Distress among Adolescents with Cancer

MARIANN HEDSTRÖM
Dissertation presented at Uppsala University to be publicly examined in Aulan, Döbelnsgatan 2, Uppsala, Thursday, April 21, 2005 at 13:15 for the degree of Doctor of Philosophy (Faculty of Medicine). The examination will be conducted in Swedish.

Abstract

The primary aim was to investigate the distress perceived by adolescents with cancer treated in paediatric oncology. In Study I, especially distressing events for children/adolescents with cancer were identified by interviews with children/adolescents/parents and nurses. Data were analysed with content analysis. A range of physical and emotional concerns was identified. Physical concerns are of a rather similar nature across age groups. Emotional concerns vary more between age groups. For children 8-12 years, emotional concerns are rather frequent. In Study II distressing and positive aspects related to some care situations for adolescents with cancer were identified by interviews with adolescents and nurses. Data were analysed with content analysis. A range of negative aspects, e.g. fear of alienation, fear of dying, altered appearance and physical concerns, as well as positive aspects, e.g. positive relations to staff and being well cared for were identified.

The aspects of distress identified in Studies I and II formed the basis for a structured interview-guide, used in Studies III and IV. Adolescents, recently diagnosed with cancer, physicians and nurses were interviewed by telephone about distress, anxiety, depression and well-being experienced by the adolescents. Adolescent ratings of prevalence, levels and worst aspects of distress do not necessarily agree, however, worry missing school and mucositis are among those rated with the highest prevalence, levels and those perceived as the overall worst. The findings from Study IV demonstrate that physicians and nurses underestimate the distress caused by worry missing school and mucositis. The accuracy of physician and nurse ratings of physical distress is acceptable, however, this is not the case for psychosocial distress. It can be concluded that it is crucial to consider how questions are asked when interpreting the significance of the answers, and that action on adolescent problems in relation to cancer diagnosis and treatment need to rely on direct communication.

Keywords: adolescents, anxiety, cancer, children, depression, distress, staff ratings, positive experiences, well-being

Mariann Hedström, Department of Public Health and Caring Sciences, Caring Sciences, Uppsala Science Park, Uppsala University, SE-75183 Uppsala, Sweden

© Mariann Hedström 2005

ISSN 1651-6206
ISBN 91-554-6176-X
urn:nbn:se:uu:diva-4832 (http://urn.kb.se/resolve?urn=urn:nbn:se:uu:diva-4832)
I should have liked to begin this story in the fashion of the fairy tales. I should have liked to say: "Once upon a time there was a little prince who lived on a planet that was scarcely any bigger than himself, and who had need of a friend..."

To those who understand life, that would have given a much greater air of truth to my story.

Antoine de Saint-Exupéry: The Little Prince
List of papers

This thesis is based on the following papers, which are referred to in the text by their Roman numerals:


Papers I-III have been reprinted with the kind permission of the publishers.
## Contents

<table>
<thead>
<tr>
<th>Section</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Introduction</td>
<td>9</td>
</tr>
<tr>
<td>Distress</td>
<td>10</td>
</tr>
<tr>
<td>Physical distress</td>
<td>10</td>
</tr>
<tr>
<td>Psychological distress</td>
<td>11</td>
</tr>
<tr>
<td>Social problems</td>
<td>11</td>
</tr>
<tr>
<td>Factors that may be related to perceived distress</td>
<td>12</td>
</tr>
<tr>
<td>Information</td>
<td>12</td>
</tr>
<tr>
<td>The life-threat</td>
<td>12</td>
</tr>
<tr>
<td>Resilience</td>
<td>13</td>
</tr>
<tr>
<td>Support</td>
<td>14</td>
</tr>
<tr>
<td>Care</td>
<td>14</td>
</tr>
<tr>
<td>Comments to the introduction</td>
<td>15</td>
</tr>
<tr>
<td>Aims</td>
<td>16</td>
</tr>
<tr>
<td>Methods</td>
<td>17</td>
</tr>
<tr>
<td>Overview of thesis</td>
<td>17</td>
</tr>
<tr>
<td>Studies of experiences of having cancer while being young</td>
<td>17</td>
</tr>
<tr>
<td>Studies I and II</td>
<td>17</td>
</tr>
<tr>
<td>Participants</td>
<td>18</td>
</tr>
<tr>
<td>Procedure</td>
<td>21</td>
</tr>
<tr>
<td>Interviews</td>
<td>21</td>
</tr>
<tr>
<td>Data analysis</td>
<td>22</td>
</tr>
<tr>
<td>Studies of distress among adolescents recently diagnosed with cancer</td>
<td>23</td>
</tr>
<tr>
<td>Studies III and IV</td>
<td>23</td>
</tr>
<tr>
<td>Participants</td>
<td>23</td>
</tr>
<tr>
<td>Procedure</td>
<td>26</td>
</tr>
<tr>
<td>Measures</td>
<td>27</td>
</tr>
<tr>
<td>Data analysis</td>
<td>28</td>
</tr>
<tr>
<td>Ethics</td>
<td>30</td>
</tr>
<tr>
<td>Results and Comments</td>
<td>31</td>
</tr>
<tr>
<td>Studies of experiences of having cancer while being young – summary of Studies I and II</td>
<td>31</td>
</tr>
<tr>
<td>Especially distressing aspects of disease and treatment (Study I)</td>
<td>31</td>
</tr>
<tr>
<td>Physical aspects</td>
<td>31</td>
</tr>
</tbody>
</table>
Introduction

Approximately 150 teenagers are diagnosed with cancer each year in Sweden. Some of these will be treated in adult oncology settings, e.g., many of those with testis cancer, thyroid cancer and malignant melanoma. Others, as most of the teenagers with diagnoses more common among children e.g. lymphoma, acute lymphatic leukaemia, osteosarcoma and Ewing sarcoma, will receive treatment in paediatric oncology settings. Forty years ago, almost no adolescents with cancer could be cured, while the current survival rate for adolescents treated in paediatric oncology settings is about 75% (1-3).

In Sweden, when an adolescent below the age of 18 years is suspected to have a malignancy, he or she will be referred to a paediatric oncology centre for definitive diagnosis. The treatment against cancer will start as soon as possible after the diagnosis. Depending on diagnosis and stage, treatment will be given according to international treatment study protocols, often identical for the Nordic countries. The adolescent will most often receive treatment at a paediatric oncology centre, either admitted to the hospital or at the outpatient clinic. Shared care is sometimes used, meaning that the adolescent at times will be treated at his or her local hospital in cooperation with physicians at the paediatric oncology centre. Most commonly, cancer treatment will include chemotherapy, alone or in combination with radiotherapy, surgery or bone marrow transplantation. Chemotherapy treatment duration usually varies between 3-4 months to 2 1/2 year.

The term adolescence has been defined in many ways, but it generally refers to the period between childhood and adulthood (4, 5). For the purpose of this thesis, adolescents and teenagers are considered as synonymous terms covering the period from 13 to 19 years. The overall developmental tasks of adolescence aim at preparation for adulthood, e.g. establishing identity and self-image, becoming autonomous from parents, developing a positive gender role and making career plans. When diagnosed with cancer, the adolescent will need to face a life-threatening disease, and lengthy, arduous treatments will lead to increasing dependence on others which may challenge the achievement of these tasks (6). A cancer diagnosis has been proposed to be especially problematic during adolescence (6-8). This is the reason for which we chose to focus the research presented in this thesis on distress in adolescents with cancer. In addition, treatment and other cancer related circumstances change continuously and the demands on medical care increase.
Thus, it is important to perform regular investigations, as results from older studies may no longer be valid.

Distress

Physical distress

Symptoms from disease and side-effects from treatment vary tremendously depending on disease and treatment, but some problems are nevertheless common and frequently reported among adolescents with cancer. Physical side-effects of treatment are often regarded as the worst aspects of disease, especially pain from treatments and medical procedures (9-12). Other commonly reported physical side-effects from treatment are nausea, vomiting, fatigue, mouth sores and low blood counts, which may lead to susceptibility to infections, bleeding tendency and anaemia. Changes in appetite and various gastrointestinal and skin changes may also occur (13-15).

Teenagers with cancer have been reported to be more troubled by physical restrictions and changes than younger children (16). Hair-loss has been reported as a major cause of concern in young people, not only as a transient cosmetic effect, but with a deep influence on peer relationship (17). Looking different than peers can evoke feelings of inadequacy which may lead to avoiding one’s peer group, and consequently isolation and loneliness (18).

Some studies compare the perceived distress of various aspects of disease- and treatment, e.g. Hinds et al (19), using the Symptom Distress Scale (20), by means of which 33 newly diagnosed adolescents rated how distressing various aspects were to them. The top three sources of distress were feeling tired, concern with appearance and lessened ability to get around. Collins et al (21), using an adapted version of the Memorial Symptom Assessment Scale (MSAS 10-18) among 159 children and adolescents, 10 to 18 years of age, reported prevalence, frequency and severity of symptoms as well as distress caused by symptoms. The most common symptoms were lack of energy, pain, drowsiness, nausea, cough, lack of appetite and psychological symptoms as sadness, nervousness, worrying and irritability. The prevalence of a symptom was not necessarily related to its frequency, severity and the distress caused by it. Symptoms causing high distress were difficulty swallowing, insomnia, mouth sores, “I don’t look like myself”, hair loss, skin changes, vomiting and problems with urination. In these studies (19, 21), as in most other studies aiming at investigating paediatric symptom distress, instruments designed for use with adults were adapted and used (22).
Psychological distress

Fears and worries related to disease and treatment include e.g. painful medical procedures, side effects of chemotherapy, effects on sexuality and reproduction, relapse and death (23, 24). Most estimates of self-reported psychological distress in adolescents on treatment for cancer have been consistent in demonstrating similar levels of anxiety (25-27) and depression (25-28) as reported by healthy controls. However, the prevalence of self-reported depression has varied somewhat between studies. Tebbi et al (29) reported a depression prevalence of 13% among adolescents, on or off treatment for cancer, whereas Canning et al (30) reported a prevalence of 6%. The depression prevalence was 25% among a healthy control sample in the Canning and co-workers study. von Essen et al (27) reported a depression prevalence of 6% among children/adolescents on cancer treatment and 17% among youngsters off treatment. Some investigators have reported that self-reported psychological distress among children and adolescents with cancer did not change with sex and age (26, 30, 31). Others have demonstrated that girls reported higher levels of anxiety (25) and depression (25, 29) than boys.

When associations between physical and psychological distress have been studied, it has been reported that among children and adolescents with cancer, if children/adolescents perceived their physical appearance as better, they were more likely to rate lower levels of depressive symptoms and social anxiety and higher self-esteem (32). It has also been demonstrated that pain intensity was associated with levels of depression and anxiety among children with chronic pain (33). Recently, in a longitudinal study of children and adolescents with cancer, Varni et al (34) reported that pain and emotional distress were associated cross-sectionally, but did not predict each other.

Social problems

The lengthy treatments, susceptibility to infections and poor health status will lead to social problems for many adolescents, e.g., interruption of school and decrease in relationship with others (24, 35). The disruption of school work has been reported as a major stressor to adolescents with cancer (35-37). In a literature review on school experiences of children with cancer, Vance and Eiser (38) state that although school absences generally decrease over time, missing school remains a great concern for many children and adolescents with cancer. However, the authors conclude that the majority of research has focused on school attendance, and that more consideration needs to be given to the child's other activities, e.g. participation in physical activities.
Factors that may be related to perceived distress

Information

The International Society of Paediatric Oncology (SIOP) Working Committee on psychological issues in paediatric oncology has published guidelines that encourage physicians to share developmentally relevant information with paediatric patients. For adolescents of legal age, the guidelines call for a full and legally mandated power to make their own decisions regarding treatment. Younger children have a moral right to, at an age appropriate level, participate in decisions (39). Previous studies have shown that adolescents wanted information about all aspects of disease and treatment (40), particularly on diagnosis, prognosis and treatment (24, 41). Open information about diagnosis and prognosis has been demonstrated to be negatively associated with anxiety and depression among 8-16 year old children with cancer (42). In a recent study, 74 newly diagnosed adolescents and 39 adolescents 1-3 years from diagnosis were asked to rate the importance of certain categories of cancer-related information. The newly diagnosed adolescents rated importance of information about getting back into school with the highest level, followed by ways to deal with procedures, relationships with friends and family, finish treatments and adjusting to the cancer diagnosis. For adolescents 1-3 years from diagnosis, ratings of importance of information were generally higher, especially information about depression, death and helping others with cancer (43). This finding is consistent with results by Bradlyn et al (44), who concluded that among paediatric oncology professionals, information was considered more important during treatment than at the time of diagnosis. In addition, information about medical topics was generally considered more essential than information about psychological topics. When young Australians, 12 to 24 years of age, on or off treatment for cancer, were asked about their needs of and preferences for information, approximately two thirds wished more information, especially about the likelihood of recurrence, side effects, emotional reactions, long-term effects and ability to have children (45). As cancer treatments usually are lengthy, the importance of renewed, age appropriate information during the disease trajectory cannot be overlooked.

The life-threat

Even if the survival rates are immensely more positive now than some decades ago, cancer is a threat to life. Adolescents have usually developed an adult understanding of death in the sense that they comprehend that death is irreversible and inevitable. However, an orientation towards the future, and a sense of invulnerability, both typical aspects of adolescence, make it difficult for some adolescents to acknowledge that they might die young from cancer.
Stuber and coworkers (48) investigated the appraisal of life-threat and trauma responses among children and adolescents, scheduled for BMT with a mean 5-year survival prognosis of 50%. The appraisal of life-threat increased significantly with age, but there was no correlation between appraisal of life-threat and reported symptoms of traumatic stress. In concordance, Neville (26) reported that presence of metastasis or stage of illness were not related to psychological distress among adolescents with cancer. These findings are supported by those of Grootenhuis and Last (31) who compared ratings of anxiety, depression, control strategies and defensiveness between 43 children in remission and 41 children not in remission, aged 8-18 years. It was demonstrated that remission vs. not remission did not explain levels of depression or anxiety, many children not in remission reported positive expectations about the course of their illness. Hence, findings indicate that there is no relation between the objectively estimated survival perspective and self-reported levels of anxiety and depression among adolescents treated for cancer.

Resilience

Despite the challenges adolescents with cancer face, it has been reported that many adolescents try to respond to cancer by being positive and making efforts to normalise their social world (49). Resilience, the process of identifying or developing resources and strengths to manage stressors, is generally recognised as a multidimensional concept (50, 51). Factors enhancing resilience include deriving meaning from the experience and using confrontive and optimistic cognitive strategies (51). Hopefulness is reported as essential for adolescents with cancer to the efforts to survive or to accept loss (52). It has been reported that, when compared to younger children, adolescents with cancer employ a greater repertoire of cognitive strategies to cope with distress (16, 53) and report using them in a wider range of stressful situations related to their illness (16). Landolt and co-workers (54) found that among children with cancer, the most frequently used strategies were avoidance, cognitive restructuring and distraction. Older children used more active coping, distraction and social support than younger. Lower physical status was associated with the strategies avoidance and seeking support. Frank, Blount and Brown (55) found that avoidance coping was associated with higher ratings of anxiety and depression among children and adolescents with cancer. Thus, it appears as if many adolescents use a great repertoire of strategies to manage cancer stressors, however, the results of the two latter studies imply that avoidance strategies are associated with physical and emotional strain for adolescents with cancer.
Support

Adolescents have reported that during treatment for cancer, their circle of friends has become smaller, but the friends who have remained have become closer (35, 37). It has been shown that parents and a close friend are reported as the greatest sources of social support for adolescents with cancer (56-58). Compared to healthy adolescents, adolescents with cancer perceive relations to parents as more supportive (56). Still, all adolescents with cancer may not perceive the closeness to parents as entirely positive. Parents can act overprotective, and adolescents may find it hard to gain privacy and independence (35, 37). Continuation of school and social activities as soon as medically feasible is recommended as important opportunities to normalise a very difficult experience and prevent future social and peer problems (36, 59).

Care

In order to provide adolescents with cancer with good care it is necessary to know what aspects of care the adolescents perceive as important. Kelly, Mulhall and Pearce (60) argue that there is a need of a more robust knowledge base to optimise adolescent cancer service. In a study by Wilkinson (61), adolescents with cancer were asked to prioritise six items according to importance. The hierarchy of the items was: 1. getting better; 2. being managed in a specialist centre; 3. keeping up with education; 4. accessibility for parents; 5. accessibility for friends, and 6. being close to home. According to the adolescents, facilities as television and computers were important, but most important was the expertise of the staff.

In a Delphi study, paediatric oncology nurses were asked to identify behaviours facilitating children’s (0-18 years of age) coping efforts with disease and treatment (62). Nurses assigned the highest importance to patient education, open and honest communication, parents’ participation in care, being the child’s advocate and positive reinforcement. Australian adolescents with cancer have identified qualities of health professionals that facilitate communication as the ability to listen, genuine concern, expertise and honesty (45). Evaluating an adolescent cancer unit by interviewing adolescents, parents and staff, Kelly and co-workers (63) reported that the physical and social structure of the unit, the sense of "being in the same boat", the family-like atmosphere and the expertise of professionals contributed to a therapeutic milieu. Ritchie (64) has concluded that 'the goal of a healthy relationship with adolescents with cancer is to help them achieve a realistic sense of normalcy and to support their continued psychosocial development toward young adulthood', p 173.
Comments to the introduction

While reviewing the current literature on the situation of adolescents with cancer, a more complete picture emerges, than what was known a few years back while the studies forming the basis of this thesis were planned. However, there are still not many studies investigating the perceived distress among adolescents with cancer, and in most studies the instruments measuring various aspects of distress were not constructed to specifically investigate distress experienced by adolescents with cancer. The primary aim was to explore what aspects of disease and treatment that adolescents perceive as distressing and thereafter to investigate the prevalence and levels of these aspects. The aim was not to investigate which factors that possibly can explain if and to what extents adolescents perceive various aspects as distressing. My personal reflection is that no study, no matter how well-designed, can catch the entire spectrum of emotions, needs and experiences of a single adolescent's cancer trajectory. However, study results can supplement each other and patterns of common and uncommon experiences can emerge. This knowledge can help healthcare providers to identify individual adolescents' needs.
Aims

The general aim was to explore experiences of having cancer during adolescence with regard to distress, positive experiences and important aspects of care (Studies I and II) and to use the inductively derived findings to further investigate the distress perceived by adolescents recently diagnosed with cancer (Studies III and IV). The specific aims were:

- To study what aspects of disease and treatment are perceived as especially distressing for children/adolescents with cancer according to children/adolescents/parents and nurses (Study I).

- To study a) what aspects with regard to being told the diagnosis, receiving chemotherapy and being admitted to the ward are perceived as distressing and positive, and b) what aspects of care are perceived as important for adolescents, according to adolescents and nurses (Study II).

- To study a) prevalence and levels of disease- and treatment-related aspects of distress, anxiety and depression, b) levels of well-being, and c) which disease- and treatment-related aspects of distress are perceived as the worst, according to adolescents recently diagnosed with cancer. Additional aims were to investigate differences between investigated variables and between investigated variables and background variables (Study III).

- To compare ratings of physicians and nurses with ratings of adolescents, recently diagnosed with cancer, with regard to a) ratings of presence and levels of disease- and treatment-related aspects of distress, b) which aspects of distress are perceived as the worst for the adolescents, and c) presence and levels of anxiety and depression (Study IV).
Methods

Overview of thesis

In 1995, when the first two studies of this thesis were designed, the psychosocial situation of children and adolescents with cancer in Sweden had been investigated in a very limited number of studies. Therefore, the studies were exploratory, aiming to identify distressing and positive aspects of disease-and treatment, and important aspects of care for children and adolescents with cancer. By interviewing children/adolescents/parents as well as nurses we thought it possible to achieve a quite adequate picture of children's and adolescents' experiences in relation to disease and treatment. Thereafter, we chose to focus the remaining two studies of this thesis on distress in adolescents. The inductively derived aspects of distress, identified in Studies I and II, formed the basis for the interview-guide used in Studies III and IV. While Studies I and II were cross-sectional, including recently diagnosed children and adolescents as well as those who had been off treatment for a number of years, efforts were made to reduce the variability in the adolescent sample in Studies III and IV, as only recently diagnosed adolescents were included.

Studies of experiences of having cancer while being young

Studies I and II

Studies I and II have a cross-sectional, explorative design and are parts of a research project entitled "Care of children and adolescents with cancer" for which children and adolescents with cancer and their families were recruited from two of the six paediatric oncology centres in Sweden (Linköping and Uppsala). The overall aims for the project were to study a) self-esteem, anxiety, depression and social competence (using questionnaires), aspects of distress and positive experiences (using interviews) in relation to disease and treatment among children and adolescents on or off treatment for cancer, b) important aspects of care and assistance at home for children and adolescents and their families (using interviews), and c) quality of life and perceived support among parents of children and adolescents with cancer (using questionnaires). Data collection took place during 1995 in Uppsala and during the spring of 1996 in Linköping.
Participants

Eligible were Swedish speaking children and adolescents, diagnosed with a malignancy at least one month before potential inclusion, and spending enough time at the ward at the time of data-collection to be interviewed and to complete questionnaires. Mentally retarded children were excluded. Children 8 years or older were to answer for themselves and parents were asked to answer for younger children. One nurse or nurse assistant (non-registered staff with two years of education) was to be interviewed about each participating child/adolescent or child for whom a parent was interviewed. In the following, nurses and nurse assistants will be referred to as nurses. During the inclusion period for participation in the interviews, 131 children and adolescents (0-19 years of age) were eligible, see Figure 1 for a presentation of participants and attrition. Fifty interviews with children/adolescents, 65 with parents, and 118 with nurses were analysed and reported in Study I. Forty-four of the interviewed parents were mothers (mean age: 32 years, range 21-41) and 21 were fathers (mean age: 35 years, range 25-45). Thirty-seven nurses were interviewed about 118 children/adolescents, range 1-10 interview per nurse. For six interviews with children/adolescents/parents that were missed by administrative failure (Figure 1), nurse interviews were however performed and included. Twenty-five were registered nurses and 12 nurse assistants. Mean age was 37 years (range 23-60), all were women but two. The nurses had worked in health care for a mean of 17 years (range 3-26) and at the particular unit for a mean of 8 years (range 0,3-26). The clinical and demographic characteristics of the participating children/adolescents and children for whom a parent and/or a nurse was interviewed are presented in Table 1.
Figure 1. Participants and attrition in Studies I and II.
Table 1. Demographic and clinical characteristics of the children and adolescents in Studies I and II.

<table>
<thead>
<tr>
<th>Study centre</th>
<th>Linköping</th>
<th>Uppsala</th>
</tr>
</thead>
<tbody>
<tr>
<td>Study centre</td>
<td>35/7</td>
<td>86/16</td>
</tr>
<tr>
<td>Sex</td>
<td>Boy 62/15</td>
<td>Girl 59/8</td>
</tr>
<tr>
<td>Age</td>
<td>0-3 46*</td>
<td>4-7 34*</td>
</tr>
<tr>
<td></td>
<td>8-12 18*</td>
<td>13-19 23*/23*</td>
</tr>
<tr>
<td>Diagnosis</td>
<td>CNS tumour 3/1</td>
<td>Ewing sarcoma 2/2</td>
</tr>
<tr>
<td></td>
<td>Leukaemia 54/3</td>
<td>Lymphoma 17/8</td>
</tr>
<tr>
<td></td>
<td>Neuroblastoma 8/0</td>
<td>Rabdomyosarcoma 7/1</td>
</tr>
<tr>
<td></td>
<td>Osteosarcoma 5/5</td>
<td>Wilms' tumour 7/0</td>
</tr>
<tr>
<td></td>
<td>Other 18/3</td>
<td></td>
</tr>
<tr>
<td>Relapse</td>
<td>No 113/23</td>
<td>Yes 8/0</td>
</tr>
<tr>
<td>Time since diagnosis / relapse</td>
<td>1 month-1 year 51/13</td>
<td>&gt;1 year-3 years 40/7</td>
</tr>
<tr>
<td></td>
<td>&gt;3 years-5 years 20/3</td>
<td>&gt;5 years 10/0</td>
</tr>
<tr>
<td>Therapy status</td>
<td>Treatment 51/11</td>
<td>Follow-up 70/12</td>
</tr>
</tbody>
</table>

* Refers to age at the time of the described event.

For the purpose of Study II, 23 interviews with adolescents and 21 with nurses (each nurse answering for one adolescent, two nurses took part in two interviews) were analysed, see Figure 1. The demographic data for the subsample of nurses participating in Study II were as follows. Fourteen were registered nurses and five nurse assistants, mean age was 38 years (range 23-51), all were women but one. The nurses had worked in health care for a mean of 17 years (range 6-26) and at the particular ward for a mean of 9 years (range 0,3-23).

**Age groups**

For a number of children a rather long period of time had passed since they got their diagnosis or received active treatment. However, the respondents
answered for the time of active illness. Therefore, we chose to present the answers of the respondents in the age group the child/adolescent belonged to at the time of diagnosis or, if the child/adolescent had suffered one or more relapses (n=8 in Study I, none in Study II), at the time of the latest relapse. E.g., if a boy was 8 years at the time of the interview but was 5 years when he was treated for cancer, his statements are presented in the age group 4-7 years. A certain respondent’s answers are presented in one age group only. Study I provides a full presentation of the number of respondents presented in the various age groups.

Procedure
Children/adolescents, in case the child was 8 years or older, and parents, in case the child/adolescent was younger than 18 years of age received an information letter one week ahead of the child's/adolescent's next visit (treatment or follow-up) to the hospital. The aim and the procedure of the study were presented in the letter, which was signed by the oncologist in charge and a coordinating nurse at the respective ward. Children/adolescents/parents were informed that participation was voluntary and that non-participation would not affect the care of the child/adolescent. On admission in Uppsala, children/adolescents/parents received further information about the study from a coordinating nurse, who asked for participation. In case participation was accepted an appointment with the researcher (Louise von Essen) was scheduled. In Linköping the children/adolescents/parents were contacted by phone and asked for participation by a coordinating nurse. In case participation was accepted an appointment was scheduled at the next visit to the ward when the interview took place (performed by Karin Enskär). In Uppsala one of the nurses who had cared for a certain child/adolescent on at least three working shifts was matched to him/her and interviewed on the same day or close to the child/adolescent/parent interview. In Linköping one of the nurses that had previously taken care of the child/adolescent was matched and interviewed on the same day or close to the child/adolescent/parent interview.

Interviews
Semi-structured interviews with open-ended questions were conducted at the ward. The interviews were audiotaped and transcribed verbatim. All respondents were asked the same questions in the same order. Questions were asked regarding experiences at the time of diagnosis, an especially distressing event, receiving chemotherapy, being admitted to the ward, coming for follow-up (for children and adolescents off treatment), important aspects of care and assistance at home. Follow-up questions were sometimes asked in order to help the respondents elucidate or develop their answers. Answers to the following question were chosen for presentation in Study I:

- Has there been any especially distressing event for you/your child/the child with regard to disease and treatment?
For presentation in Study II, answers to the following questions were chosen:

- Has there been anything distressing/positive for you/the adolescent about being told the diagnosis?
- Has there been anything distressing/positive for you/the adolescent about receiving chemotherapy?
- Has there been anything distressing/positive for you/the adolescent about being admitted to the ward?
- What caring aspects are important for you/the adolescent to feel cared for?

For each question, the respondents were allowed, if they wished, to tell about more than one event. Background data for the participants were collected and a coordinating nurse collected medical data for the adolescents, such as diagnosis, and time since diagnosis, from the medical records.

**Data analysis**

Content analysis (65) was used to analyse the interview data. According to Weber (66), content analysis can be used to draw valid conclusions about a message in a communication by objective and systematic identification of communication characteristics. Answers to open ended questions are suitable for this technique. Words and sentences in the interviews were classified into mutually exclusive categories, which are supposed to reflect central messages in the interviews. Sentences classified in the same category are presumed to have a similar meaning, either based on the precise meaning of the words or on words sharing similar connotations. The content analysis was performed in the following steps:

1. The entire transcribed text was read and reread.
2. Sentences or part of sentences that contained information relevant to the study question were identified and defined as recording units.
3. Recording units were grouped into mutually exclusive categories reflecting central text messages. Criteria for distinguishing the categories were that they were positively different from one another. After a discussion in the research team of the categorisations, some of the categories were judged to be misleading and were rewritten.
4. Boundaries of each category were defined and descriptions of the central characteristics of each category were developed.
5. In Study I, categories were grouped into dimensions according to content.

No matter how many times a certain recording unit was mentioned by a certain person, it was calculated as mentioned once by that specific person in the presentation of the results.
**Interrater agreement for categories**

With access to the recording units, the categories and the descriptions of the central characteristics of each category, an additional assessor independently assigned the recording units to the categories. Boundaries between categories and their content were discussed (by the research team and the additional assessor) and a few changes were made in order to clarify the coding system and the categories. Finally, a comparison of the assessors’ (the one by the research team and the one by the additional assessor) categorisations of recording units was made with the Kappa method (67). The Kappa values varied between 0.84 and 1.00 indicating excellent agreement (68).

**Studies of distress among adolescents recently diagnosed with cancer**

**Studies III and IV**

Studies III and IV are the first reports of an ongoing research project entitled “How do adolescents with cancer cope with disease- and treatment-related aspects of distress?”, a longitudinal, prospective study for which adolescents were recruited from three of the six paediatric oncology centres in Sweden: Lund, Umeå and Uppsala. The overall aims for this project are to investigate psychosocial function among adolescents with cancer at four to eight weeks, six, twelve and eighteen months, and two, three and four years after diagnosis. The reports presented in this thesis are based on data collected from adolescents (Studies III, IV) and physicians and nurses (Study IV) four to eight weeks after diagnosis. The inclusion period for Study I was from June 1999 in Uppsala and March 2000 in Lund and Umeå until October 2003, and for Study IV from February 1999 in Uppsala and from March 2000 in Lund and Umeå until September 2003.

**Participants**

Eligible were: Swedish speaking adolescents (13-19 years of age), diagnosed with cancer (or a relapse after a minimum of one year off treatment; Study IV), treated with chemotherapy, and considered emotionally, cognitively, and physically capable to participate by the project coordinating nurse or the treating physician at the centre where the adolescent was treated and cared for. This assessment was done subjectively according to the nurse's and the physician's personal knowledge of the adolescent. Eleven adolescents were excluded: 4 did not speak Swedish well enough to participate and 7 were considered too physically or cognitively affected by the disease or a neurological comorbidity to participate (5 of whom died shortly after the diagnosis). None was considered emotionally incapable to participate. During the inclusion periods, 92 adolescents were eligible for Study III and/or Study IV,
see Figure 2 for a presentation of participants and attrition. The demographic and clinical characteristics of the participating adolescents are presented in Table 2. The age distribution varies slightly between the samples in Studies III and IV, therefore limits for the age groups differ between studies (Table 2).

One physician and one nurse were to be telephone interviewed for each included adolescent (Study IV). Twenty-six physicians were interviewed about 48 adolescents (5 interviews were not performed as the workload on the ward did not allow time for physicians to participate), range 1-7 interviews per physician (mean 1.8, SD 1.5). Mean age was 45 years (range 28-62), 9 were women and 17 men. The physicians had worked in health care for a mean of 17 years (range 4-35), and with adolescents with cancer for a mean of 8 years (range 0.2-31 years). Forty-four nurses were interviewed about 53 adolescents, range 1-3 interviews per nurse (mean 1.2, SD 0.5). Thirty-six were registered nurses and 8 were nurse assistants (non-registered staff with two years of education). Mean age was 37 years (range 25-59), 38 were women and 6 men. The nurses had worked in health care for a mean of 14 years (range 1-35) and with adolescents with cancer for a mean of 7 years (range 0.3 months-30 years).
Figure 2. Participants and attrition in Studies III and IV.

Eligible adolescents

92

- 24 Refused

Accepted

68

- 2 Administrative failure
- 1 Chose to withdraw from study
- 2 Became too ill before interview

Not eligible for Study III:
- Diagnosed with relapse 5
- Did not match inclusion period 2

Not eligible for Study IV:
- 7 Mainly treated at local hospital
- 3 Did not match inclusion period

63

One physician and one nurse to be inter-
viewed for each participating adolescent

56 included adolescents

Study III
adolescents

53 included adolescents

Study IV
adolescents, physicians and nurses

53 included nurses

48 included physicians
Table 2. Demographic and clinical characteristics of the adolescents in Studies III and IV. The majority of the adolescents are included in both samples.

<table>
<thead>
<tr>
<th></th>
<th>Study III (n=56)</th>
<th>Study IV (n=53)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Centre</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lund</td>
<td>19</td>
<td>17</td>
</tr>
<tr>
<td>Umeå</td>
<td>17</td>
<td>17</td>
</tr>
<tr>
<td>Uppsala</td>
<td>20</td>
<td>19</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Boy</td>
<td>32</td>
<td>36</td>
</tr>
<tr>
<td>Girl</td>
<td>24</td>
<td>17</td>
</tr>
<tr>
<td>Age</td>
<td></td>
<td></td>
</tr>
<tr>
<td>13-15/13-14</td>
<td>35</td>
<td>36</td>
</tr>
<tr>
<td>16-19/15-19</td>
<td>21</td>
<td>17</td>
</tr>
<tr>
<td>Diagnosis</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CNS tumour</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>Ewing sarcoma</td>
<td>4</td>
<td>3</td>
</tr>
<tr>
<td>Leukemia</td>
<td>18</td>
<td>18</td>
</tr>
<tr>
<td>Lymphoma</td>
<td>20</td>
<td>16</td>
</tr>
<tr>
<td>Osteosarcoma</td>
<td>8</td>
<td>8</td>
</tr>
<tr>
<td>Other</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Relapse</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No</td>
<td>56</td>
<td>48</td>
</tr>
<tr>
<td>Yes</td>
<td>–</td>
<td>5</td>
</tr>
<tr>
<td>Mainly treated at</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Local hospital</td>
<td>7</td>
<td>–</td>
</tr>
<tr>
<td>Paediatric oncology centre</td>
<td>49</td>
<td>53</td>
</tr>
<tr>
<td>Prognosis as estimated by physician</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Better</td>
<td>44</td>
<td>–</td>
</tr>
<tr>
<td>Worse</td>
<td>12</td>
<td>–</td>
</tr>
</tbody>
</table>

Procedure
All adolescents who met the inclusion criteria and their parents received oral and written information about the study approximately three weeks after diagnosis from a coordinating nurse at the respective centre. After a few days, the coordinating nurse contacted the adolescent and asked him/her if he/she accepted participation, and asked parents for consent if the adolescent was ≤17 years. If the adolescent and the parents, in case the adolescent was ≤17 years of age, or the adolescent, in case he/she was 18 years or older, accepted participation, the researcher who was to conduct the interview was contacted. This person then in turn contacted the adolescent by telephone. The majority of the interviews were performed by Mariann Hedström, some interviews were performed by Kristina Haglund and Elisabet Mattsson. For the purpose of Study IV, the coordinating nurse matched one of the nurses and one of the physicians who knew the adolescent the best to be interviewed as close in time to the adolescent interview as possible. Data were collected by structured telephone interviews 4-8 weeks after the diagnosis. All adolescents had started chemotherapy at the time for data collection. At the end of each interview, the adolescent was asked whether he/she accepted to be contacted again for a second interview four to five months later. The adolescents received a small present in return for their participation.
Measures

Prevalence, levels and worst aspects of distress (Studies III and IV)

A structured interview-guide, developed for the study, was used to investigate prevalence and, for most aspects, levels of some disease- and treatment-related aspects of distress since diagnosis (4-8 weeks ago). The aspects were identified through semi-structured interviews with adolescents on or off cancer treatment and nurses caring for them (Studies I and II). The aspects were arranged in four areas: physical concerns, personal changes, feelings of alienation and disease- and treatment-related worries. For 16 aspects, prevalence and levels were identified using a six-grade verbal scale, range not at all to very much, coded 0-5. For 4 aspects prevalence was identified through a dichotomised yes/no response alternative. Questions referred to the time since diagnosis. Physicians and nurses answered a staff version (Studies IV), which included the same aspects, but with questions directed to the staff, asking them to reply what they thought their matched adolescent had experienced since diagnosis. For all aspects, staff had the possibility to answer "I don’t know". The respondents were then asked to identify the aspect within each area that had been the most distressing since diagnosis (Study III). Finally the respondents were asked to identify the aspect of distress that had been the worst of all aspects since diagnosis (the overall worst aspect) (Studies III and IV). The respondents were encouraged to identify one aspect only, but were allowed to report more than one aspect as the worst in each area/the overall worst.

Prevalence and levels of anxiety and depression (Studies III and IV)
The Hospital Anxiety and Depression Scale (HADS) (69) was used to investigate prevalence and levels of anxiety and depression. Depression is often difficult to diagnose in cancer patients as physical symptoms of depression such as disturbances in sleep, appetite, concentration and decreased energy levels may occur as a consequence of cancer and its treatment (70). HADS was designed for screening of anxiety and depression among somatically ill patients. Hence, the constructors omitted physical indicators of anxiety and depression as they could be due to disease and/or treatment (71). HADS refers to the preceding week and consists of two subscales, one assessing anxiety (HADS-A, 7 items) and one depression (HADS-D, 7 items). Answers are given on a four-grade verbal scale, coded 0 to 3. Subscale scores range from 0 (no distress) to 21 (maximum distress). Physicians and nurses answered a staff version (72), which includes the same items, but with questions directed to the staff, asking them to reply as they thought their matched adolescent had felt during the past week (Study IV). Chronbach alpha values for the Swedish version have been reported as satisfactory when used for adults, 0.84 to 0.85 for HADS-A and 0.81 to 0.82 for HADS-D (73, 74). The HADS has been found to possess adequate test-retest reliability and sensitiv-
ity for use with adolescents (75), and to correctly identify a majority of clinical cases of psychiatric mood disorders among adolescents with cancer as identified by a psychiatric evaluation (76).

Levels of well-being (Study III)
To measure levels of well-being, we first considered the SF-36 (77). However, the complete instrument, in combination with the interview-guide and HADS, were estimated to be too demanding, for adolescents undergoing chemotherapy treatment, to answer. Therefore, two subscales from the SF-36, Vitality (4 items, pep/life, energy, worn out, tired) and Mental Health (5 items, nervous, down in dumps, peaceful, blue/sad, happy), were selected (78). Responses are given on a six-grade verbal scale, coded 1 to 6. SF-36 refers to the four previous weeks. The SF-36 has been translated to Swedish and tested for its psychometric qualities on 8930 Swedish persons, aged 15-93. Chronbach alpha values for Vitality and Mental Health were 0.85 and 0.87, respectively (79). The Vitality subscale has recently been reported to be associated to ratings of functional mobility among childhood survivors of osteosarcoma, aged 10-27 years, indicating convergent validity for physical performance (80). The Mental Health subscale has been reported to be highly associated with HADS-A (r=0.75) and HADS-D (r=0.68) among men with testicular cancer (81). This is a potentially useful information as there do not exist norm-values for HADS for Swedish adolescents, while norm-values for SF-36 exist for the Swedish population aged 15-24 (79).

Background variables (Studies III and IV)
Respondents were asked to report background variables and the coordinating nurse collected clinical data for the adolescents from the medical records. For the purpose of Study III, one of the treating physicians was asked to estimate the prognosis for the adolescent on a five-grade verbal scale, very good, good, quite good (in the analysis classified as "better"), somewhat and not good ("worse"). For 13 adolescents for whom an estimate of prognosis had not been done, the prognosis was estimated retrospectively by an experienced paediatric oncologist, with access to the medical data for the adolescents from the time of the collection of data but with no information of the outcomes for the adolescents.

Data analysis
Prevalence, levels and worst aspects of distress (Studies III and IV)
It was assumed that if an adolescent answered quite much, much or very much in response to a question with continuous response alternatives, or yes to a question with dichotomised response alternatives, about prevalence/level of an aspect, the same aspect was, at least to some extent, experienced as distressing by the same individual. This assumption was the starting point
for calculations of prevalence of aspects. Prevalence is defined as quite much, much and very much answers, coded 3 to 5, and yes answers to questions with dichotomised response alternatives. Levels and worst aspects of distress are reported with descriptive statistics.

Adolescent-staff agreement with regard to aspects of distress (Study IV)
Adolescent-staff agreement with regard to disease- and treatment-related distress was analysed with sensitivity/specificity calculations. Sensitivity was defined as the proportion (%) of staff being able to detect an aspect as distressing, i.e. answering quite much, much or very much to questions with continuous, and yes to questions with dichotomised, response alternatives, when the adolescents also provided any of these answers. Specificity was defined as the proportion (%) of staff being able to detect an aspect as not distressing, i.e. answering quite little, little or not at all to questions with continuous, and no to questions with dichotomised, response alternatives when the adolescents also provided any of these answers. Calculations of sensitivity and specificity are based on an idea presented by Brunelli et al (82). Overestimation was defined as staff giving a rating more than one score above and underestimation as a rating more than one score below the rating by the adolescent. Calculations of over- and underestimation could not be performed for questions with dichotomised response alternatives. Perceptions of worst aspects of distress are reported with descriptive statistics.

Prevalence and levels of anxiety and depression (Studies III and IV)
For calculations of prevalence of anxiety and depression the cut-off scores ≥9 for anxiety (HADS-A) and ≥7 for depression (HADS-D), indicating a potential clinical case, recommended for use with adolescents by White et al (75) were employed. Levels of anxiety and depression are reported with descriptive statistics. Missing values were substituted by the mean value of the adolescent’s answers to the remaining scale items, providing that six of the seven items had been answered. This procedure was performed for 3 values in Study III and 9 values in Study IV.

Adolescent-staff agreement and differences with regard to anxiety and depression (Study IV)
Sensitivity and specificity were calculated using the cut-off points presented above. For calculations of associations, Pearson product moment correlation coefficients (PPM) were applied. For calculations of differences, paired, two-tailed t-tests were used. If more than one item per scale was missing, that subscale was treated as missing data in the prevalence, PPM and t-test calculations. This was the case for one depression and one anxiety subscale for adolescents, four depression and four anxiety subscales for physicians, and one depression and three anxiety subscales for nurses. Furthermore,
eight physicians and three nurses were not asked to answer HADS as more than one week had elapsed since they had met the adolescent.

*Differences between possible cases and possible non-cases of anxiety and depression with regard to prevalence of aspects of distress and background variables (Study III)*

Possible differences between adolescents scoring as potential clinical cases for anxiety and/or depression and those not scoring as potential clinical cases with regard to prevalence of aspects of distress and background variables were analysed using Fisher's exact tests.

*Levels of well-being (Study III)*

Levels of well-being are reported with descriptive statistics and compared to Swedish norm data with one-sample t-tests.

*Differences between background variables with regard to prevalence and levels of aspects of distress and levels of anxiety, depression and well-being (Study III)*

Differences between background variables with regard to disease- and treatment-related aspects of distress with continuous response alternatives, and levels of anxiety, depression and well-being were analysed using Mann-Whitney U-tests. Differences between background variables with regard to aspects of distress with dichotomous response alternatives were analysed using $\chi^2$ tests

**Ethics**

Ethical approval was obtained from the local ethics committees at the faculties of medicine at the universities in Linköping and Uppsala (Studies I and II) and Lund, Umeå and Uppsala (Studies III and IV). The ethical issue of asking under age children and adolescents for participation in a research study was discussed thoroughly in the research group. As a result, it was decided that when the coordinating nurses asked the children and adolescents for participation, this should be done very cautiously and great consideration should be taken to the children's and adolescents' integrity and wishes. Especially in Studies III and IV, when the adolescents were recently diagnosed and therefore presumably under considerable strain, this procedure may have increased the number of adolescents not accepting participation.
Results and Comments

Studies of experiences of having cancer while being young – summary of Studies I and II

Especially distressing aspects of disease and treatment (Study I)

The aim was to investigate what aspects of disease and treatment that were perceived as especially distressing for children and adolescents with cancer according to the children/adolescents/parents and nurses. Fifty interviews were performed with children and adolescents, 65 with parents and 118 with nurses. The answers were analysed with content analysis, which revealed twenty categories of especially distressing events with regard to disease and treatment. These were grouped into a Physical and an Emotional dimension. A presentation of the participants’ responses by category and dimension is reported in Table 3.

For children 0-3, 4-7 and ≥13 years respondents most often mentioned aspects referred to the Physical dimension, whereas for children 8-12 respondents most often mentioned aspects included in the Emotional dimension.

Physical aspects

The most frequently mentioned aspects of distress referred to the Physical dimension were categorised as pain from diagnostic procedures and treatments, nausea and fatigue. For children in different age groups the categories with most included statements were as follows. For children 0-3 years old: pain from diagnostic procedures and treatments, nausea and pain from disease. For children 4-7 years old: pain from diagnostic procedures and treatments, fatigue and nausea. For children 8-12 years old: pain from diagnostic procedures and treatments, nausea and oral medication and for adolescents 13-19 years: nausea and pain from diagnostic procedures and treatments.
Table 3. Participants’ responses by category and dimension to the question “Has there been any especially distressing event for you/your child/the child with regard to disease and treatment?” (Study I).

<table>
<thead>
<tr>
<th>Dimension</th>
<th>Category</th>
<th>0-3 years</th>
<th>4-7 years</th>
<th>8-12 years</th>
<th>13-19 years</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical</td>
<td>Diarrhoea and constipation</td>
<td>0/4/1</td>
<td>0/0/0</td>
<td>0/0/1</td>
<td>0/0</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>Fatigue</td>
<td>0/2/3</td>
<td>1/7/3</td>
<td>2/0/0</td>
<td>2/1</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Fever</td>
<td>1/1/2</td>
<td>0/2/2</td>
<td>0/1/0</td>
<td>3/0</td>
<td>12</td>
</tr>
<tr>
<td></td>
<td>Hunger and thirst</td>
<td>0/3/1</td>
<td>0/0/0</td>
<td>1/0/0</td>
<td>3/0</td>
<td>8</td>
</tr>
<tr>
<td></td>
<td>Mouth sores</td>
<td>0/1/1</td>
<td>0/4/1</td>
<td>1/0/0</td>
<td>0/0</td>
<td>8</td>
</tr>
<tr>
<td></td>
<td>Nausea</td>
<td>0/8/4</td>
<td>1/3/5</td>
<td>1/1/2</td>
<td>7/5</td>
<td>37</td>
</tr>
<tr>
<td></td>
<td>Neuromuscular disorders</td>
<td>0/3/2</td>
<td>0/0/1</td>
<td>0/0/0</td>
<td>0/0</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>Oral medication</td>
<td>1/0/2</td>
<td>0/0/0</td>
<td>3/0/1</td>
<td>0/0</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Pain from diagnostic procedures/treatments</td>
<td>5/9/10</td>
<td>3/5/7</td>
<td>1/1/4</td>
<td>4/5</td>
<td>54</td>
</tr>
<tr>
<td></td>
<td>Pain from disease</td>
<td>0/5/7</td>
<td>0/1/0</td>
<td>0/0/0</td>
<td>0/0</td>
<td>13</td>
</tr>
<tr>
<td></td>
<td>Respiratory distress</td>
<td>0/0/1</td>
<td>0/3/3</td>
<td>0/0/2</td>
<td>1/0</td>
<td>10</td>
</tr>
<tr>
<td></td>
<td>Other physical concerns</td>
<td>2/2/3</td>
<td>1/4/2</td>
<td>0/1/1</td>
<td>0/2</td>
<td>18</td>
</tr>
<tr>
<td></td>
<td>Total physical</td>
<td>9/38/37</td>
<td>6/29/24</td>
<td>9/4/11</td>
<td>20/13</td>
<td>200</td>
</tr>
<tr>
<td>Emotional</td>
<td>Changed appearance</td>
<td>1/1/1</td>
<td>1/3/1</td>
<td>2/0/1</td>
<td>5/3</td>
<td>19</td>
</tr>
<tr>
<td></td>
<td>Confinement</td>
<td>1/6/9</td>
<td>1/2/4</td>
<td>2/2/5</td>
<td>1/1</td>
<td>34</td>
</tr>
<tr>
<td></td>
<td>Feeling of alienation</td>
<td>1/4/2</td>
<td>1/7/3</td>
<td>3/1/2</td>
<td>3/2</td>
<td>29</td>
</tr>
<tr>
<td></td>
<td>Feeling of uneasiness</td>
<td>0/5/4</td>
<td>0/4/3</td>
<td>1/1/1</td>
<td>1/1</td>
<td>21</td>
</tr>
<tr>
<td></td>
<td>Lack of family support</td>
<td>0/0/4</td>
<td>1/0/5</td>
<td>0/0/2</td>
<td>0/1</td>
<td>13</td>
</tr>
<tr>
<td></td>
<td>Loss of integrity</td>
<td>1/0/2</td>
<td>0/1/0</td>
<td>0/0/0</td>
<td>1/1</td>
<td>6</td>
</tr>
<tr>
<td></td>
<td>Worry before medical procedures</td>
<td>0/3/6</td>
<td>0/3/4</td>
<td>1/0/4</td>
<td>0/6</td>
<td>27</td>
</tr>
<tr>
<td></td>
<td>Worry about death</td>
<td>0/0/2</td>
<td>0/1/2</td>
<td>2/2/6</td>
<td>0/1</td>
<td>16</td>
</tr>
<tr>
<td></td>
<td>Total emotional</td>
<td>4/19/30</td>
<td>4/21/22</td>
<td>11/6/21</td>
<td>11/16</td>
<td>165</td>
</tr>
<tr>
<td>Do not know</td>
<td>Do not know/remember</td>
<td>0/2/12</td>
<td>0/1/6</td>
<td>0/0/2</td>
<td>0/4</td>
<td>27</td>
</tr>
<tr>
<td>Nothing</td>
<td>Nothing distressing</td>
<td>0/0/0</td>
<td>0/1/1</td>
<td>0/0/0</td>
<td>2/1</td>
<td>5</td>
</tr>
</tbody>
</table>
The results with regard to physical distress varied little between age groups. Pain from diagnostic procedures and treatments was, for all age groups, one of the two most frequently mentioned categories referred to the Physical dimension. This finding supports earlier findings by Ljungman et al (12) and McGrath et al (11), who have concluded that pain from treatment and procedures is a greater problem than pain from the disease itself. Pain from diagnostic procedures and treatments was by far the most frequently mentioned physical problem for children 0-3 years, followed by Pain from disease. Ljungman et al (12) demonstrated that younger children (<5 years) were more concerned about pain than older children. These findings might be related to results by Reissland (83) who suggested that younger children lack the cognitive maturity to develop autonomous strategies and therefore might be less able than older children to cope with pain.

For adolescents, 13-19 years, nausea was the most frequently mentioned problem. Dolgin and co-workers (84) concluded that adolescents exhibit consistently higher levels of nausea/vomiting than younger children. Psychological as well as biological factors might contribute to this circumstance. Adolescents may attach more concern to side effects such as nausea due to their increased understanding of the disease, and younger children often metabolize chemotherapy more rapidly than adolescents and adults.

Emotional aspects

The most frequently mentioned aspects of distress referred to the Emotional dimension were categorized as confinement, feeling of alienation and worry before medical procedures. For children/adolescents in different age groups the categories with most included statements were as follows. For children 0-3 years old: confinement, feeling of uneasiness and worry before medical procedures. For children 4-7 years old: feeling of alienation, confinement, feeling of uneasiness and worry before medical procedures. For children 8-12 years old: worry about death, confinement and feeling of alienation and for adolescents 13-19 years: changed appearance, worry before medical procedures and feeling of alienation.

The results concerning emotional aspects vary to some extent between age groups. For the youngest children, 0-3 years, confinement, e.g. physical restraint in relation to various disease- and treatment-related procedures, was the most often mentioned emotional problem. For children 4-7 years, feeling of alienation was the emotional aspect most often mentioned. The result implies that even very young children miss their everyday life and feel different and left aside. This issue is not, to our knowledge, substantially described in the literature. In fact, most studies investigating distress among children with cancer focus on children over 10 years of age (22).

For children 8-12 years, worry about death was the most frequently mentioned aspect of distress, and emotional aspects of distress were more often mentioned than physical aspects of distress for this age group. This was not
the case for the other age groups. Children about 7-11 years old begin to understand death as permanent and non-reversible and might thus understand the possible implications of a life-threatening disease (6). It has previously been argued that being diagnosed with, and living with cancer during adolescence might be especially distressing since adolescents have the cognitive ability to understand the implications of cancer, but may lack the personal resources to contend with the problems associated with the situation (7, 49). Younger children may have had even less opportunity to develop adequate strategies to cope with anxiety (55).

Even though it is assumed that adolescents experience considerable psychological strain to cope with the cancer threat (6-8), the adolescents participating in this study did not describe events connected with fear. Berard and Boermeester (76) have argued that adolescents tend to mask distress, partly since emotional vulnerability often is understood as weakness. For adolescents in this study, changed appearance is the most often mentioned emotional aspect of distress. It is well-known that physical appearance has an important impact on self-image, especially during adolescence (85).

Distressing and positive aspects of disease- and treatment and important aspects of care (Study II)

The aim was to investigate distressing and positive experiences with regard to being told the diagnosis, receiving chemotherapy and being admitted to the ward, and important aspects of care for adolescents with cancer according to adolescents and their nurses. Twenty-three interviews were performed with adolescents and 21 with nurses. The answers were analysed with content analysis, categories derived from the answers are presented in Tables 4 and 5.

**Being told the diagnosis**

Six categories of distressing, and four categories of positive experiences were developed from the answers to the questions about being told the diagnosis. Distressing aspects were mostly related to fears, most often mentioned were fear of alienation, fear of altered appearance and fear of dying. As a positive aspect, more than half of the adolescents and nurses mentioned good to know, as opposed to uncertainty and secrets. Some adolescents expressed a hope for the future, and that understanding that something could be done against pain was experienced as a relief. Last and van Veldhuizen (42) have argued that information about diagnosis, prognosis and treatment is important as hope becomes realistic when the possibilities for treatment are explained.
Table 4. Categories derived from answers to the questions about distressing and positive experiences.

<table>
<thead>
<tr>
<th>Questions about</th>
<th>Distressing aspects</th>
<th>Positive aspects</th>
</tr>
</thead>
<tbody>
<tr>
<td>Being told the diagnosis</td>
<td>Distress due to previous experiences</td>
<td>Good to know</td>
</tr>
<tr>
<td></td>
<td>Fear of alienation</td>
<td>Hope</td>
</tr>
<tr>
<td></td>
<td>Fear of altered appearance</td>
<td>It could have been worse</td>
</tr>
<tr>
<td></td>
<td>Fear of medical treatments</td>
<td>Physical relief</td>
</tr>
<tr>
<td></td>
<td>Feeling of uneasiness</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fear of dying</td>
<td></td>
</tr>
<tr>
<td>Receiving chemotherapy</td>
<td>Altered self-image</td>
<td>Better than expected</td>
</tr>
<tr>
<td></td>
<td>Confinement</td>
<td>To get well</td>
</tr>
<tr>
<td></td>
<td>Physical concerns</td>
<td></td>
</tr>
<tr>
<td>Being admitted to the ward</td>
<td>Confinement</td>
<td>Amusement</td>
</tr>
<tr>
<td></td>
<td>Fear of interventions and treatments</td>
<td>Physical ambience</td>
</tr>
<tr>
<td></td>
<td>Feeling of alienation</td>
<td>Positive relations to the staff</td>
</tr>
<tr>
<td></td>
<td>Lack of integrity</td>
<td>To get well</td>
</tr>
<tr>
<td></td>
<td>Lack of comfort</td>
<td>Well cared for</td>
</tr>
<tr>
<td></td>
<td>Memories and concerns</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Small children</td>
<td></td>
</tr>
</tbody>
</table>

*Categories are alphabetically ordered

**Receiving chemotherapy**

Three categories of distressing, and two categories of positive experiences were developed. Distressing aspects were categorised as physical concerns, e.g. nausea and mouth sores, altered self-image e.g. hair-loss and fatigue and confinement. The majority of the adolescent and nurse statements of positive experiences were related to the understanding that treatment was necessary, to get well.

**Being admitted to the ward**

Seven categories of distressing, and five categories of positive experiences were developed. Distressing aspects were mostly related to restrictions of freedom, most often mentioned were confinement, lack of integrity and feeling of alienation. However, 11 of the 23 interviewed adolescents answered that nothing was distressing about being admitted to the ward. The positive aspects most often mentioned were positive relations to the staff, to get well and well cared for.

Most adolescent and nurse statements with regard to positive experiences in relation to being admitted to the ward concern positive relations to the staff. A number of adolescents mentioned being treated with knowledge and experience, i.e. being well cared for, as a positive experience. When the ado-
lescents participating in Study III and/or IV were asked about how well at ease they felt at the ward, 69% answered very well or well, 23% quite well and 5% somewhat or not at all (unpublished results). In an interview study with healthy adolescents, Adamson et al (86) conclude that all respondents expressed a need for adult contacts as sources of knowledge and experience, but that contacts outside the closest family are rare. Since a cancer diagnosis during adolescence usually implies lengthy contacts with the health care system, many adolescents with cancer have great possibilities to close and meaningful contacts with other grown-ups than their parents.

**Important aspects of care**

Twelve categories were developed. Most often mentioned were *clinical competence, social competence, emotional support, information and respect for integrity*. Adolescents, but not nurses, mentioned *physical ambience, continuity and family participation* as important in order to feel cared for, whereas nurses, but not adolescents, mentioned the importance of *flexible routines, and participation in decision making*. Clinical competence was one of the two caring aspects mentioned by most adolescents and nurses, but has not been demonstrated as any of the most important aspects of care for younger children with cancer (87, 88). However, clinical competence has previously been identified as the most important care aspect to make adult cancer patients feel cared for (89). The findings may reflect developmental factors, young children will rarely understand that not all adults are good at their jobs, and hence not have reasons to appreciate skills or fear ignorance. Adolescents, on the other hand, will probably carefully evaluate the skills of health care professionals.

Ritchie (57) has recognised emotional support as a significant determinant of adolescents’ psychosocial response to the experience of cancer and treatment. The importance of information for children and adolescents with cancer has been identified in earlier studies (62, 87, 90).

The statements included in the categories continuity, flexible routines and time are related to how health professionals organise their daily work, e.g. provide adolescents with the possibility to meet the same physician and nurse, spend time with the adolescents and facilitate the adolescent’s everyday life by trying to offer flexible routines. Other categories, i.e., amusement, family participation and physical ambience are related to equipment and possibilities at the ward, e.g. computers, videos and TV-games, nice and cosy interior decorations and possibilities for parents to stay at the ward.
Table 5. Categories derived from answers to the question about important care at the ward.

<table>
<thead>
<tr>
<th>Important aspects of care</th>
</tr>
</thead>
<tbody>
<tr>
<td>Amusement</td>
</tr>
<tr>
<td>Clinical competence</td>
</tr>
<tr>
<td>Continuity</td>
</tr>
<tr>
<td>Emotional support</td>
</tr>
<tr>
<td>Family participation</td>
</tr>
<tr>
<td>Flexible routines</td>
</tr>
<tr>
<td>Information</td>
</tr>
<tr>
<td>Participation in decision making</td>
</tr>
<tr>
<td>Physical ambience</td>
</tr>
<tr>
<td>Respect for integrity</td>
</tr>
<tr>
<td>Social competence</td>
</tr>
<tr>
<td>Time</td>
</tr>
</tbody>
</table>

* Categories are alphabetically ordered

Studies of distress among adolescents recently diagnosed with cancer – summary of Study III and IV

**Prevalence and levels of aspects of distress, anxiety, depression and well-being (Study III)**

The aims were to investigate prevalence and levels of disease- and treatment-related aspects of distress, anxiety, depression, levels of well-being and which disease- and treatment-related aspects of distress are perceived as the worst. Fifty-six adolescents diagnosed with cancer since 4-8 weeks participated in a structured telephone interview.

**Aspects of distress**

A presentation of prevalence, levels and worst disease- and treatment-related aspects of distress is reported in Table 6.
Table 6. Prevalence of aspects of distress, levels of aspects, worst aspects belonging to each area and the overall worst aspects of distress (N=56) (Study III). NA= Not Applicable.

<table>
<thead>
<tr>
<th>Area</th>
<th>Aspect of distress</th>
<th>Prevalence n (%)</th>
<th>Mean (range 0-5)</th>
<th>SD</th>
<th>Worst aspect belonging to each area</th>
<th>Overall worst aspect</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical</td>
<td>Infections</td>
<td>21 (38)</td>
<td>1.70</td>
<td>1.73</td>
<td>6</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Mucositis</td>
<td>30 (54)</td>
<td>2.45</td>
<td>1.85</td>
<td>16</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Nausea</td>
<td>23 (41)</td>
<td>2.32</td>
<td>1.83</td>
<td>16</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Pain from disease</td>
<td>18 (30)</td>
<td>1.45</td>
<td>1.80</td>
<td>5</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Pain from procedures/treatments</td>
<td>17 (30)</td>
<td>2.14</td>
<td>1.67</td>
<td>12</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>10 (18)</td>
<td>NA</td>
<td>NA</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Personal changes</td>
<td>Changed temper</td>
<td>28 (50)</td>
<td>NA</td>
<td>NA</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Fatigue</td>
<td>35 (62)</td>
<td>2.80</td>
<td>1.40</td>
<td>22</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>Losing hair</td>
<td>51 (91)</td>
<td>NA</td>
<td>NA</td>
<td>23</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>Round face</td>
<td>25 (45)</td>
<td>NA</td>
<td>NA</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Weight loss/gain</td>
<td>45 (80)</td>
<td>NA</td>
<td>NA</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>7 (12)</td>
<td>NA</td>
<td>NA</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>Feelings of alienation</td>
<td>Experiencing lower self-esteem</td>
<td>5 (9)</td>
<td>0.52</td>
<td>1.08</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Feeling different than friends</td>
<td>17 (30)</td>
<td>1.34</td>
<td>1.52</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Feeling left-out by friends</td>
<td>8 (14)</td>
<td>0.82</td>
<td>1.31</td>
<td>6</td>
<td>3</td>
</tr>
<tr>
<td></td>
<td>Missing leisure activities</td>
<td>47 (84)</td>
<td>3.75</td>
<td>1.63</td>
<td>40</td>
<td>5</td>
</tr>
<tr>
<td></td>
<td>Not wanting others to see me</td>
<td>12 (21)</td>
<td>1.25</td>
<td>1.55</td>
<td>8</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>11 (20)</td>
<td>NA</td>
<td>NA</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Disease- and treatment-related worries</td>
<td>Worry being left-out by friends</td>
<td>10 (18)</td>
<td>0.85</td>
<td>1.42</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Worry for changed appearance</td>
<td>21 (38)</td>
<td>1.87</td>
<td>1.73</td>
<td>6</td>
<td>0</td>
</tr>
<tr>
<td></td>
<td>Worry for pain from procedures/treatments</td>
<td>14 (25)</td>
<td>1.50</td>
<td>1.41</td>
<td>8</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>Worry missing school</td>
<td>35 (62)</td>
<td>2.84</td>
<td>1.79</td>
<td>14</td>
<td>7</td>
</tr>
<tr>
<td></td>
<td>Worry not getting well</td>
<td>19 (34)</td>
<td>1.91</td>
<td>1.63</td>
<td>23</td>
<td>10</td>
</tr>
<tr>
<td></td>
<td>Other</td>
<td>11 (20)</td>
<td>NA</td>
<td>NA</td>
<td>4</td>
<td>1</td>
</tr>
</tbody>
</table>

a Aspects are alphabetically ordered within each area. b Adolescents could report more than one aspect as the worst. c Two adolescents stated that nothing was distressing with regard to personal changes. d Two adolescents stated that they did not experience any disease- and treatment-related worries.
The results indicate some correspondence between prevalence and mean values for aspects and some correspondence between mean values for aspects and the number of adolescents rating a certain aspect as the worst. However, this does not apply for all aspects, losing hair and weight loss/gain are reported as some of the most prevalent aspects, but are not among the aspects given the highest mean values or considered worst. Missing leisure activities was assigned the highest mean value even though only five adolescents considered it as the worst aspect. On the other hand, ten adolescents considered worry not getting well as the worst aspect but the item was only assigned the seventh highest mean value. The aspects worry missing school and mucositis were rated among those with the highest prevalence, highest levels and perceived as the overall worst. These aspects probably need special consideration from health care professionals.

### Anxiety and depression

The mean value for HADS-A was 4.68 (SD 3.03) and for HADS-D 4.32 (SD 2.80). Seven adolescents (12%) reached the cut-off score for anxiety indicating a potential clinical case and 12 (21%) reached the cut-off score for depression indicating a potential clinical case, a higher clinically relevant self-reported depression prevalence than previously reported (27, 29, 30). Three adolescents scored as potential clinical cases for both anxiety and depression. The Pearson product-moment correlation between the two scales was \( r = 0.34 \) (\( p < 0.01 \)). The Cronbach alpha values were 0.66 for HADS-A and 0.54 for HADS-D.

More adolescents among those scoring in the clinical range of anxiety and/or depression reported experiencing a number of aspects of an emotional nature, weight changes and pain from procedures and treatments than among those not scoring in the clinical range of those problems. Results demonstrating that higher pain intensity was associated with higher depressive and anxious symptoms among children with chronic pain have been reported (33). Recently, Varni et al. (34) reported that pain and emotional distress among children and adolescents with cancer, although associated cross-sectionally, did not predict each other. It was concluded that it is essential to intervene against both pain and emotional distress.

In concordance with previous findings, adolescents with a worse prognosis did not report different levels of anxiety, depression or mental health than adolescents with a better prognosis (26, 31, 48). It should be remembered however, that it is not known how the adolescents perceived their own survival perspective. Thus, it can be speculated that subjective survival perspective, e.g. influenced by the adolescents’ characteristics, ways of handling difficult situations and personality, is associated with psychological distress.

More adolescents mainly treated at a local hospital scored as potential clinical cases for anxiety than adolescents treated at a paediatric oncology centre. In fact, 4 out of 7 adolescents (57%) who were treated at a local hos-
hospital scored in the clinical range of anxiety, while only 3 of 49 (6%) of the adolescents mainly treated at a paediatric oncology centre, did so. In Study II, we reported that many adolescents treated at a paediatric oncology centre considered clinical competence as important in order to feel cared for. It can be speculated that the expertise of the staff at paediatric oncology centres provides a certain sense of security to the adolescents. Further, the presence of other children and adolescents with cancer may help normalising the abnormal, thus reducing insecurity.

Well-being
The mean values for Vitality and Mental Health were lower than Swedish population norms for ages 15-24 (79). The mean value for Vitality was 47.8 (SD 21.9) (norms M 70.8, SD 19.4) ($t_{[55]} = 7.9$, $p< 0.0001$), an expected finding as the adolescents were on chemotherapy treatment. The mean value for Mental Health was 65.9 (SD 18.7) (norms M 81.8, SD 16.2) ($t_{[55]} = 6.4$, $p< 0.0001$). The Cronbach alpha values were 0.75 for Vitality and 0.80 for Mental Health.

Accuracy of assessment of distress, anxiety and depression by physicians and nurses (Study IV)

The aims were to compare the ratings of physicians and nurses with the ratings of adolescents, regarding prevalence and levels of disease- and treatment-related aspects of distress, which aspects of distress that were experienced as the worst for the adolescents and presence and levels of anxiety and depression. Fifty-three structured telephone interviews were conducted with adolescents diagnosed with cancer or a relapse since 4-8 weeks, 48 interviews were conducted with physicians and 53 with nurses.

Agreement on aspects of distress
A presentation of prevalence and staff sensitivity, specificity, overestimation and underestimation for disease- and treatment-related aspects of distress is reported in Table 7. Sensitivity and specificity of around 60% and higher were approximately equivalent to significant associations. Thus, as a basis for discussing the accuracy of the staff ratings, 60% was chosen to indicate reasonable agreement.
Table 7. Prevalence of, and physician and nurse sensitivity, specificity, overestimation and underestimation for aspects’ of distress (Study IV). Numbers indicate %. NA= Not Applicable.

<table>
<thead>
<tr>
<th>Area</th>
<th>Aspect of distress</th>
<th>Adolescents (N=53)</th>
<th>Physicians (N=48)</th>
<th>Nurses (N=53)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physical concerns</td>
<td>Infections</td>
<td>42</td>
<td>59</td>
<td>84</td>
</tr>
<tr>
<td></td>
<td>Mucositis</td>
<td>55</td>
<td>60</td>
<td>83</td>
</tr>
<tr>
<td></td>
<td>Nausea</td>
<td>36</td>
<td>71</td>
<td>77</td>
</tr>
<tr>
<td></td>
<td>Pain from disease</td>
<td>30</td>
<td>71</td>
<td>79</td>
</tr>
<tr>
<td></td>
<td>Pain from procedures/treatments</td>
<td>40</td>
<td>60</td>
<td>57</td>
</tr>
<tr>
<td>Personal changes</td>
<td>Changed temper</td>
<td>49</td>
<td>60</td>
<td>17</td>
</tr>
<tr>
<td></td>
<td>Fatigue</td>
<td>68</td>
<td>84</td>
<td>35</td>
</tr>
<tr>
<td></td>
<td>Losing hair</td>
<td>94</td>
<td>84</td>
<td>67</td>
</tr>
<tr>
<td></td>
<td>Round face</td>
<td>45</td>
<td>52</td>
<td>80</td>
</tr>
<tr>
<td></td>
<td>Weight loss/gain</td>
<td>87</td>
<td>49</td>
<td>14</td>
</tr>
<tr>
<td>Feelings of alienation</td>
<td>Experiencing lower self-esteem</td>
<td>6</td>
<td>0</td>
<td>67</td>
</tr>
<tr>
<td></td>
<td>Feeling different than friends</td>
<td>28</td>
<td>73</td>
<td>23</td>
</tr>
<tr>
<td></td>
<td>Feeling left-out by friends</td>
<td>17</td>
<td>29</td>
<td>39</td>
</tr>
<tr>
<td></td>
<td>Missing leisure activities</td>
<td>90</td>
<td>60</td>
<td>20</td>
</tr>
<tr>
<td></td>
<td>Not wanting others to see me</td>
<td>17</td>
<td>11</td>
<td>69</td>
</tr>
<tr>
<td>Disease- and treatment-related worries</td>
<td>Worry being left-out by friends</td>
<td>13</td>
<td>0</td>
<td>50</td>
</tr>
<tr>
<td></td>
<td>Worry for changed appearance</td>
<td>30</td>
<td>60</td>
<td>36</td>
</tr>
<tr>
<td></td>
<td>Worry for pain from procedures/treatments</td>
<td>24</td>
<td>45</td>
<td>43</td>
</tr>
<tr>
<td></td>
<td>Worry missing school</td>
<td>57</td>
<td>25</td>
<td>79</td>
</tr>
<tr>
<td></td>
<td>Worry not getting well</td>
<td>32</td>
<td>79</td>
<td>21</td>
</tr>
</tbody>
</table>

* Aspects are alphabetically ordered within each area. ** Number of respondents range 36-47, † number of respondents range 41-52.
Physicians rated six aspects with a sensitivity and specificity close to or above 60%: infections, mucositis, nausea, pain from disease, pain from procedures and treatments and losing hair, indicating reasonable accuracy for detection of physical problems. Nurses rated five aspects with a sensitivity and specificity close to or above 60%: infections, mucositis, nausea, losing hair, round face and weight loss/gain, thus in lower agreement than physicians concerning pain, but higher concerning round face and weight changes. For aspects belonging to the areas feelings of alienation and disease- and treatment-related worries, sensitivity/specificity were generally unbalanced, with either a sensitivity or specificity around 30% or lower. The findings indicate that it is difficult for staff to identify problems of a psychosocial nature, a finding in concordance with previous results (91).

From a clinical point of view and in the present setting, it seems reasonable to assume that sensitivity is more important than specificity to certify the identification of problems in need of consideration for some kind of intervention. Low ability to identify absence of distress may not have as serious consequences. Therefore, it seems especially important that staff show high sensitivity to aspects that adolescents consider as the worst. For five of the six aspects that most adolescents considered overall worst, staff showed high or reasonable sensitivity, i.e. for mucositis, nausea, pain from procedures and treatments, missing leisure activities and worry not getting well. For worry missing school, physicians showed a low sensitivity (25%) and nurses a sensitivity that was only marginally better than chance.

For most aspects, staff tended to overestimate rather than underestimate the distress experienced by the adolescents. However, physicians underestimated levels of distress for more adolescents with regard to mucositis, worry missing school and missing leisure activities than for whom they overestimated distress. Nurses underestimated levels of distress for more adolescents with regard to mucositis, worry missing school and infections than for whom they overestimated distress. In Study III, we reported that mucositis and worry missing school were among the six aspects that adolescents rated with the highest prevalence, levels and considered as the worst. Hence, staff tended to underestimate distress caused by some of the aspects that distress most adolescents the worst. Thus, mucositis and problems related to school need increased attention from staff.

The fact that staff tended to overestimate the distress experienced by the adolescents may not be a problem for the adolescents, but if problems are overestimated, staff may feel overwhelmed or reluctant to approach the adolescents out of misdirected consideration. As adolescents may try to avoid to burden their parents with their concerns (92), it is important that adolescents, if they wish, get regular opportunities to talk with physicians and nurses without parents being present. The staff may then get a truer picture of the adolescents' concerns, and lack of concerns, which may help them to prioritise care and recourses.
Worry not getting well was the aspect most often mentioned as the overall worst by adolescents, physicians and nurses. Still, the number of adolescents mentioning the problem was low compared to physicians and nurses (Table 8), a result that indicates that even though many adolescents consider the life threat to be very distressing, this is not as outstanding for as many adolescents as staff seem to believe. Rather, the adolescents’ distress seems to be more evenly distributed among the investigated distress areas.

Table 8. The top five aspects of distress considered as the overall worst according to adolescents (n=53), physicians (n=48) and nurses (n=53). Respondents could report more than one aspect as the worst.

<table>
<thead>
<tr>
<th>Adolescents (n)</th>
<th>Physicians (n)</th>
<th>Nurses (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Worry not getting well (11)</td>
<td>Worry not getting well (32)</td>
<td>Worry not getting well (23)</td>
</tr>
<tr>
<td>Pain from procedures/treatments (9)</td>
<td>Pain from procedures/treatments (5)</td>
<td>Mucositis (6)</td>
</tr>
<tr>
<td>Mucositis (7)</td>
<td>Pain from disease (2)</td>
<td>Pain from procedures/treatments (5)</td>
</tr>
<tr>
<td>Nausea (7)</td>
<td>Losing hair (2)</td>
<td>Pain from disease (4)</td>
</tr>
<tr>
<td>Worry missing school (6)</td>
<td>Feeling different than friends (2)</td>
<td>Fatigue (4)</td>
</tr>
<tr>
<td></td>
<td>Missing leisure activities (2)</td>
<td></td>
</tr>
</tbody>
</table>

**Agreement on anxiety and depression**

Five adolescents (10%) were identified as potential clinical cases for anxiety and ten (19%) were identified as potential clinical cases for depression. For anxiety, physician ratings were more accurate for the presence (75%) than for the absence (54%) of adolescent self-reported anxiety whereas nurses were more accurate for the absence (82%) than for the presence (60%) of adolescent self-reported anxiety. Staff ratings of depression were more accurate for the absence (60%, 71%) than for the presence (56%, 50%) of adolescent self-reported depression. There were statistically significant correlations between staff and adolescent ratings concerning level of anxiety. However, staff reported significantly higher levels of anxiety and depression than did adolescents.
Methodological discussion

Perspectives

In Study I and II, our aim was to describe the investigated phenomena as entirely as possible. Therefore, we assigned equal value to each informant's statement, no matter from a child/adolescent, parent or nurse. In consistency with the explorative purposes of the studies, we argued that disparate reports might reflect child/adolescent situation and behaviour under different conditions. In Study IV, we changed the perspective as we took the adolescent's reports as the point of reference. As physicians and nurses are involved in medical treatment, decision-making, and prioritise care and use of health care resources, we thought it imperative to find out whether staff's assessments of a certain adolescent's physical and emotional function agree with the adolescent's assessment of his/her own function.

In adult care, it is commonly accepted that patients are the best raters of their function in different regards, however, in paediatric care, the issue is not indisputable. Some argue that when possible with regard to age and health status, self-report is preferable as there is little concordance between child and proxy assessment (93). Others call for caution that children/adolescents may not disclose their concerns when asked about sensitive/difficult matters (29, 94).

The participating nurses and physicians sometimes mentioned the proxy problem, i.e., whose assessment that should be regarded as most "true". In connection to an interview, a nurse said: "I answer this way, because that's what I think he's been through, but I think he will answer differently. Teenagers don't want to admit that they've got problems". Parsons and co-workers (92) have argued for the need to reframe the question about proxy reporting from "Who is right?" to "What does each rater contribute to our understanding?". Even if the issue of gold standard among raters is complex (92, 95), we believe that assigning the adolescent's ratings the value of gold standard was justifiable in Study IV as it gives an idea of the accuracy of physicians' and nurses' assessments of physical and emotional function among adolescents with cancer.
Design and Samples

In a recent review on symptom experiences of children and adolescents with cancer, Docherty (22) concluded that since paediatric cancer is rare, many studies struggle with methodological problems such as small and heterogeneous samples. Most studies employ cross sectional designs, include children at various ages, and at various time points on and off treatment. There is a lack of longitudinal studies on distress among children and adolescents with cancer (22, 96-98). As a consequence, the generalisability of the findings from many studies is limited. In Studies I and II, many of the above mentioned limitations exist. We did our best to avoid the impact of the limitations by dividing the sample in age groups and by presenting the answers of the respondents in the age group the child belonged to at the time of active treatment. Still, there might be a problem of recall bias. Some data concern distress experienced several years ago whereas other data concern recent or present experiences. Naturally, one might recall an experience differently, depending on if the experience is recent or more distant. However, we believe that the results are valuable in identifying patterns of common and uncommon experiences of cancer in childhood/adolescence and that the derived knowledge can help healthcare providers to identify individual child and adolescent needs.

In Studies III and IV, we consider the samples strengths of the studies, as the adolescents were relatively homogenous in terms of time from diagnosis (4-8 weeks), age group and treatment. As cancer in adolescence is uncommon, consists of a multitude of diagnoses which are treated in numerous ways, the sample still is heterogeneous and relatively small which limits the possibilities for subgroup analyses. Only a small number of adolescents suffering from CNS-tumours were included in the studies as chemotherapy treatment was an inclusion criterion. This circumstance can speculatively affect the results of perceived distress, as youngsters suffering from CNS-tumours have been reported to be especially affected by physical and social problems (99). Similarly, as adolescents suffering from leukaemia or lymphoma were in majority, it is not an unexpected finding that pain from procedures and treatments are more prevalent than pain from disease. However, in order to get a sample of the present size we included adolescents with cancer for over four years at three of the six Swedish paediatric oncology centres. The inclusion period could have been halved had all centres agreed to participate. In the future, in order to get sample sizes big enough to draw valid conclusions, and at the same time optimise the utilisation of time and money, many research projects should be national, including all paediatric oncology centres. Thus, all centres should make joint decisions about which projects to prioritise.

The adolescent non-participation rate of 31% in Study III and 33% in Study IV might be considered substantial. It has been argued that adoles-
cents have a tendency to mask distress (76) which may be a reason why some adolescents chose not to participate. After a range of unwelcome information and musts in relation to the cancer diagnosis, to decline the invitation to participate in a study was maybe the first opportunity to reject a request. As mentioned in the methods section, when the coordinating nurses asked the adolescents for participation, this was done very cautiously and great consideration was taken to the adolescents’ integrity and wishes, which may have increased the number of adolescents not accepting participation.

When we analysed for potential differences between participants and eligible non-participants in Study III concerning age, sex or survival, the findings indicated a certain equality between groups on these variables. However, in Study IV, in which adolescents with a relapse were eligible, more participants than eligible non-participants were alive in May 2004 ($\chi^2 [1] = 4.11$, p< 0.05) indicating that eligible non-participants were worse off than participants.

In Studies I, II and IV, registered nurses and nurse assistants are joined together as “nurses”, and we have not aimed to analyse for differences between these groups. It is possible that nurses and nurse assistants do not share the same perceptions about the situation for children and adolescents with cancer. However, in Sweden, nurses and nurse assistants, to a great extent, enact in the same nursing tasks and communicate much with each other. Therefore, it is not unreasonable to believe that nurses and nurse assistants would agree on many aspects. Still, there might be different perceptions due to educational background.

Telephone interviews

Due to the long distances between the three participating centres in Studies III and IV, the data were collected by telephone interviews, as, for practical reasons, it was difficult to perform face-to-face interviews. Our impression is that most adolescents appreciated the relative anonymity of the telephone contact. Agreement between telephone and face-to-face assessment of anxiety and depression have previously been shown to be very good for young adults (100). However, health ratings have been shown to vary by method of administration, ratings being more favourable for telephone and face-to-face administration than for postal response administration (101, 102). As our telephone interview-administered results of the SF-36 subscales Mental Health and Vitality were lower than postal-administered norm values, it may be speculated that differences between our data and norm data would have been greater if methods of administration had been the same for both samples.
Measures

Distress

One strength of Studies III and IV, is the use of an interview guide, developed from results from Studies I and II, specifically constructed to investigate disease- and treatment-related distress among this population while most studies aiming to investigate the distress of children and adolescents with cancer have used adapted versions of questionnaires developed for use with adults (22, 43). The content validity of the interview-guide could be regarded as supported by the inductive procedure from which it was developed. However, the data through which the aspects were derived were collected approximately ten years ago. We cannot conclude whether this circumstance has any consequence for the content validity of the interview-guide. It could however be speculated that some aspects would not be identified and that some new aspects would be identified if adolescents with cancer were interviewed today about what aspects of disease and treatment that bothers them.

When adolescents in Study I were asked to mention an especially distressing event, none described an event connected with fear. In Study III, the adolescents assigned the highest mean value to the aspect missing leisure activities, while the aspect worry not getting well was assigned the seventh highest mean value. However, when the adolescents were asked which aspect they considered overall worst, most adolescents answered worry not getting well. A discussion about which result that is most "true" is naturally tentative, but probably necessary. Canning and co-workers (30) have reported that a higher proportion of adolescents with cancer than healthy adolescents were identified as employing a repressive adaptation style, i.e., reporting low levels of distress and high levels of defensiveness, and concludes that adaptive style may influence research findings as repressors seek to convince themselves and others that they are doing well. In addition, Berard and Boermeester (76) have argued that many adolescents may experience emotional vulnerability as weakness. In an interview situation, when adolescents are asked an open question such as "Can you tell me about an especially distressing event", it is not unlikely that they will choose to tell about something less personal than existential worries. Similarly, when adolescents are asked to rate their concerns about not getting well on a verbal scale, some adolescents will probably choose to give a low rating, possibly to protect him- or herself from unwelcome thoughts. Contrasting items is a way of asking the respondent to consider the distress impact of an item in relation to other items. As an example, a boy answered that he was little worried about not getting well and very much worried for changed appearance. He commented his ratings by saying: "It seems unnecessary to worry about not getting well as I tell myself I am going to be well. That's why I worry about my
looks, looks are not as important. However, when he was asked to rate which aspect that was the worst of the Disease- and treatment-related worries, he mentioned both aspects. The results imply that conclusions about which aspects that are considered the worst can not be drawn from comparisons of mean values. Thus, it is crucial to consider how questions are asked when interpreting the significance of the answers.

Anxiety, depression and well-being
The Cronbach's alpha value of 0.54 for HADS-D in Study III requires some consideration when interpreting the results for depression. Items nr 10 and 14 do not fit into the structure of the depression scale, with correlation coefficients of -0.02 and 0.08, respectively. Many adolescents commented in relation to item 10 ("I have lost interest in my appearance"), that they had changed their view upon certain matters, and thought that looks were not that important as before the cancer diagnosis. Therefore, they might have given a high score on that particular item. This score may more indicate a constructive coping strategy instead of a sign of depression. The problem with item 14 ("I can enjoy a good book or a radio or TV program") might be related to that, due to decreased energy levels, distraction as watching TV is one of the few "normal", and thus constructive, things an adolescent can do during cancer treatment. A score on this item might thus be poorly associated with the total depression score. Thus, there is a risk for false positive cases on depression. Still, the cut-off score of 7 for HADS-D recommended for use with adolescents was employed as others have demonstrated that it provides optimal sensitivity and specificity for adolescents (75). In Study IV, the low Cronbach's alpha values for the adolescent ratings of HADS-A and HADS-D (0.64 and 0.56, respectively) make the interpretation of the staff assessments for HADS tentative. As Eiser and Morse have pointed out, if the reliability of a measure is low, agreement between raters can never be high (103). Taken together, the findings indicate that HADS-D might not be suitable for screening of depression among adolescents on cancer treatment. Furthermore, as there do not exist norm-values for HADS for Swedish adolescents, the results on levels of anxiety and depression are difficult to interpret. The fact that the mean value on the Mental Health subscale (MH) was lower than corresponding Swedish norm data might indicate higher levels of anxiety and depression for adolescents in this study than for adolescents without cancer. Scores on HADS and MH have been reported to be associated (81). In this study, the correlation coefficients between MH and HADS-A and HADS-D were −0.49 (p= 0.001) and −0.56 (p< 0.001) respectively.

Adolescents scoring in the clinical range of anxiety and depression were in minority. However, the findings indicate a low mental well-being among the adolescents, implying that all was not well. Well-being may be a more appropriate measure of psychosocial function among this population than
anxiety and depression. When interpreting the scores of well-being for the adolescents in Study III, caution must be taken, as there exist no Swedish norm data for children <15 years old. However, norm data for SF-36 and HADS for Swedish youngsters 13-23 year, are presently collected by von Essen and co-workers.

Analyses

Content analysis

When considering how the interrater agreement for the categories was established in Study I, one might argue that there is a risk of inflated Kappa values. The risk should be due to the fact that the categorisations of some recording units were changed in order to clarify the coding system after an additional assessor had assigned the recording units to categories but before the Kappa values were calculated. However, we consider the risk minimal as very few changes were done as a result of this procedure.

Mass significance

In Study III, we highlighted some statistically significant findings. However, a great number of statistical analyses were performed on the same data, which increases the risk for type one errors. Still, to avoid the risk that clinically significant results would go undiscovered, we chose to set the alpha level at 0.05. If meaningful relationships are found, hypotheses can be formulated and investigated in future studies.

Sensitivity and specificity calculations

There are no absolute cut-off points for what is considered good sensitivity and specificity for a test, as this is highly dependent on the clinical situation in which the test is used. In general, both sensitivity and specificity should be satisfactory, as a high sensitivity alone may indicate a systematic overestimation and a high specificity alone may indicate a systematic underestimation. Sensitivity/specificity percentages can be expected to be 50% by chance alone. Sensitivity and specificity of screening and diagnostic tests used in routine health care are often in the 70 – 100% range, but lower values are not infrequent (104). In Study IV, sensitivity and specificity values of around 60% and higher were accompanied by significant associations between adolescent and staff ratings when significance analyses were performed. Thus, as a basis for discussing the accuracy of the staff ratings, we established coexisting percentages for sensitivity and specificity around 60% and higher as indicating reasonable agreement. In contrast to traditional sig-
nificance analyses most commonly used to analyse similar data sets, sensitivity and specificity calculations do not correct for chance or provide significant \( p \) values. However, the choice to express the accuracy of staff ratings in terms of sensitivity and specificity has the advantage of making it possible to discuss staff ratings of adolescent distress, anxiety and depression in terms of properties of many tests used in health care.
Conclusions and Implications

The results from Studies I and II, aiming at exploring experiences of having cancer while being young, indicate that the major physical aspects of distress are of a rather similar nature across age groups, i.e. pain from diagnostic procedures and treatments, nausea and fatigue. The emotional aspects of distress vary more between age groups. The results indicate that for children 8-12 years emotional concerns were frequent, especially worrying about death.

Cancer during adolescence is connected with a range of aspects of distress such as fears of alienation, altered appearance and dying, and various physical concerns. Positive aspects related to being treated for cancer are positive relations to staff and being well cared for. Good care for adolescents with cancer mainly consist of meeting nice, friendly, supportive and competent staff. The inductively derived aspects of distress formed the basis for an interview-guide, used in Studies III and IV, aiming at studying distress experienced by adolescents recently diagnosed with cancer. The conclusions from Studies III and IV are presented below:

Conclusions with methodological implications:

- Ratings of prevalence, levels and worst aspects of distress do not necessarily agree. Therefore, it is crucial to consider how questions are asked when interpreting the significance of the answers. These results are of value for future development of questionnaires.

- Adolescents scoring in the clinical range of anxiety and/or depression were in minority. However, the findings indicate a low mental well-being among the adolescents, implying that all was not well. Well-being may be a more appropriate measure of psychosocial function among this population than anxiety and depression.

- The Cronbach's alpha values of 0.54 and 0.56 (Study III and IV, respectively) for HADS-D, with two items correlating poorly with the depression score, indicate that HADS-D might not be suitable for screening of depression among adolescents on cancer treatment.
Conclusions with clinical implications:

- The aspects worry missing school and mucositis, are rated among the top six aspects regarding prevalence, levels and aspects perceived as the overall worst according to the adolescents. At the same time, these aspects are more commonly underestimated than overestimated by physicians and nurses. Thus, school concerns and mucositis need increased attention from staff.

- Physicians and nurses are reasonably accurate at identifying physical distress in adolescents, it seems however more difficult for them to correctly identify problems of a psychosocial nature. They frequently overestimate the distress caused by these aspects. The results imply that identification of and action on adolescent problems in relation to cancer diagnosis and treatment need to rely on direct communication.

- Physicians and nurses are reasonable accurate in identifying the presence of the majority of the aspects that adolescents consider as the overall worst, i.e., mucositis, nausea, pain from procedures and treatments, missing leisure activities and worry not getting well. Thus, staff may be able to identify problems in greatest need of consideration for some kind of intervention.

- Pain from procedures and treatments is more common among those who scored as potential clinical cases of depression. These adolescents seem particularly vulnerable, scoring in the clinical range of both physical as well as emotional strain. It is thus important to intervene against both pain and depression.

- Adolescents treated at a local hospital more often scored as potential clinical cases of anxiety. This finding is tentative due to the small number of adolescents treated at a local hospital, but worth keeping in mind when meeting these adolescents, and possibly worth exploring further.
Ongoing studies

The project “How do adolescents with cancer cope with disease- and treatment-related aspects of distress?” is a prospective, longitudinal study, in which the participating adolescents are interviewed by telephone at approximately four to eight weeks, six, twelve and eighteen months after diagnosis. Besides reporting on distress, anxiety, depression and well-being, the adolescents are asked to answer questions about behavioural and cognitive strategies used to cope with distress. At two, three and four years after diagnosis, the adolescents are telephone interviewed about negative and positive consequences of having had cancer during adolescence, and are asked to answer questions measuring anxiety, depression and health-related quality of life. At four years after diagnosis, the adolescents are asked to answer questions measuring their health-economical situation. To our knowledge, there is no other study, national or international, in which adolescents with cancer longitudinally are studied from four to eight weeks after diagnosis with regard to these variables. It is our hope that this data, when reported, will provide valuable information about the disease trajectory of cancer during adolescence.
Acknowledgements

I’d like to thank the following people:

First of all, all the children and adolescents with cancer who participated, as well as their parents, nurses and physicians. You have all taught me so much.

Louise von Essen, my chief supervisor, for asking me to work on the projects, for generously sharing her time and vast scientific knowledge, and for all her hard work and help.

Gustaf Ljungman, my co-supervisor, for his clinical wisdom and his time, enthusiasm, support and advice.

Anders Kreuger, my co-supervisor, for his encouragement, support and invaluable knowledge in the field of paediatric oncology.

The late professor PO Sjödén, head of the unit for most of my time at the Section of Caring Sciences, who taught me to see possibilities rather than obstacles.

Marianne Carlsson, head of the department, for revealing the world of science to me when I was a student nurse, and for continuously being such an extraordinary role model over the years.

Per Lindberg, head of the unit, for always being there with help and sound advice.

Coordinating nurses Britt Fällman, Yvonne Håkansson, LizBeth Blennnerup Persson, Lise-Lott Pettersson and Lotta Öijen for their invaluable help with recruitment and for the constructive discussions.

Peter Nygren and Inger Skolin, my co-authors. Working with you has been a pleasure.

Thanks to everyone at the Section of Caring Sciences, in particular: Senior researchers Eva Denison, Kerstin Gustafsson, Claudia Lampic, Karin Nordin, Anne Söderlund and Lena Wettergren, for willingly sharing their time and knowledge, and for their shrewd advice on a number of scientific matters.

Majbritt Sundelin for her administrative support and for being patient with all my questions. I do find economics a mystery.
Camilla Fröjd, Kristina Haglund, Susanne Lorentz, Elisabet Mattsson and Ulrika Pöder for being the most amusing, brilliant, clever, supportive and beautiful work mates a woman could ever wish for.

Frida Anteson, Cilla Arving, Karin Eriksson, Elisabet Wasteson and Jeanette Winterling for being not bad either.

Pernilla Åsenlöf, for friendship and emotional handholding when we were in the parallel process of completing a thesis.

Gunnel Larsson, Birgitta Johansson and Lena-Marie Peterson, who came and went before me, for their guidance on being a PhD student.

Thanks also to:

Helene Almqvist and Lena and Jonas Rönnbäck for being there.

My parents Bertil and Evy Hedström, to whom I owe everything.

My brother Tomas, his wife Helene and their children Anna and Eric, for obvious reasons.

My children Joel and Mira, for being precious beyond belief.

My beloved Claes; If I was anything at all into sentimental romantic comedies, I'd probably call you my match made in heaven, but as I'm a serious researcher, I'll just say: Thank you.

This thesis was supported by grants from the Swedish Children Cancer Foundation and the Swedish Cancer Society.
References


78. Gandek B. Personal communication. 1999 Jan 25.
Acta Universitatis Upsaliensis

Digital Comprehensive Summaries of Uppsala Dissertations from the Faculty of Medicine 18

Editor: The Dean of the Faculty of Medicine

A doctoral dissertation from the Faculty of Medicine, Uppsala University, is usually a summary of a number of papers. A few copies of the complete dissertation are kept at major Swedish research libraries, while the summary alone is distributed internationally through the series Digital Comprehensive Summaries of Uppsala Dissertations from the Faculty of Medicine. (Prior to January, 2005, the series was published under the title "Comprehensive Summaries of Uppsala Dissertations from the Faculty of Medicine").

Distribution: publications.uu.se
urn:nbn:se:uu:diva-4832

ACTA UNIVERSITATIS UPSALIENSIS UPPSALA 2005