106 (44)	
Males	66 (50)	133 (56)	
Time since diagnosis years, median (range) <sup>c</sup>	12 (5-23)	13 (5-24)	
Diagnosis, n (%)			0.036 <sup>d</sup>
Tumours of the CNS	30 (23)	80 (33)	
Hodgkin's lymphoma	32 (24)	49 (21)	
Rabdomyosarcoma	20 (15)	26 (11)	
Neuroblastoma	19 (14)	53 (22)	
Osteosarcoma	18 (13.5)	18 (8)	
Ewing/Ewing like sarcoma	14 (10.5)	13 (5)	

<sup>a</sup> Non-participants: those who actively or passively declined and one participant (female; tumours of the CNS) not included in the analysis because being alone in that particular online discussion.

<sup>b</sup> Tested for differences in proportions by Chi-square test. Degree of freedom: 1.

<sup>c</sup> Time since diagnosis at day of inclusion.

<sup>d</sup> Tested between tumours of the CNS vs. all other diagnoses.

The analysis of the written online focus group discussions resulted in one main category *Sex as a given part of life* and four generic categories *Sex considered to be good; Feeling insecure and falling behind; Relating sex to a stable relationship* and *Physical concerns*, including sub-categories (Table 9). The main category illustrates that sex is something natural, important and taken for granted in life for the participants. Participants described and reasoned about sex and sexuality in a relaxed way and talked about problems and worries quite openly.

Table 9. The sub-categories and generic categories forming the main category:

***Sex as a given part of life***

<b>Sub-categories</b>	<b>Generic categories</b>
Positive sexual experience No sexual problems	<i>Sex considered to be good</i>
Limited sexual experience Insecure about sex Concerns around ever finding a sex partner Peer influence Later sexual debut Sex as a proof of being sexually desirable	<i>Feeling insecure and falling behind</i>
Focus on the relationship Waiting with sex	<i>Relating sex to a stable relationship</i>
An altered body Altered desire Feeling dry Using aids Difficulties reaching orgasm Difficulties related to erection and ejaculation	<i>Physical concerns</i>

*Sex considered to be good*

Most commonly participants described sex in a positive way and they had good experiences without any problems related to sex. Many participants stated also that they had not reflected upon any possible impact on sex and sexual activities from the disease and/or treatment received.

*Feeling insecure and falling behind*

Feeling insecure and falling behind were described and participants were relatively open when revealing their thoughts and sometimes worries. In other cases, participants more or less stated that “I’m still a virgin”.

Below is an example where a man describes his worries in relation to being behind and also the influence from peers.

“In my class, it’s very open when it comes to sex, everybody knows what’s happening to everybody else and this feels tough when it’s not happening to me that often.” (Man, 22 years, group 34)

In some discussions, other participants replied, asked additional questions and/or acted in a supportive way if someone had worries or concerns.

A later sexual debut was described by one participant in a way as being a “late bloomer”. Part of one interaction illustrated on the one hand being behind others but on the other hand that peers were the ones being behind instead, not having the same experience. Some women mentioned that they used sex or had one-night stands as proving to them that they were sexually desirable.

“I had many one-night stands during my teens, then nobody noticed anything really [signs from the cancer and/or treatment]. Now I live with a partner, it was hard in the beginning but now it works fine.” (Woman, 23 years, group 35)

### *Relating sex to a stable relationship*

In some cases, sex was discussed focusing on the relationship. Wanting to be in a relationship with someone when having sex was mentioned and a desire for stability due to the cancer experience, something that had limited the number of sex partners. The importance of waiting with sex until marriage was also stressed by one man and one woman.

### *Physical concerns*

Here participants described an altered body, altered desire, feelings of dryness, using aids and difficulties related to reaching orgasm and erection and ejaculation difficulties. In some cases, the concerns were connected to the cancer experience, such as operation in the genital area, back pain or a lower sensibility making it difficult reaching orgasm. Some participants described their own situation as a matter of fact while others were more emotional. Further, some replied supportively towards other participants in the same online discussion while other participants just described their own situation.

Vaginal dryness was described as problematic and worrying by some women while it was usually not related to the cancer experience or treatment received. By large, sexual problems were not spontaneously associated with medication received. However, when we explicitly asked about this, one man mentioned that changes in medication have had an impact on desire and general well-being and one woman stated that her medication (due to phantom pains) have had an impact on her sexual desire and without the medication she would have been even more sexual.

Difficulties related to erection and ejaculation was described by some men and either it was due to an operation or performance anxiety. It was expressed during one online discussion that talking about this subject was difficult and not something you wanted to do with anybody.

## 7 DISCUSSION

The overall results from this thesis reveal that the majority of young persons with a childhood cancer experience get along well in terms of self-reported HRQoL with special focus on independence and sexuality. However, a small group seem to be in need of extra support, which is demonstrated by lower ratings of the included aspects of HRQoL. In the online focus group discussions, experiences and thoughts about sexuality were, by some participants, related to the cancer experience, such as ever finding a partner, being a “late bloomer” or having physical concerns like vaginal dryness and erectile dysfunction.

The results revealed that persons with a childhood cancer experience rated their HRQoL as similar or higher (*School Environment*) (study II) when compared to ratings from the comparison group. The positive attitude towards school was reflected in the semi-structured interviews as well (Doukkali, et al., 2013), where the survivors described that changed priorities due to the cancer experience influenced performance and motivation in school. However, females and older participants (17-23 years), both among the survivors and when analysed together with the comparison group, rated their HRQoL as lower, in one or more KIDSCREEN-dimensions (study II). It has been proven in previous research (McDougall & Tsonis, 2009) as well as in national and international reports (investigating children and adolescents up to the age of 19) that females and older individuals rate their well-being and life satisfaction lower than males and younger individuals (Currie et al., 2012; Petersen et al., 2010). Our findings, with older individuals rating their HRQoL as lower, might not be very surprising, since adolescence is a period of developmental transition which include changes with regard to relationships, and physical and emotional development connected to a move into a mature adult life (Currie, et al., 2012; Im, 2010).

At the same time, it is promising that those with a cancer experience, a median time of five years after diagnosis, rated their independence as high, and that the level had increased since time of diagnosis (study III). However, the results were reflected in mean values (not revealing the entire range of responses among individuals) and it is therefore important to note that not all survivors might have reached a satisfactory level of independence, five years post- diagnosis. It is also well-known that persons treated for childhood cancer have a life-long risk of increased morbidity and mortality, with a possible effect on independence, among other aspects (Robison & Hudson, 2014). This is why studies with in an even longer time perspective are warranted. Previous research regarding independence is limited but existing reports (measuring different aspects of independence) have indicated both higher and lower as well as similar independence compared to controls (Koch, et al., 2006; Stam, et al., 2005; J. van Dijk, et al., 2007). Still, the high ratings of independence can reflect satisfaction with one’s position in life and be related to a changed view on life. This was also reflected in one of the online focus groups (study IV) where a woman stated that the cancer experience was something unique and she felt that peers were the ones being behind, not her. In line with the illustration from this young woman are findings from a study analysing illness blogs (Kim & Gillham, 2013) where one young man described that although he would always be a childhood cancer survivor, he still had the possibility to change his lifestyle. He had gained a different view of life due to the cancer experience which meant that he no longer took anything for granted (Kim & Gillham, 2013). This message is further evident when reading the literature, where previous findings from interviews with survivors of cancer during childhood or adolescence, at least one year

or more since diagnosis, have shown that they report an increased psychological maturity, optimism and hope, new values and priorities, new strengths, good self-esteem, increased compassion and empathy, good relations and broader perspectives (Enskär & Berterö, 2010; Karian, Jankowski, & Beal, 1998; Mattsson, Ringner, Ljungman, & von Essen, 2007; Parry & Chesler, 2005). These previous findings can be seen in light of the description of health by Dahlberg and Segesten (2010) and the fact that an individual can experience health despite an event of a disease. The overall high ratings of HRQoL (study II) might be connected to an increased psychological maturity, optimism, hope and new strengths, some years after a cancer experience. The high ratings regarding independence and the increased levels of independence compared to diagnosis (study III), might as well be related to an increased maturity, new strengths and a good self-esteem. Acting with compassion and empathy was evident from the interactions in the focus groups (study IV), where participants acted supportive and caring when someone else in the discussion revealed sensitive issues, reflecting an understanding for others that have suffered from a difficult experience. The importance of good relations, new strengths and a high self-esteem were also evident during the online discussions. For instance, one young woman expressed her desire of really wanting to be in a relationship with someone and where the cancer experience would not be a hindrance. The cancer experience was part of her history and something related to her personal identity.

A concept related to these descriptions from previous research and from the persons in the present studies, is thriving: despite an experience of trauma, positive adaptation or growth can occur (Parry & Chesler, 2005). Thriving is a complex process, which involves making a positive meaning out of a challenging life event together with an integration of both gains and losses in ways that promote growth. At the bottom line is the achieved ability to live fully and richly and embracing struggle along the way (Parry & Chesler, 2005). Apart from thriving is another aspect, worth mentioning here, and labelled as self-deception response bias (O'Leary, Diller, & Recklitis, 2007), where it is hypothesized that the individual deny problems and does not report negative symptoms. Self-deception response bias is believed to be unconscious, so the individual suffering from it is not aware of negative feelings, attitudes or behaviours. O'Leary et al. (2007) found that self-deception response bias was significantly related to self-reported QoL (using a multiple regression model); higher self-deception response bias contributed to higher ratings of psychological well-being among childhood cancer survivors, with five years or more since diagnosis (O'Leary, et al., 2007). Since only one study has been found regarding this aspect and the complexity in investigating something that is unconscious, it needs to be further evaluated. However, while it is evident that both thriving and self-deception apparently can exist among survivors, it has also been shown that self-reports of late effects (e.g. hearing deficit, renal abnormality, visual impairments, motor deficits) are higher than late effects reported by health care professionals (Casagrande, et al., 2013) why further investigation can be warranted.

The findings from the written online focus group discussions revealed that the majority of participants experienced no sexual problems and quite easily and relaxed discussed and interacted with each other regarding sex and sexual experiences. However, worries and thoughts about limited sexual experiences, falling behind peers and having a later sexual debut were expressed. These thoughts may reflect young person's thoughts in general, even without a disease experience. A survey conducted in Sweden in 2009 among adolescents and young adults, aged 15-29 years (Tikkanen, Abellsson, & Forsberg, 2011), showed that those with lower self-esteem described their latest sexual

encounters in more negative terms than the others. It has also been shown that lower self-esteem and psychological well-being were associated with lack of preventive measures at latest sexual encounter (pregnancy or sexual transmitted infections [STIs] prevention) among childhood cancer survivors (Klosky et al., 2013). Still, the majority of the Swedish adolescents and young adults described their sexual debut and their latest sexual experience in positive terms (Tikkanen, et al., 2011) which is similar to the participants in the online focus group discussions.

Independence and sexuality are important aspects of a person's experience of health (Dahlberg & Segesten, 2010; National Institute of Public Health, 2012) and the sexual perspective becomes more visible during the adolescent period along with the pubertal development (Epelman, 2013; Helmius, 2010). Some recent research results, with regard to sexuality and sexual experiences among young people in general, are presented here, in light of the findings from the online focus groups (study IV). The trend among young female high-school- and university students reveal that they are willing to take risks reflected by, for instance, more sexual partners and increased self-reports of STIs. The young women expressed more liberal attitudes concerning casual sex while they at the same time were considered to take more responsibilities for contraceptive use, including compliance, than young men (Ekstrand, Larsson, Von Essen, & Tyden, 2005; Larsson & Tyden, 2006; Tyden, Palmqvist, & Larsson, 2012). There seem to be fewer studies with young men, however, one study which compared male and female high-school students, showed a trend with more female students having experienced partner sex while male students had more experience of watching pornography and masturbation. However, an overall significant increase was seen over time regarding ever had experienced sexual intercourse (Häggström-Nordin, Borneskog, Eriksson, & Tyden, 2011). The participants in the online focus group discussions expressed both liberal and more conservative views regarding sex and sexual behaviour, although the majority appeared to have quite a liberal view, and one may speculate that it is not very different from young people without a disease experience.

The overall liberal picture is supported in the National Public Health Report (Danielsson et al., 2012) which reports that the percentage of young persons (aged 16-24) who have sex on the first night have increased since 2000, with the greatest increase being among young female adolescents (16-17 years of age). The number of sexual partners has continued to rise while the age for sexual debut has not changed, it is slightly over the age of 16 and not very different from other Nordic countries (Danielsson, et al., 2012). In a study investigating sexual risk-taking among childhood cancer survivors, aged 15-20 years and at least 5 years post-diagnosis, those with a longer time since diagnosis or those having better psychological health reported significantly less unprotected sex at their last sexual encounter. In addition, a high psychological well-being was significantly connected to less likelihood of an early sexual debut (Klosky, et al., 2013). According to their self-reports, the majority of the participants in the online discussions have had their sexual debut, slightly more females than males, but at what age is unknown. A study comparing childhood cancer survivors to their siblings (aged 14-20 years) revealed no significant differences regarding having had sex or not and age at first sexual encounter (Klosky et al., 2012).

A satisfactory sexual life is important for well-being and health and it is part of our role as health care professionals to focus on factors that maintain health, including providing relevant information (National Institute of Public Health, 2012). Therefore, it is noteworthy that when we explicitly asked about the role of the health care sector, many

participants in the focus groups mentioned that they had received a limited amount of information and support regarding sexuality and sex (results will be presented elsewhere). This needs to be taken into account in the future and were health care professionals, e.g. registered nurses, can play an important role in approaching young persons about sexual health, especially considering that research has shown that adolescents themselves do not bring up sexual matters at health visits to physicians (Alexander, et al., 2014). The participants in the focus groups also described physical concerns, such as vaginal dryness and erectile dysfunction, referring to definitions of sexual dysfunction (Fugl-Meyer & Fugl-Meyer, 2010). However, most often they did not relate these concerns to the cancer experience, which could mean a lack of information and support from health care professionals in this area. Some previous research has shown that survivors with sexual dysfunction report lower levels of HRQoL and higher levels of depression and anxiety (Bober, et al., 2013; Zebrack, et al., 2010) and that those feeling worse engage in more sexual risky behaviours (Klosky, et al., 2013). Therefore, efforts and further research is warranted, preferably with a quantitative design where conclusions could be drawn with regards to the prevalence and knowledge of sexual dysfunction and if differences exist within different diagnoses.

## **7.1 METHODOLOGICAL CONSIDERATIONS (STUDIES I-III)**

An overall strength within this thesis is the multi-method approach, which was used in order to investigate the topic from different perspectives. To test the psychometric properties of KIDSCREEN-27 a Rasch analysis was performed in study I; a combination of qualitative interviews and data from the HRQoL instrument (KIDSCREEN-27) was used in study II; quantitative methods were utilised in study III; and finally study IV contains written online focus group discussions.

The importance of using valid and sensitive measurements, and ensuring the reliability and validity of the measure relevant for the population under study, was achieved by performing a Rasch analysis (study I) before further evaluations were performed. However, the development of the KIDSCREEN instrument was ambitious throughout Europe, with satisfactory cross-cultural validity in the 13 countries tested, including Sweden (Robitail, et al., 2007). Since it is a generic instrument it was suitable to use for both the survivors and the comparison group. Still, the limited use among persons with a childhood cancer experience compelled us to further test the instrument psychometrically. Among other analyses, internal scale validity, person response validity, i.e. construct and content validity (Bond & Fox, 2007; Kazdin, 2010) and item reliability index (person versus item map) were examined with acceptable results. KIDSCREEN-27 was further validated in the sense that self-reported ratings of HRQoL were compared to interview data (study II) and revealed that data were in congruence with each other. It has been recommended, when using a distribution-based method (i.e. effect size calculations) for investigating clinical significance, to use it together with a meaningful external anchor, in this case interviews (Wyrwich, et al., 2005).

A further strength with the present study is that results are based on a representative sample from a national cohort consisting of all school children diagnosed with cancer during a 2-year period (2004-2006) and compared to an age-matched comparison group, randomly drawn from the general population. The distribution of diagnoses in the sample used for studies I-III shows an almost similar distribution (Table 2) to the



overall Swedish and Nordic distribution (Gustafsson, et al., 2013). Furthermore, the slightly higher incidence among boys (ratio 1.17:1) (Gustafsson, et al., 2013) is to some extent exaggerated in the sample at follow-up (ratio 1.4:1, 37 males and 26 females), however not reaching the ratio at the time of diagnosis (1.5:1, 87 males and 58 females). The slight difference, although not significant, between time at diagnosis and follow-up is probably due to the fact that more males than females were among non-participants at follow-up (19 males and 10 females). In general, incidence differs from year to year and the sex distribution ratio (1.17:1) is calculated among all individuals diagnosed from 1984–2010 (Gustafsson, et al., 2013) and therefore does not show short term fluctuations, with regard to sex distribution, that might occur.

However, there are limitations to the study. Although the sample is from a national cohort it is a small sample, leading to low power which increases the risk for type II errors, especially in subgroups analyses (*threat to statistical conclusion validity*) (Kazdin, 2010). As evident from previous research, that certain diagnoses and treatment types (e.g. tumours of the CNS and those treated with intensive treatments) contribute to lower ratings of HRQoL (Bradley Eilertsen, et al., 2012; McDougall & Tsonis, 2009; Mört, et al., 2011a, 2011b; Sundberg, Lampic, Björk, Arvidson, & Wettergren, 2009) was found in study II, regarding treatment (Table 4). Table 6 shows that the majority of those treated for solid tumours received combined treatment why the beta-values were relatively high, when including both type of diagnosis and treatment in the same multiple regression model (study III). However, these variables (type of diagnosis and type of treatment) did not reach significant influence over self-rated independence, as shown in the multiple regression model (study III). In addition, the  $R^2$ -values were in general small (study II and III), still *Physical well-being* and *Psychological well-being* showed somewhat higher values (11.6%; 17.4%; 18.9%), both when analysing survivors separately and survivors and the comparison group together. However, it indicates that the included independent variables (sex, age, diagnosis, treatment, group; survivors or comparison group) did not explain that much of the variance in the ratings by the participants, for the dependent variables (the Independence dimension and the four KIDSCREEN-27 dimensions) (Pallant, 2010).

When performing a study with a longitudinal approach, i.e. several data collections over time, there is always a risk of attrition and thereby *internal validity* due to subjects declining further participation, having moved to another city or have died (Kazdin, 2010). Since the reasons for non-participation are mostly unknown, one could only speculate on the reasons. On the one hand it may reflect the person's decision to move on with life or on the other hand, those feeling more troubled about their condition may have declined participation. Therefore, survivors that chose to participate could have been those feeling at their best and those most willing to reveal their story. Conversely, those who participated may have wanted to reveal their story due to having a troublesome life. However, no significant differences with regard to event (relaps or secondary malignancy) were seen among participating and non-participating survivors, a median time of five years post-diagnosis.

Since the sample for studies I-III is relatively heterogeneous with regard to different diagnoses, ages and treatment given, there are additional difficulties in sub-group analyses (*threat to statistical conclusion validity*) (Kazdin, 2010). However, from an *external validity* point of view, a heterogeneous sample is favoured with regards to generalizability to other countries and other studies focusing on childhood cancer survivors. There is always a balance between internal and external validity and to what degree one should enhance one or the other. Some of the limitations found across

studies, as stated in the introduction section, included small sample sizes and heterogeneous samples (Klassen, et al., 2011), are also present in the current studies. Therefore, large-scale studies where robust sub-groups analyses could be performed, encompassing several countries and using the same measurements would be beneficial.

A limitation and a threat to *external validity* is the representativeness of the comparison group and the response rate, 54%. The sample was randomly drawn from the general population and from the whole of Sweden but the representativeness is mainly unknown. Those feeling at their best might have been the majority of those who chose to participate and therefore, not representative with regard to the general population. Some of the eligible participants stated why they chose not to participate. These reasons included not having enough time, not being interested or their parent/parents did not want them to participate. However, since the group was randomly drawn, health problems that generally exist among people in that age range (11-23 years) such as asthma, diabetes, hay fever, depressive symptoms and neuropsychiatric disabilities, were present. We also compared the present comparison group to European norm data, collected in 13 European countries, approximately ten years ago, for KIDSCREEN-27 among those aged 12-18. The results from the collected European data showed acceptable representativeness in order to provide norm values for the KIDSCREEN instrument (Berra et al., 2007; Robitail, et al., 2007; The KIDSCREEN Group Europe, 2006). Regarding our comparison group, when compared to the European norm data, for the dimension *Physical Well-being* there were no significant differences when comparing the T-value means with independent samples t-test between females and males. For *Psychological Well-being* and *School Environment*, our comparison group (both females and males) displayed significantly higher T-value means than did the European norm data. For *Social Support & Peers* there were no differences between females while males in our comparison group had significantly higher T-value means than males in the European sample. In conclusion, the comparison group used in studies I-III generally showed higher T-value means than the European norm data and the reasons for this are not totally clear. However, when comparing only to The Netherlands and their norm data (Engelen et al., 2011), one dimension (*Physical Well-being*) displayed somewhat higher T-value means than our comparison group did while the dimension *School Environment* showed a little higher values in our comparison group. The other two dimensions showed similar T-value means. Still the collected norm data within the KIDSCREEN project is ten years old, something which might have an impact. More recently published data (Statistics Sweden, 2011-2012) showed that 85% of children and adolescents (n=1492, aged 10-18 years) considered their health to be very good or good. When comparing to a similar question (In general, how would you say your health is?) embedded in the KIDSCREEN-27, approximately 90% of participants in our comparison group answered excellent, very good or good, indicating rather similar figures.

Several steps were taken to increase the response rate for the comparison group. Reminders were sent by mail and additional telephone calls were made. However, as of today, response rates from surveys may vary from 50-75% (Trost, 2007). Therefore 54% can be considered acceptable. Additionally, collecting data from control- or comparison groups is more difficult than that of collecting from patients groups or other special interest groups (Trost, 2007) due to lower incentives for controls.

Efforts were also made to compare the results obtained from the DCGM-37 (Independence dimension) from the survivors to European norm data for DCGM-37.

However, that norm data was not considered relevant since it is old (from 2003) and it also refer to diagnoses other than cancer (The European DISABKIDS Group, 2006).

The data collection for studies I-III was telephone-based, which was favourable with regards to less missing data. Also, clarifications could be given to participants if they perceived certain questions as difficult. On the other hand, there is a risk of “helping” the participant too much and telephone-based interviews could be considered more anonymous, since they lack non-verbal interaction and body language. However, it is a suitable method when wanting to reach many geographically, it saves time and is less expensive. A study with Swedish adolescents and young adults (13-23 years), randomly selected, showed that type of data collection (telephone interview or postal questionnaire) only had limited impact on their self-reported health and emotional distress (Wettergren, Mattsson, & von Essen, 2011). Significantly higher mean values were seen for the telephone administered interviews in four dimensions: physical functioning, vitality, social functioning and mental health, but with small effect sizes. The samples used in studies I-III showed no clear pattern with regard to those that preferred to receive the questionnaire by mail instead. In the sample of survivors, two of them preferred to answer the questionnaire at home, one female, 17 years, and one male, 14 years. The female expressed balance difficulties while the male did not express any long-term disability. In the comparison group, 27 individuals preferred to answer the questionnaire at home, with an equal distribution of females and males, of different ages and the majority (n=25) without any long-term disability. The recommended age limit was exceeded by a number of participants for KIDSCREEN-27 and DCGM-37 but there were no particular reservations expressed by participants during the interviews.

## **7.2 METHODOLOGICAL CONSIDERATIONS (STUDY IV)**

Earlier research has indicated that sexuality could be a sensitive topic and difficult to discuss in a focus group format (Kent et al., 2012). Yet we found that participants in the online focus group discussions interacted well giving the study a rich material. This was also evident in a study by Flynn et al. (2011) where focus groups regarding sexual function were performed with adult cancer patients during or after treatment. Among our participants, different views were expressed but there were seldom disagreements. The format can have a facilitating effect, i.e. participants can feel relief when hearing that others have had similar issues and/or problems (Flynn, et al., 2011). On the other hand, participants may answer with what they think is expected of them or other participants may influence their responses (Bredart, Marrel, Abetz-Webb, Lasch, & Acquadro, 2014). Nonetheless, this method for data collection seems to be relevant for young persons and allows for the possibility of reaching many people. A pilot focus group was performed with young relatives of the moderators before data collection started, and no particular problems were encountered. Other research has also pointed out the benefit of using the internet for young adults affected by cancer, with the ability to stay connected to friends and families (Kim & Gillham, 2013). Kim and co-workers analysis of illness blogs revealed that the content provided good insight with regard to young people’s experiences of their cancer disease. However, no sexual matters were mentioned in the blog content (Kim & Gillham, 2013). Moreover, as expressed in evaluations by participants, more than one online discussion for each group would have been beneficial. On the one hand, this would have provided more in-depth knowledge and the participants might have felt more comfortable when participating a second time with the same group members. On the other hand, participating more than once might

not be beneficial to achieve a spontaneous discussion. To consider is the fact that older participants generally discussed more in detail than did younger participants. This may have been because older participants are more mature than younger participants and can more easily express their thoughts and experiences in writing. Efforts were made to form focus groups with participants of approximately the same ages; however this was not always possible.

To discuss aspects of *trustworthiness*; efforts were made to have representation of both sexes and different diagnoses in each online focus group discussion, along with the moderators different experiences and backgrounds. This contributed to a greater variation of the aspect under study, i.e. the *credibility* (Graneheim & Lundman, 2004; Patton, 2002). It was noted that none of the participants explicitly revealed if they were homo- or bisexual. It is not known if they did want to do it or if they did not find it important or relevant. Therefore, the variation in the sample regarding sexual orientation is unknown and could be a threat to the credibility of the study (Graneheim & Lundman, 2004). However, sexual orientation was not brought up by moderators as a topic for discussion and throughout the focus groups the more gender-neutral term “partner” was used. To enhance transparency and thereby trustworthiness; the authors were presented to the participants and the readers, including their occupation and experiences and representative quotations from different focus groups were used, including interactions between participants (Sandelowski, 1994; Tong, Sainsbury, & Craig, 2007). Furthermore a continuous discussion was held among all authors during the data collection process regarding saturation of data and during the analysis procedure regarding the development of sub-categories and categories in order to make sure that the categories covered the data (Graneheim & Lundman, 2004) and were mutually exclusive. A seminar presentation was held for other researchers to receive input regarding less obvious parts and/or possible missed parts. However, there is a risk for *selection bias* and lack of variation, i.e. those that chose to participate were the ones most willing to reveal their experiences, either having more or less problems. Still, eligible participants might have declined participation due to that they felt the topic being too sensitive to discuss, which could be an explanation to the relatively low response rate (36%). Some of the eligible participants gave reason why they did not want to participate including that they felt they had left their experience behind and did not want to talk about it any longer. Other reasons included being so young at diagnosis that they did not think they could contribute with anything, while others gave no reason. Most often, response rate is not taking into considerations in qualitative studies, still it was found relevant here due to the considerably large sample (n=400) and the fact that some quantitative analyses were performed with regard to participants vs. non-participants (see under *transferability*).

Regarding *dependability*, the process of performing online focus group discussions, as with any other interviews, involves acquiring new knowledge of the proposed subject which influences what follow-up questions that will be posed (Graneheim & Lundman, 2004). Our topics and main questions were the same for all participants but the posing and timing of questions and follow-up questions developed during the process, in a dialogue among moderators, meaning that not all discussions were the same. It can be considered that gaining new knowledge during data collection enriches the study and each focus group contributed to a variation in the data material. Most of the moderators had limited experience in using this chat format, but were experienced in interviewing. Still, there might be a risk of not always having posed the relevant follow-up questions or sometimes having misinterpreted the intended meaning, i.e. *investigator effect*, (Patton, 2002) due to the fast interactions between participants in some groups.

Furthermore, oral discussions, performed live, may on the one hand have revealed a richer material due to the fact that we express ourselves more easily in oral communication along with the presence of body language. On the other hand, considering the sensitive nature of the topic, the written format may have been the one in favor. Still, more survivors of tumours of the CNS declined participation and reasons for that might be perceived cognitive difficulties (Boman, Lindblad, & Hjern, 2010) within this group or other obstacles possibly related to this specific diagnosis. Some eligible participants were excluded due to explicitly stated having cognitive difficulties and the majority of those were survivors of tumours of CNS. We could therefore have missed participants due to that they may have felt the online chat format being demanding, and requiring them to respond rather quickly. Still, some of the participants were open with that they, for instance, had difficulties such as with dyslexia.

Finally, *transferability* (Graneheim & Lundman, 2004) can be achieved by describing the context and the method, including participants, data collection and analysis as well as a clear description of the findings. We tried to provide a clear description of our participants and also present the number of non-participants including some comparisons between the groups (Tong, et al., 2007). Our notion is that the findings from the online focus group discussions could be transferred to other young persons, both to those without but also to those with a long-term health conditions, where there is a the risk of experiencing complications affecting sex and sexuality. Furthermore, online focus group discussions can play an important role when constructing new measures that accurately could evaluate sexuality and sexual experiences among young persons with an experience of childhood cancer. These instruments could be of help for registered nurses and other health care professionals, meeting young survivors of childhood cancer and it would further make it possible to achieve a clearer picture and provide stronger evidence regarding important aspects of HRQoL in this group.

## 8 SUMMARY AND CONCLUSIONS

Health care professionals meeting persons with a childhood cancer experience require extended knowledge and understanding to be able to meet their needs and deliver appropriate care. This is essential and relevant in light of the description of health; as encompassing the experience of well-being, to feel well and to be able to fulfil smaller and larger life projects. Health care professionals from different areas of the health care sector will encounter persons with a childhood cancer experience, due to the increased survival rates in this group. Therefore it is of great importance to investigate survivors own perceptions of their health and life situation including health-related quality of life (HRQoL). The overall aim was to investigate self-reported HRQoL with special focus on independence and sexuality among young persons treated for cancer during childhood.

KIDSCREEN-27 seems to, overall be an appropriate and feasible instrument to use for young persons treated for childhood cancer, although unidimensionality was not totally supported for one dimension. Clinically relevant thresholds were established for the instrument and KIDSCREEN-27 could be further useful in clinical practice with the aim of identifying survivors in need of support, using mean values of 45 or less as thresholds. Overall, the self-rated HRQoL was in parity or higher (*School environment*) among those treated for cancer during childhood compared to those in the comparison group, which is promising with regard to the survivors. Still, a small group expressed that their cancer experience influenced their daily lives and that they felt different from others, which was shown through lower self-reports of HRQoL.

One of the studied aspects was independence, important for young people developing their identity. The findings revealed that, a median time of five years after diagnosis, young persons with a cancer experience have reached an acceptable level of independence, measured by the DISABKIDS Chronic Generic Module (DCGM-37) and compared to the levels during initial cancer treatment. The survivors also rated their independence higher than the comparison group. However, none of the clinical or demographic variables had any impact on self-rated independence. The results are promising since they reflect survivors' confidence in the future and show that they consider themselves as being able to live an autonomous life without being impaired due to their cancer experience.

Regarding how young persons with a cancer experience from childhood think and reason about sexuality and sexual experiences; the result showed that the majority of the participants in written online focus groups described sex as a given part of life. The discussions revealed that participants overall were relatively comfortable discussing and reason around the topic. However, some of them described worries, feelings of shyness, being insecure and falling behind peers and physical concerns including vaginal dryness and erectile dysfunction. Therefore, these online focus groups can be seen as a point of departure for further research with a quantitative approach, focusing on psychological aspects and physical concerns related to sexuality in this group of young people.

## 9 IMPLICATIONS FOR NURSING

The course of life for young persons with a childhood cancer experience, as of any adolescent and young adult, includes aspects related to the experience of health such as establishing and creating their own identities and lives where partner relationships, sexuality, and having families and children become important.

Central in the discipline of nursing is to meet the needs of another human being and build a relationship to him or her. The importance is the 'with' rather than the 'for'. It is not only something that the nurse does for the patient, or the young person with a cancer experience; it is something mutual between them (Wiklund Gustin & Lindwall, 2012). To bear in mind is the description of health as encompassing the experience of well-being, to feel well and to be able to fulfil smaller and larger life projects (Dahlberg & Segesten, 2010). As a registered nurse, by listen to the person's own experience how he or she views and interprets life the story shapes and enriches with both their experiences (Wiklund Gustin & Lindwall, 2012). With this as a framework, an honest and strong relationship can be created, crucial to understand thoughts, desires and needs of a particular individual and when designing the follow-up care. When working with young persons with a cancer experience from childhood, knowledge about that the majority of them get along quite well and their increased levels of independence from time of diagnosis up to 4-6 years after diagnosis, can be of additional importance in these meetings.

Registered nurses are suggested to take an active role in the follow-up care with regard to the aspects studied in this thesis. Young persons with a cancer experience perceive their experience differently; some may thrive while others suffer from their problems and shortcomings and need support. By letting young persons with a cancer experience during childhood assess their own HRQoL in the follow-up care, using KIDSCREEN-27, those in need of extra support and interventions can be identified. A recommendation is to use the KIDSCREEN dimensions mean values of 45 or less as thresholds. It is of importance to identify those in need of support; not least since a number of late effects after cancer diagnosis and treatment are well-known, will increase with time and the fact that over- and/or underreporting of late effects and problems may exist in this group.

Further, registered nurses are suggested to take an active part with regards to information and support of sexual matters in the follow up care, as well as in research, especially since there is an association between sexual dysfunction and lower levels of HRQoL. Information meetings in groups, with persons with a similar experience of cancer during childhood, along with individual support and information, might be appropriate, and possibly together with other professions, such as mid-wives. To be open, understanding, creative and confirmative is crucial (Dahlberg & Segesten, 2010), and when dealing with aspects such as sex and sexuality, potentially sensitive for both the nurse and the survivor, this might be of extra importance. Special skills in communication and counselling might be needed, still by starting to listen and engage in the meeting, is a first step.

## 10 SUMMARY IN SWEDISH

Medicinska framsteg har inneburit att samhället har en växande grupp av unga människor som genomgått behandling för cancer i barndomen. Detta innebär i sin tur att fler yrkeskategorier inom hälso- och sjukvården kommer att möta dem. Hälsa är en viktig del av en individs nuvarande och totala livssituation, en integrerad del av livet och den viktigaste aspekten att ta i beaktande inom hälso- och sjukvården. Mot bakgrund av detta är det viktigt att undersöka dessa unga människors upplevelser av sin livssituation, innefattande hälso-relaterad livskvalitet, självständighet och sexualitet, där de två senare områdena är viktiga för unga människor, på väg ut i livet. Det övergripande syftet med denna avhandling var därför att undersöka självrapporterad hälso-relaterad livskvalitet med speciellt fokus på självständighet och sexualitet hos unga människor behandlade för cancer under barndomen.

Den första delstudien är en metodologisk studie och de tre efterföljande har en tvärsnittsdesign men där en studie (studie III) också innehåller longitudinella data, insamlade vid flera tillfällen över en längre tidsperiod. Studie I-III baseras på data från en svensk kohort av unga personer som behandlats för cancer under barndomen (n=63; 12-22 år). Totalt samlades data in vid fyra datainsamlingstillfällen, från tiden vid diagnos fram till 4-6 år efter diagnos. Vid sista tillfället ingick också en jämförelsegrupp som bestod av ungdomar i samma åldersspann men utan cancererfarenhet (n=257). Studie IV baseras på data från en annan datainsamling, genomförd med en större grupp ungdomar som hade genomgått cancerbehandling under barndomen eller ungdomsåren (n=133; 16-25 år).

I studie I testades mätgenskaperna och användbarheten hos frågeformuläret (KIDSCREEN-27) i gruppen barn och ungdomar som behandlats för cancer med hjälp av modern testteori, så kallad Rasch-analys. KIDSCREEN-27 består av fem områden som mäter hälso-relaterad livskvalitet avseende fysiskt välbefinnande, psykologiskt välbefinnande, autonomi och föräldrarelationer, socialt liv samt skolmiljö. Formuläret utvärderades också utifrån om de olika frågorna fungerade olika för de som haft cancer och de utan cancererfarenhet.

I studie II jämfördes intervjuresultat med självskattningar av hälso-relaterad livskvalitet utifrån frågeformuläret (KIDSCREEN-27) i gruppen barn och ungdomar som haft cancer, 4-6 år efter diagnos. Skattningar avseende livskvalitet jämfördes också mellan de som haft cancer och jämförelsegruppen, där variablerna kön och ålder togs i beaktande.

I studie III mättes deltagarnas självständighet med ett annat livskvalitetsinstrument (DISABKIDS Chronic Generic Module, DCGM-37). Resultat jämfördes mellan tiden vid diagnos och 4-6 år efter diagnos samt med skattningar från jämförelsegruppen.

I studie IV genomfördes fokusgrupper online, i skriven form, och fokus var kring sexualitet och sexuella erfarenheter. Deltagarna chattade i smågrupper med en eller två moderatorer som ställde frågor.

Resultaten visade att KIDSCREEN-27, till stora delar förutom dimensionen autonomi och föräldrarelationer, är användbart i en grupp av barn och ungdomar, behandlade för cancer (studie I). Vidare visade resultaten (studie II och III) att majoriteten av de unga som behandlats för cancer mår väl, enligt deras självskattningar av sin hälso-relaterade



livskvalitet (fyra av de fem KIDSCREEN-27 dimensionerna samt självständighet) samt från resultatet av de semi-strukturerade intervjuerna. Resultaten visade även att nivån av självständighet steg från tiden vid diagnos fram till 4-6 år efter diagnos samt att den då var högre än hos jämförelsegruppen. En mindre grupp av de som behandlats för cancer uttryckte dock att de kände sig annorlunda och skattade sin hälso-relaterade livskvalitet lägre än övriga. Generellt skattade äldre deltagare (17-23 år) samt kvinnor sin hälso-relaterade livskvalitet lägre.

Fokusgrupperna (studie IV), genomförda online, gav ett rikt material där ungdomarna, som genomgått cancerbehandling under barndomen eller ungdomsåren, diskuterade, interagerade och delgav sina tankar och erfarenheter rörande sexualitet och sexuella erfarenheter. Mestadels beskrev de sexualitet och sexuella erfarenheter i positiva ordalag men de delgav även blygsel, begränsade erfarenheter, rädsla för att hamna efter kompisar samt fysiska problem/åkommor såsom torra slemhinnor och erektionssvårigheter.

Sammanfattningsvis visade resultaten att majoriteten av unga människor med en cancererfarenhet från barndomen mår ganska bra med avseende på självskattad hälso-relaterad livskvalitet och med speciellt fokus på självständighet och sexualitet. En mindre grupp verkar vara i behov av ytterligare stöd, vilket visade sig i lägre skattningar av hälso-relaterad livskvalitet samt avseende sexuella aspekter såsom psykologiska och fysiska bekymmer. I framtiden är det viktigt att fortsätta följa dessa unga människor då det är väl känt att långtidseffekter av behandlingen kan ge hälsoproblem av olika slag på längre sikt. Vidare är det viktigt att fortsätta studera sexualitet och sexuella problem då kunskap inom området är begränsad. Här kan sjuksköterskor spela en viktig roll, både i omvårdnadsarbetet såväl inom forskningen.

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