ON SURVIVING CHILDHOOD CANCER

Late psychosocial consequences for patients, parents, and siblings

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ON SURVIVING CHILDHOOD CANCER

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Genezen van kinderkanker: late gevolgen voor het hele gezin

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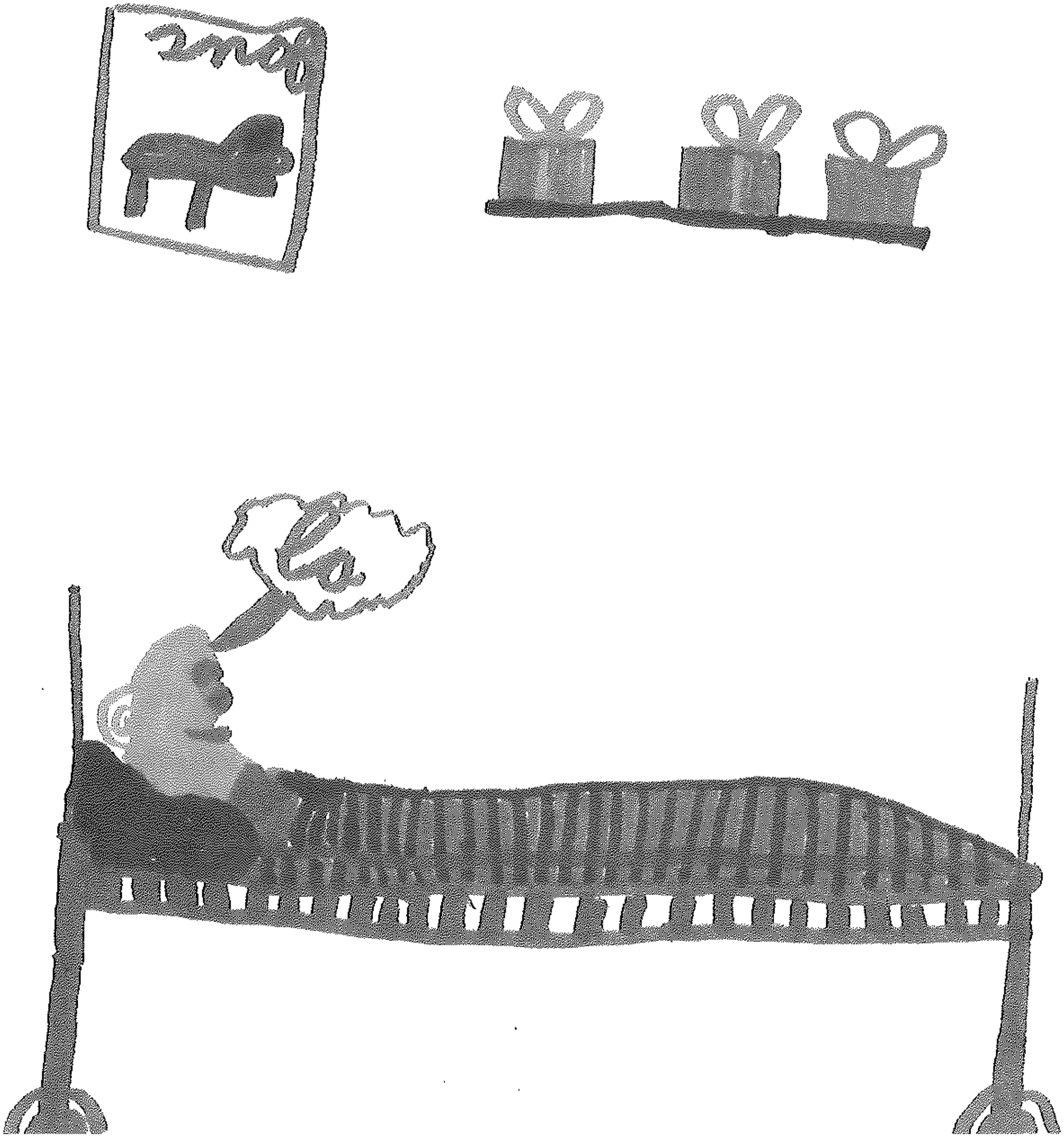
From Sybil. Daniel Petri, USA, 1976, 198'.

One day Dr. Wilbur asked Sybil:
"How do you prepare yourself for the misfortunes in your life?"
"You never are prepared for catastrophes", replied Sybil, "They just happen;
And that's what makes you prepared."

Opgedragen aan ouders van een kind dat kanker heeft gehad, zoals de ouders van

Ian, Sylvia, Bart, Vincent, Manon, Rudi, Roel, Tessel, Astrid, Gert-Jan, Arjan, Cindy, Richard, Giaco, Leonard, Jerry, Johan, Ramon, Marjolijn, Martin, Martijn, Hessèl, Maikel, Yvonne, Marvin, Rob, Esther, Hubert, Björn, Chantal, Jeroen, Mattias, Debbie, Alexander, Jonathan, Claudia, Derk, Charissa, Willemien, Jos, Miranda, Annick, Marijna, Dino, Ellen, Wouter, Björn, Peter, Mark, Fleur, Martin, Patrick, Martine, Sandra, Erik, Jan, Margreet, Richard, Karin, Martin, Jack, Marjolein, Valerie, Chris, Erica, Peter, Niels, Femke, Natascha, Johan, Anita, Dennis, Bas, Elke, Frouke, Joost, Dirk-Jan, Paul, Alicia, Pieter, Ilse, Chantal, Henk, Louise, Pieter, Ludo, Arjan, Karla, Manuel, Akko, Richard, Andrew, Ronald, Roy, David, Charlton, Marie-Louise, Sjoerd, René, Diana, Marielle, Sanne, Evelien, Janneke, Robert-Jan, Gerbrand, Dennis, Sharissa en Jordy.



Arjan, 8 yrs (sibling of a childhood cancer survivor)

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Chapter 1: General introduction

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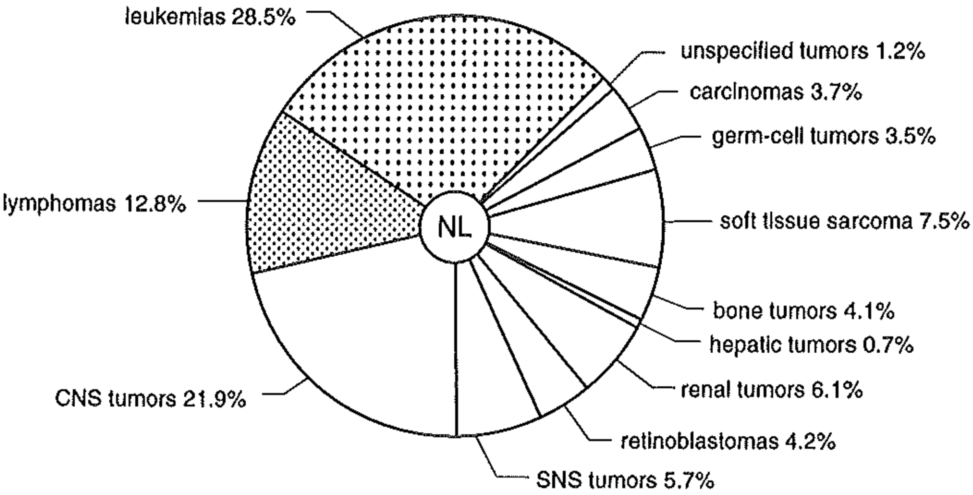
CHAPTER 1.1

MEDICAL ASPECTS OF CHILDHOOD CANCER

INCIDENCE

Childhood cancer has an incidence of ~13 per 100,000 children younger than 15 years of age (1-3). In the Netherlands, every year approximately 370 children are diagnosed with cancer (1,2). Although childhood cancer is a relatively rare disease, it is the most common cause of death from disease in children older than 1 year in industrialized countries (4,5).

The main types of malignancies in the pediatric population differ significantly from those of adults (6). The malignancies that affect children are rare in adults and vice versa. The common childhood malignancies are of embryonic origin, whereas in adults the principal types are carcinomas (6). The distribution of childhood malignancies in The Netherlands (1) and the U.S.A. (3) are presented in Figure 1. Overall, the distribution pattern is comparable between the two countries, although there is some variation in the incidence for particular types of malignancies Differences in incidence between the two countries can be attributed to various causes: Firstly, the data in The Netherlands and the U.S.A. are collected in different time periods and time trends in incidence of cancer are reported (3,5,7). Secondly, there are geographic and racial differences between the two populations. Thirdly, there might be a selection of cases in the U.S.A. as a result from registration procedures. In The Netherlands with a population of 15 million people annually approximately 370 cases of childhood malignancies are registered which represents a virtually complete registry (1). In contrast, in the U.S.A. with a population over 250 million, approximately 620 cases per year are registered (3,8). Considering the above mentioned causes for variation in incidence of childhood cancer, the differences between the two countries are surprisingly small.



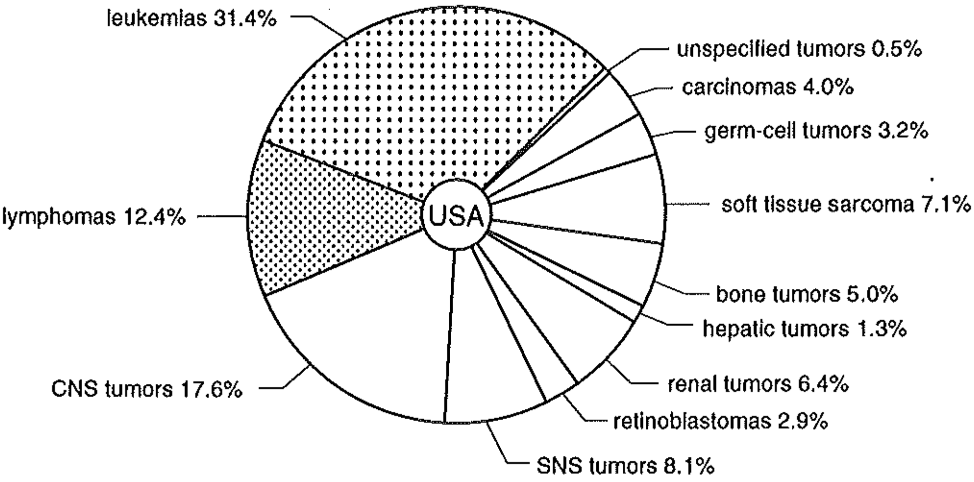


Figure 1. Distribution of malignancies in children less than 15 years of age in The Netherlands (period 1989-1991; based on ref. 1 and the U.S.A. (period: 1983-1991; based on ref. 3. Shaded area: hematopoietic malignancies. Abbreviations used: CNS, central nervous system; SNS, sympathetic nervous system.

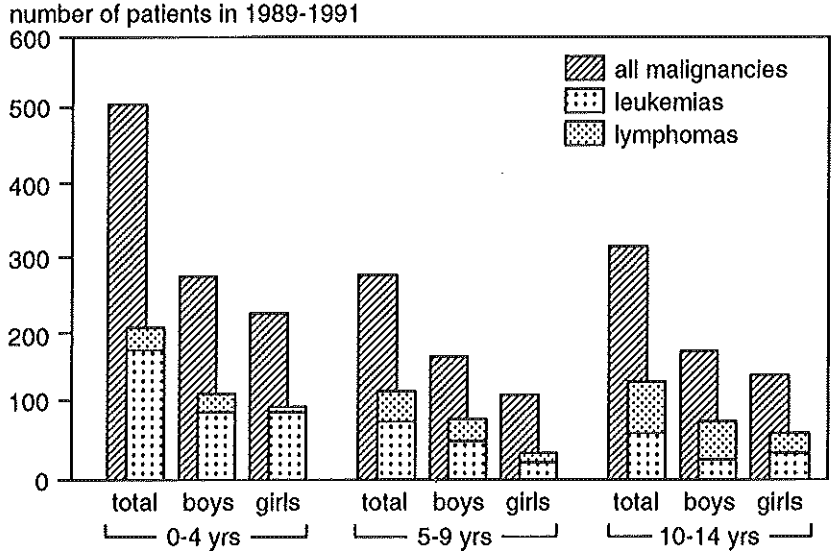


Figure 2. Total number of malignancies in children less than 15 years of age according to age at diagnosis and gender in 1989-1991 in The Netherlands (based on ref. 1).

The two main categories of childhood malignancies are hematopoietic malignancies (consisting of leukemias and lymphomas) and solid tumors consisting of all other types of cancer. The most frequent type of cancer is leukemia accounting for approximately 30% of all malignancies (3,9), followed by tumors of the central nervous system and lymphomas (Figure 1). Most types of cancer can be divided into several subtypes. Leukemia includes two main subtypes: acute lymphoblastic leukemia (ALL), which is the most common subtype, and acute non-lymphocytic leukemia (ANLL).

Young children up to 5 years of age are more frequently diagnosed with cancer than children older than 5 years of age and more boys than girls are affected (Figure 2). Leukemias and lymphomas are good examples of the higher incidence in males as compared to females (Figure 2). For each type of cancer, however, the incidence rate differs with age and sex (4,5). For instance, the peak incidence for leukemias (Figure 2) and kidney tumors is before the age of 5, whereas the incidence of lymphomas and bone tumors increases with age (Figure 2) (4,5,7).

SURVIVAL RATES

Many childhood malignancies respond much more favorable to treatment than adult cancers (6,10). Progress in the biomedical sciences and technology have resulted in a

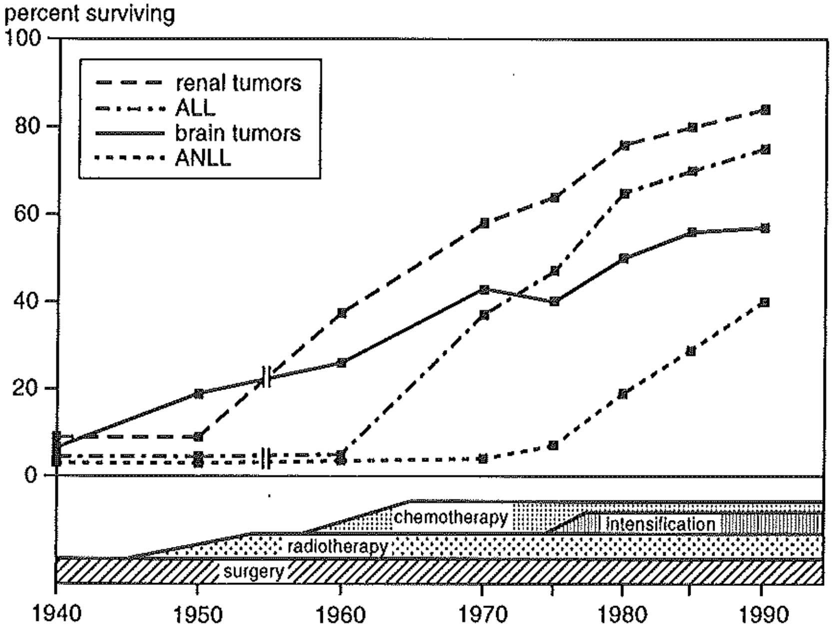


Figure 3. Survival rates for ALL, ANLL, brain tumors, and renal tumors in children < 15 years of age. During the last five decades combined and improved treatment modalities have gradually increased survival rates from less than 20% to ~70% (for 1940-1950 2-year survival rates are given, for 1960-1990 5-year survival rates are given; first three decades represent estimated values from the literature; adapted from refs. 6,11,12,45).

spectacular improvement of survival rates for children with cancer (Figure 3). In the 1940s and 1950s less than 20% of children survived their disease for more than 2 years (6,10). In the late 1960s and early 1970s progress started as a result from the combined application of surgery, radiotherapy, and chemotherapy. During the last two decades further improvement was obtained by more aggressive and intensive treatment regimens (11). With modern cancer treatment, between 60% and 70% of all children with cancer survive their disease over 5 years (12). This implies that today cure is not an exception but the norm in childhood cancer (13).

Survival rates differ for each (sub)type of childhood cancer. Among the cancers with the best 5-year survival rate are solid tumors, such as retinoblastoma (~95%) and renal tumors (~85%), whereas children with ANLL have by far the poorest 5-year survival rate (~25%) (3,12). Current results from clinical trials on ALL show a 5 year event-free-survival rate ranging from 60% to 75% (9,14). In spite of the advances in treatment, most childhood cancer patients die from leukemia (all types), accounting for ~40% of all childhood cancer deaths (15).

TREATMENT

To achieve cure in childhood cancer chemotherapy, surgery, and irradiation are used. The nature and duration of treatment is dependent on the (sub)type of cancer and its clinical and biological features (6). The aim of treatment is not only to eradicate the primary tumor but also its micrometastases in patients with a solid tumor (16) and 'minimal disease' i.e. low numbers of circulating malignant cells in patients with leukemia (17). These low frequencies of malignant cells generally remain undetected and represent a concealed tumor load (16,17). These micrometastases have to be eliminated with chemotherapy (16) because low frequencies of malignant cells can cause a relapse of the disease during or after treatment. At relapse, generally treatment is reinitiated, but the chance for cure decreases with each relapse. Because of the likelihood of relapse, patients are monitored over a prolonged period of time, also after cessation of treatment.

Leukemia

In The Netherlands, treatment protocols for children with ALL are designed by the Dutch Childhood Leukemia Study Group (DCLSG). The last two protocols (ALL-VII and ALL-VIII) have been developed in close cooperation with the Berlin-Frankfurt-Münster (BFM) Study Group. Although treatment protocols for childhood leukemia may differ per center or per country, all current treatment protocols comprise four basic elements: remission induction, central nervous system (CNS) preventive therapy or CNS prophylaxis, intensification, and maintenance therapy (9). In recent Dutch protocols (ALL-VII and ALL-VIII), induction treatment consists of 5 weeks chemotherapy aiming at inducing complete remission, which is defined as the absence of clinical signs and symptoms of the disease in blood, bone marrow or elsewhere in the body. With modern chemotherapy, complete remission is achieved in almost 98% of children with leukemia (9).

During induction therapy, CNS prophylaxis is given to eradicate leukemic cells which passed the blood-brain barrier. CNS prophylaxis has made a major contribution to the increased survival rates in leukemia. Most children receive either intrathecal methotrexate or intrathecal triple-agent therapy, consisting of methotrexate, hydrocortisone, and cytarabine (9). Soon after remission induction, intensification of treatment starts. The dose of chemotherapeutic agents is increased or new agents are administered to tackle the problem of drug-resistance. In protocol ALL VII and VIII, patients receive new combinations of chemotherapeutic agents until 28 to 32 weeks after diagnosis. Then maintenance therapy is administered over a prolonged period of time. It has been shown that treatment protocols for leukemia with a relative short duration of treatment (less than 16 months) resulted in a high relapse rate, whereas prolonging treatment over 3 years showed no significant improvement in survival rates (9). Apparently, the optimal duration of treatment ranges within this interval; for current Dutch and BFM protocols, the duration of treatment is 2 years.

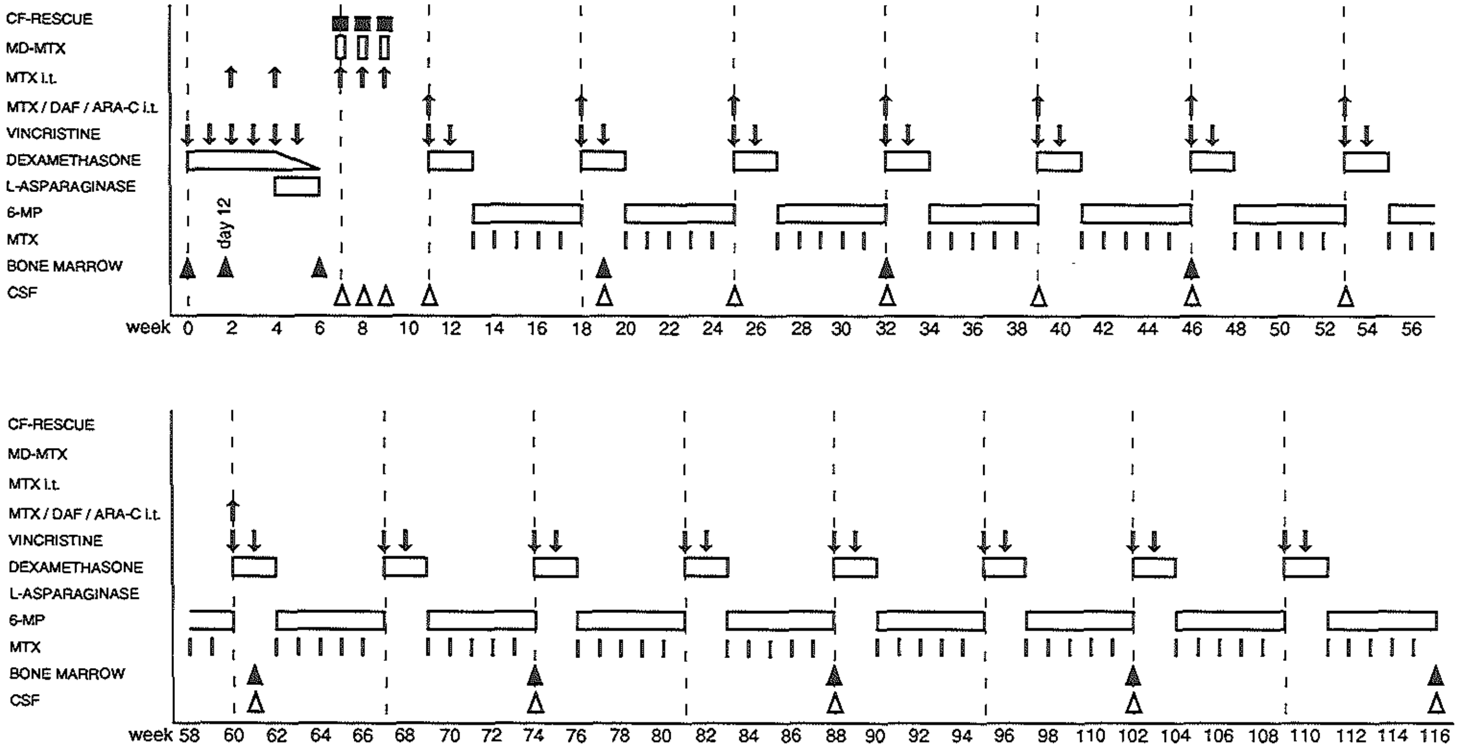


Figure 4. Treatment regimen of the Dutch Childhood Leukemia Study Group (DCLSG) for ALL patients who were diagnosed in 1984-1988 (ALL VI). Protocol ALL VI: Onderzoek en behandeling van kinderen met acute lymfatische leukemie zonder tekenen van verhoogd risico bij diagnose (ALL-NHR). DCLSG, The Hague, The Netherlands, 1984.

Chemotherapy is the primary option in treating childhood leukemia, but children who respond unfavorably to chemotherapy (18) or children who relapse (9) may be treated with bone marrow transplantation (BMT). Preparation of patients for BMT consists of intensive chemotherapy most often combined with total body irradiation, which totally destroys the bone marrow. Afterwards the patient is transplanted with bone marrow from an appropriate donor by intravenous infusion. Autologous BMT is an alternative option for patients without an appropriate donor (19).

Solid tumors

The treatment of solid tumors is dependent on the (sub)type, site, and progression of the tumor (20). In contrast to the treatment of leukemia, surgery makes a major contribution to the treatment of solid tumors in combination with chemotherapy and radiotherapy. These latter two treatment modalities are administered to eradicate microscopic extensions of the disease and to prevent metastases.

Surgery is generally used for diagnostic purposes such as taking biopsies for staging of the tumor i.e. characterizing the extent of the disease. If possible, surgery is also used for removal of the tumor at the primary site and for the management of metastatic disease (20). Multiple operations may be required. Surgery is aimed at excising the primary tumor completely, preferably with a margin of normal tissue surrounding the tumor. Depending on the bulk of the tumor, large resections may be required, such as removing affected organs or limbs. However, not always can surgery be used in the treatment of solid tumors. In case of an irresectable tumor or metastases, irradiation and/or chemotherapy might be applied (20).

A number of tumors are responsive to chemotherapy and/or radiotherapy. In the majority of solid tumors preoperative chemotherapy is administered to reduce tumor mass. Consequently, less extensive surgery can be performed (20,21). After removing the tumor, intensive courses of 'adjuvant' chemotherapy are administered when the risk of recurrence at distant sites is known to be high. In most of the common cancers, including Wilms' tumor, Ewing sarcoma, rhabdomyosarcoma, and osteosarcoma, adjuvant chemotherapy is used (21). The total duration of these treatment regimens varies from 6 to 9 months (22-24).

DIRECT SIDE EFFECTS OF TREATMENT

Treatment for cancer is long and intensive which is illustrated by the DCLSG ALL-VI treatment protocol in Figure 4. This treatment protocol was used between 1984-1988 and is demonstrated, because many patients in our study have completed this protocol. The treatment protocols for cancer necessitate frequent hospital visits for check-ups and treatment. The medical procedures used in cancer treatment such as bone marrow aspirations, lumbar and venous punctures, and injections or infusions with chemotherapeutic agents are painful and distressing for the child.

A number of acute toxicities are associated with chemotherapy (21). These side effects are common, when effective doses of these agents are administered. Direct side effects of chemotherapy include nausea and vomiting, allergic reactions, mouth ulcers, loss of hair, weight gain, and weight loss. In addition, chemotherapy can induce serious suppression of the immune system. As a result normal childhood infections such as varicella (chicken-pox) can be life-threatening. Without special precautions, infections are a common cause of death in children with malignancies (25). To reduce this chance, children are protected from infections by isolated treatment in the hospital, avoiding contact with other children which are or might be infected, and adhering to special (germ-poor) diets. More recently in special cases G-CSF can be applied to stimulate the maturation of granulocytic cells to shorten the period of susceptibility for bacterial infections (26-28).

Side effects related to radiotherapy are reactions of the skin, food aversion, nausea and vomiting, hair loss, and fatigue (29). Also visceral effects have been noted, including problems of lungs, liver, and kidneys and effects on the CNS. After irradiation of intracranial tumors an exacerbation of neurological symptoms can be observed. A serious effect on the CNS is post-irradiation necrosis which might occur between 6 months and 2 years (29).

LATE SIDE EFFECTS OF TREATMENT

With increasing survival rates, the price of progress in medical treatment of childhood cancer became apparent. There is a growing body of literature documenting the late effects of cancer and its treatment. In a study on residual disabilities in former childhood cancer patients, 73% of the patients had residual effects of disease and treatment, of which 41% were severely affected (30). The type of late effects is related to the cancer site, the age of the patient, and the specific therapeutic modalities to which the patient has been exposed (31).

Physical side effects

Several disorders of the endocrine system due to irradiation and chemotherapy have been found (32). Growth failure is a result of cancer treatment inducing endocrine dysfunction (33-36). Short stature is associated with cranio(spinal) irradiation and therefore frequently found in children treated for leukemia and brain tumors. Severe growth retardation, defined as a standing height below the 5th percentile has been found in as many as 35% of patients with brain tumors (31). Also a loss in final height is observed in leukemia patients (34,35,37-39). Besides growth failure, obesity is a common problem in children successfully treated for brain tumors and leukemia (37,40-44). Not only cranial irradiation but also steroids, are responsible for weight gain. In a recent study from our center on obesity in children treated for ALL, the highest prevalence of obesity was found in patients treated with a combination of steroids i.e. prednisone and dexamethasone (44).

Gonadal dysfunction and reproductive failure have been observed in male and female survivors of childhood cancer caused by irradiation or chemotherapy (45,46). Permanent sterility is induced by irradiation of the gonads, e.g. total body irradiation for bone marrow transplantation or testicular irradiation in case of recurrence of leukemic cells in the testes. Furthermore, impairment of the gonadal system may require hormone supplementation for a prolonged period of time. In a study of 2283 long-term survivors, relative fertility varied considerably according to sex, site of cancer, and type of treatment (47). Whereas a depressed fertility up to 60% was found in male survivors who have been treated with chemotherapy with alkalylating agents, for women this association could not be demonstrated. For those survivors who are fertile after treatment, concern remains about their ability to have normal pregnancies and normal children (31). Studies have shown that offspring of childhood cancer patients have neither an increased risk of cancer (31,48) nor an increased frequency of congenital abnormalities (49).

A variety of other sequelae has been documented, such as musculoskeletal abnormalities, absence of limb, cardiopulmonary dysfunction, hepatic and renal failure, blindness, and hearing loss (30,31,50,51). Besides the functional handicaps inherent in a number of these disabilities, they may have cosmetic implications. Also teeth damage, problems with hair growth, scars, facial deformities, short stature, and obesity have a direct effect on the child's appearance.

Second malignancies

A recent serious concern is the development of second malignancies in children who have had cancer. Survivors are at an increased risk for cancer because of a possible genetic predisposition to cancer and because of the exposure to radiation and chemotherapy (5,52). Paradoxically these treatment modalities not only cure but also cause cancer because their cytotoxicity affects not only the tumor cells but also normal cells (6,53). In a frequently cited study of Neglia and co-workers (54), a substantial excess of second malignancies in survivors of ALL was found. The overall estimated cumulative proportion of patients with a second malignancy was 2.5% at 15 years after diagnosis. Compared to the risk in the general population, there was a 7-fold excess of all cancers and a 22-fold excess of CNS tumors. Especially children treated with cranial irradiation who were ≤ 5 years of age at diagnosis, were prone to developing CNS tumors.

Neuropsychological side effects

Disease and treatment can have potential deleterious effects on future intellectual functioning. Children treated for brain tumors declined significantly in intellectual ability and academic performance, a substantial number showing severe disabilities (55-58). Another group at risk are children with leukemia who receive CNS prophylaxis to prevent CNS relapse. The first effective type of CNS prophylaxis consisted of cranial irradiation with 2400 cGy plus intrathecal administration of methotrexate (59). As it became apparent that cranial irradiation at a dose of 2400 cGy had adverse side effects on neuropsychological functioning (58,60-62), the dose was reduced to 1800 cGy. However, it has been shown that irradiation with doses even as low as 1800 cGy had an undesirable effect on IQ (63,64). Especially children treated younger than 3 years of age have an increased risk (58,60,63,65). These cognitive impairments have negative effects on the child's academic achievement and educational late effects are frequently observed (40,58,66,67). It was shown that up to 40% of leukemic patients developed learning disabilities (30,68), necessitating special education (67).

The documentation of late effects has led to an increased awareness of the potential deleterious side effects of cancer treatment and the need to refine treatment to prevent late sequelae in children. Since adverse effects of cranial irradiation used in CNS prophylaxis became apparent in children with ALL, the Dutch treatment regimens for children with standard risk ALL were adapted in 1984. Cranial irradiation was eliminated from the treatment protocols and instead children were treated with chemotherapy administered intrathecally and intravenously. Currently cranial irradiation is only reserved for those with a high risk of CNS relapse (9). This adaptation in CNS prophylaxis did not negatively affect survival rates, showing that this new type of treatment was as effective as cranial irradiation (69). As a result most children in The Netherlands and the U.S.A. receive nowadays chemotherapy instead of irradiation as prevention of CNS involvement. However, also these new treatment protocols are potentially neurotoxic and their adverse sequelae remain to be evaluated. Late effects of new treatment protocols cannot be assessed rapidly, because their effects can be delayed or can only be measured at a later developmental stage.

Decision making and ethical dilemmas

Traditionally in pediatric oncology, there is great concern about the ethical issues in treating children with cancer. New developments in childhood cancer treatment have direct implications for the patient and create new ethical dilemmas (70). Randomization in clinical trials, options for choice in treatment, innovative treatment protocols, and more aggressive treatment for those with poor prognosis can have far reaching consequences for the quality of life of the survivor. One is faced with the dilemma which side effects are acceptable for the child's immediate *and* later future. Another issue is whether a further improvement in survival rate is acceptable, if increased sequelae must be tolerated. Or can one choose for less adverse side effects but higher mortality rates? These dilemmas are further complicated by the fact that the child has a limited capacity to make decisions about treatment, because of his psychological, cognitive, and physical development (71). Children are primarily focused on the immediate consequences of cancer and its treatment (71,72). Most children are therefore unable to evaluate or incorporate implications of treatment for later life (71). Finally, who defines and decides what is acceptable for a minor who cannot choose for him- of herself? According to Pinkel (53) the risk of each drug or treatment modality negatively affecting the quality of survival must be carefully weighed against its contribution to cure. Studying the late consequences of cancer and its treatment is an essential step in the process of defining the 'risk/benefit' ratio in childhood cancer.

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CHAPTER 1.2

PSYCHOSOCIAL ASPECTS OF CHILDHOOD CANCER: A REVIEW OF THE LITERATURE*

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ABSTRACT

The literature concerning the psychosocial aspects of childhood cancer is reviewed. The emphasis of the literature is on the impact of the disease on the child and family and depicts the life-situation when faced with childhood cancer and responses to the disease. The literature strongly indicates that families experience serious difficulties and are a population at risk of developing psychosocial problems. The approach of studying the child with cancer and family is discussed and suggestions are made for future research.

INTRODUCTION

Progress in the field of paediatric oncology has drastically raised the rate of survival in childhood malignancy (1). Childhood cancer, once regarded as an acute fatal illness, has become a chronic life-threatening disease (2). The psychosocial aspects evolving from the improving medical outlook of childhood cancer are reflected in the literature. The early literature, written at a time when the child with cancer nearly always died, dealt almost exclusively with the problems of the fatally ill. Diagnosis was almost synonymous with death and the child was treated as a dying patient from diagnosis onward. Progress in medical therapy transformed the psychological dilemma from one of adapting to the imminence of death to one of coping with uncertain survival (3). Along with this change, the attention of the literature shifted to the new needs of this population of cancer patients with the focus on the preparation for living with childhood cancer. Modern cancer treatment, albeit very sophisticated, cannot guarantee cure in every case and therefore the early literature on the fatally ill child with cancer is still relevant today.

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The aim of this paper is to review the literature on the psychosocial aspects of childhood cancer, delineating the major areas of interest. The emphasis is on the impact of disease and treatment on the child and family. Although childhood cancer is a relatively rare disease, it yields significance for understanding child development and family functioning in general.

There is a considerable body of literature on the psychosocial aspects of childhood cancer (4), depicting the life situation of the children and their families exclusively and in a detailed manner. Although much of this picture is based on anecdotal descriptions and clinical impressions, investigative studies are being reported with increasing frequency and the emphasis will be on them. The papers reviewed in this article are selected on their: (a) contribution to the psychosocial field of childhood cancer (measured by the number of times referred); (b) exploration of a new issue associated with childhood cancer; (c) addition of views relevant to the field (including articles on child development and on other diseases); and (d) accessibility to the original source, which meant a practical limitation. The structure of this paper is related to the course of the disease. First some notes are made on aetiology. Then diagnosis and treatment are separately discussed for the child and family. Later the issues related to the result of the treatment will be dealt with: first, the dying child and after-effects on the family; and secondly, the child surviving the disease, the long-term survivor.

AETIOLOGY

Childhood malignancies are a heterogeneous group of diseases and have varying aetiologies attributed to genetic, immunological and environmental events (5). Among the possible factors related to cancer incidence, psychological factors and stress have been mentioned (6). Very few studies have explored the psychogenic etiological factors in relation to childhood cancer, suggesting that stressful life events may constitute one of the multiple conditions determining the development of childhood malignancies (7-10). However, studies investigating psychological factors and stress in relation to illness onset have been firmly criticized on several methodological grounds (6,11-13). On the basis of the present data it is argued that there is little if any evidence that psychosocial factors are associated with the occurrence of cancer in children (5,12).

THE CHILD AND THE DIAGNOSIS OF CANCER

A major topic in the early literature was not the reaction of the child to the diagnosis but whether the child should be told the diagnosis. The key to resolving the controversy of telling the child involved the extent to which the child was able to perceive the seriousness of the disease and impending death. This resulted in different approaches to the ill child.

The proponents of the protective approach based their arguments on existing literature of the child's cognitive understanding of death. This literature revealed a developmental trend in the child's understanding of death (14-17). Children under the age of 6 usually view death as reversible. Older children gradually get a more mature conception of death, but not until the age of 10 does the child understand its universal and irrevocable nature (19-21). These immature conceptions of death convinced the proponents of the protective approach that the child has no fear of death, unless told of the potential fatal disease and as a consequence had to be shielded from the diagnosis (22-24). At the same time it was believed that the immature ego of the child was unable to cope with the fear of death (25). Furthermore, a lack of concern about the disease on the part of the child was assumed, based on the observation that children seldom ask questions about their illness (26,27).

In challenging the protective approach, Vernick and Karon (28,29) argue that trying to shield the child from the seriousness of the disease results in feeding the unrealistic fantasies of the child causing depression, withdrawal, and a heightened anxiety, which has also been observed by others (30-33). In addition, children will not ask further questions when they observe how upset parents become by the questions. Older children may even remain silent in an attempt to protect the adult (24,34). Due to misconceptions of the child 's awareness of death, the adult will remain silent, thereby overtly or covertly giving cues to the child not to talk about death. Karon and Vernick (29) refer to this situation of blocked communication as a 'conspiracy of silence'. To avoid the negative effects of this collusion the open approach advocated open communication in which the child feels free to express concern (34). It was noted that adjustment problems decreased when children could talk about their disease (28,29,35). In addition, observations demonstrated that even at an early age, children are aware of the life-threatening nature of their disease (36-41). Consequently, Spinetta (42) attacks the relevance of the existing literature on the awareness of death as studied in healthy children, suggesting that these findings cannot be generalized and might not apply to the fatally ill. A number of carefully controlled studies provide convincing evidence for the rationale of the open approach, demonstrating that even young children with cancer are aware of the seriousness of their illness and the threat of death (43-46).

Disease-related communication

In the literature in favor of the open approach, the impression is created that telling the child about the potentially shortened life-span may be accomplished in a single statement. This is due to the emphasis put on the child's awareness of the fatal disease, which showed the need for an open approach. However, the incorporation of this knowledge into disease-related communication has been neglected. Very few authors bridge the gap between empirical evidence and the way of approaching the patient. In an excellent article Spinetta (47) describes how best to talk to a child about a life-threatening disease while taking into account several factors concerning parent and child which may influence this communication. Furthermore, the literature seems to suggest that the open approach has to be employed in every case. Spinetta (48) thinks a word of caution is appropriate, lest the results in favor of the open approach are interpreted as a blank support for open communication in all families and all circumstances. Others have also discerned the limitations of an open communication (49,50).

Informing parents and children of a potentially fatal disease is a difficult task but it is also considered to be critical in establishing a basis for ongoing support during the treatment period. Basic outlines for the initial interview are described, focusing on the essential information to be discussed as well as on the attitude conveying this information (31,51-57). The importance of communication between doctors and parents is demonstrated by Johnson, Rudolph and Hartmann (58), who reported that 39% of all parents found 'unanswered medical questions' a major source of distress during the disease. Research has focused on the analysis of the complex verbal and non-verbal communication patterns between medical staff, parents and patients (59-62). Findings suggest that disagreement and misunderstanding in this communication may be responsible for seemingly unusual reaction patterns in the patient and family (63).

THE IMPACT OF THE TREATMENT PERIOD ON THE CHILD

When a malignancy is diagnosed treatment will be initiated because an untreated malignancy is fatal. Treatment is aimed at inducing a remission, characterized by the absence of signs and symptoms of the disease (2). Dependent on the nature of the malignancy, the treatment varies according to the combination of chemotherapeutic agents, surgical intervention and radiotherapy. The more complex the therapeutic regimen the more likely this may interfere with the child's rehabilitation (64). Most often children have to undergo aggressive treatment for a considerable time. A relapse may occur during or after treatment. Then treatment has to be re-initiated but the prognosis of the disease becomes worse after each relapse. During treatment the child is faced with serious threats which involved the following:

The life-threatening nature of the disease

The most striking aspect of cancer is the life-threatening nature of the disease. As pointed out in the previous section concerning the diagnosis, the child is aware of the seriousness of the disease. This awareness is associated with an increase in anxiety (42). It is demonstrated that children with cancer experience significantly more anxiety related to the seriousness of their illness than children with non-fatal chronic illnesses or healthy controls, even though this concern may not always take the form of overt expressions about death: "Whether or not one wishes to call this nonconceptual anxiety about the child's fatal illness 'death anxiety' seems to be a problem of semantics rather than of fact" (44, p. 844). How the child becomes aware of the potential shortened life-span is described by Bluebond-Langner (37,38), outlining different stages of awareness the child passes through. She argues that this process is not directly related to the child's age but a function of the child's experience with the disease and treatment. While some 4-year-olds have reached the final stage of awareness, e.g. the internalization of the prognosis, some older children, still in first remission with few clinic visits and hence less experienced, passed through the first stages only. Also, Spinetta and Maloney (43) noted that the child with cancer experiences more anxiety with the progression of the disease and with each clinic visit. It is assumed that this increase in anxiety makes the child more liable to psychosocial problems (33). Howarth (65) compared a group of children with fatal disorders, including leukemia, and non-fatal disorders. He found a 40% prevalence of psychiatric disorders in the fatally ill group as against 8% in the non-fatally ill group. In the same way the relation between fear of death and separation is evident. Lansky and Gendel (66) observed in children with malignancies an extreme separation anxiety with regressive behavior which resulted in a symbiotic relationship with the mother. Remarkably, in the history prior to the diagnosis there were no signs which could predict the pathological interaction between mother and child.

With the progress in medical outlook the imminence of death is replaced by uncertain survival. As remission lengthens there tends to be a continued movement towards health and life. However, this change is accomplished within the shaky context of ever-present doom (67,68). Even though children in remission indicated that they had overcome the threat of death, it appeared that the anxiety associated with it was not dispelled (69). It is this chronic uncertainty about relapse and survival that poses a significant threat for the child living with cancer (70,71).

Physical discomfort

The modern methods of cancer treatment are not without side effects. A major complication is the physical discomfort caused by the treatment, such as pain, weakness and illness (72). For many children feeling relatively well at diagnosis, the treatment seems worse than the disease (64,73). Cancer treatment involves a high degree of acute distress, generally caused by (a) anxiety and pain associated with medical procedures, such as bone marrow aspirations, lumbar and venous punctions and injections with chemotherapeutic agents, and (b) nausea and vomiting resulting from chemotherapy and from conditioned anxiety associated with treatment (74). These recurrent procedures pose a significant problem in the adjustment to childhood cancer which may manifest itself in a variety of symptoms such as insomnia, nightmares, anorexia, withdrawal, and depression (75-78), and which may increase the risk of non-compliance with the medical regimen (74,79,80). No habituation pattern in the distress caused by medical procedures could be established, which indicated a need for clinical intervention in order to reduce this particular type of distress (76). Several interventions are proposed to ameliorate anxiety, pain and discomfort associated with cancer treatment, which include hypnosis (81-85), drugs (85), behavioral techniques (77,86), procedural preparation (87), biofeedback and other forms of counseling (88).

Bodily changes

Children undergoing cancer treatment experience a number of bodily changes. These side-effects of treatment may be reversible, such as weight gains or losses, loss of hair, mouth ulcers and even acute psychosis (89), or may be permanent, such as amputation, sterility, organic brain damage and skeletal abnormalities. The latter will be discussed in the section on long-term survivors. The impact of visible disfigurements, especially the loss of hair, has received considerable attention in the literature. The actual change in physical image is a constant reminder for the child with cancer that he or she is different from other children (72,73). The side effects of treatment continually change the child's body image and a number of authors express their concern about the potential loss of self-esteem induced by changes in body image (58,91-93). A lack of self-confidence resulting from physical appearance might lead to regressive behavior, a withdrawal from peers and a fear about going to school, thus affecting emotional and social adjustments and academic performance (73).

Hospitalizations

Usually children with cancer are hospitalized many times during their illness. Although there is a considerable amount of literature on the effects of illness and hospitalization on children, this hardly includes children with cancer. It has been well documented that repeated and long-term hospital admissions can cause a specific pattern of emotional reactions (94-97). In the early literature on the fatally ill child with cancer the child's reactions were particularly related to the fatal character of the disease. Hospitalization was not only experienced as a temporary separation but also as a reminder of the final separation. Clinging, signs of distress, despair, withdrawal and depression were associated with this separation anxiety (26,30,31,98). Only recently have issues concerning hospitalization entered the literature as a result of the introduction of protected environments in the treatment. These are used to reduce the rate of bacterial infection, a potentially life-threatening complication to chemotherapy (99). It has been noted that patient care in reverse isolation may increase the risk of psychological disturbance because it implicates a reduction of social contacts and sensory stimulation (100). Although psychological problems are reported, including anxiety, depression and behavioral changes in sleep and play (78,100), the change in patients' emotionality seemed to be influenced by the severity of the disease rather than the degree of isolation (101,102). It could not be concluded that reverse isolation inevitably leads to psychosocial problems, but this favorable outcome may be due to the high degree of involvement of parents and medical staff, especially with young children, who are less capable in generating their own sensory stimulation than older children (99,100,103,104).

Outpatient care

With modern cancer treatment patients are less hospitalized and after initiation of treatment medical care will often be provided on an outpatient basis. This is advocated in order to decrease the emotional side-effects associated with hospitalization and to facilitate the patient's return to family, friends, school, and community while still undergoing rigorous treatment. There is an emphasis on having the child return to a lifestyle as normal as possible because only a physically and emotionally successful rehabilitation will result in a truly cured child (70,73,93,105-107). In this respect the re-entry to school is highlighted because of its central role in a child's normal day-to-day life (108,109).

There are a number of factors that can hamper a good reintegration in schoollife. The child may be reluctant to go to school for several reasons: worries about the reaction to the changes in physical appearance, the loss of contact with peers, and difficulties in following the curriculum (73,110). Extreme separation anxiety may also prevent a successful return to school. High rates of school phobia among children with a malignancy as compared to the normal school population have been reported by Lansky et al. (111). Their observations could not be confirmed by other studies, which found that school phobia was not typical of paediatric cancer patients and that they go to school willingly (112,113). However, children with cancer do have difficulties at school, but these are not of a pathological nature. Deasy-Spinetta (112) found subtle differences between cancer patients and their healthy controls in school-related behavior. On the emotional level they are less likely to express freely their positive as well as their negative feelings and in social contacts they tend to play a more passive role than their controls. With respect to their academic performance, a higher rate of learning disabilities was detected as compared to their peers (112,114). Apart from emotional problems, a decline in academic performance can be attributed to factors such as side effects of treatment, the attitude of the teacher and parents, and the frequent absences from school (107,114-117).

Many authors offer suggestions to facilitate the academic re-entry of the child under treatment, e.g. establishing communication between parent, hospital, and school (58,73,111). The teacher's need of both emotional support and information concerning the child's illness and treatment is emphasized because the teacher's attitude to the child influences the success of the child's re-integration at school to a high degree (93,117). Some programs for teachers are designed to remove misconceptions of cancer and to give practical advice about the management of the child at school (112,118).

The meaning of cancer to the child

The stresses inherent in cancer have overwhelming effects on the child. According to Brunnquell and Hall (119) it seems that almost every aspect of cancer and the ensuing treatment has implications in the areas of separation-loss and control-competence. These mutually influencing areas recur in the various stages of development. The life-threatening nature of the disease implies a separation from and loss of loved ones. Not only does the separation from important people induce loss but also the illness itself (120,121), such as a loss of health, a loss of body parts through surgery and a loss of contributing to family life (72,119,122). The child's new role of being sick or being different involves a change in nearly every aspect of the child's life: they will experience a change in relation to other people and in the perception of themselves (123). This change can be regarded as a loss associated with illness in general and has often been overlooked in the literature in favor of the loss associated with death. Furthermore, illness has major consequences for control and competence, since from infancy onwards the major developmental changes have always been directed towards a greater control of the self and to autonomy. The illness deprives the child of control of daily routine, privacy, relationships with family and friends, and control of life itself (124). In addition, the greater dependency on others, the restrictions in mobility and activity, the invasion of treatment and the constant medical attention all effects the child's sense of control and competence (58,119).

The implication of cancer for the child varies with the course of development. Cancer and its treatment affect different aspects of control-competence and separation loss in the various age-groups. Although increasing attention is paid to the specific interference of cancer with the various stages of development, investigative studies of these issues are few. Often the impact cancer will have in a certain stage of life is described (69,73,105,119). Most authors focus on the problems of the adolescent with cancer (72,122,125-134).

THE CHILD COPING WITH CANCER

The term 'coping' is a core concept in the literature concerning the impact of childhood cancer on the child and family, and is used to denote how children adapt to their disease and treatment. Lazarus (135) defines coping as a process activated when threat is perceived, intervening between threat and observed outcome, and aimed at regulating emotional distress and eliminating the threat. In attaining this goal coping behavior or strategies operate to decrease the disturbing feelings associated with the threat. The extent to which the negatively tuned effect is reduced is a measure of the effectiveness of the coping process. In this respect the importance of coping strategies have been emphasized because the ways people cope with stress may be even more important to psychosocial functioning than the frequency or severity of episodes of stress themselves (136,137). Not surprisingly, the literature tried to identify coping behavior related to the outcome of the coping process (138). Patients, as well as their parents, were divided in 'good' and 'poor' copers depending on their use of effective or ineffective coping strategies, as was evaluated against criteria of problem behavior or psychopathology. The most important strategies related to positive outcome were open communication and honesty (139,140), maintaining hope and the effective use of defense mechanisms, in particular denial. Denial, once considered as a destructive defense mechanism in the case of illness (127), is now regarded as playing a crucial role in facing day-to-day reality and in maintaining a positive outlook for the future (62,63,68,141-143).

With this approach both the dynamics of coping (135,144) and factors influencing the coping process got too little attention. In an excellent review on stress, coping and development, Rutter (145) states that it would be an error to assume that the only source of difference in the outcome following stress events concerns strategies or tactics dealing with these events. There is no most successful strategy but modes used will vary in effectiveness along with the type of stress, situational context and the characteristics the individual brings into the situation (145). Among the major individual and situational determinants of the child's adaptation to cancer, characteristics related to the disease, the child's developmental level and social environment are mentioned (64,69,121). With respect to the latter, it is important to study the child within the context of the family because this provides the framework for the major adjustments made by the child (146-148). Although for ease of survey the issues of the child and family are separately dealt with, it is demonstrated that they are closely connected (29,66,75,149).

The child 's adaptation to cancer

The outcome of the child's coping efforts with the total set of threats associated with cancer has drawn much attention because of its role in the development of psychosocial problems. Behavioral problems and emotional disturbance have often been reported, such as anxiety, fear, depression, extreme dependency on the parents, sleep disturbances, regression, anger and withdrawal (23,29,31,33,69,74-76,78,82,150-152). According to Susman et al. (153), the emphasis on identifying pathology and the discussion of the child's behavior in terms usually reserved for psychiatric patients resulted in a pessimistic view of the child with cancer. Futterman and Hoffman (113) were the first to suggest that the usual criteria for assessing pathological behavior cannot easily be applied to the observed behavior in a child with cancer because this behavior might have adaptive value in extreme situations. The criteria for assessing psychopathology are based on a physically healthy population and normalcy is often defined as an absence of symptoms. In contrast, the symptoms of the child with cancer may be regarded as appropriate in the circumstances (123). Consequently, there is a need to establish base rates of this behavior, providing a criterion against which the responses of the child with cancer can be evaluated (153-155).

Several studies have demonstrated that the psychosocial functioning of patients with a chronic illness, including cancer, is one of psychologic normalcy rather than deviance, although the pediatric cancer patient experiences additional problems (131,143,156-162). In refutation of the suggestion that psychosocial maladjustment is inevitably associated with chronic illness, including cancer, some studies (143,158) overemphasize the similarity between physically healthy and ill adolescents in overall psychological functioning. Spinetta (155) points to the limitations of the research design as used in these studies. While investigations demonstrate that chronic illness does not inevitably preclude psychosocial adjustment, they show at the same time that the nature and duration of stress associated with cancer put the child at risk (68). In addition, Rutter (145) points out the necessity of investigating whether adverse experiences alter the course of subsequent development. Deasy-Spinetta (112) has demonstrated subtle differences, albeit non-pathological, between cancer patients and healthy controls which could not be detected by the usual standardized tests which discriminate normal from pathological populations of children. This shows the need to study the child's behavior from a non-pathological developmental perspective. A useful study approach, proposed by Susman et al. (104), assumes that the basic developmental processes still proceed under stressful conditions such as cancer, although these processes might be altered by the cancer experience, resulting in an atypical course of development. A deduction of this approach is that the reactions to the illness are likely to involve age-related developmental concerns and need not be pathological or synonymous with problems of adjustment.

Care for the child with cancer

In the literature two ways of management can be distinguished. First, supplying direct emotional support by important persons for the child, namely the family and the health care team. Second, arranging conditions to promote optimal developmental growth. This is reflected in the encouragement of treating the patient as normal as possible (discussed in the section of child-rearing), and of having the child resume normal activities (discussed in the section of outpatient care).

Emotional support by the family is provided by talking about feelings, fears,and hope, and by sharing concerns related to the disease. According to Spinetta and Maloney (140), children from families who openly discuss the disease "exhibit a non-defensive personal posture and express a long range close relationship with the parents and a basic satisfaction with the self" (p. 1541). Thus open communication can facilitate effective coping and give access to the intrafamilial sources of support.

When the child is frequently hospitalized the health care team tries to prevent any adverse effects of hospitalization (122,163-167). As evidence accumulated in favor of the open approach its philosophy spread to many aspects of the child's management and is reflected in articles describing how an empathic and supportive environment for the child with cancer is created (28,151,168-175). In stimulating open communication the various ways by which the child may communicate are acknowledged and age-related ways of communication are offered, such as play programs, art therapy, group-sessions, drawings and counseling (90,122,168,170,176-178).

THE FAMILY OF THE CHILD WITH CANCER

The stresses inherent in the cancer experience are different for child and family. For the child cancer means having a life-threatening disease and being confronted with own potential death and for the family it means living with a seriously ill child and the threat of losing a member of the family. The impact on the family has mostly been discussed in relation to the individual family member. Consequently, the issues for parents and siblings will be separately dealt with.

THE PARENTS AND THE DIAGNOSIS OF CANCER

The initial reactions of the parents have frequently been studied in the early literature and have to be interpreted while bearing in mind the fact that the child was expected to die within a relatively short period of time. Parents reacted to the diagnosis with shock (26,151,179). Anxiety, grief, anger, hostility, guilt, and disbelief are among the responses most often observed in parents (26,30,31,98,180,181). Despite the improvement in medical outlook, most parents display practically identical responses to the diagnosis of cancer (58,73,182-184), implying that for most parents a diagnosis of cancer in a child is still inextricably connected with death. Therefore parents have to be re-educated in cancer and its treatment in order to give them a realistic expectation of the future (58,59).

THE IMPACT OF THE TREATMENT PERIOD ON THE PARENTS

The life-threatening nature of the disease

The threat of losing a child varies in intensity and is related to the child's medical condition. Learning the diagnosis for parents is the first serious confrontation with the potential death of their child. The initial parental reactions to the diagnosis have a striking resemblance with the emotional and behavioral effects of normal grief at or after a death of a loved one as described by Lindemann (185). The term 'anticipatory mourning' is introduced to refer to parental reactions of mourning and grief prior to the final loss (186). Many authors observe this process, which starts after the parents learn the diagnosis (23,26,187-189). Viewed from the parental perspective, it is generally assumed that anticipatory mourning serves an adaptive function in case the child dies because the parent relinquishes the attachment to the dying child over time (188,190). Parents who had difficulties in accepting the fatal prognosis and as a consequence failed to mourn in anticipation found no relief after the death of the child (191). It has been suggested that the adaptive value of anticipatory mourning is related to time. There seems to be a minimal time required to complete the process of anticipatory mourning (26,191). This was important in the time when the child was expected to die after a short disease-course, but there is also a maximum time period up to which anticipatory grief has salutary effects, becoming actual with modern cancer treatment. Parents whose child died after a long illness had subsequent poorer adjustment (192). From the child's perspective the parental anticipatory mourning can be destructive for the relation between parents and child (193). This has been noted in those instances in which the child that has been realistically mourned for lived on (194,195). Easson (196) reported that families who had adjusted to a future without their child had serious difficulties when their child survived cancer. Professional help was required to re-integrate the child into the family.

With the improved prognosis the inevitable threat of loss is replaced by the uncertainty about the future. The impending death can no longer be considered as the primary stress with which the family has to cope but living with a chronic life-threatening illness (197). When the child is medically doing well the immediate threat of loss becomes remote and parents move from a state of anticipatory mourning to an increased hope for cure (69,147,198,199). It has been noted that hope appears to be inversely related to the presence of anticipatory mourning (31). With continued remission, affirmation of life becomes stronger, and most parents whose child was in remission experienced few difficulties and returned to normal life (200). However, when the child relapses the threat of loss becomes very actual and the process of anticipatory mourning is relived (30,31,201). Parents find the first relapse even worse than the initial diagnosis because it decreases the hope built on an improved treatment and the outlook of a better prognosis (179). Although parents continue to hope, their hope narrows with the progression of the disease, finally converging in living on a day-to-day basis (31).

Child-rearing

The parents' emotional reaction to the life-threatening nature of the disease influences their attitude of parenting. It is commonly advised that the child be treated as normal as possible (53), but many authors remark that parents overprotect and overindulge their child (31,58,91,151,202,203). Maguire (183) seriously doubts if it possible for a parent to treat the child as normal while being aware of the potential shortened life-span. In addition, rearing a child with a life-threatening disease makes extra demands on parenting skills, for example talking with the child about the disease, supporting the child, taking care of the child's physical condition and preparing the child for death as well as for living. The competence of parents is further challenged when the child bypasses them as a consequence of a transition of authority from parents to the medical staff (31). Therefore the need to reinforce parents in their role as educators is underlined (73,204).

The child 's medical treatment

For parents the child's treatment involves emotional as well as practical burdens in addition to the threat of loss. They must accept the rigors of treatment without any guarantee that it will cure the disease (72). Parents have to deal with informed consent procedures (58,197), their child's suffering, hospitalizations, repeated clinic visits (183), considerable financial expenses (205-207) and the disruption of family routines (72). Especially the change in family life is considered to be a crisis for families with a child with cancer (148,208). With the diagnosis of cancer each family member is not only faced with their own individual reaction to the child's illness, but must also adapt to the changing family rules and roles. In addition, the illness requires a re-orientation on family values, goals and expectations for the future. In terms of family system theory, a change in a part of the system affects other parts as well as the system as a whole. The family's integrity and stability is threatened because the existing interactive homeostasis will be redefined by the illness (116,204,209). The task for the family is to regain a state of equilibrium.

The stability of the family is often assessed by the intactness of the parents' marriage, e.g. rate of divorce among parents of a child with cancer. It has been noted that childhood cancer aggravates already existing frictions and causes marital problems (210-212). It was generally assumed that the rate of divorce was high in these couples (151). In contrast, it appears that parents of cancer patients have a slightly lower incidence of divorce than control groups (213). This corresponds with the findings of other studies (159,193,214,215). However, the assessment of the quality of the marital relationship revealed that parents of cancer patients experienced significantly more disharmony in their marriage. These findings indicate that the disease does not bring couples closer together nor does it increase the likelihood of divorce in spite of the very real marital problems (213).

PARENTS COPING WITH CHILDHOOD CANCER

From diagnosis onward parental behavior can be regarded as coping behavior, being defined as the total sum of all strategies employed by the individual to meet a significant threat (31). Parental behavior in response to the threat of loss as well as to the stresses inherent in caring for the child have been incorporated in describing stages of psychosocial adaptation to the disease (26,31,147,180,188,191,199,204,209,210, 216). Each stage has its own set of problems, and in outlining the specific stresses parents have to cope with, these stresses are redefined in adaptive tasks to accomplish. Thus coping with the stresses of childhood cancer implies accomplishing the following tasks: maintaining confidence, equilibrium and a sense of control (53,200), anticipatory mourning, and balancing emotional, physical and financial resources in order to meet the immediate needs as well as any future needs of the patient, siblings, and themselves (183,188). These tasks are specified in, for instance, accepting the reality of the diagnosis, informing siblings and other people of the diagnosis, establishing relations with the medical staff, and creating a normal family life. Coping behavior, used to accomplish these tasks, serves two functions: an externally directed one enabling the individual to master the situation, and an internally directed one protecting the individual from disruptive degrees of anxiety (31,187). The most often observed coping strategies are seeking information, searching for a meaning of the disease often related to religion, increased motor activity, communication, avoiding visits to the patient, seeking support and comfort, and the use of defense mechanisms including isolation of affect, rationalizing, and denial (30,31,72,180,188,200,204,217-219).

The literature concerning parental coping has almost the same research problems as outlined in the section of the child's coping process. Therefore only some issues will be elaborated further. The different definitions of coping and psychological disturbance complicate comparison of the various studies. Parental responses may be an indication of the coping strategy used as well as an indication of the outcome of coping with the disease. Furthermore, for some authors adaptive coping is synonymous with a complete absence of distress, for others signs of distress are regarded as a natural response in the circumstances (220). In addition, it is noted that coping strategies can have conflicting results (221). Parents' use of denial to ward off their own fear resulted in a decrease of anxiety but also in a limited perception of the needs of the other family members, leading to social isolation (220). Consequently, the observed adjustment or maladjustment is dependent on the psychosocial dimensions evaluated.

The parents ' adaptation to cancer

The parental response to the diagnosis can be characterized as a state of shock that may last for several weeks. Severe neurotic symptoms can easily develop during this phase. Anxiety and depression may occur in up to 50% of the parents, although most of these symptoms subside or spontaneously disappear during treatment (26,184,211). After recovering from the first shock many authors report that most parents are able to take care of their child and adequately fulfil their responsibilities (36,30,31,106,179, 182,215). Systematic follow-up studies confirmed that 1 year after the initial diagnosis parents cope well with the disease of their child (159,200). In contrast, several studies found serious problems for parents. Although there is a reduction in the level of distress as compared to the initial diagnosis, levels of anxiety, sleep disturbance, and depression remained high (184,222). Maguire (183) reported that 12-18 months after the initial period 28% of the mothers of leukaemic children were depressed and anxious to a morbid degree, while only 8% of the control mothers were similarly disturbed. In addition, it was found that the stress of coping with childhood cancer may cause health problems, alcohol abuse, social isolation, sexual and marital difficulties, and problems at work (69,152,183,190,203,210,223,224).

The discrepant findings of the studies may be attributed to the extent authors focus either on social functioning or on the emotional adaptation. Studies emphasizing the strength and resilience of families point to their capability of living with the disease and coping ability, their return to normal life and the intactness of the family. Studies reporting problems point to the psychological distress of these parents. Powazek et al.

(184) noted that although high levels of emotional distress were experienced, most families continued to function adequately. Thus, despite adequate day-to-day functioning, there does seem to be considerable emotional problems. Even long after the termination of therapy parents experience anxiety about the possibility of recurrence (69,212,225,226). Furthermore, there can be a discrepancy in observed and actual problems. In this respect the concept of hidden morbidity is of particular importance. Only one of the six mothers who developed anxiety, depression or sexual difficulties disclosed their problems to a member of the medical or psychosocial staff (183,211). Also, Magni et al. (222) found that it is difficult to uncover depression.

Unfortunately, in describing parental responses to the illness, situational and personal characteristics are frequently not taken into account, although several factors have been identified as major determinants of such responses. These include the physical condition of the child (49,71,200,227,224), the concurrent stresses for the family in addition to the stresses inherent in the child's illness (228), availability of support and psychosocial services (197,207,229), the existing interaction pattern within the family (220), the cultural context (229-231) and severe psychopathology in the parents prior to the illness (232). These characteristics as well as parental responses to the illness, such as the inability to communicate the seriousness of the illness and the ongoing experience of high levels of distress, may serve as predictors to identify families at risk of developing difficulties (155,200,211,233). Further studies are needed in order to provide corroborative evidence of whether such factors are reliable in predicting families most at risk (211).

Care for parents

In order to ameliorate distress, care for parents focuses on providing practical help, information and emotional support. Several programs have been designed to prevent or reduce parental problems (98,146,202,211,234-238). Special attention is directed to the role of fellow sufferers to enhance parental coping efforts and many authors advocate group sessions for parents of children with cancer (170,203,216,239-248). However, little is known about the effectiveness of the various methods, pointing to the need of studies that critically evaluate these programs (183).

SIBLINGS OF THE CHILD WITH CANCER

The main focus of the literature has been on the patient, parents and the patient-parent dyad, but increasing attention is now being paid to emotional problems in siblings (249-251). In a study Lavigne and Ryan (252) found that siblings of chronically ill children, including children with cancer, were more likely to show symptoms of irritability and social withdrawal than a control group of siblings of healthy children. Problems frequently noticed by parents are feelings of jealousy and guilt, academic underachievement, enuresis and acting-out behavior (73,184,212). Additional problems are observed in siblings who are donors in the case of bone marrow transplantations (150). Studies of siblings of chronically ill children have mainly been based on the observations of parents and health care professionals (250). Only recently have the siblings themselves been examined in these studies. Cairns, Clark, Smith and Lansky (253) found striking similarities in emotional distress between patients and their healthy siblings as to anxiety, social isolation, vulnerability to illness and injury, and self-esteem. In some areas the siblings showed even more signs of distress than the patients. A study of Spinetta (254) confirms these results.

The major theme in discussing the siblings' concerns is the disruption they experience in interpersonal relationships, especially with their parents (91,253,255). Siblings notice that parents treat the ill child differently and find themselves neglected (73,223,253). Siblings of leukaemic children felt more frequently the lack of a close relationship with their parents than siblings of other chronically ill children and simultaneously felt isolated from supportive systems outside the family (256). It was demonstrated that within the family the needs of the siblings are met least of all (254). Thus feelings of isolation, reported by many authors, are very real. Feelings of isolation, guilt and jealousy may be aggravated when the child has no reliable information to explain the change in parental attitude. Half of the families gave no or insufficient information about the disease to the siblings (91). Therefore strengthening the parents' awareness of the needs of the siblings will lead to greater support for them (254). In the same way, attention of health care professionals might directly focus on the siblings' needs (255,257,258).

THE DYING CHILD

The literature about the dying child deals mainly with the care for the dying child and family and the role of the health care team. In taking care of the child the critical aspects involve the amount of information the child ought to receive and how to respond appropriately to the emotional needs of the dying child (39,47,169,175,259-263). Anxiety and emotional isolation of dying children have been observed (32,33,45,90,264). The initiation of discussions about fear and fantasies related to death revealed that children respond eagerly, facilitating their adaptation to death (28,29,265,266). Kübler-Ross (267) and Bluebond-Langner (38) emphasize the children's need of sharing their knowledge about death and their need for emotional and physical proximity with their parents. Many authors underline the realistic view the child has about death, which may result in having the child in the final stages of cancer participate in discussions of therapy choice (268). Nevertheless, despite this realistic view, all kinds of defense mechanisms against the idea of death may still develop (266).

Many parents experience a renewal of anger and fear when the terminal stage is reached (269). The emotional responses of parents to the impending death are very similar to the dying patient's own reactions as described by Kübler-Ross (270), involving the stages of denial, anger, bargaining, depression and acceptance (203,242). In addition to emotional support, parents may need help in preparing the child as well as his siblings for death (25,198,267,269,271,272). When death draws near, information about the way the child may die as well as information about relieving the child's discomfort and pain will be helpful to parents (273-275). Often there is a need of close physical contact with the child and parents should be allowed to stay with their dying child in the hospital (276). An alternative is to offer parents the possibility to let the child die at home and to participate in a 'home care' program (277-281).

The literature describes how the health care team can offer substantial emotional support as well as help the parents to make practical arrangements before and after the child's death (22,25,260,261,282). Another aspect is the impact of the potential and actual confrontation with a dying child on the health care team and their response to it (236,264,284-291).

BEREAVEMENT

For the family the death of a child is a devastating experience and the literature is concerned with adaptation to this tragic event, in particular of the parents. In one of the most often cited studies Binger et al. (36) reported a high incidence (50%) of emotional disturbance in at least one family member after the child's death. Also, others have noticed various psychosocial problems among the parents as well as the siblings (191,192,224,233,271,292-294). In contrast, other studies (181,188,215,276) failed to find evidence of a pathological mourning process or other forms of psychosocial maladjustment, pointing to the coping resources of the families. Although pathology and maladjustment are not inevitable after the death of a child (295), some studies claimed that parents fully recovered to normal, ignoring the inevitability of change following bereavement (229,296,297). Other explanations that might account for the discrepant findings of the various investigations are the different methods of data collection and the different time intervals between loss and the moment of investigation because the affective and behavioral effects of grief may vary in onset and duration (185). Although the expression of grieving may alter with the lapse of time its intensity may not diminish, as was generally assumed. Studies showed that families can experience ongoing difficulties in coping with the loss (192,295). Another concern is the replacement of the child by a new baby, deliberately planned to substitute the deceased child as a result from unresolved parental grief (298-300). Therefore several authors advocate a continuing involvement with the family after the child has died (198,201,225,247,301-303).

LONG-TERM SURVIVORS

As the period of survival lengthens, the physical and psychosocial consequences of cancer and its treatment become increasingly important issues for the paediatric cancer patient.

Physical sequelae

Multiple long-term physical side effects have been reported for survivors (304-306), but only those will be discussed which have been studied in relation to psychosocial functioning. Some investigators (307,308) have suggested a possible relationship between psychosocial adjustment and a wide range of physical impairments in childhood. It has been shown that survivors who adjust well do so regardless of their physical handicap. However, there appeared to be a significant relationship between physical limitations or visible impairments and the marital status of the long-term survivor (68,309). Women with a higher degree of physical disability were less likely to marry, while the opposite seemed true for men. It was suggested that these unmarried women were unable to attain the cultural standards of feminine attractiveness. For men the critical factor in getting married is probably the ability to work and to earn a living rather than the presence of a physical impairment (309).

Another concern is the potential deleterious effects of disease and treatment on future intellectual functioning. Children treated for brain tumors declined significantly in intellectual ability and academic performance, young children being most affected (310,311). A substantial proportion of these patients show mental disabilities of severe magnitude (312-315). Another group of patients at risk are children with acute lymphoblastic leukaemia (ALL) because they receive preventive central nervous system (CNS) treatment. Many studies have been undertaken to assess the neurotoxicity of leukaemia treatment and it is well documented that children with ALL, receiving preventive CNS treatment, consisting of cranial irradiation and intrathecal chemotherapy, decline in overall IQ although they still function within the normal range (316-324). Young children had significantly lower IQ scores than patients who received the same treatment at older age (115). In addition, several studies report learning disabilities (326-327) and specific patterns of neuropsychological dysfunction, including disturbances in psychomotor speed and motor skills (328-330), distractibility and memory deficits (331,332). In contrast, some studies found no evidence of disturbances in intellectual functioning as a result of CNS prophylaxis (330,333,334). Apart from the several methodological problems inherent in evaluating cognitive functioning carefully outlined by several authors (325,335-339) and to which discrepant findings may be attributed, in their investigations the time that has elapsed between the initiation of CNS treatment and the psychological testing was much shorter than in those studies that did reveal changes in intellectual functioning. Consequently, there seems to be a latency period before deficits associated with CNS prophylaxis become manifest (340,341). In comparing several modes of CNS prophylaxis it is suggested that in particular cranial irradiation in addition to methotrexate therapy results in a decline of cognitive function (321,342-344). As a consequence alternative effective forms of CNS prophylaxis have been developed (345).

Psychosocial sequelae

Few studies have been undertaken to evaluate the psychosocial functioning of children surviving childhood cancer. Studies primarily dealing with the life-style of survivors concluded that most survivors had made good (346) or acceptable (347) adjustments. However, the global appreciations on which their findings are based limit their conclusions. A very extensive and well-designed study by Koocher, O'Malley and associates (68), involving a large number of survivors, indicates that cancer survivors have a high rate of psychological adjustment problems. Fifty-nine per cent of their former patients were found to have at least mild psychiatric symptom formation, such as depression, anxiety, problems of self-esteem and fluctuations in mood (348). By most survivors the emotional impact of the experience of cancer is perceived as considerable. Compared to a small control group of ex-patients with chronic non-life-threatening illness, cancer survivors are at a higher risk of developing emotional disturbance as a late side effect (68). A number of variables have been identified in differentiating between the well-adjusted and maladjusted survivors. Factors which seem related to optimal long-term psychosocial adjustment include a short treatment course, no relapse of the disease, an early knowledge of the diagnosis, a supportive and openly communicating family, the ability of effectively using denial and the onset of the disease at a young age (349). It is a remarkable finding that extreme youth at the onset of the disease is related to a good psychosocial adjustment. According to Koocher et al. (349) this can be partly attributed to the fact that two types of cancer which develop predominantly during infancy and early childhood, Wilms' tumor and neuroblastoma, have a good prognosis, if detected early. On the other hand, many writers have pointed out the importance of early life experiences to future psychosocial development (94,350). Therefore one would expect young children to be more vulnerable in developing adjustment problems. However, Rutter (166) emphasizes the resilience of young children and argues that substantial recovery after early adverse experiences is not only possible, but common. The findings of Koocher and O'Malley (68) and Fergusson (351) provide further corroborative evidence in this respect.

In conclusion, it is clearly demonstrated that survivors of childhood cancer are at a real risk of developing emotional problems but at the same time there are a considerable number of survivors who are in good psychological health, showing the resilience of children (68). Finally, it should be noted that the survivors studied had cancer at a time when survival was less probable than today. With advancing medical treatment it might be expected that children receiving treatment today will have a better psychosocial outcome because the odds are in their favor (3).

CONCLUDING REMARKS

The fact that the literature on the psychosocial consequences of childhood cancer has evolved from progress in the medical discipline has a profound impact on the study approach of the child with cancer and the family. The literature on the fatally ill child with cancer can be characterized as fixed on *loss*, with an emphasis on the anxiety of a threatened loss, separation and (anticipatory) mourning. The aspects studied are related to death and dying, and the preparation for the imminence of death of the patient and parents. Little attention has been paid to emotional aspects of *living* with the disease, e.g. the value of the period between diagnosis and death (235,352,353). In contrast, the literature on the child with cancer with an extended life-expectancy, the chronically ill, focused on *living* with cancer, and its main research themes are issues previously neglected in the literature about the fatally ill: coping with and adjusting to the illness aimed at maintaining as many aspects of normal day-to-day living as possible. Therefore the child who will die of cancer should be studied based on this 'cure-approach' because a child who will die of cancer will also *live* with illness. Accomplishing developmental tasks, having a rewarding family-life and planning a future, all within the context of death, are issues equally important for the child who will die of cancer as for the child who will survive. At the same time, research on the child who will survive cancer may take into account the approach and concepts of the literature about dying, because the literature has failed to take into account the loss and subsequent mourning processes involved in the illness, such as the loss of self-esteem and the loss of control over daily living. Furthermore, the disease period for the chronically ill has been described from the cure option resulting in both a disregard of the group of children that relapse and die of their disease and in an underrating of the life-threatening nature of the disease. In emphasizing living with uncertain survival as a reaction to the early literature that focused only on death, the fatal aspect of the disease has been blurred. As long as childhood cancer is not a completely curable disease, and it is unpredictable whether a child will die or will be cured, aspects directly related to the loss of the child will be present. As clearly pointed out in the literature, these aspects evolve from the changes in medical outlook. Therefore new knowledge is needed concerning the life-threatening aspects in modern cancer treatment, such as knowledge of the child's course of becoming aware of the life-threatening aspect of the disease and the parents' process of anticipatory mourning. Consequently, it will be useful to replicate former studies on the fatally ill in our present time. In conclusion, with changes in medical therapy two separate lines of psychosocial research have been developed. Different concepts and approaches to studying the child and family are used, focused either on living or on dying. However, despite the new issues that will evolve from improvements in medical therapy, the same fundamental psychosocial processes in response to a life-threatening illness are still active. Several factors, like situational and personal characteristics, will influence the extent and the form of such processes and their manifestation. Therefore it is advocated that both approaches be integrated in order to get a deeper understanding of the complexity of the responses of the child and family to childhood cancer (354).

Methodological and theoretical shortcomings, frequently jeopardizing existing investigative studies, need to be avoided in studying the responses to childhood cancer. Methodological shortcomings include a biased selection of children and families, an incomplete description of sample group and design, and inappropriate use of design and of research instruments. Some methodological problems are due to a lack of theory. Concepts are often operationalized differently and various measurement techniques are used to measure the same concept. Simultaneously, different concepts are used to describe the same kind of responses of the child and family. In order to integrate empirical data, theory is needed to provide guidance for systematic research (71). However, there is no overriding theory concerning the impact of childhood cancer on the child and family. Mostly, the child and family have been studied in order to determine in what way childhood cancer affected normal life. This led to an exploration of several important fields in the life of child and family, evaluating the nature and the extent to which a particular field is affected. As a result, research efforts have been fragmentary and a diversity of many intriguing problems are reported in the psychosocial literature about the child with cancer. Although many of these problems are interrelated, no clear structure has evolved. Consequently, it is suggested that the impact of childhood cancer be studied within a theoretical model meeting the requirements of: (a) describing the problems the individuals experience when faced with a life-threatening situation and how they cope with these problems, from fundamental psychological or psychosocial theories of human behavior; (b) studying the child and family as normal individuals under stress aiming at re-integration into every-day-life; and (c) incorporating individual and situational characteristics in explaining responses to cancer, in particular the psychosocial development of the child, disease-related variables and significant others in the life of child and family. Such a model will yield valid insight into the many psychosocial aspects related to childhood cancer.

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CHAPTER 1.3

INTRODUCTION TO THE STUDY

Three decades ago childhood cancer was a disease with a poor prognosis. Nowadays, childhood cancer is still regarded as a serious life-threatening disease but with a real chance of cure. As the period of survival extends, the long-term physical and psychosocial consequences of cancer and its treatment become increasingly important. It is emphasized that disease-free survival is not synonymous with cure (1). Cure implies that the child will be restored to its premorbid condition without any residual limitation in present or later adult functioning. The introduction of a new concept in pediatric oncology 'the truly cured child' (2) reflects the recognition that successful treatment must extend beyond biological cure to the quality of survival. Only a physical and psychosocial successful rehabilitation will result in a truly cured child (2,3). Therefore, after cessation of treatment the main concerns are (a) how the stresses during cancer and its treatment affect late psychosocial functioning and (b) which new problems evolve from the illness after cessation of treatment.

Childhood cancer affects the child and those directly around the child: parents and siblings, but also the family system as a whole. Just as childhood cancer is a family-disease, so is long-term survival or cure an issue that impinges upon the entire family system. On the other hand, the family has major influence on the quality of life of the child, because the child is highly dependent on the family. The family provides the framework for the major adjustments made by the child. Thus, the concept of the 'truly cured child' needs to include the child's family (4). With disease-free survival not only the psychosocial consequences for the patient need detailed investigation but also those for the family.

It has been estimated that 5 years from now, by the year 2000, 1 in 1000 young adults between the ages 20 to 29 has suffered from childhood cancer, given an average cure rate of 60% (5). Other calculations predict that by the turn of the century, given the incidence of childhood cancer and current survival rate, 1 out of every 900 individuals between 16 and 44 years of age will be a survivor of childhood cancer (6). The potential implications for society of having such large numbers of survivors in the population is evident and point to the increasing need of obtaining insight into the long-term consequences of childhood cancer.

Although information is accumulating on the long-term effects of childhood cancer, most existing research has focussed primarily on the medical sequelae of disease and treatment. Considerable less information is available on the psychosocial outcome for survivors. Since the publication of our review on the literature (Chapter 1.2.), few studies on the long-term sequelae of childhood cancer have been reported. Those published have yielded inconsistent results. Surprisingly, the lack of information on parents and siblings of children surviving cancer remains to date (see reference 7). This underlines the need to get insight into their current psychosocial functioning.

Therefore the aim of this thesis was to get more insight into the unique problems and needs of children surviving cancer and their family. The research objectives were:

1. to describe in a systematic way the psychosocial functioning of children surviving cancer *and* their families

2. to explore the relationship between psychosocial functioning and specific variables related to disease, child, and family characteristics in order to identify children and families 'at risk'

3. to formulate recommendations concerning care for the survivor and family

Chapter 2 summarizes the methods of the study. The study approach, sample, and procedure are delineated. To investigate the psychosocial functioning of the child surviving cancer *and* his family, a variety of methods is used including in-depth interviews, psychological tests, questionnaires, and observations. We developed an instrument to measure the specific problems of parents after cessation of their child's treatment from a theoretical framework. This conceptual framework of coping with childhood cancer is described as well as the instrument evolving from the model. For patients and siblings, we adapted an existing instrument to measure self-concept and examined its psychometric properties before it was used in our study.

Chapter 3 describes the impact of childhood cancer on the parents. To obtain insight into this new field, we applied a qualitative study method to obtain insight what it is like to live through childhood cancer. A wide range of variations is described how parents perceive the impact of childhood cancer on their life and how they cope with it. The themes emerging from the analyses are meant to refer back to the life-world of the parents and provide a rich and differentiated picture of parental experiences and coping strategies. Also information on parental problems using standardized measures are presented. We investigated which problems parents reported on the instrument designed to measure illness-specific problems and whether these problems were related to the lapse of time since cessation of treatment. The relation between reported problems and demographic, personality, disease-related, and family variables was investigated to identify those parents who are most 'at risk' of experiencing late problems. Finally, one specific and new theme that evolved from the study, namely family planning, was investigated in more detail, using a combination of qualitative and quantitative methods.

Chapter 4 describes the late consequences of childhood cancer on the children in the family: the former patient and the sibling. To gain insight into the late effects for the survivor, the impact of the cancer experience on academic and psychosocial functioning was investigated. Firstly, we described the prevalence of learning problems in children with cancer differing in diagnosis and/or treatment regimen. Secondly, we studied the nature and severity of late consequences of childhood cancer by comparing the psychosocial functioning of children surviving cancer with that of healthy peers. Reports by parents and children were used to describe the child's psychosocial functioning. To explore why some children had more problems than others, the relation between risk factors and psychosocial problems was investigated. In this way insight was obtained into those factors which contribute to psychosocial adjustment. In the last part of this chapter, the siblings' psychosocial functioning was investigated. In this study the impact of the cancer experience on the sibling's psychosocial functioning was assessed, using parents and siblings as informants.

Chapter 5 discusses the significance of the presented data and the implications for patient care. By integrating the results of our extensive study, insight is obtained into the late consequences of childhood cancer for patients, parents, and siblings, and the factors that contribute to it. Proposals are made for application of the results in its clinical context and for further research studies.

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Chapter

2

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CHAPTER 2.1

DESIGN OF THE STUDY

INTRODUCTION

Studying the psychosocial consequences of childhood cancer is complex due to a number of methodological issues. These are not unique for children with cancer but pertain to children with chronic disease in general. In several articles (including Chapter 1.2), these methodological problems in studies on children with cancer and their families have been outlined (1-5). The most frequent methodological criticisms concern sample (including sample size, subject selection, and compliance), type of informants, and study method (including choice of instruments and comparison groups). These concerns have guided our study design and I will describe below how we approached these problems.

Sample: sample size, subject selection, and compliance

One of the problems in pediatric oncology concerns the size of the sample. Because childhood cancer is a relatively rare disease the number of children is limited. Apart from the availability of subjects, other factors such as subject selection, compliance and study method affect *sample size* as well. These will be successively discussed.

With respect to *subject selection*, sample size will be reduced when more selection criteria are included in the study. Homogeneity of subjects and sample size are inversely related. This is illustrated by the sample size of studies in the different fields of psychosocial aspects of childhood cancer. Research on the adverse sequelae of treatment regimens on cognitive functioning in ALL patients requires a specific diagnosis and treatment regimen as selection criterion. In a review on these studies (2), the median number of irradiated patients with ALL was 20. Samples of long-term survivors, where the only selection criterion was having terminated treatment (reviewed by 1), have a median size of 42 subjects.

A strategy to increase sample size is to have liberal inclusion criteria, for instance, including children both on and off treatment (see for example 6). In studies on long-term survivors the sample is regularly expanded by including children across a wide age-range (1,7). For instance, in the Koocher and O'Malley study (8) the age of the subjects ranged from 5 to 32 years, whereas in another study on the late effects of brain tumors, the age of the subjects varied even more: from 8 to 51 years (9). Mixing young children with adults in their thirties to fifties creates a significant problem, because the implications of cancer for the child varies along its developmental level. Problems also arise when different instruments have to be applied for different age groups. This complicates the comparability of the results within the sample. By studying late consequences of cancer in discrete age groups, insight is obtained how cancer affects the developmental processes within a particular stage. This developmental approach is not only most appropriate to study the impact for the child, but also for its parents. Parents may experience different problems and worries related to the child's developmental stage. With a limited age range, responses can be compared within the sample, because age-related issues are the same. In this way, insight is obtained into which factors facilitate or hamper psychosocial functioning in a particular developmental stage. Although it has been advocated in the literature to investigate the responses to childhood cancer at a specific developmental level (4-6,10), there are few studies that focus on a particular developmental stage. Therefore, we focussed on patients in the age range from 8 to 12 years old. This age range was chosen, because most research has focussed on adolescents and young adults. Probably these subjects are better able to verbalize their experiences and more instruments are available for this age group. Consequently, less is known about how childhood cancer affects developmental processes in middle childhood.

Another factor which affects the size of the sample is the rate of *compliance* of eligible subjects. The importance of an unbiased sample is often underemphasized, although it is a vital aspect. Representativeness affects directly the validity of the conclusions. In addition, it is important to get insight into the reason for refusal to have an indication whether results are underrepresented or overrepresented. Our experiences with the present study indicate that families who have difficulties in coping with the disease are less inclined to comply. Consequently, most studies will underreport problems, even if the compliance rate is as high as 90% or 95%.

As indicated, the need for an unbiased sample is crucial for the validity of the conclusions. Therefore we carefully designed a procedure which maximizes the possibility for participation *and* which gave insight into the reason of refusal. An individual approach was used to adjust the procedure to the subject's preferences and to overcome a number of practical obstacles for the participants. Furthermore, open, free-attitude interviews as study method might have increased the motivation to participate, because of its personal approach to the subjects' experiences. This approach resulted in a high compliance rate (see section on procedure). Our sample included 95 survivors, which represents the largest sample to date with such a restricted age range. Recruiting and motivating subjects to maximize compliance is a very time and energy consuming effort, which is frequently underestimated in planning an investigation. Time pressure to complete a project usually results in rushing over this phase. However, ensuing analysis on a less optimal data base will not improve the outcome of the study. Therefore, efforts for data collection and data analysis should be in balance.

Informants

Substantial criticism has been directed against the way data on children with chronic illness and their siblings are obtained (3,4,11). Often data on the children, especially the siblings, are obtained from parents instead of examining the children themselves. With respect to parental experiences, often only mothers are included in the study. Because this study is on the late-effects of childhood cancer on the family, data were obtained from *all* family members (fathers, mothers, survivors, and siblings).

In child assessment the risk of obtaining an incomplete picture on psychosocial functioning increases, when one source of information is used. It has been shown that data from different sources reveal different information. Each informant makes a unique contribution to psychosocial functioning, which cannot be provided by another source (12-15). Low correlations between reports of informants should be regarded as a reflection of the different perspectives of each informant rather than an indication that the informant's reports are invalid or unreliable (12). In this study parent and child (sibling) reports were used to give a more comprehensive picture of the psychosocial functioning of patients and siblings.

Finally, to evaluate the patient's and sibling's adjustment to cancer, also raters outside the family were used. Parents' own functioning and strategies of coping may influence their perception of the children (16,17). Because assessment of the child's functioning is also dependent on the method used (14,16), the non-familial informant used a different method to assess psychosocial adjustment than parent and children. Two expert judges were employed to rate independently the subjects' level of adaptation from the interview by a standardized rating scale. Both judges were psychologists and underwent extensive training before analyzing the interviews. The interviews of the parents were assessed in a similar way.

Study methods: what to measure and how to measure it

As delineated in Chapter 1.2, the first studies on children with cancer examined whether children with cancer differed from healthy children. For this purpose, often instruments were used designed to discriminate emotionally healthy from disturbed children. Children with cancer were compared on these instruments to a comparison group or normative data. This 'general' study approach was questioned, because its design implicitly communicated the message that a more negative outcome was expected, thereby reinforcing a deficit orientation on the child with cancer (18,19). It has been stressed that patients and their families can be generally viewed as normal individuals confronted with an extreme stressful situation (18-21; Chapter 1.2). Several studies demonstrated that the psychological functioning of pediatric patients is one of psychological normalcy rather than deviance and that psychopathology is not a common response to cancer (4,18,20).

Most *general instruments* measure broad areas of psychosocial functioning and are not designed to measure the specific problems associated with childhood cancer. In studying the psychosocial consequences of childhood cancer, first an understanding is needed what happens to the child with cancer and its family. A systematic and detailed description of the field provides knowledge on which variables have to be assessed and in which combinations. Without such description there is a risk of overinterpretation of findings or investigating irrelevant questions to the problem under study. This is the source of most validity errors in clinical research (22) and refers directly to the ecological validity (23). Therefore, there is a need to describe the nature of responses associated with surviving cancer to provide insight into what type of responses are present and which are not.

Interviews are well suited to identify the nature of problems specific to the cancer experience, because they allow a broad spectrum of issues to emerge. However, other types of instruments are preferred, because they are able to measure the illness-related issues in a more standardized way. Unfortunately, there is a scarcity of such *illness-specific instruments*. This is a major problem for researchers is pediatric psychology (24), and therefore available illness-specific instruments have great appeal. One should be aware that there are also pitfalls in using these measures, because illness-specific instruments do not automatically guarantee scientifically valid conclusions. A prerequisite is that the psychometric properties have to be acceptable. Because of their instant appeal, however, psychometric research to these illness-specific instruments is often neglected (25).

TABLE 1. Possibilities and limitations of general and illness-specific study approaches to children with physical illness and their families.

Characteristics	General study approach	Illness-specific study approach
Ecological validity	limited	high
Availability of instruments	numerous	few
Psychometric properties	well investigated	poorly investigated
Type of study design	between-group design	within-group design
Standardization	well standardized	less standardized

An issue related to study approach is the use of a *comparison group*. In a general approach, a comparison group provides a criterion to which the results can be evaluated. There has been much debate concerning the use of a comparison group in pediatric oncology (21,26). A between-group design which use a comparison group may not be meaningful, because healthy children do not have equivalent experience with the critical aspects of childhood cancer (21,27,28). For instance, comparing uncertainty about survival chances between cancer survivors and healthy children is not relevant. Sometimes children with other chronic illnesses are used, however, they do not provide a comparison for all aspects of the disease. Therefore, within-group comparisons are used in which the cancer population functions as its own control. With this approach, factors can be identified which are related to well and less well adaptation (19,21). At the same time, to interpret the results some measure of comparison is needed. Comparison or control data often provide such standard. Consequently, the determination of an appropriate comparison group in pediatric psychology remains methodologically unresolved (28).

Whether a general of illness-specific study approach has to be chosen is dependent on the research question being asked. Both approaches have their advantages and limitations. They are in fact complementary to each other (Table 1). Consequently, both approaches have their value for studying psychosocial issues in pediatric oncology. The advantages of a general approach is that many instruments are available, which are administered in a standardized way. They can provide reliable and valid data, that can be compared with a normative sample or healthy children. In this way relevant data on the psychosocial functioning and behavioral manifestation of stress are obtained. Studies using this approach are important in identifying the childhood cancer survivors' position on a continuum of mental health (21). As outlined above, the major limitation of these instruments is their limited ecological validity.

In our study, we combined both study approaches, using general assessment techniques and illness-specific measures, to meet the different research needs in our study. As indicated, instruments measuring the illness-specific problems are scarce. Although some have been developed for childhood cancer patients (29-31), specific instruments for their parents are not available. Therefore, we developed such an instrument for parents from a conceptual framework of the impact of a stressor developed by Van den Borne and Pruyn (32). In Chapter 2.2 it is demonstrated that this conceptual framework is also applicable to childhood cancer and that the many psychosocial problems experienced by parents can be assigned to the problem categories of the model. The model outlines the domains which are negatively affected by childhood cancer and provides guidance in operationalizing the relevant concepts needed to assess late sequelae. In Chapter 2.3 we describe how we adapted the concepts of the model to assess the specific psychosocial problems of parents of childhood cancer survivors and present the items of the questionnaire. As indicated, a potential weakness of illness-specific instruments is that often their psychometric properties are not clear. Therefore, we performed extensive psychometric analyses, demonstrating that the instrument measured the specific problems of parents of children surviving cancer in a reliable way (see Chapter 2.3).

Because there is an emphasis on studying the child with cancer from a non-deficit approach, there is a need for instruments that can detect mild adjustment problems or differences in behavioral symptoms within the normal range (20). The Self-Perception Profile for Children (SPPC) (33) is such an instrument. The SPPC measures self-concept. Self-concept is a central concept in theories on child development, which relates to adaptation, competence, and mental health. However, before we could use this instrument, it had to be adapted for and tested in Dutch children. In Chapter 2.3 we report satisfactory indices of reliability and validity demonstrating that the instrument holds cross-cultural generalizability.

So far, I have outlined the study approach of the investigation. In the following sections the materials and methods of the study will described: subjects, procedure, instruments, and data analysis, in that order.

SUBJECTS

Children who had been diagnosed with a malignancy and meeting the following criteria were included in the study: (a) successful termination of cancer treatment and in symptom free-remission, (b) 8 to 12 years old (grades 3-6), and (c) Dutch as the native language. All children were treated in the Division Hematology/Oncology, Department of Pediatrics of the Sophia Children's Hospital/University Hospital Rotterdam, one of the four pediatric cancer centers in the Netherlands.

To get a more homogenous sample with respect to disease-related characteristics, all eligible children who had been treated for leukemia and lymphoma were approached. Leukemia is the most common type of cancer in childhood and belongs together with lymphoma to the group of hematological malignancies (see Chapter 1.1). Both types of cancer resemble each other concerning prognosis and treatment. In addition, we included children with a solid tumor, who resembled children with hematological malignancies with respect to prognosis, treatment protocol, difficulty of treatment, length of treatment, and type of late sequelae.

Both parents of the survivor were approached to participate in the study. In case of divorce, only the parent living with the child was enrolled in the study. Also a sibling of the survivor was approached. To exclude overrepresentation of family or patient variables related to adjustment, only one sibling per family was selected. This concerned the sibling closest in age to the former patient.

PROCEDURE

Families of the survivors were approached by the pediatric oncologist during follow-up appointments in the outpatient oncology clinic. The oncologist introduced the study to the family and invited the family to have an initial meeting with the investigator. If parents agreed, the investigator was introduced by the oncologist. Then the investigator explained the purpose of the study and its procedure to the family. This information was also made available in printed form. It was stressed that the family had not to reach for an immediate decision. Instead, permission was asked to contact the family by telephone a week later to inquire whether they would be willing to participate. By giving some time to parents to make up their minds and to provide the possibility to convey their decision by telephone, they had the opportunity to distance themselves from the request. In this way, parents would feel less obliged to participate. No insistence was made that all family members had to participate. If members of a family did not want to participate, their wish was respected. If family member(s) agreed to participate, informed consent forms were mailed and an appointment was scheduled.

Each family was offered to be visited at home. Except for two children, who preferred to come to the hospital, each family member was visited at home. We presumed that participants would feel more comfortable at home. The first appointment was made with the parents. In this way parents got acquainted to the way in which their experiences were investigated and were able to make a well-considered decision whether they let their children participate. During this first visit, parents were interviewed in an in-depth way about the impact of the disease on their personal life, giving freedom to discuss their experiences and areas of importance in their own way. All interviews were audiotaped. Duration of the interview ranged from 2.5-4.5 hours. After the interview, parents received questionnaires and were requested to mail the completed forms to the investigator. If incomplete or ambiguous data sets were obtained, the investigator contacted parents. Some parents preferred to give information by questionnaires only, others indicated that the interview caused emotional upheaval and they decided not to return the questionnaires. No insistence was made on giving information by both interview and questionnaire. These parents were not excluded from the study in order to reduce bias and because they provided valuable information obtained by one method. At the same time insight was obtained into the reason of refusal and into the 'natural history' of how family members cope with the request for information about their experiences.

If patients and siblings were willing to participate, a second appointment was scheduled. Survivors and siblings underwent exactly the same procedure: first they were interviewed about their experiences with the illness followed by completing psychological tests and questionnaires. Survivors and siblings were separately examined by the same investigator. The interviews of the children were audiotaped. Duration of the interviews and tests varied from 1.0 to 2.5 hrs.

To all parents a follow-up appointment was offered. Sometimes, the investigation had caused emotional disturbance and follow-up appointment(s) were needed. In addition, a number of parents wanted to discuss the impact of the illness on the children. During these meetings the experiences of all family members, including the impact of participating in our study, were discussed. A follow-up appointment was made with 35 of the 85 families.

An interview with all family members had a positive effect on both the motivation to participate and the accuracy with which the questionnaires and tests were completed. A substantial number of parents expressed gratitude for having the opportunity to share their feelings and concerns and to put their experience in perspective after the passage of time. Parents especially appreciated having been interviewed in their homes and at a time convenient for them, for instance after-work hours. In this way, problems concerning job responsibilities, baby-sitters, transportation, and their related financial expenses were overcome. The here described individual and personal approach of subjects made a substantial contribution to the low rate of drop-out. Of the 87 families approached for the study, including the pilot study, only two families refused to participate, which resulted in a compliance rate of 98%. Both non-complying families indicated that they feared a set-back in their emotional equilibrium as a result from the study. In addition, in subsamples of the investigation a high compliance rate was obtained: for parents: 98% (n=162; Chapter 3.1); 98% (n=133; Chapter 3.2), 96% (n=130; Chapter 3.3), for survivors: 96% (n=100; Chapter 4.1), 96% (n=95; Chapter 4.2), and siblings: 95% (n=60; Chapter 4.3). These high compliance rates indicate that our samples can be considered as representative.

INSTRUMENTS

In-depth interviews, questionnaires, tests, and observations were used to describe the psychosocial functioning of parents, patients, and siblings. Details of the instruments are described below.

Psychosocial functioning of parents

Interview

With parents open-ended, free-attitude interviews were conducted focussing on their experience with their child's illness from the first physical symptoms leading to the diagnose of cancer until the moment of the interview. The topics explored during the interview were: positive and negative changes in personal, marital and family functioning, and their ways of coping with these changes, sources of support, changes in the ill child or its siblings. In particular, parents were questioned about their current attitudes towards the illness, their current and future concerns for the survivor and the sibling(s) in the family, and to which extent current family functioning was affected by the cancer experience. The interviewer asked the question in an open-ended manner, allowing parents to talk at length about feelings and concerns they considered relevant (see Chapter 3.1).

Questionnaire for parents

This questionnaire was developed to measure the specific experiences with childhood cancer. The questionnaire consists of a number of questions related to survivor, sibling, and the parent's own functioning, including questions concerning parental problems (see Chapter 2.3 and 3.2).

Observation

For scoring general adaptive functioning of the parents, the Global Assessment Scale (GAS) was used. The GAS is a comprehensive rating of general functioning designed by Endicott et al. (34). Scores on the GAS can range from 0 to 100 giving a summary statement of psychosocial functioning along a continuum from the most severely mentally ill to the hypothetically healthiest individual. The GAS allows the rater to assimilated and synthesize his or her knowledge about many different aspects of the subject's social and psychosocial functioning and condense it into a single clinically meaningful index of severity of disturbance (see Chapter 5). Two expert-judges (psychologists) listened independently to the audiotaped interviews. On the basis of the interview and written observations by the interviewer, the judges gave independently a GAS rating. The Global Assessment Scale for interview-based ratings has demonstrated good interrater reliability in clinical settings, with intra-class correlation coefficients (ICC) ranging from .76 to .92 (34-36). For record based ratings ICC of .62 was found (37). Concurrent validity (34-36,38,39) and predictive validity (34-36) were supported.

It is not clear whether the psychometric properties of the GAS generalize to non-patient samples, as our parent sample, where defining variations in normal functioning is more important than rating signs of psychopathology. Consequently, we computed reliability and validity coefficients of the GAS for our sample. The ICC's between two raters, respectively .99 for mothers and .93 for fathers, were rather high in comparison with other studies. Correlations of the GAS scores with four scales measuring psychosocial functioning (see Chapter 2.3) ranged from -.12 to -.38, showing the same levels of concurrent validity generally found in relation to specific measures of mental health (34,35,38).

Empirical data on the ability of the GAS to discriminate between disturbed and normal subjects are sparse. Endicott et al. (34) stated that the majority of individuals in psychiatric treatment was expected to be rated below 71. Ferdinand et al. (40) showed that for the strongly to the GAS related Global Assessment of Functioning Scale ($r=.96$; Ferdinand and Verhulst, unpublished data), the proportion of subjects classified as moderate to marked impaired was higher for a cut-off point of 61 than for a cut-off point of 71. We applied both cut-off points to divide parents in three categories: parents showing good mental health (GAS ≥ 71); parents with mild adjustment problems (GAS 61-70), and parents with moderate to marked adjustment problems (GAS ≤ 60).

Psychosocial functioning of patient and sibling

Interview

Open-ended, free-attitude interviews with children were conducted covering their experience during the illness until the moment of interview, their knowledge of the disease including diagnosis and prognosis, coping with the disease and sources of support, positive and negative changes in their lives and in parent-child interaction, current concerns about health and illness, and the perceived impact on their present way of life.

Questionnaires and tests

. Child Behavior Checklist (CBCL) (41)
The CBCL is a standardized instrument to obtain parents' report on their child's behavioral and emotional problems. The CBCL has been translated into Dutch and its good reliability and discriminative validity were replicated in Dutch studies (15,42-43). Procedure and scoring is described in detail in Chapter 4.1.

. Self-Perception Profile for Children (SPPC) (33).
This instrument is considered as the most promising instrument measuring self-concept in children during middle childhood (44). It examines the perceived competence in various domains important for children during this developmental stage. However, because no validated Dutch version was available, we had to investigate the psychometric properties of the SPPC before we could use it in our sample (45). For detailed information see Chapter 2.4.

. Amsterdam Biographic Questionnaire for Children (ABV-K) (46).
This instrument is a forced choice (yes/no) personality questionnaire measuring psychoneurotic complaints, somatization of psychoneurotic complaints, extroversion-introversion, and social desirability. For details see Chapter 3.1.

. Children's Depression Scale (CDS) (47).
This instruments measures depression by a Q-sort technique. Children sort cards with descriptive statements on 5 piles ranging from very true to very untrue. For detailed description see Chapter 4.1.

. Projective measure.
Children were asked to draw or to write about their most significant or ingrained experience with the illness. On the cover of this thesis as well at the beginning of each chapter, some of these drawings are shown.

Observation

For scoring the child's general adaptive functioning, the Children's Global Assessment Scale (CGAS) was used. The CGAS is a scale of adaptive functioning designed by Shaffer et al. (48) based on the Global Assessment Scale for adults. As with the GAS, the scoring format ranges from 1 representing the most functionally impaired child, to 100, representing the healthiest. The CGAS is divided into 10 equal intervals and each decile on the scale is accompanied by descriptions and examples of behaviors and functional levels typical of that decile. The highest interval 91-100 indicates good functioning while lower scores indicate problems in functioning. Scores between 61-70 can be used to indicate some difficulty in functioning or mild problems, while the interval between 51 and 60 represents moderate impairment. The CGAS score was given by two expert-judges (psychologists), who listened independently to the audiotaped interviews. On the basis of the interview and written observations by the interviewer, the judges independently gave a CGAS rating (see Chapter 5).

The CGAS has shown good interrater reliability (49-51). In our sample, ICC's of .86 and .87 were found for patient and siblings, respectively. In addition, good discriminant and concurrent validity have been reported (49-51). These psychometric properties indicate that it is an instrument of considerable strength in providing a measure of impairment (49-51). For the CGAS the cut-off points of 61 and 71 were empirically supported (50,51). Therefore, we divided patients and siblings into three groups: children without adjustment problems (CGAS > 70); children with mild adjustment problems (CGAS 61-70), and children with moderate to marked adjustment problems (CGAS ≤ 60).

QUALITATIVE AND QUANTITATIVE DATA ANALYSIS

For analysis of the data a qualitative and quantitative approach was used depending on our research questions. Quantitative studies can give insight into intensities of variables associated with childhood cancer. The qualitative method gives insight into what it is like to live through childhood cancer in all its aspects. Each approach has its own method of data collection and data analysis (Table 2). In the quantitative approach, *a priori* hypotheses are formed from pre-existing theory. The variables and interrelations between variables to be investigated are chosen by the investigator and are tested under controlled conditions. Subjects are assigned to groups in such a way as to minimize the potential impact of intervening variables (22,52). The observations are usually translated into figures and answers are provided by *statistical analysis*.

In the qualitative approach, analyzing and hypothesizing starts during and after data collection. Data collection and analysis are intertwined in a process of generating

TABLE 2: Differences between qualitative and quantitative study approach.[1]

	Qualitative study approach	Quantitative study approach
Research design	-generation of hypotheses and theory -emphasis on understanding behavior from the subject's own frame of reference -data collection and analysis are intertwined -use of investigator as research instrument -emphasis on free interplay of variables as they occur in natural setting -notation of data is narrative -textual analysis	-testing of hypotheses and theory -emphasis on verification of causal relationships -data collection and analysis are separated -use of standardized instruments -control of intervening variables -notation of data is numerical -statistical analysis
Validity	Whether the material reflects a faithful representation of the phenomena under study	Whether the measurement instrument is able to represent the phenomena under study
Methods to establish validity	-validity of material: high involvement of subjects, freshness of material; new exploration during interview. -validity of analysis: search for rival explanations; critical appraisal by another neutral investigator -validity of interpretation: positive exemplification	-predictive validity -content validity -construct validity
Reliability	The extent to which the material is accurate	The extent to which measurement instrument is accurate
Methods to establish reliability	-accurate description and recording of material and analyzing process -articulating the context of investigation and investigator -no conflicting findings when study is replicated -involving other investigators in the research process	-test-retest reliability -interrater reliability -internal consistency
Representativeness	Emphasis on the representativeness of concepts in their varying forms	Emphasis on selecting subjects which are representative for the entire population to which one wants to generalize
Methods to establish representativeness	Sampling techniques concern material: -variational sampling: to maximize findings of differences at the dimensional level	Sampling techniques concern subjects: -a-select sampling, -sample large enough -non-biased sample

[1] Based on references 22,52-59.

categories, comparing concepts, and constructing theories as suggested by the data. In this way it is tried to understand phenomena from the subject's own frame of reference (22,52-55). In the qualitative approach, language is used as the main mode of obtaining and processing data. As such the investigator him- or herself has a crucial role in the process (55). Thus, answers are provided by *textual analysis*. The scientific value of quantitative and qualitative studies are judged according to the same criteria with respect to internal validity (including aspects of reliability) and external validity (including representativeness). These concepts have similar meaning for each method, but their application and methods to accomplish them differ (Table 2).

The qualitative analysis of the interview data is described in Chapter 3.1, whereas in the other chapters a variety of statistical techniques and methods are used to analyze questionnaires, psychological tests, and observations.

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CHAPTER 2.2

COPING WITH CHILDHOOD CANCER: A CONCEPTUAL VIEW*

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ABSTRACT

This article presents a conceptual framework for studying the impact of childhood cancer on the psychological and social functioning of the patient and the family. A model based on the literature about coping with stress, especially with respect to cancer, and on two psychosocial theories of human behavior (the attribution theory and the social comparison theory) is used as a starting point. The model (a) describes the problems people experience when confronted with a threatening situation, (b) provides a classification of problems that has a manageable number of categories to which problems can be assigned, and (c) gives insight into some important coping strategies that children and their parents use to lessen the impact of those problems. The problem categories and coping strategies of the model are illustrated mainly from findings described in the literature and from information the authors obtained during interviews with patients and parents.

INTRODUCTION

During the past few years, a considerable body of literature on the psychosocial aspects of childhood cancer has accumulated. From an extensive review of this literature (1), the following conclusions can be drawn: first, research on the serious problems that the child and family experience when faced with cancer has rarely been conducted within a theoretical framework. As a result, a clear outline of the interrelations between the problems experienced and ways of coping with them has not evolved. Second, the studies that have been done have several methodological shortcomings which can be attributed, in part, to the lack of a theoretical framework.

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This article presents a conceptual framework for studying the impact of childhood cancer on the psychological and social functioning of the child and the family. The model developed by Pruyn and associates is used as a starting point (2-7). This model is based on the literature about coping with stress, especially with respect to cancer, and on two psychosocial theories of human behavior: namely, the attribution theory (8,9) and the social comparison theory (10,11). It describes the problems people experience when confronted with a threatening situation and the coping strategies they use to lessen the impact of those problems. It also provides a classification of problems that has a manageable number of categories to which problems can be assigned. We will show that the many problems experienced by children with cancer and their families can be assigned to those categories. The model also gives insight into some important coping strategies that children and their parents use to cope with the impact of childhood cancer. The problem categories and coping strategies of the model will be illustrated mainly from findings described in the literature and from information obtained during our interviews with patients and parents.

PROBLEMS ASSOCIATED WITH CHILDHOOD CANCER

A stimulus that is a threat to a person is called a stressor. Every stimulus is a potential stressor, but whether an individual perceives a stimulus as a threat depends on personal and situational characteristics (12). Some important personal variables in a child are age and levels of cognitive and psychosocial development. For example, a loss of hair as a side effect of chemotherapy will be disturbing to an adolescent girl, whereas it is likely to be only a minor problem for a toddler (13). Some important situational characteristics for a child are the degree of support provided by the family and the health care team and the variables related to the disease and its treatment. As a consequence of differences in personal and situational characteristics, the extent to which children will perceive a stimulus as a stressor will vary.

Obviously, cancer presents a complex of potential stressors, including the life-threatening nature of the disease, the side effects of medical treatment, the number of hospitalizations, the tone of the physician's voice, and the reactions of family members, friends, and others. According to the model, a stimulus is perceived as a stressor when it causes (a) uncertainty, (b) loss of control, (c) threat to self-esteem, and (d) negative feelings. These four aspects of stress can vary in intensity, and they can occur simultaneously.

Uncertainty

Uncertainty is defined as a lack of information about a value system that is important to a person. This can result in an inability to predict forthcoming events. Uncertainty is perhaps the most striking feature of childhood cancer. Some decades ago, when childhood cancer was almost invariably fatal, uncertainty centered on when the terminal phase would occur rather than on whether it would occur (14). As a result of progress in medical treatment, childhood cancer has become a disease with an increased chance of disease-free survival and cure. Consequently, the course of the disease has become extremely difficult to predict. This increases the uncertainty of parents, who express this feeling in remarks such as "The real hell of this illness is that you just don't know" (14).

The psychological dilemma of children with cancer and their families has been transformed from one of adapting to the imminence of death to one of coping with uncertain survival (15). Because individuals in this situation do not know whether to prepare for death or for life, living with the disease, by definition, means living with uncertainty (16-18). In addition to uncertainty over the outcome, there are other questions related to the disease and its treatment. For example, patients may ask questions such as "What illness do I have?" "Will I lose my hair?" "When can I leave the hospital?"

Apart from questions related to disease and treatment, uncertainty can be experienced about oneself and one's feelings. Patients may wonder what the deeper source of their anxiety is (19) or whether they will be able to get their degree at school. Parents may ask themselves whether their depression is normal and whether other parents react to the disease in the same way.

Young cancer patients and their parents also can experience uncertainty about interpersonal relationships. Children often worry about how their peers will react to them (20). For example:''How will my classmates react to my baldness?" ''Will my boyfriend still want to see me?" "Do the other students treat me this way because they like me or because they pity me?" Parents may be uncertain about how much to tell the patient about the illness, how to discipline the patient, or how to give the patient's siblings the attention they need, or they may wonder where to turn for help (21-26).

Loss of control

Loss of control is defined as the inability to manage or influence events. Although loss of control is closely related to uncertainty, it is a distinct aspect of stress. Control is a central concept in psychological theories of child development and is a necessary element in maintaining mental health (27).

Almost all aspects of childhood cancer and its treatment have a major impact on control (28). For example, because the disease requires constant medical care and attention, patients are confronted with increased dependence on others, loss of privacy, submission to hospital rules, restrictions on their way of life, and overprotection by parents. Furthermore, they must permit invasive treatment procedures and accept the side effects.

For children, control of the body has special significance because the body is an essential tool for accomplishing developmental tasks and therefore mastering the environment, individuating from parents, and forming one's own identity (28). The child with cancer, however, faces loss of this control.

Loss of control also can be experienced in relation to emotions. One patient said that he had frequent outbursts of aggression toward his parents and felt sad and ashamed afterward because he was unable to control these feelings. Losing control of one's emotions may be facilitated or induced by drugs used to treat childhood cancer (29). For example, parents of children treated with prednisone often mention that all the child's emotions are amplified to the extreme. Other problems that can be experienced as a loss of control include changes in therapeutic schedules, changes in everyday life, difficulties in planning for the future (27), loss of financial resources (30), and loss of friends.

Threat to self-esteem

Self-esteem is related to the image an individual has about his or her body, psychological state, and social functioning. Children with cancer face a loss of self-esteem because they are in a new role of being ill or different, which means that their relationships with other people and their perceptions of themselves will change (31). The importance of this problem is underlined by the fact that many authors view the maintenance of self-esteem as a primary task for individuals faced with a threatening situation such as childhood cancer (32-35).

Because cancer and its treatment often alter the patient's physical appearance and cause pain, weakness, and sickness, the child's body image can change continually. A number of authors have expressed concern about the potential loss of self-esteem induced in children by cancer treatments (36-40). Actual changes in physical appearance are constant reminders that the patient is different from other children (34). Adolescent patients worry about their physical appearance and compare themselves with peers (16,41,42). Children who lack self-confidence because of their physical appearance may withdraw from their peers and be afraid to go to school, which will further affect their emotional and social adjustment (20).

Self-esteem also is related to one's performance and capabilities. A child's sense of integrity is closely related to mastering and accomplishing developmental tasks. When a child's academic career is jeopardized by frequent absences from school (43,44) or by a decline in IQ score as a side effect of treatment (45), his or her self-esteem may be affected. The ability to perform a specific job or role also can diminish a child's self-esteem: for example, an adolescent who had a testicular tumor refused local radiotherapy because he felt that his life would be worthless if he could not be a natural father.

Finally, self-esteem is related to the quality of interpersonal relations. Geist (46) noted that children with cancer experience a loss of contributing to family life, and this affects their self-esteem. Their feeling of being an emotional and financial burden to their parents also may lower their sense of competence (34).

Negative feelings

Negative feelings such as isolation, anger, guilt, shame, loneliness, apathy, bitterness, and confusion are often observed in patients and parents (see, for example, 38,47-49). Moreover, the loss of health or ability to plan for the future may initiate the process of mourning (50). Not surprisingly, the symptoms of depression have frequently been observed in pediatric cancer patients and their parents (41,51-56) and in parents (57,58). Maguire (24) found that 28 percent of mothers of leukemic children were anxious and depressed to a morbid degree.

In both patients and parents, negative feelings such as anxiety and fear are often related to recurrence of the disease and its potentially fatal outcome. Children, even young ones, can experience the fear of death (see, for example, 59-63). Also, children can experience fear and anxiety about the recurrent medical procedures and the side effects of treatment (41,64,65).

COPING WITH CHILDHOOD CANCER

When individuals are faced with problems, they are motivated to use coping strategies to lessen or eliminate those problems. The most important coping strategies are discussed here: seeking information, seeking support and comfort, attributing events to causes, attempting to change the situation, using denial and avoidance, and accepting the situation. These strategies are used to (a) reduce uncertainty, (b) gain and maintain some control of the situation, (c) maintain and protect self-esteem, and (d) reduce negative feelings. Some of the strategies have been deduced directly from the theories on which the model is based; others originate primarily from the literature on coping with stress.

Seeking information

Seeking information is a strategy that is used primarily to reduce uncertainty and negative feelings such as anxiety and fear. When people feel uncertain about an illness, i.e. when they feel they lack information about it, they prefer to consult formal sources such as the hospital medical staff or social worker, their family physician, books, or television programs. However, when these sources do not or cannot provide the desired information, they will seek information from informal sources. One important example is obtaining information by comparing one's own situation to that of others who are in a comparable situation (10): e.g., when a physician is unable to give information about a child's prognosis, parents often try to obtain it from the case histories of other cancer victims (14). Especially, the impact of questions about oneself and one's relatives can be reduced by the process of social comparison. As one parent said: "When I was depressed, I would have liked to know if other parents went through the same process and how they coped with it."

Children also seek information when they feel uncertain. For example, if they are shielded from the diagnosis by formal sources and do not obtain information they require to reduce uncertainty about their condition, they will turn to informal sources: e.g., they will notice changes in emotional climate, hospital routine, or treatment schedule or compare themselves with fellow sufferers (see, for example, 59). Several studies have documented that accurate information is a powerful method of reducing uncertainty and depression (see, for example, 52,66,67). As several authors have said, fear of the unknown is worse than fear of the known (25,59,68)

Seeking support and comfort

The strategy of seeking support and comfort is often used to reduce negative feelings. Schachter (11) showed that people who are in a threatening situation prefer to be with others who are in a similar situation. For example, an anxious person feels supported when other people also feel anxious. The idea that fellow sufferers can have positive effects is supported in the literature advocating group sessions for children with cancer (19,69-73) as well as for their parents (74-81). That fellow sufferers may exert a strong influence is illustrated by the observation that parents in group meetings are more receptive to taking advice from each other than from a professional (21). In some situations, however, social comparison may aggravate negative feelings. This is illustrated by one parent's remark about being with other parents in the clinic waiting room: "Waiting hours were difficult. When I heard parents talking about their child's illness, I felt an urge to pick up my child immediately and run away from the hospital because my own misery returned so overwhelmingly." Social comparison may provoke negative feelings, especially when children and parents compare their situation with one that is worse: e.g., when a fellow sufferer dies (see, for example, 82). Consequently, patients and parents tend to select their information by stressing the similarities between the patient and other victims who appear to be doing well and by avoiding identification with those who have not responded well to therapy (14).

Support and comfort can be obtained not only from fellow sufferers but also from other persons including nurses, friends, and social workers. However, the amount of support and comfort that friends, nurses, and social workers provide will depend on their understanding of the situation. Parents can be a powerful source of support for the child with cancer by talking about the child's feelings, fears, and hopes and by sharing their concerns about the disease. Spinetta and Maloney (67) found that patients from families that openly discussed the illness had fewer problems and were more satisfied with themselves. Receiving support and comfort may strengthen the patient's sense of control and, in turn, enhance his or her feelings of self-esteem.

Attributing events to causes

People construct reality in an idiosyncratic way and interpret events within this frame of reference. By attributing to causes, they create a logical, structured world-one that is understandable and predictable and, to some extent, controllable (8,9). Thus, the confrontation with childhood cancer calls for an explanation resulting in a search for the meaning of the disease (14). In searching for a meaning, one can attribute the cause of the disease inside oneself (internal attribution) or outside oneself (external attribution). One example of internal attribution is that parents often blame themselves for their child's illness (see, for example, 16); another is the young child who looks upon his illness as a punishment for wrongdoing (see, for example, 28,38).

Attributing the cause of an illness to an external agent can have positive effects because it allows one to maintain self-esteem. For instance, one couple, looking for a causal relationship, said that their child's illness was caused by living near a nuclear power station.

Changing the situation

Attempts to change the situation, the most active coping strategy, include all activities aimed at eliminating the anticipated harmful condition that constitutes the threat (12). In the case of illness, changing the situation may mean taking medicine, going to the hospital, and complying with the therapeutic regimen. Most parents regard medical treatment as a tool to regain some control over the illness and thereby reduce feelings of helplessness. As one parent said: "They showed us the treatment protocols for the Wilms's tumor of my son. I was glad there was something we could do about it."

In the case of a life-threatening illness, the strategy of changing the situation has limited value because attempts to attack the illness increase the chance of survival but give no guarantee for cure. However, this strategy can be effective in the process of physical and psychological rehabilitation because attempts to remove the negative consequences of the illness enhance the sense of mastery and restore self-esteem. Examples of active strategies in the process of physical rehabilitation are adhering to a diet, participating in treatment decisions, and learning to use a prosthesis. In the process of psychological rehabilitation, efforts such as arranging conditions that will promote optimal development are important (21,38). Then, the child can use active strategies such as returning to school and planning for the future.

Using denial and avoidance

Denial of an illness presumes a lack of awareness of the illness. Avoidance implies that a person is aware of the illness but attempts to suppress or avoid confronting its implications. Although both are distinct strategies, they resemble each other in their manifestations and function. Both may be appropriate ways of coping with a stressor that realistically cannot be alleviated: e.g., when the primary stressor is a life-threatening disease. The function of both denial and avoidance in this instance is to ward off disruptive degrees of anxiety so that the person can function as normally as possible and preserve his or her self-image. Such strategies are often observed soon after a diagnosis of cancer (see, for example, 83,84) and continue to operate until the person is able to handle the stressor better. This is illustrated by Natterson and Knudson (85), who observed the following response pattern in parents who had just been informed that their child had cancer: (a) shock and denial of the diagnosis, (b) acceptance of the diagnosis but not of the poor prognosis, and (c) acceptance of both the diagnosis and the prognosis.

Denial often permits the child with cancer to live with the illness. Several investigators have confirmed that this use of denial results in better psychosocial adjustment and a more positive outlook on the future (42,46,53,86-89).

Accepting the situation

Because one has only limited influence in the case of a life-threatening illness, children with cancer and their families must find a way of coping with this fact. One method of coming to terms with the situation is to accept it; this strategy is often observed after a period of denial and emotional discomfort (85,90). Once patients have accepted the situation, they are able to perceive reality from the perspective of their new role of being ill, which enables them to adapt themselves to the new reality of day-to-day living.

Other strategies

Two additional methods of coping with childhood cancer should be mentioned briefly: impulsive action and cognitive restructuring. Impulsive action is characterized by sudden outbursts of emotion such as crying or anger. Outlets such as these, which are often observed in parents soon after they hear the diagnosis (24), can be regarded as a method of reducing severe negative feelings and the sense of loss of control.

Cognitive restructuring also may help to reduce the negative concomitants of a stressor such as a life-threatening illness. For example, a child may use self-instruction to reduce anxiety while undergoing medical procedures.

FINAL REMARKS

In this article, we have tried to demonstrate how a model for coping with a threatening situation developed by Pruyn and associates can be used to deal in a structured way with the psychosocial problems of pediatric cancer patients and their parents (2-7). The model is based on the literature about coping with stress, especially stress associated with cancer, and on two psychosocial theories of human behavior: the attribution theory (8,9) and the social comparison theory (10,11).

The article represents a start in describing the impact of childhood cancer on the psychosocial functioning of the child and the family. The subject needs further elaboration, however, especially concerning two issues. First, problems involving uncertainty, loss of control, threat to self-esteem, and negative feelings should be described for each stage of the disease. Several authors have reported that specific problems are associated with each phase of childhood cancer (16,18,56,91,92). As a consequence, questions concerning the disease and its treatment that arise during the initial phase of the illness will differ from those that arise in later phases. These differences can be explored by means of the model. Second, the problems that children with cancer experience will vary, depending on their stage of development (93). The impact of cancer on adolescents has been described in detail by a number of authors (94-98) and can be reflected in the model. However, the problems and coping strategies of younger cancer patients have not been examined thoroughly. Furthermore, the problems experienced by the patient's siblings and their coping strategies also can be delineated by means of the model.

From a methodological perspective, the concepts of the model can be put into operation easily because the model is based on fundamental theories of human behavior. Recent studies indicate that valid and reliable instruments can be developed to measure these concepts in adult cancer patients (5-7).The model seems to be a promising contribution to the study of coping with childhood cancer as well.

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CHAPTER 2.3

PSYCHOMETRIC PROPERTIES OF AN INSTRUMENT MEASURING PSYCHOSOCIAL FUNCTIONING OF PARENTS OF CHILDREN WHO SURVIVED CANCER*

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ABSTRACT

Described the development of an instrument from a conceptual framework of coping with stress, measuring the specific stressors for parents of children surviving cancer, involving the issues of uncertainty, loss of control, threat to self-esteem, and negative feelings. The instrument consisted of eleven a priori scales. The concepts of uncertainty and self-esteem were each measured by one questionnaire, the concepts of loss of control and negative feelings by three and six questionnaires, respectively. In a sample of 133 parents the homogeneity of the four concepts was studied using confirmatory factor analysis and reliability analysis. Results revealed satisfactory indices of homogeneity for uncertainty, self-esteem, and negative feelings. Homogeneity was found to be higher for loss of control if the subscale which measures loss of control with respect to the sibling was deleted.

INTRODUCTION

Behavioral research on the psychosocial aspects of childhood cancer has a relatively short research tradition. One of the most common problems in studying the impact of childhood cancer in parents is the lack of specific instrumentation. Although existing, highly standardized instruments measuring general personality factors and psychopathology in adults allow for reliable judgements, they do not measure the problems specific for parents of a child with cancer. In addition, instruments which are developed and standardized to differentiate between psychopathological and normal adult samples are inappropriate because, in general, parents of a child with cancer can

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be regarded as normal individuals under extreme stress. By focussing on identifying psychopathology the whole range of responses remains concealed. Another obstacle in developing instruments is the lack of a theoretical framework in studying the psychosocial responses of parents to childhood cancer because such a framework provides guidance and structure in operationalizing relevant concepts (1-4). In the psychosocial literature on childhood cancer the need for specific and reliable instruments to detect variations within the normal range of responses to childhood cancer has been underlined (1,3-5). In this chapter we will describe our approach to the problem of instrumentation.

We developed instruments to study the specific impact of childhood cancer in parents from a general conceptual framework of coping with stress (6-11). This model, based on the literature on coping with stress and two psychosocial theories of human behavior, namely the attribution theory and the social comparison theory, describes the problems people experience when confronted with a threatening situation. According to the model, a stimulus is perceived as a stressor when it causes (a) uncertainty, (b) loss of control, (c) threat to self-esteem, and (d) negative feelings (Figure 1). These four aspects can vary in intensity and they can occur simultaneously. Uncertainty is defined as a lack of information about a value system that is important for a person.

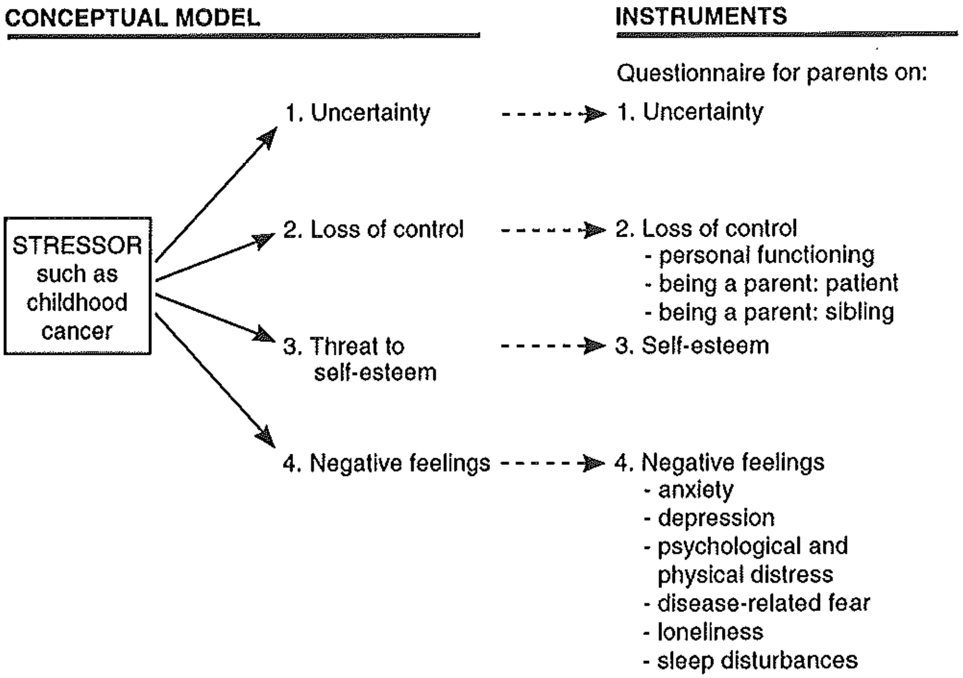

Figure 1. Conceptual model of coping with cancer and instruments measuring the concepts of the model.

People strive to experience as less uncertainty as possible in order to predict and influence forthcoming events. This implies that people want to have information when they experience uncertainty. Loss of control is defined as the inability to manage or influence events. Loss of control can be experienced regarding a number of different areas that are important for the individual including the individual's daily, physical, emotional, social, and general functioning. Self-esteem is related to the image an individual has about oneself. Finally, negative feelings include all other feelings by the individual perceived as unpleasant such as loneliness, depression, and fear. It has been demonstrated that the many psychosocial problems experienced by parents of children with cancer can be assigned to these categories (12). Thus, to assess the psychosocial problems of parents of children with cancer by standardized instruments at least these four domains should be covered.

The aim of this chapter is to outline the development of a standardized instrument for parents of children with cancer, measuring (a) uncertainty, (b) loss of control, (c) threat to the self-esteem, and (d) negative feelings. These four problem categories of the model were made specific for parents of children who survived cancer, that is, specific issues of uncertainty, loss of control, threat to the self-esteem, and negative feelings were formulated for parents after their child's treatment has ended. These issues were assembled in eleven a-priori scales measuring the four problem categories: one questionnaire for uncertainty, three questionnaires to measure loss of control, one questionnaire to measure self-esteem, and six questionnaires to measure negative feelings (Figure 1). To assess whether the instrument can be used to measure the concepts of the model, the homogeneity of the scales measuring the four problems categories was explored using confirmatory factor analysis and reliability analysis. Because loss of control and negative feelings are measured by several questionnaires, first analyses were performed to determine the homogeneity of the separate a priori subscales. To investigate whether each of these subscales measured the same underlying concept, confirmatory factor analysis and reliability analysis were performed on the subscale scores. In this way the homogeneity of the overall scales of loss of control and negative feelings was assessed.

MATERIALS AND METHODS

Subjects

Parents of 8-12-year-old children (3 children were older than 12 years, but still attending elementary school) diagnosed with childhood malignancy, who were in continuous remission and had completed chemotherapy, were the subjects of this study. Termination of chemotherapy had occurred 6 months to 7 years and 9 months (median 4 years and 1 month) beforehand. All patients were of Dutch cultural background and represented all eligible children with leukemia and lymphoma at the Sophia Children's

Hospital/University Hospital Rotterdam (one of the four pediatric cancer centers in The Netherlands) and four children with a solid tumor, who were consecutively selected according to the same criteria. These latter four patients resembled the children with lymphoproliferative diseases with respect to prognosis, treatment protocol, difficulty of treatment, length of treatment, and type of late medical sequelae.

Seventy-three families were approached to participate in the study. Two families refused because they feared a disturbance in their psychological equilibrium as a result from the study. In total 71 families participated with one (*n*=6) or both parents (*n*=65). In these former six families, only the mother participated because she was single (*n*=4), or the father refused to participate (*n*=2). Of a total of 136 parents, 133 (98%) parents from 70 families returned the questionnaires. Reasons for not returning questionnaires were: psychological upheaval (*n*=1), physical handicap (*n*=1), and loss of questionnaire (*n*=1). In Table 1 the demographic characteristics of the parents are summarized.

TABLE 1: Demographic characteristics of parents of childhood cancer survivors (*N*=133).

Sex:		*Religion:*	
-Father	48% (*n*=64)	-Catholic	25% (*n*=33)
-Mother	52% (*n*=69)	-Protestant	28% (*n*=37)
		-Other	9% (*n*=12)
Marital status		-None	37% (*n*=49)
-Married	97% (*n*=129)	-Missing	1% (*n*= 2)
-Divorced or single	3% (*n*= 4)		
		Educational level:	
Age (median, range):		-Low	59% (*n*=78)
-Age at diagnosis:		-Medium	23% (*n*=31)
.father	34 y (24 y - 54 y)	-High	17% (*n*=22)
.mother	32 y (23 y - 43 y)	-Missing	1% (*n*= 2)
-Age at investigation:			
.father	40 y (32 y - 58 y)	*Family size:*	
.mother	39 y (31 y - 55 y)	-One child	9 % (*n*= 13)
		-More than one child	91% (*n*=120)

Instruments measuring the concepts of the model

Item pool

As a starting point we used the items developed by Van den Borne and Pruyn (10,11) measuring the four problem categories of the model in adult cancer patients. To get insight into the specific problems of parents, a pilot study was performed in 14 families. Parents were interviewed in-depth about their experiences with their child's illness and their present situation. These open, free-attitude, interviews gave parents the freedom to discuss their own areas of importance in their own way. The duration of the interviews ranged from 2.5-4.5 hours and all interviews were audiotaped. From the recorded verbatim protocols and observations during the interview problems were identified (see also 13). An exhaustive list of items was formulated, some of them being direct verbalizations of the parents. Items were subsequently assigned to one of the problem categories of the model. All available items from both the pilot study and those developed by Pruyn and Van den Borne (10,11) for adult cancer patients covering a problem category were assembled. Then a selection among these items was made according to the following criteria: the item (a) was relevant for parents as measured by the recurrence of the issue in the interviews; (b) covered a new issue which was considered important in the context of the model; (c) covered an issue reported in the literature (d) was not redundant. In this way eleven a priori scales were constructed to measure the four problems categories of the model. These scales were then used in a pilot study of twelve parents to get insight into the formulation of items, its unambiguous content, response format and length of the questionnaire. Consequently, some items were adapted or eliminated and some changes in lay-out were made resulting in a manageable and attractive questionnaire to fill in by parents.

Uncertainty
Uncertainty in parents of childhood cancer survivors was measured by one *a priori* scale containing 21 items.

Loss of control
Loss of control was measured in relation to (a) the parents' own functioning and in relation to his/her role of being a parent, (b) with respect to the patient, and (c) sibling. Therefore, three *a priori* scales were developed to measure this concept. Loss of control concerning parents' personal functioning was measured by a questionnaire containing 10 items. Two a priori scales, measuring loss of control in their role as parents i.e. with respect to the cancer survivor and his/her sibling, consisted of nine and ten items, respectively.

Self-esteem
Self-esteem was measured by one *a priori* scale containing 4 items.

Negative feelings
In this study six *a priori* scales were used to measure the concept of negative feelings: anxiety, disease-related fear, sleep disturbances, loneliness, depression, psychological and physical distress. These feelings are most often reported in the literature on psychosocial functioning of parents of children with cancer during treatment (4). *Anxiety:* Because of our interest in parent's present feelings of anxiety an adaptation of the state version of the State-Trait Anxiety Inventory (14) was used consisting of seventeen items. *Depression*: Depression was measured by an adaptation of the Zung Depression Scale (15) and consisted of ten items. *Psychological and physical distress:* For psychological and physical distress the Rotterdam Symptom Checklist was used (16). *Disease-related fear*: In this study an eleven item questionnaire was composed to measure disease-related fear in parents based on the literature on the late physical and

emotional consequences in childhood cancer (17,18) and on parental concerns (19). *Loneliness*: A nine item loneliness scale was used containing four items regarding specific issues for parents of childhood cancer survivors. *Sleep disturbances*: In this study a five item scale was used.

Response format

Responses could be displayed in open boxes next to each item. Above these boxes the degree to which an item was applicable was indicated. Uncertainty was measured by a 4-point Likert scale (nothing at all; a little; fairly much; a great deal). Loss of control and sleep disturbances were measured by a 2-point Likert scale (yes; no); self-esteem by a 3-point Likert scale (correct; not correct/not wrong; wrong); and all other problems by a 4-point Likert scale: fear (not at all worried, a little worried, rather worried, extremely worried); loneliness (I strongly agree; I agree; I disagree; I strongly disagree); depression (a little of the time, some of the time, good part of the time, most of the time); psychological and physical distress (not at all; a little; somewhat; very much); and anxiety (not at all; somewhat; moderately so; very much so).

Procedure

Initial contact for the study was established during regular appointments at the outpatient clinic. To give parents time to make up their minds, informed consent was obtained from parents one week after the initial contact. Parents were interviewed at their homes about their experiences with the disease. After the interview, they received a booklet containing a section with questions about the survivor, the sibling, and themselves. The questionnaire of the present study was integrated into these sections. Parents were requested to mail the completed forms to the investigator.

RESULTS

To determine whether the eleven scales were unidimensional, we estimated and fitted a 1-factor model for each a priori scale using the LISREL program (20) to the polychoric correlations among the scale items. Polychoric correlations were used because of the categorical nature of the response format. In tables 2 to 5 the estimated factor loadings from confirmatory factor analysis are reported as well as the goodness-of-fit-index (GFI) and adjusted-goodness-of-fit-index (AGFI). These indexes indicate the degree to which the one-factor model fitted the scale scores. If GFI $>.90$ the one-factor model was accepted as a reasonable approximation of the data. Furthermore, factor loadings $\geq .40$ on the latent factor were considered as an indication that these items measured the defined area and were reliable (21,22).

To explore the reliability of scale items, Cronbach's alpha and item-total correlations were computed separately for each scale. Both statistics are reported in Appendixes 1

4. Items with an item total correlation ≥ .30, considered of medium magnitude (23), were assessed to have an acceptable reliability. If an item had both a factor loading < .40 on a scale measuring a particular concept and an item-total correlation < .30 this item was excluded from the scale. All other items were included into the scale.

To determine whether one underlying loss of control concept was measured by the three separate subscales of loss of control and one underlying negative feelings concept by the six separate subscales of negative feelings, we investigated the unidimensionality of these two concepts applying confirmatory factor analysis and reliability analysis to the three and six subscale scores measuring loss of control and negative feelings, respectively.

Uncertainty

Estimation of the 1-factor model yielded factor loadings ≥. 40, which are presented in Appendix 1. GFI and Cronbach's alpha for the total scale were .95 and .93, respectively. The Pearson correlations between items and the total score for uncertainty ranged from .46 to .77.

Loss of control

Results for the three separate a priori loss of control scales and for the overall loss of control scale are summarized in Appendix 2. The GFI values resulting from confirmatory factor analysis were .98 for the subscale loss of control with respect to the parent, .97 for the subscale with respect to the long-term survivor, and .98 for the subscale loss of control with respect to the sibling. Factor loadings were all ≥ .40, except for item 6: *'I have gone downhill financially'* of the subscale loss of control with respect to the parent and two items of the subscale loss of control with respect to the sibling: item 5: *'I have the feeling that I put too much weight on his/her shoulders'*, and item 9: *'I have to take care that this child does not feel neglected for his/her brother or sister that has been ill'*.

Reliability analysis replicated these findings, revealing correlations of ≥ .30 for all items except for the items *'I have gone downhill financially'*, *'I have the feeling that I put too much weight on his/her shoulder'*, and *'I have to take care that this child does not feel neglected for his/her brother or sister that has been ill'*. These items were excluded from their respective subscales. Cronbach's alpha was .77 for the ten items of the subscale loss of control with respect to the parent, .80 for the nine items of the subscale for the loss of control with respect of the survivor and .74 for the 10 items constituting the subscale for the loss of control with respect to the sibling.

Confirmatory factor analysis and reliability analysis of the overall loss of control scale revealed factor loadings and subscale-total correlations for the subscales loss of control with respect to the parent and to the survivor ≥ .40 and ≥ .30, respectively. However, for the subscale loss of control with respect to the sibling, a factor loading of .24 and a subscale-total correlation of .19 was found, which indicates that this subscale does not belong to the overall concept of loss of control. Cronbach's alpha was .53 for the three subscales and .64 if the subscale loss of control with respect to the sibling was deleted.

Self-esteem

Estimation of the 1-factor model showed factor loadings of ≥ .40 for all items (see Appendix 3) and a GFI of .99. The item-total correlations resulting from reliability analysis ranged from .47 to .58. Cronbach's alpha of .73 in our study was somewhat higher than the .67 for the same self-esteem scale used in the adult cancer patients study (11).

Negative feelings

For all six scales, estimation of the 1 factor model showed GFI indices ≥ .90 (see Appendix 4). Factor loading were ≥ .40 for all items constituting the six scales, except for item 1 *'worry that the illness will come back in the future'*, that had a loading of .30 on the scale measuring disease-related fear. Reliability analysis replicated these results, revealing item-total correlations ≥ .30 for all items, except for *'worry that the illness will come back in the future'* which yielded a correlation of .25 with the total score of disease-related fear. Cronbach's alphas for the fear, anxiety, loneliness, depression, sleep disturbance, and psychosomatic complaints scale were .86, .95 (.95), .84, .87 (.83), .70, and .93 (.93), respectively. Between brackets the coefficients alphas from the Van den Borne and Pruyn (11) study are presented if exactly the same scale was used.

Confirmatory factor analysis (GFI=.99) and reliability analysis of the overall scale measuring negative feelings showed six factor loadings ≥ .40 and six subscale-total scale correlations ≥ .30. Cronbach's alpha was .82 for the six subscales constituting the overall scale for negative feelings.

Intercorrelations between the scales

The theoretical model implies that the four problem categories are not independent from each other but can occur simultaneously as the result of a stressor. Therefore we investigated the intercorrelations between the scales which are presented in Table 2. All intercorrelations between subscales were significant ($p < .001$) except for the correlation between uncertainty and self-esteem. Especially, the subscale negative feelings showed high correlations with other subscales.

TABLE 2. Subscale correlations between the four problems categories of the model for the total sample ($N=133$).

	Uncertainty	Loss of control	Self-esteem	Negative feelings
Uncertainty	-	-	-	-
Loss of control	.36*	-	-	-
Self-esteem	.12	.41*	-	-
Negative feelings	.34*	.66*	.73*	-

* $p<.001$

DISCUSSION

The aim of our study was to develop an instrument to measure the specific late consequences for parents of childhood cancer in the areas of uncertainty, loss of control, threat to the self-esteem, and negative feelings. The results of this study are encouraging. There was evidence for the internal validity of the four concepts as measured by the instrument. Uncertainty, self-esteem, and negative feelings showed unidimensionality including all a priori scales. With respect to the concept of loss of control, however, only two of the three scales belonged to the overall concept of loss of control. It seems that the *a priori* scale with respect to the sibling measures a distinct aspect of loss of control, although the scale can be used in a reliable way to measure this specific aspect of loss of control. Whereas parental feelings of loss of control with respect to their own functioning is closely related to their feelings of loss of control with respect to the survivor, this is not the case with feelings of control experienced with respect to the sibling. This might be explained by the fact that parents regard the survivor as the central issue in their family on which the emotional atmosphere within the family is dependent (24). Consequently, the survivor is the family member to which parental well-being is closely related. Corroborative evidence for the distant role of the sibling within the family is provided by studies demonstrating a strong bond between parents and the child with cancer and a looser relationship between parents and sibling (25) and showing that within the family the needs of the child with cancer and the parents are best fulfilled whereas the needs of the sibling are met least of all (26). Observations during interviews with parents of pediatric cancer survivors concerning their experiences with childhood cancer also revealed, that despite the attempts to focus on the impact of the disease on the sibling, these issues were generally mentioned only briefly (13).

With respect to internal consistency, both measures of unidimensionality indicated acceptable to high internal reliability for all scales. However, the scores obtained from parents were not completely independent, which may have affected the reliabilities. Although parents were requested to report their own experience, on some scales, this experience may be closely linked to the status of the child, which is the same child for both parents. To explore this effect, separate analyses for mothers and fathers revealed that the mean deviation of Cronbach's alpha from the reliability computed for the total sample was .02 and .03, respectively. This finding indicates that in the present study the dependence of the scores had a negligible effect on reliability. The satisfactory internal consistency found in measuring the concepts of the model in both parents of pediatric cancer survivors and adult cancer patients (10,11) indicate that the problem categories of the model can be reliably measured in different samples. However, there were also problems in establishing reliability in our sample. First, no direct control was exerted over the independence of completing the questionnaires in couples. Second, we did not establish test-retest reliability in our sample, because of our study design. As indicated in the Procedure section parents were first in-depth interviewed. This interview lasted several hours and parents generally indicated to be emotionally involved (13). Because of the impact of the interview on the parents, we decided for clinical reasons not to approach these parents again for retesting for only scientific reasons. Although we did not retested parents in our study, we feel that establishing test-retest reliability is a feasible option when a different procedure is used f.i. by using only questionnaires.

With respect to the instrument's validity, to judge if the applied instruments cover the domain of problems existing in our sample of parents, the high internal consistency of the problem scales found in this study is a first and necessary, but not sufficient, indication for construct validity. One of the problems in establishing construct validity in an instrument measuring disease-specific issues is the uniqueness of the experience. A suitable approach would be to study the relation between constructs and a different set of observables (27). A way of finding indications for external validity is to explore whether problem scores obtained on this instrument correlate with other indices of stress. Indices which can be used for this purpose are, for instance, absence from work, use of services for mental health, loss of social contact, visits to the general practitioner, medicine intake, and marital satisfaction. If problem scores on the here presented questionnaire are high, indicating a greater burden for parents, other indices of stress should be elevated as well. Therefore, a next step in exploring the validity of the instrument would be to investigate its relation with other indices of stress.

Problems associated with childhood cancer vary and change during the course of the illness and treatment. Each phase of the disease has its own characteristic problems (28-30). The variations and differences in problems over time can be explored by means of the presented theoretical model. In this study the instruments are made specific for the disease stage after treatment ends. Although this is a strength of the instrument, it poses also some limitations to the application of the instrument during other stages of the disease process. Although a number of scales can be used in their current form during treatment such as physical and emotional complaints, anxiety, depression, loss of control, self-esteem, some scales need some slight adaptations such as the uncertainty and loneliness scale. In addition, items may be added which are specific for the phase under study.

Our study illustrates that reliable instruments can be developed from a conceptual framework of coping with stress to measure the specific impact of childhood cancer on parents. Because the model from which the scale domains were derived is a general framework of describing the impact of a stressor, it can be applied to other diseases and made specific for the type of stress evolving from a particular disease. The items selected in this study to assess cancer-related stress can be used as a starting point to adapt these instruments for another stressor under study.

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APPENDIX 1: Estimated factor loadings and item-total correlations of the uncertainty scale.

Description of instruction and items	Loadings	Item-total correlation
Parents of children who have been ill are often left with a number of questions after treatment has been completed, questions they would like to have more information about.		
To what extent would you like to know more about:		
1. the cause of the disease	.51	.46
2. the present physical condition of your child	.71	.57
3. the long-term chances of survival of your child	.72	.58
4. possible side effects of the treatment in the long run	.67	.50
5. the extent to which your child has been physically impaired by the treatment of the disease	.75	.59
6. the purpose of the follow-up examinations	.74	.65
7. when your child is considered cured	.82	.74
8. how to talk or deal with a doctor	.71	.59
9. which people you come in contact with, you have to inform about the disease	.63	.56
10. where to go with questions and problems around the disease of your child	.78	.71
11. where to obtain good information material or literature about the period after completion of treatment	.74	.65
12. the extent to which the illness left its marks on the child's emotions	.78	.64
13. what is going on in your child's mind in connection with the disease	.66	.53
14. how you can help your child in getting over the disease and treatment	.84	.70
15. whether you should talk about the disease with your child or not	.85	.77
16. in how far you should adapt your child's upbringing to what he or she experienced because of the disease	.67	.61
17. which demands you may make as to your child's learning at school	.55	.48
18. how you can help your child to mix with peers	.76	.68
19. what questions other parents have about their child who has been ill	.67	.61
20. how other parents try to solve these questions	.69	.64
21. how other parents cope with the disease of their child personally	.62	.55

GFI: .95; AGFI: .94
Cronbach's alpha: .93

APPENDIX 2: Estimated factor loadings and item-total correlations of the three loss of control subscales and of the overall score for loss of control.

Description of instruction and items	Loadings	Item-total correlation
Loss of control with respect to the parents personal functioning		
Because of the disease of my child:		
1. I find it more difficult to have contact with friends, acquaintances, and relatives	.67	.37
2. more things are prying on my mind	.76	.52
3. I feel like having less grip on my family	.72	.40
4. I have my emotions less under control	.79	.54
5. I have less contact with my partner	.69	.39
6. I have gone downhill financially	.39	.25
7. I am not yet my old self again	.84	.56
8. I have become more dependent on others	.86	.52
9. the mutual relations in the family are more easily disturbed	.81	.50
10. I find it still difficult to pick up the daily rhythm	.75	.47
GFI: .98; AGFI: .96 Cronbach's alpha: .77		
Loss of control with respect to the patient		
Because of the disease of my child and its treatment:		
1. it is harder for me to decide what is right for this child	.81	.60
2. it is less easy to take time off for myself	.87	.59
3. I have less opportunity to go out and have a nice time with my partner	.80	.51
4. I cannot get a grip on what's going on in my child's mind	.71	.44
5. bringing up my child goes less as smoothly as before	.63	.41
6. I have the feeling that my child's happiness is less in my own hands	.68	.47
7. I have the feeling that I am less able to get at the child	.85	.53
8. it is less easy to take a job	.76	.42
9. I am less relaxed with my child	.81	.48
GFI: .97; AGFI: .95 Cronbach's alpha: .80		

(continued) →

APPENDIX 2 (continued).

Description of instruction and items	Loadings	Item-total correlation
Loss of control with respect to the sibling		
Because of the disease of the brother or sister of this child:		
1. I was not able to spend enough time this child	.57	.30
2. I have the idea that I have missed a period from the life of this child	.74	.48
3. bringing up this child goes not as smoothly as before	.73	.40
4. I cannot get a grip on what's going on in this child's mind	.90	.56
5. I have the feeling that I put too much weight on his/her shoulders	.31	.22
6. it is as if the child has slipped away from me	.91	.56
7. contact with the child is less easy than in the past	.61	.39
8. I have the feeling that I am less able to get at this child	.99	.65
9. I have to take care that this child does not feel neglected for his/her brother or sister that has been ill	.35	.23
10. I worry more about this child	.85	.51
GFI: .98; AGFI: .97 Cronbach's alpha: .74		
Overall score of loss of control		
1. with respect to the parents	.85	.48
2. with respect to the patient	.56	.38
3. with respect to the sibling	.24	.19
GFI: 1 Cronbach's alpha: .53		

APPENDIX 3: Estimated factor loadings and item-total correlations of the self-esteem scale

Description of instruction and items	Loadings	Item-total correlation
Below you find a number of statements. Please indicate to what extent they are correct; not correct, not wrong; wrong.		
1. I feel cheerful	.64	.47
2. I lack self-confidence	.67	.50
3. I can handle my own problems quite well	.77	.54
4. I find it very hard to cope with setbacks	.80	.58
GFI: .99; AGFI: .99		
Cronbach's alpha: .73		

APPENDIX 4: *Estimated factor loadings and item-total correlations of the six negative feelings subscales and of the overall score for negative feelings.*

Description of instruction and items	Loadings	Item-total correlation
Anxiety[*]:		
1. feeling calm	.91	.76
2. feeling secure	.91	.74
3. feeling tense	.83	.71
4. feeling strained	.92	.79
5. feeling at ease	.94	.80
6. feeling worried over possible misfortunes	.70	.60
7. feeling satisfied	.87	.70
8. feeling frightened	.72	.53
9. feeling comfortable	.93	.78
10. feeling self-confident	.92	.79
11. feeling nervous	.72	.59
12. feeling relaxed	.95	.83
13. feeling content	.92	.73
14. feeling worried	.72	.63
15. feeling confused	.71	.58
16. feeling steady	.85	.71
17. feeling pleasant	.99	.84
GFI: .95; AGFI: .93		
Cronbach's alpha: .95		
Disease-related fear		
Below you find a number of concerns expressed by parents who have a child who has been ill. Could you indicate to what extent you are worried:		
1. that the illness will come back in the future	.30	.25
2. about the late after-effects of treatment	.68	.64
3. about the growth in height of your child	.49	.40
4. about his/her sexual development	.89	.61
5. about his/her (later) fertility	.83	.54
6. about the school performance of your child	.59	.53
7. about your child's possibility of finding a good job in the future	.68	.60
8. about the way your child associates with other children	.72	.55
9. about the emotions of your child	.77	.65
10. about the chances for your child to find a partner in life	.84	.73
11. about a complete decline of your child	.81	.67
GFI: .91; AGFI: .87		
Cronbach's alpha: .86		

(continued) →

APPENDIX 4 (continued).

Description of instruction and items	Loadings	Item-total correlation
Loneliness:		
How strongly do you agree or disagree with the following statements:		
1. it is hard to find real friends	.63	.50
2. when you really need someone, there is no one there	.70	.57
3. because of the disease of my child, we as a family have become left on our own	.81	.66
4. sometimes I feel very lonely	.66	.47
5. sometimes I feel that one is alone in the world after all	.79	.65
6. because nobody understands, it is better to keep your worries about the child to yourself	.73	.62
7. people just do not understand there are still a number of worries left after treatment has ended	.66	.54
8. sometimes it is very difficult to make long-lasting contacts	.71	.57
GFI: .93; AGFI: .87		
Cronbach's alpha: .84		
Sleep disturbance:		
Could you indicate whether you have often been disturbed in your sleep of late by:		
1. awaking early	.76	.50
2. during sleep re-experiencing illness	.62	.32
3. awaking tired	.82	.52
4. awaking often during night	.75	.45
5. not able to sleep	.90	.53
GFI: .99; AGFI: .98		
Cronbach's alpha: .70		

(continued) →

APPENDIX 4 (continued).

Description of instruction and items	Loadings	Item-total corelation
Depression:		
Below you find a number of statements that may describe the way you feel during the past few days. Could you indicate to what extent these statements apply to you:		
1. I feel down-hearted and blue	.88	.64
2. I have crying spells or feel like it	.78	.55
3. I get tired for no reason	.61	.49
4. I feel hopeful about the future	.62	.52
5. my mind is clear as it used to be	.77	.58
6. I find it easy to do the things I used to do	.80	.64
7. I am more irritable than usual	.66	.59
8. I still enjoy the things I used to do	.73	.64
9. I feel despondent	.93	.68
10. I fail to see the purpose of a lot of things	.77	.64
GFI: .94; AGFI: .91		
Cronbach's alpha: .87		
Physical and psychological distress:		
Below you find a number of complaints. Could you indicate to which extent you suffered from these complaints during the past few days.		
1. irritability	.69	.54
2. tiredness	.77	.61
3. worrying	.70	.59
4. sore muscles	.73	.57
5. depressed mood	.89	.75
6. low back pain	.69	.56
7. nervousness	.81	.67
8. desperate feelings about the future	.86	.68
9. headaches	.64	.57
10. dizziness	.82	.60
11. feeling lonely	.66	.52
12. tension	.88	.73
13. anxiety	.83	.69
14. heartburn/belching	.79	.66
15. shivering	.74	.52
16. tingling hands or feet	.62	.50
17. burning (or sore) eyes	.73	.53
18. shortness of breath	.72	.58
19. lack of energy	.85	.70
20. dry mouth	.75	.57
GFI: .93; AGFI: .92		
Cronbach's alpha: .93		

(continued) →

APPENDIX 4 (continued).

Description of items	Loadings	Item-total corelation
Total negative feeling-score		
1. anxiety	.82	.72
2. disease-related fear	.41	.41
3. loneliness	.53	.52
4. sleep disturbances	.65	.59
5. depression	.91	.77
6. physical and psychological distress	.81	.71
GFI: .99		
Cronbach's alpha: .82		

*:Exact item description and instructions for completing questionnaire have to be obtained by Consulting Psychologist Press, Inc. P.O. Box 10096, Palo Alto CA, 94303.

CHAPTER 2.4

CROSS-CULTURAL VALIDATION OF HARTER'S SELF-PERCEPTION PROFILE FOR CHILDREN IN A DUTCH SAMPLE*

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ABSTRACT

The Self-Perception Profile for Children (SPPC) was administered to 300 school-aged children (8-12 years) in order to investigate its psychometric properties in a non-American sample. Exploratory and confirmatory factor analyses supported the multi-factorial structure of the SPPC. The internal-consistency and test-retest reliability of the SPPC were found to be acceptable. Significant gender differences were found, showing that boys perceive themselves as more competent than girls. The results of this study closely match the results obtained in American samples, indicating that the SPPC is a reliable and internally valid instrument in measuring the self-concept in children.

INTRODUCTION

Self-concept is an important construct in theories on child development and is related to adaptation, competence, and mental health. Although its theoretical standing and clinical relevance have been firmly established, problems with operationalization and measurement of self-concept are numerous (1,2). Self-concept has been defined as a unidimensional construct, stressing the global nature of self-esteem (3) but others have argued that self-concept is a construct with multiple dimensions (4,5). In addition, Hughes (2) outlined major problems in the instrumentation of self-concept such as lack of theoretical basis, little construct validity, and a substantial influence of social desirability.

Harter (6,7) acknowledged these problems and developed the Self-Perception Profile for Children (SPPC), a self-report instrument for measuring self-esteem or perceived competence in children. This instrument differs from existing scales for measuring

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self-esteem or self-concept, in that it assesses the child's sense of competence across several specific domains as well as his or her general sense of self-worth, instead of viewing perceived competence as an unitary concept. Underlying the construction of the SPPC is the assumption that an instrument providing separate measures of perceived competence in different domains, as well as an independent assessment of global self-worth, would provide a richer and more differentiated picture of one's self-concept than instruments providing only a single self-concept score. In addition, tapping children's perception of themselves in various domains of their lives makes it possible to examine differences across and relationships among these domains.

The SPPC is a revised version of the Perceived Competence Scale for Children (PCSC; 6) which contained four subscales. Harter (7) added two new subscales to the PCSC, i.e., physical appearance and behavioral conduct, revised a number of items from the domain-specific and global self-worth subscales and renamed the original scales. The SPPC contains six subscales, tapping five specific domains: (a) scholastic competence, (b) social acceptance, (c) athletic performance, (c) physical appearance, and (e) behavioral conduct. A sixth subscale, global self-worth, measures the global judgment of the self as a person, in contrast to domain specific evaluations of competence or adequacy. It is assumed that this latter subscale is independent of any particular skill measured in the other subscales. The questionnaire consists of a total of 36 items (six items for each subscale).

During the last decade, the SPPC has become a widely used instrument for measuring the self-concept in various samples of children in different countries. Studies on the psychometric properties of Harter's perceived competence scales (6,8,9), however, concerned only the earlier version of the SPPC, the PCSC. On the revised version, the SPPC, no such studies have been reported in the international literature, since the publication of the manual (7). Therefore, in the present study the psychometric properties of the SPPC were investigated in a Dutch sample. The aims of this study were: (a) to investigate the multi-factorial structure of the SPPC; (b) to investigate the internal consistency and test-retest reliability of the SPPC scales; this is the first study to report on the test-retest reliability of the competence scores on the SPPC; and (c) to describe gender and age differences in mean scale scores and reliabilities.

MATERIALS AND METHODS

Subjects

A sample of 300 children from two primary schools in a suburban and a rural area were enrolled in the study. In total, 143 boys (47.6%) and 157 girls (52.3%) from the 3rd to 6th grade (Grade 3, *n*=82 (27.3%), age *M*=8.5 y; Grade 4, *n*=69 (23.%), age *M*=9.4 y; Grade 5, *n*=82 (27.3%), age *M*=10.4 y; Grade 6, *n*=67 (22.3%), age *M*=11.4 y) completed the SPPC. The mean age of Grades 3 and 4 (younger children), was 8 years and 10 months, and of Grades 5 and 6 (older children), 10 years and 10 months. To obtain test-retest results, the SPPC was completed four weeks after the first assessment by 129 children from one school (56 boys [44.2%], 73 girls [55.8%]; Grades 3 - 6).

Instrument

The SPPC (7) was translated into Dutch and a pilot-study was performed with 32 children (age 8-12 years; eight respondents in each Grade 3-6). For the SPPC, Harter (6) devised a 'structured alternative format' for each question in order to reduce children's tendency to give socially desirable responses. The child is presented with the following type of questions:

really true for me	sort of true for me	some kids often forget what they learn	BUT	other kids can remember things easily	sort of true for me	really true for me
☐	☐				☐	☐

It is assumed that this formulation of questions provides an opportunity for the child to identify himself or herself with either group of children without feeling deviant, thus reducing response bias.

In the pilot study, although the content of the questions was easily understood by all children, the questionnaire appeared difficult for them to complete as a result of the question and response format used. The problem was how the child's choice had to be represented in the response format. The decision making process for completing this type of item requires several steps: (a) relating (and applying) the content of the questions to one's own competence, (b) deciding to which broad category one belongs, and (c) finally making a discrete judgment on a continuum from low to high competence. In the pilot-study, the following problems emerged concerning the scoring: (a) children had problems perceiving the response format as a continuum of competence (some subjects scored both one box on the left and one on the right side of the statement) and (b) children had problems relating the statement concerning the competence of other children to themselves. The latter was enhanced by the fact that the statements above the box (i.e., *really true for me* and *sort of true for me*) were only listed at the top of each page and not above each subsequent item. Our experiences with this format were confirmed by van den Bercken (personal communication, January 13th, 1987), who used the earlier version of the SPPC (6) with Dutch children (10).

In order to facilitate the multi-step decision making process, an effort was made to visualize some of these steps so as to more easily guide the child's decision-making. Consequently, several alternative response formats were tried, resulting in a Dutch

TABLE 1. Factor pattern (oblique rotation) for the SPPC obtained from exploratory and confirmatory factor analyses (decimal points omitted).

		Loadings from exploratory analysis[a]					Loadings from confirmatory analysis[b]				
Item description		Scholastic competence	Social acceptance	Athletic competence	Physical appearance	Behavioral conduct	Scholastic competence	Social acceptance	Athletic competence	Physical appearance	Behavioral conduct
1	Good at schoolwork	77 (62)					68				
7	Just as smart	67 (64)					58				
13	Schoolwork quickly	68 (64)					55				
19	Remember things easily	59 (59)					58				
25	Do well at classwork	76 (67)					77				
31	Can figure out answers	70 (60)					63				
2	Easy to make friends		66 (69)					39			
8	Have a lot of friends		83 (70)					66			
14	Easy to like		76 (41)					73			
20	Do things with a lot of kids		42 (56)					35			
26	Most kids like me		59 (62)					80			
32	Popular with others		57 (43)			34(–)			67		
3	Do well at sports			88 (80)					58		
9	Good enough at sports			71 (77)					60		
15	Good at outdoor activity			81 (49)					59		
21	Better than others at sports			35 (72)					63		
27	Play rather than watch			37 (41)					46		
32	Good at new outdoor games			52 (73)					62		
4	Happy with the way I look				73 (71)					68	
10	Happy with height and weight				50 (64)					53	
16	Like body the way it is				62 (52)					61	
22	Like physical appearance as is				79 (65)					79	
28	Like face and hair as is				62 (28)					57	
34	Are attractive or good looking				80 (49)					81	
5	Like the way I behave	33				–(36)					39
11	Usually do the right thing	50				–(57)	61				--
17	Act the way supposed					59(69)					54
23	Don't get in trouble	38				–(69)					62
29	Don't do the things shouldn't					64(82)					60
35	Behave myself					76(50)					45

Note. Factor loadings less than .30 are not included. In parentheses factor structure (sample C_1; $n = 100$) from Harter (1985). Decimal points have been ommitted.

[a] Loadings obtained from principal components analysis.

[b] Loadings are unweighted least squares LISREL estimates.

version of the SPPC (see Appendix 1) with the following adaptation of the question and response format:

Some kids often forget what they learn BUT Other kids can remember things easily

What children are you like?

really true for me	sort of true for me	really true for me	sort of true for me
☐	☐	☐	☐

This modification was the only adaptation made. The content and order of the items, the scoring key (positively and negatively keyed items), and the scoring remained identical to Harter's (7). Each item is scored from 1 to 4, where a score of 1 represents low perceived competence and a score of 4 indicates high perceived competence.

RESULTS

Factor structure

Exploratory factor analysis

A principal components analysis was performed on all items except those included by Harter (7) in the general self-worth scale. According to Harter (7), a distinctive factor for global self-worth is not likely to emerge because the value of the particular domain for one's self-worth varies among individuals and as a consequence bears a different relationship to self-worth for different subjects. The analysis was restricted to a 5-components solution and was followed by varimax and oblique rotation. The varimax rotation showed a factor pattern with numerous loadings above .30. The oblique rotation revealed a structure with a better fit to the five factors described by Harter (7). The results of the oblique rotation for the five specific domains are presented in Table 1. This 5-factor solution accounted for 50.1% of the variance. Before rotation, Factors F1 to F5 accounted for 23.2%, 8.1%, 6.0%, 7.5% and 5.3% of the variance, respectively. After rotation, the eigenvalues ranged from 4.74 to 2.65 and the explained variance for Factors F1 to F5 was 15.8%, 12.7%, 14.1%, 12.6% and 8.8%, respectively. The correlations between factors ranged from -.30 to .24.

The results of the present study were compared with those from Harter's study (7) for samples that were comparable, namely Harter's sample C (*N*= 247; Grade 3, 24 boys and 36 girls; Grade 4, 32 boys and 25 girls; Grade 5, 27 boys and 29 girls; Grade 6, 26 boys and 28 girls). Harter's Sample C was divided into C1: Grades 5 and 6, and C2: Grades 3 and 4. For some comparisons Harter's C1 sample was used and for others, C2. Compared with Harter's C1 Sample, the factor structure in the present study matched the first four reported by Harter (7), except that Item 32 ("popular with others"), originally designed for Social Acceptance, had a .34 loading on Behavioral Conduct, but a higher loading (.57) on Social Acceptance. Factor 5, Behavioral Conduct, however, was less well replicated. Half of the items, originally designed for Harter's Factor 5 (Items 5, 11 and 23), had loadings of .18, .10, and .29, respectively in the present study on Factor 5, Behavioral Conduct, but had higher loadings of .33, .50 and .38, respectively, on Factor 1, Scholastic Competence.

Confirmatory factor analysis

In order to test the factor structure obtained from Harter's study (7), a confirmatory factor analysis was performed using version 7.16 of the LISREL program (11). Since the items were of an ordinal level of measurement, maximum likelihood estimation was not justified and unweighted least squares estimates were used instead. As an initial model, Harter's model (see Table 1) was fitted to the data with each item loading only on the factor as specified by Harter (7), and all other loadings fixed at zero. Although the goodness-of-fit (GFI) indices for this model were quite acceptable (GFI=.959, AGFI=.951, RMR=.058, *df*=395), the exploratory analysis suggested a better fit for the model if Items 5, 11, and 23 were allowed to load on the first instead of the fifth factor. Furthermore, the exploratory analysis suggested Item 32 to have a loading on Factor 5. A model was fitted leaving all items comprised in Factor 5 as well as Item 32 free to load on any factor. This resulted in Items 17, 29, and 35 having high loadings on Factor 5, and item 11 having a high loading on Factor 1. Items 5 and 23, however, had only loadings below .30 on Factors 1, 4, and 5, and Factors 1 and 5, respectively. The loading of Item 32 on Factor 1 was more than two times as high as its loading on Factor 5. Subsequent testing of models in which loadings of Items 5, 23, and 32 were specified as suggested by these findings, however, indicated no improvement of the initial model. Therefore, a final model was fitted which was the same as the initial model except that Item 11 loaded on Factor 1 instead of Factor 5. This resulted in a slight but significant improvement of fit (GFI=.961, AGFI=.954, RMR=.057, *df*=395), with a significant decrease of 28 points in χ^2. It was concluded that Harter's model was significantly improved without loosing degrees of freedom. The LISREL estimates of the factor loadings obtained for the final model are given in Table 1.

Intercorrelations among subscales

Overall results

The correlations between subscales of the SPPC are presented in Table 2. The intercorrelations among subscales were generally lower than the correlation of each subscale with the global self-worth scale. The only exceptions were the correlations obtained between scholastic competence and behavioral conduct and between scholastic competence and general self-worth (.52 and .48, respectively). This finding indicates

TABLE 2. SPPC subscale intercorrelations for the total sample (N=300), grades 3-4 (n=151, below diagonal) and grades 5-6 (n=149, above diagonal), and for boys (n=143, below diagonal) and girls (n=157, above diagonal) (decimal points omitted).

Subscale	Scholastic competence	Social acceptance	Athletic competence	Physical appearance	Behavioral conduct	General self-worth
Total Sample						
Scholastic competence		29	34	34	52	48
Social acceptance			42	34	31	51
Athletic competence				33	29	38
Physical appearance					40	64
Behavioral conduct						47
Grade Level						
Scholastic competence		18[b](31)	22[a](18)	26(31)	42(29)	44(46)
Social acceptance	40(63)[c]		43(31)	36(29)	24[a](22)	54(41)
Athletic competence	43(52)	42(45)		28(43)	21[a](08)	40(35)
Physical appearance	39(48)	33(51)	37(50)		41(12)[c]	64(62)
Behavioral conduct	58(45)	38(29)	32(28)	35(38)		54(42)
General self-worth	50(61)	50(58)	36(52)	63(73)	40(57)	
Gender						
Scholastic competence		25	32	31	55	40
Social acceptance	33		44	36	34	53
Athletic competence	32	41		35[a]	26	42
Physical appearance	30	30	23		51	67
Behavioral conduct	53	28	35	31		52
General self-worth	54	48	29	58	44	

Note. All intercorrelations for the total sample were significant at $p < .001$. In parentheses data (Sample C_1: grades 3-4; n= 110) and Sample C_2: grades 5-6; n= 100) from Harter (7). All intercorrelations significant at $p < .01$ except [a] $p < .05$, [b] ns; [c] not significant after Bonferroni correction.

that children who are functioning well at school also report fewer behavioral problems. This high correlation was also reflected by the fact that half of the items of the behavioral conduct subscale also loaded on the scholastic competence subscale. Of the correlations between each specific domain and general self-worth the highest correlation was between physical appearance and global self-worth. Harter (7) also found that these subscales showed the strongest correlation.

The intercorrelations between the subscales reported by Harter (7) for the American sample and the intercorrelations found in the present study were compared, using Fisher's-Z, and are presented in Table 2. Only two significant differences were found. In the sample of young children (Grades 3 and 4), the intercorrelation between scholastic competence and social acceptance was higher in the Dutch sample than in the American sample. In addition, physical appearance and behavioral conduct were correlated higher in the American sample than in the Dutch sample of older children

(Grades 5 and 6). However, these differences were not significant after Bonferroni-correction for number of comparisons.

Gender and age effects

The subscale intercorrelations for gender and grade are presented in Table 2. For boys the strongest correlation (.58) was between general self-worth and physical appearance, closely followed by scholastic performance (.54). The weakest correlation (.29) was between athletic competence and general self-worth. Similar to boys, the correlations between the specific domains and general self-worth showed that also for girls physical appearance had the strongest correlation with general self-worth (.67), whereas scholastic competence in girls had the lowest correlation (.40). This finding indicates, that for girls physical appearance was most and school and athletic performance were least important for their general sense of self-worth; whereas for boys, physical appearance and school performance were equally important for their sense of self-worth. Similar to girls, for boys competence in sports was also the least important.

In younger as well as older children physical appearance showed the highest correlation with general self-worth (.63 and .64, respectively), followed in older children by social acceptance and behavioral conduct (both .54), and in younger children by scholastic competence and social acceptance (both .50). In both age-group, the weakest correlation between general self-worth was with the subscale athletic competence (.36 and .40, respectively for Grades 3-4 and 5-6). For the older children, the correlations among subscales were lower than the correlation of each subscale with the global self-worth scale, except for athletic competence. This subscale had a correlation of .43 with social acceptance, whereas the correlation with general self-worth was .40. For younger children, however, only social acceptance and physical appearance had the highest correlations with general self-worth, .50 and .63, respectively. Athletic competence and behavioral conduct had the highest correlation with scholastic competence, .43 and .58, respectively (vs .36 and .40 with general self-worth, respectively) and the reverse, scholastic competence had the highest correlation of .58 with behavioral conduct vs .50 with general-self worth.

Subscale Means and Standard Deviations

Overall results

The mean scale scores and standard deviations for the total sample are presented in Table 3. The means of the subscales for the total sample were all above the midpoint of the scale. Means ranged from 2.81 on scholastic appearance to 3.28 on general self-worth. The mean scale scores for American children in the normative sample provided by Harter (1985) were compared with those obtained in this study, using *t* tests. Dutch children had higher scores, indicating higher perceived competence, on social acceptance, athletic competence, physical appearance, and general self-worth. After applying

TABLE 3. Means and standard deviations of subscales SPPC for the total sample and means for age and gender.

Subscale	Mean total sample (*N*=300)	SD	Boys (*n*=143)	Girls (*n*=157)	Grade 3-4 (*n*=151)	Grade 5-6 (*n*=149)
Scholastic competence	2.81 (2.82)	.69 (.69)	2.91	2.70***	2.97	2.65[a]
Social acceptance	3.08 (2.87)***	.68 (.76)	3.15	3.02	3.08	3.09
Athletic competence	3.07 (2.89)[a]	.62 (.73)	3.22	2.93***	3.14	2.99[a]
Physical appearance	3.16 (2.91)***	.75 (.76)	3.32	3.03***	3.29	3.04[a]
Behavioral conduct	2.89 (3.04)[a]	.58 (.56)	2.88	2.89	3.03	2.75***
General self-worth	3.28 (3.04)***	.59 (.69)	3.38	3.19[a]	3.35	3.21[a]

Note. In parentheses data (Sample C, *N*= 210) from Harter (7).
[a] not significant after Bonferroni correction.
***$p<.001$

the Bonferroni correction, the difference on athletic competence failed to reach statistical significance. In contrast, American children scored higher on behavioral conduct, although this difference was not significant after the Bonferroni correction. Standard deviations varied across the total sample from .58 to .75. Harter (1985) found corresponding values for standard deviations ranging from .58 to .70.

Gender and age effects

Table 3 presents the mean scores for the different subscales for both sexes and the two age-groups. A two-way analysis of variance (ANOVA) was performed with age and gender as independent factors. Gender effects were significant for four subscales with boys scoring systematically higher than girls on scholastic competence, athletic performance, physical appearance, and general self-worth, although the latter difference did not remain significant after the Bonferroni correction. Systematic grade effects were obtained favoring younger children on 5 of the 6 subscales. However, after the Bonferonni correction, the only significant difference was on the subscale behavioral conduct. No significant interactions were found.

Reliability

Test-retest

The 4-week test-retest correlations for the different subscales are presented in Table 4. The correlations ranged from .66 for behavioral conduct to .83 for scholastic competence, indicating that the majority of the subscales were stable over time. On only one subscale, scholastic performance, the scores obtained at retest were significantly higher than those obtained at the first administration of the SPPC ($p<.01$).

TABLE 4. Four-week SPPC subscale test-retest reliabilities (r) and Cronbach's alpha's.

Subscale	Test-retest reliabilities				
	Total sample (*N*=129)	Boys (*n*=56)	Girls (*n*=73)	Grade 3-4 (*n*=65)	Grade 5-6 (*n*=64)
Scholastic competence	.83	.79	.85	.86	.81
Social acceptance	.80	.74	.83	.83	.76
Athletic competence	.78	.70	.80	.72	.84
Physical appearance	.76	.74	.76	.69	.82
Behavioral conduct	.66	.64	.67	.70	.59
General self-worth	.71	.62	.75	.69	.74
	Cronbach's alpha's				
	Total sample (*N*=300)	Boys (*n*=143)	Girls (*n*=157)	Grade 3-4 (*n*=151)	Grade 5-6 (*n*=149)
Scholastic competence	.80 (.82)	.77	.82	.79	.81
Social acceptance	.77 (.75)	.69	.83	.76	.79
Athletic competence	.75 (.81)	.67	.77	.75	.75
Physical appearance	.81 (.76)	.83	.79	.79	.83
Behavioral conduct	.65 (.73)	.65	.68	.65	.59
General self-worth	.75 (.78)	.66	.79	.69	.79

Note. In parentheses data (sample C; *N*= 210) from Harter (7).

Gender and age effects

Table 4 also shows test-retest correlations for each sex and age-group. Although girls tended to have more stable scores over time on all subscales (ranging from .67 to .85; median value=.78) than boys (ranging from .62 to .79; median value=.72), these differences failed to reach statistical significance. Test-retest correlations for younger children did not differ significantly from those for older children.

Internal consistency

The internal consistency reliabilities (Cronbach's alphas) for all six subscales are presented in Table 4. The reliabilities ranged from .65 to .81. The lowest reliability score was found for the fifth subscale (behavioral conduct) as was the case in Harter's (7) study. Reliability improved only slightly for the subscale social acceptance (from .77 to .79) if Item 20 ("do things with a lot of kids") was deleted. In addition, deletion of Item 10 ("happy with height and weight") marginally improved the reliability of the subscale physical appearance (from .81 to .82).

Gender and age effects

Subscale alphas, in general, were lower for boys (ranging from .65 to .83; median value=.68) than for girls (ranging from .68 to .83; median value=.79). Differences between younger and older children in subscale alpha's were generally very small. For younger children these reliabilities ranged between .65 to .79 (median value=.76) and for older children between .59 to .83 (median value=.79).

DISCUSSION

The psychometric properties of the SPPC found in the present study closely matched those reported for an American sample of 3rd to 6th graders (7). The results of the exploratory and confirmatory factor analyses supported the multi-factorial structure of the SPPC. The factor structure provided by Harter (7) was nearly perfectly replicated in the Dutch sample despite differences in language and culture. Intercorrelations among subscales supported Harter's hypothesis that children do not only make discrete judgments about their competence in different domains, but also construct a view of their global self-worth over and above specific domains. A study of the self-perceived competence in Chinese children provides corroborative evidence in this respect (9). All studies on the factor structure with the SPPC or PSCS until now supported the use of a differentiated construct of self-esteem to study the child's perceptions of the self.

The factor structure in this study resembled the one reported by Harter (7). Of the two new subscales of the SPPC, physical appearance showed perfect congruence with Harter's (7) results, whereas behavioral conduct did not as was shown both by the results of the factor analysis and reliability measures. Also in Harter's study (7) behavioral conduct appeared to have the weakest psychometric characteristics. In examining the content of the items of this subscale "like the way I behave," "usually do the right thing," and "don't get in trouble," it can be assumed that they measure a school-related behavioral component. The child perceives his behavior within the context of school and assesses it accordingly, taking the school situation as a point of reference. This may be related to the fact that the SPPC was administered to the children at school. For the Dutch sample, it seems that scholastic competence includes cognitive abilities as well as a behavioral aspects. This outcome may be attributed to both the limited theoretical background for including this factor and the conceptual confusion of the content of the items of this subscale, resulting in an uncertainty of the child as to which norms the child has to evaluate its behavior. These results suggest a reconsideration of the subscale behavioral conduct, for instance by more clearly specifying the situational aspects of behavior such as home- or school-related.

In identifying the domains important for the child's self-esteem, Granleese et al. (8) underlined the necessity of interpreting the intercorrelations of domains for boys and girls separately. Granleese et al. (8) showed that the combined data do not represent the true pattern of self-perceived competence for either sex. The results of the present study also showed a different pattern for boys and girls. Boys perceived physical appearance and scholastic competence as being their domain of highest competence. Girls' sense of self-worth seems to be mainly determined by the 'physical appearance' factor. This scale accounted for 44.9 % of the variance in common in an analysis of correlations between subscales. In addition, girls considered scholastic competence as least important to their overall sense of self-worth. These findings seem to reflect the traditional stereotypes of role-performance of men and women in Western society. In this respect, it is also noticeable that boys had significantly higher scores on all subscales, except for behavioral conduct. Granleese et al. (8) also found a tendency that boys systematically rated themselves as more competent than did girls. However, Harter (7) found gender-related differences in both directions. Boys scored significantly higher than girls on athletic competence, whereas girls obtained significantly higher scores on behavioral conduct. Although the importance of the different domains to the general self-worth are more or less comparable in younger and older children, the contribution of the different domains to general self-worth is different for both groups. Harter (4) hypothesized that along with the child's development, self-descriptions become more differentiated and situation-specific, combined with a reintegration of the components. The results of this investigation support such a developmental trend because the pattern of intercorrelations in younger children suggests that they make separate abstractions in particular domains but that these are not fully integrated into a higher-order abstraction of global self-worth, whereas older children construct their overall perceived competence in a more hierarchical way.

Test-retest reliabilities showed that self-concept is relatively stable over a 4-week period, both in younger and older children. Only on the subscale scholastic competence, all children scored significantly higher during the second administration of the questionnaire. A possible explanation for this finding is that between the first and second administration all children received their first report on academic achievement for the current school-year. Children may tend to evaluate their scholastic capabilities in a careful way if they have no actual information about their school performance. For the total SPPC, the mean 4-week test-retest reliability was .86. Test-retest reliability coefficients of other self-report instruments on self-esteem for children over a period of several weeks typically range between .70 and .90 (2,4). As compared to these self-report measures, the stability of the SPPC is quite respectable. In addition, the stability coefficients obtained in this study give an indication that the adapted question format does not add error variance.

In this study, the SPPC was sensitive to differences in gender but not in age. Comparing the different gender-groups medium effects were found for scholastic competence and athletic performance. The only age difference was found for behavioral conduct. Because behavioral conduct is psychometrically the weakest subscale, it is suggested that in using the SPPC for Dutch children, separate norms need to be established only for each sex but not for the different age-groups.

Results of this study may be regarded as a test of the cross-cultural (American-Dutch) comparability and generalizability of the SPPC. Just as the older version of the SPPC (9) the present instrument can measure self-esteem across cultures. In addition, the SPPC apparently is able to detect cross-cultural variations in children's self-esteem.

In summary, the present study confirmed the factor structure of the SPPC presented by Harter (7) supporting the multi-dimensional nature of the child's sense of competence. Factorial validity was found for four domains of competence: (a) scholastic competence, (b) social acceptance, (c) athletic competence, and, (d) physical appearance. The fifth domain, behavioral conduct, had the weakest psychometric properties. In addition, the child's perceived competence as measured by the SPPC was stable over time, for older as well as for younger children. The next step will be to investigate the criterion-related and construct validities of the SPPC.

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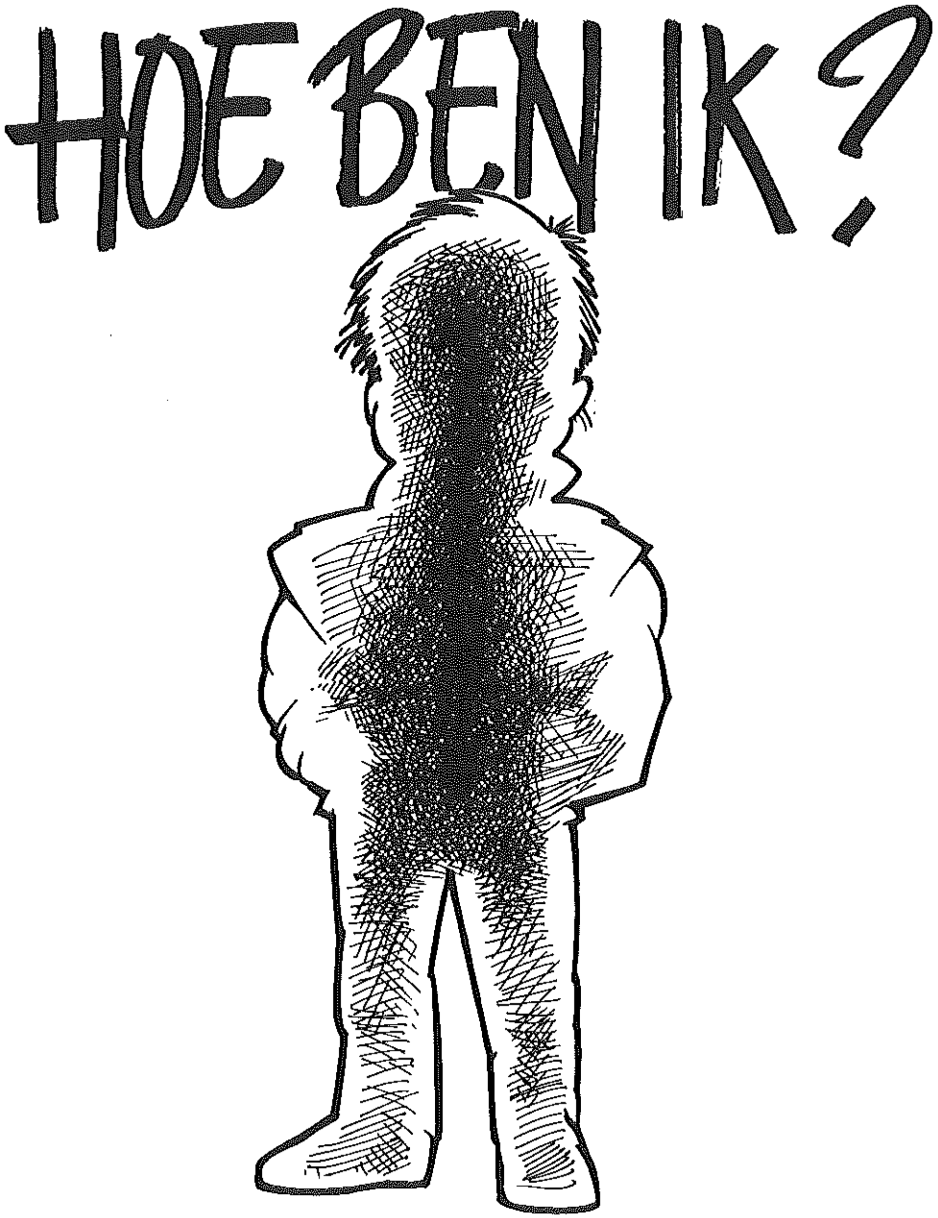
APPENDIX 1:* *Dutch version of the Self Perception Profile for Children.



Sophia Kinderziekenhuis/AZR
Gordelweg 160
3038 GE Rotterdam

Naar "Self-perception profile for children"
van Susan Harter, University of Denver, U.S.A.

APPENDIX 1 (continued):

HOE BEN IK ?

Naam ____________________ Leeftijd _____ Je verjaardag is op ____________ Groep ______
dag maand

Jongen of meisje (omcirkel welke)

Op de volgende bladzijden staan zinnetjes die iets zeggen over hoe kinderen zijn. Het is de bedoeling dat je bij elke regel kijkt **hoe jij bent.** Elke regel bestaat uit twee zinnetjes. Onder elk zinnetje staan twee hokjes, dus per regel staan er vier hokjes. Je moet van die vier hokjes er steeds **één** aankruisen. Zet een kruisje in het hokje dat het meest op jou slaat. Een voorbeeld:

Sommige kinderen spelen liever buiten		**MAAR** Zoals welke kinderen ben **jij** ?	Andere kinderen kijken liever T.V.	
Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen		Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐		☐	☐

Eerst moet je kiezen of je meer van buiten spelen òf van t.v. kijken houdt. Ook al vinden kinderen vaak beide dingen leuk (of beide dingen niet leuk), het is toch de bedoeling dat je kiest welke van deze twee zinnetjes het meest bij je past. Daarna moet je kiezen of jij **net** zoals de kinderen van dat zinnetje bent, òf dat je **een beetje** zoals deze kinderen bent. Kruis dan dat éne hokje aan dat het meest op jou slaat.
Kijk nog eens naar het voorbeeld. Als je vindt dat je liever buiten speelt kun je kiezen uit twee antwoorden, namelijk 'ik ben net zoals deze kinderen' en 'ik ben een beetje zoals deze kinderen'. Als je veel van buiten spelen houdt, kruis je dit hokje aan:

Sommige kinderen spelen liever buiten		**MAAR** Zoals welke kinderen ben **jij** ?	Andere kinderen kijken liever T.V.	
Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen		Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
■	☐		☐	☐

Als je net iets liever buiten speelt dan naar de t.v. kijkt, kruis je dit hokje aan:

Sommige kinderen spelen liever buiten		**MAAR** Zoals welke kinderen ben **jij** ?	Andere kinderen kijken liever T.V.	
Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen		Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	■		☐	☐

Het kan ook zijn dat je liever naar de t.v. kijkt dan dat je buiten speelt. Als dat zo is kun je ook weer kiezen uit twee antwoorden, namelijk 'ik ben net zoals deze kinderen' en 'ik ben een beetje zoals deze kinderen'. Als je veel van t.v. kijken houdt, kruis je dit hokje aan:

Sommige kinderen spelen liever buiten		**MAAR** Zoals welke kinderen ben **jij** ?	Andere kinderen kijken liever T.V.	
Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen		Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐		■	☐

Als je net iets liever t.v. kijkt dan buiten speelt, kruis je dit hokje aan:

Sommige kinderen spelen liever buiten		**MAAR** Zoals welke kinderen ben **jij** ?	Andere kinderen kijken liever T.V.	
Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen		Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐		☐	■

LET EROP DAT JE STEEDS MAAR ÉÉN HOKJE AANKRUIST.
Er zijn geen foute of goede antwoorden. Elk antwoord is goed als het maar je eigen mening is.

APPENDIX 1 (continued):

1. Sommige kinderen vinden dat ze goed kunnen leren **MAAR** Andere kinderen maken zich zorgen of ze hun werk op school wel goed kunnen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

2. Sommige kinderen vinden het moeilijk om vrienden te maken **MAAR** Voor andere kinderen is het nogal gemakkelijk om vrienden te maken

Zoals welke kinderen ben jij?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

3. Sommige kinderen zijn erg goed in sport **MAAR** Andere kinderen vinden dat ze niet zo goed in sport zijn

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

4. Sommige kinderen zijn tevreden over hoe ze eruit zien **MAAR** Andere kinderen zijn niet tevreden over hoe ze eruit zien

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

5. Sommige kinderen zijn niet tevreden over hun gedrag **MAAR** Andere kinderen zijn wel tevreden over hun gedrag

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

6. Sommige kinderen voelen zich vaak ongelukkig met zichzelf **MAAR** Andere kinderen voelen zich nogal gelukkig met zichzelf

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

APPENDIX 1 (continued):

7. Sommige kinderen vinden dat ze net zo slim zijn als andere kinderen uit hun klas **MAAR** Andere kinderen zijn daar niet zo zeker van

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

8. Sommige kinderen hebben veel vrienden **MAAR** Andere kinderen hebben niet zoveel vrienden

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

9. Sommige kinderen zouden willen dat ze beter in sport waren **MAAR** Andere kinderen vinden dat ze goed genoeg in sport zijn

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

10 Sommige kinderen zijn tevreden over hun lengte en gewicht **MAAR** Andere kinderen willen wel dat hun lengte of gewicht anders zou zijn

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

11 Sommige kinderen doen meestal precies de dingen goed **MAAR** Andere kinderen denken vaak achteraf: had ik het maar anders gedaan

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

12 Sommige kinderen vinden hun leven niet prettig **MAAR** Andere kinderen vinden hun leven wel prettig

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

APPENDIX 1 (continued):

13 Sommige kinderen doen nogal lang over hun werk op school **MAAR** Andere kinderen hebben hun werk snel af

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

14 Sommige kinderen zouden graag heel wat meer vrienden hebben **MAAR** Andere kinderen hebben zoveel vrienden als ze maar willen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

15 Sommige kinderen denken dat ze ieder nieuw spel buiten meteen kunnen **MAAR** Andere kinderen zijn bang dat ze niet mee kunnen spelen met elk nieuw spel buiten

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

16 Sommige kinderen zouden willen dat hun lichaam anders was **MAAR** Andere kinderen zijn tevreden met hun lichaam

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

17 Sommige kinderen gedragen zich zoals hun ouders van hen verwachten **MAAR** Andere kinderen gedragen zich anders dan van hen verwacht wordt

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

18 Sommige kinderen zijn tevreden met zichzelf **MAAR** Andere kinderen zijn vaak niet tevreden met zichzelf

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

APPENDIX 1 (continued):

19 Sommige kinderen vergeten vaak wat zij hebben geleerd **MAAR** Andere kinderen kunnen gemakkelijk onthouden wat ze hebben geleerd

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

20 Sommige kinderen doen graag dingen samen met andere kinderen **MAAR** Andere kinderen doen graag de dingen in hun eentje

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

21 Sommige kinderen vinden dat ze beter kunnen sporten dan andere kinderen van hun leeftijd **MAAR** Andere kinderen vinden dat ze niet zo goed kunnen sporten als anderen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

22 Sommige kinderen zouden graag hun uiterlijk (hoe ze eruit zien) anders willen **MAAR** Andere kinderen vinden hun uiterlijk (hoe ze eruit zien) prima

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

23 Sommige kinderen raken door de dingen die ze doen vaak in de problemen **MAAR** Andere kinderen raken meestal niet in de problemen door de dingen die ze doen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

24 Sommige kinderen vinden het goed zoals ze zijn **MAAR** Andere kinderen zouden vaak zoals een ander willen zijn

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

APPENDIX 1 (continued):

25 Sommige kinderen zijn erg goed in de klas **MAAR** Andere kinderen zijn niet zo goed in de klas

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

26 Sommige kinderen zouden wel willen dat meer kinderen van hun leeftijd hen aardig vinden **MAAR** Andere kinderen vinden dat de meeste kinderen van hun leeftijd hen wel aardig vinden

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

27 Sommige kinderen kijken liever toe met sport en spel in plaats van mee te doen **MAAR** Andere kinderen spelen liever mee dan toe te kijken

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

28 Sommige kinderen zouden wel willen dat er iets aan hun gezicht of hun haar anders zou zijn **MAAR** Andere kinderen vinden hun gezicht of hun haar goed zoals het is

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

29 Sommige kinderen doen dingen waarvan ze weten dat ze die eigenlijk niet moeten doen **MAAR** Andere kinderen doen bijna nooit zulke dingen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

30 Sommige kinderen vinden het prima zoals ze zijn **MAAR** Andere kinderen zouden graag wel wat anders willen zijn

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

APPENDIX 1 (continued):

31 Sommige kinderen hebben moeite om op school de goede antwoorden te bedenken **MAAR** Andere kinderen weten bijna altijd het goede antwoord

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

32 Sommige kinderen worden door alle kinderen van hun leeftijd aardig gevonden **MAAR** Andere kinderen worden niet door iedereen even even aardig gevonden

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

33 Sommige kinderen zijn niet zo goed in nieuwe spelletjes buiten **MAAR** Andere kinderen zijn in die spelletjes meteen goed

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

34 Sommige kinderen vinden dat ze er goed uitzien **MAAR** Andere kinderen vinden dat niet zo

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

35 Sommige kinderen gedragen zich keurig **MAAR** Andere kinderen vinden het vaak moeilijk om zich netjes te gedragen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

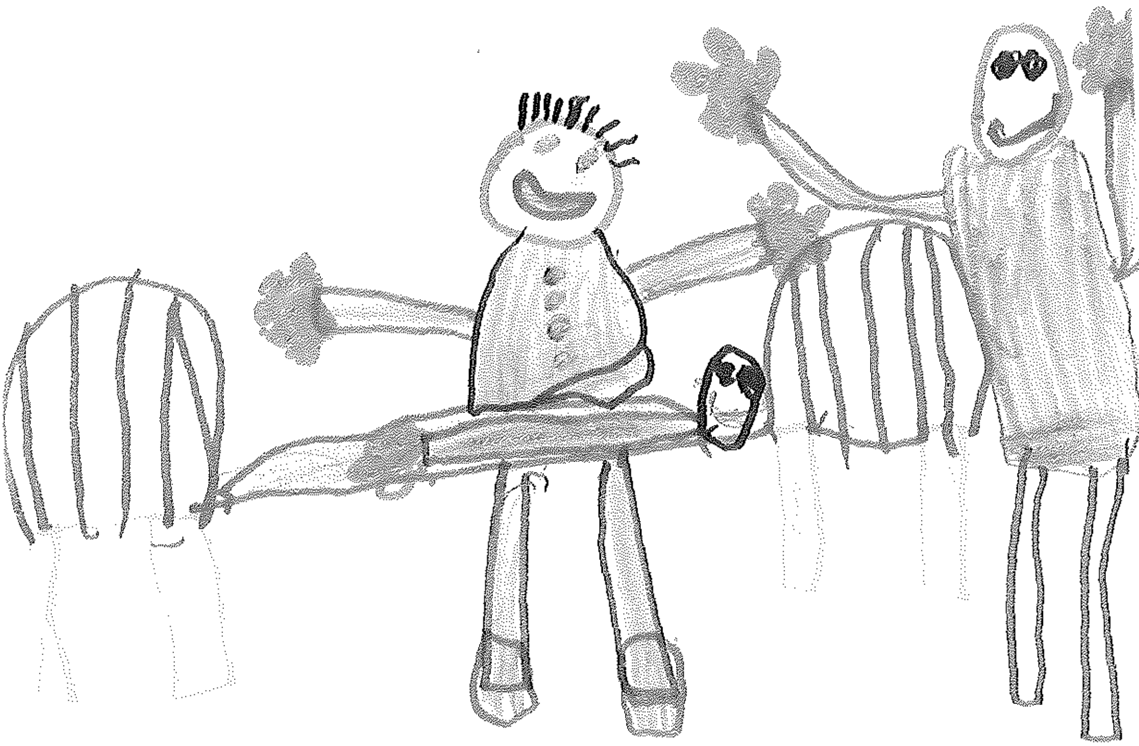
36 Sommige kinderen zijn niet erg tevreden over hoe ze veel dingen doen **MAAR** Andere kinderen denken dat ze de meeste dingen goed doen

Zoals welke kinderen ben jij ?

Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen	Ik ben net zoals deze kinderen	Ik ben een beetje zoals deze kinderen
☐	☐	☐	☐

Chapter

3



Bas, 6 yrs (sibling of a childhood cancer survivor)

Chapter 3 : Surviving childhood cancer: Impact on parents

3.1 Experiences of parents of childhood cancer survivors: A qualitative analysis
Submitted for publication. 147

3.2 Late consequences for parents of children who survived cancer
Published in: J Pediatr Psychol 1995;20:567-586. 169

3.3 Impact of childhood leukemia on family planning
Published in: Pediatr Hematol Oncol 1995;12:117-127. 189

CHAPTER 3.1

EXPERIENCES OF PARENTS OF CHILDHOOD CANCER SURVIVORS: A QUALITATIVE ANALYSIS*

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ABSTRACT

Parents whose child survived cancer were in-depth interviewed about the late consequences of the disease. Variational sampling was used to maximize the contrast in parental experience and in coping with the disease. The qualitative analysis resulted in a description of a whole range of variations in both perceived impact and the way parents cope. Parents experienced changes of a definitive and long-lasting nature as a result of cancer in their child. In spite of the child's survival, perseveration of problems and feelings of loss prevailed. Parents who used coping strategies in a non-extreme way appeared to be function well.

INTRODUCTION

During the last few decades the prognosis of childhood cancer has drastically improved. With lengthening survival, the question arises what the consequences are for the child and his parents after treatment has been terminated. Studies on the long-term consequences have mainly focused on the physical and psychosocial sequelae for cancer survivors, only a few studies have focused on the sequelae for parents. Some of these studies reported no special problems (1,2) or an improvement in adjustment to the situation over time (3,4), whereas others reported that long after the termination of therapy, parental anxiety about a recurrence of the disease remains a major issue (5-7). These discrepancies can be attributed to the different and limited areas of psychosocial functioning that have been addressed in each of these studies. Because of the sparse knowledge on the long-term consequences for parents, insight is needed into what it is

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* *Submitted for publication.*

like for them to live through childhood cancer. Instead of focussing on a particular issue, we wanted to describe their situation in all its aspects.

For this purpose we used a qualitative research strategy. A qualitative approach matches well with the specific character of the problem under study: *the complexity of human experience.* It enables the investigator to analyze the situation of parents of cancer survivors in all its complexity and richness thereby using the subject's own frame of reference (8-10). Another argument for using a qualitative study approach concerns the newness of the field. In a new area of research, the first step is to gain an understanding about what happens to parents of cancer survivors. A detailed and systematic description is needed to have knowledge which variables to measure, and in which combinations, before transforming the experiences of parents into quantitative response formats. To rush over this basic task is the source of most *validity errors* in field research (11).

The aim of our study was to describe the experiences of parents of a child surviving cancer and their way of coping with it. We analyzed in-depth interviews with parents of children who survived cancer in a qualitative way. We combined strategies from the existential-phenomenological tradition to reduce the data and transform them into meaning units (12,13) with the method of constant comparison developed by Glaser and Strauss (14,15).

MATERIALS AND METHODS

Data collection

Subjects and procedure

Parents of 87 8- to 12-year old children who had been successfully treated for a malignancy were approached. These children represented all eligible Dutch patients with leukemia or non-Hodgkin lymphoma ($n=75$) treated during 1976 and 1988 in the Sophia Children's Hospital/University Hospital Rotterdam, one of the four paediatric cancer centers in The Netherlands. Parents of twelve children with a solid tumor were included as well, consecutively selected to the same criteria. Two families (2%) refused because they feared a disturbance in mental balance. In total 85 families participated. Time off treat-ment ranged from 6 months to 7 years and 9 months (median: 4 years and 2 months).

Initial contact for the study was established during regular follow-up appointments at the outpatient clinic of the Department of Hematology/Oncology. All parents were interviewed at home by the first author during a period of 3 years. Each interview was audiotaped. In addition, observations and reflective remarks were made directly after each interview.

Interviews

The original material consisted of 85 audiotaped interviews with one (*n*=8; 9%) or both (*n*=77; 91%) parents present. The duration of the interviews ranged from 2.5 to 4.5 hours (comprising of 73 to 126 pages of written text). The interviews were conducted in an open, Rogerian-like manner, giving the parents the freedom to discuss their own areas of importance in their own way. In the opening question, parents were asked to go back in time till the moment when they first became aware of physical symptoms and to recall their experiences with the illness of their child up to the present day. The recall of experiences may have been influenced by the lapse of time or interpreted from the perspective of the child's survival. Although this retrospective information might not be a genuine reflection of past experiences, they reflect the current view of parents on their experiences with their child's illness and thus can be used to portray current experiences.

Another issue was whether the collected data represented a faithful reflection of the phenomenon under study. In the literature, the following indicators of the validity of the qualitative interview data have been mentioned: a high involvement of the interviewees, the freshness of the material, and the emergence of new explorations during the interview (16,17). In our study a high involvement was revealed by the ceaseless stream of words and the length of the interview. The material collected impressed as dramatic. Parents reported that they were emotionally involved and experienced psychological or physical symptoms of tension related to the interview. Experiences were presented in an unpolished way and many parents disclosed that it was the first time that they talked about the complete period in such an elaborate way. Parents conveyed that they had learned new things from the other parent's story. In addition, a number of parents spontaneously reported beneficial effects for themselves as a result of the interview. These observations can be considered a as positive indication for the validity of the data.

Data analysis

Selection of cases

The aim of our study was to analyze the entire situation of parents and not to focus on a restricted number of aspects. Therefore we proceeded in variational sampling, defined by Strauss and Corbin (18) as a procedure to purposefully find in the data as many differences as possible at the dimensional level. This implies that those families will be chosen that maximize the opportunity to elicit data regarding variations along the dimensions of interest. Consequently, our results will give a picture of the most salient and (extreme) variations of the impact childhood cancer may have on parents' life and the way they cope with it. With this selection procedure the representativeness of the concepts in their varying forms was maximized. However, it should be kept in mind that these results do not reflect their actual empirical frequency in the total sample in a quantitative way. A prerequisite for variational sampling is that families are being selected on the basis of foreknowledge of contrasts within the whole pool of interviews (18). Because of our acquaintance with the material and with the range of the most salient dimensions, we were able to select eight contrasting 'cases' for further comparative analysis (families A to H).

Main dimensions of contrast

The selection of cases was guided by the method of constant comparison developed by Glaser and Strauss (14). This method implies that, by comparing families, a number of dimensions emerge on which these families show differences and similarities. From these contrasts and similarities, new questions and hypotheses were formulated leading to a selection of consecutive families who would contrast on the dimension focused on. In this way there was a constant interaction between analyzing and the selection of subsequent families.

The first and most pronounced dimension of contrast that emerged was *emotional expression*. This concerns the degree of openness the parents displayed in dealing with their feelings and emotions. On this continuum families A and B could be placed at either extreme, where family A is an example of an open way of dealing with feelings and family B is an illustration of the opposite. During comparative analysis also similarities between these families appeared of which *attributional style* was the main one, i.e. whether the cause of their problems was attributed inside or outside themselves. This dimension was chosen as the next one. Whereas families A and B displayed an attributional style linked to an internal orientation of control, family C struck as a family with an external attributional style. Focus on the *perspective from which the experiences were told* was the following contrasting dimension. Family D was selected because, in contrast to families A, B, and C, the experiences of the child were the main issue during the interview.

Family E was selected as an example of a *well-balanced* and harmonious unit. During analysis it appeared that this family adopted a position in the middle on all of three above mentioned dimensions of openness of emotion, attributional style, and focus on child versus parental experiences. The other families were selected to elaborate on specific themes such as *socioeconomic status* (family F), *psychopathology* in one of the parents at the time of interview (family G), and the continuing *discrepancy in experience and coping between partners* up till the moment of the interview (family H). The demographic and disease-related characteristics of these eight families are summarized in Table 1. We did not included siblings in our final analysis because the interview text on siblings was generally very limited.

Analysis of the interview material and data reduction

The first two authors independently listened to the audiotapes of the eight interviews,

TABLE 1: Demographic and disease-related variables of eight families at the time of the interview.

Family	A	B	C	D	E	F	G	H
Demographic characteristics								
-Age[a]: father	36 y	34 y	34 y	34 y	37 y	34 y	45 y	45y
mother	32 y	32 y	37 y	35 y	37 y	37 y	34 y	34 y
-SES[b]	3	4	5	2	2	6	4	2
-Marital status[c]	1	1	1	1	1	1	1	1
-Age[a] and sex survivor	9y M	13y M	9y M	9y F	8y F	8y F	11y M	9y F
-Age[a] and sex sibling	-	11y F	7y M 0y M	11y F 4y M	5y F	0y M	14y F	-
Disease-related characteristics								
-Diagnosis	ALL	ALL	ALL	ALL	ALL	ALL	ALL	ALL
-% survival at diagnosis	35	35	70	50	70	70	50	70
-Age child at diagnosis	2y	2y	3y	3y	3y	4y	4y	1y
-Length of treatment	2y	4y	2y	2y	2y	2y	2y	2y
-Intensity of treatment[d]	1	3	2	1	2	2	1	2
-Relapse	no	yes	no	no	no	no	no	no
-Time off treatment	3y	5y	5y	4y	3y	2y	5y	5y
-Late physical sequelae	no	yes major	no	no	no	no	yes minor	no
-School hinderance[e]	2	0	1	3	0	0	2	0

Abbreviations: M=male; F=female; ALL=acute lymphoblastic leukemia.

[a] age at the time of the study. [b] SES (socioeconomic status) rated according to Dutch standards (19) on a 6-step scale (6=highest status occupation), [c] marital status: 1=married (first marriage of the parents). ; [d] intensity of treatment course: 1=mild, 2=intermediate, 3=severe; [e] school hinderance: 0=no problems at elementary school, 1=minor learning problems at elementary school, 2=special education for children with learning problems, 3= special education for mentally retarded children.

with an 'open' mind. Observations and notes were made with reference to both the content of the interview and to other verbal and nonverbal aspects. Also field notes of the interview and home visit were used. The complete verbal transcript of the eight interviews was read. Several operations were carried out to achieve a psychological comprehension of the material such as imaginative variation, utilization of an existential baseline, and grasping implicit meanings as described by Giorgi (9,12) and Wertz (13,16,20). An extensive description of each family was made, illustrated with many quotations, followed by a condensed description of the main themes emerging from the material. Finally, a cross-case analysis of the processed material was performed as described by Miles and Huberman (15). Cases were ordered and scaled into high, medium, and low, according to the variable being examined. Then the data was combined with similar data on other dimensions of interest to get insight into the clustering of cases (patterns) and to try out hypotheses.

Reducing the data through transformation involves the risk of discarding relevant information or missing information crucial in understanding the phenomenon. This risk of distortions was decreased by having two psychologists perform these operations independently and by intensive discussion and reflection on these operations afterwards. Carrying out the analysis and interpretation by independent persons will bring to light bias of each separate investigator, enhancing the reliability and validity of the analyzing process (20). Another aspect of validity concerns the interpretation of the data. The standard objection to the results of an interview study is the subjectivity of its interpretations (21). We acknowledge that other researchers might come to different interpretations by reading our interview data. The decisive criterion in this kind of research is "whether a reader who adopts the same viewpoint as articulated by the researcher can also see what the researcher saw, whether or not he agrees with it" (21: p. 29). Wertz (20) states that all psychological statements must be based on positive exemplification. Consequently, our interpretations will be illustrated with literal quotations from the interviews.

RESULTS

Results emerging from the comparative analysis of the selected families are grouped into two sections. First, the impact on the parents' present way of life is described, and secondly, the way they report upon coping with the disease and its aftereffects is summarized.

Impact of childhood cancer after treatment ends

Parents indicated that cancer in their child was the most overwhelming experience in their life. By comparing their life before and after the treatment period, parents reported a number of significant changes. These changes were not incidental, but were of a definitive, long-lasting nature. The far majority was labelled as negative by the parents. The negative changes reported can be categorized into *losses* and *perseveration of anxiety and problems*. We found few positive changes and these were reported in only a few families. A third category of secondary gains was added to describe consequences of the illness which may be secondarily reinforcing to parents. These gains were least frequently observed.

NEGATIVE CHANGES

Losses

The first main category of losses concerned *losses in the area of outlook on life.* The illness of the child was a severe shock. The outlook on life was drastically changed and this was expressed in a number of ways:

> Father A: "When your child is in danger of dying, it's really driving you crazy, believe me, you go off your head, it's the end of your world, you are pulling down your own house."
> Father G: "I really enjoy this world, but that one little thing in my own image, a little family that should run smoothly, that should be fine, that must be harmony, and that is upset. How can I make that clear to you? It's not what it used to be, it will never be like that, it has changed, those days are gone and will never come back."
> Mother B: "We cannot be light-hearted any longer, I cannot laugh anymore, I've forgotten how to laugh."
> Mother H: "I've never had anything to worry, had a very happy youth, I mean, never had any trouble and then, out of the blue: wham, you are struck by such a thing, that is a real shock, in the beginning you have hard feelings, all your spontaneity is gone. You will never be your old self, your one-time spontaneity, you will never have it back."

As a result of this existential shock parents were confronted with the loss of their usual way of life: they described that they could never be the same relaxed person again, that they had lost the experience of joyfulness, and felt a disappointment in life. Parents also reported that they felt more vulnerable for traumatic events. When confronted with new stresses they reported to be more easily disturbed in their psychological equilibrium and less able to cope with these stresses. In other words, they experienced a *loss of invulnerability*:

> Mother F: "But then, just like three months ago when something happens, then you find that, unconsciously, you have still all kind of little worries" Father F: "that you are vulnerable" Mother F: "that you are very vulnerable indeed, when only the slightest thing happens, seemingly, it evokes such clear association that you are again completely.." Father F: "Actually, you say, you cannot stand that too many things go wrong or you are thrown back to a lower level (of functioning)" Mother F: "yes exactly so, everything must be running smoothly with us, or otherwise..."

Among the practical burdens of treatment, parents had to spend a large amount of time and energy on the care for their child. Time they could have spent differently if their child had not been ill. This feeling was interpreted as a *loss of a part of their life-time*:

> Father C: "That is a period you keep going along, and you have to, but it certainly costs a few years of our life" Mother C:"and we are still making up for those lost years."

A second main category of losses concerned the *marital relationship*. When a child falls ill, parents might expect that they go through this ordeal together and share the same feelings. But, although it was their mutual child who was seriously ill, partners might experience the illness and cope with it in different ways. The illness set limits to the expectation and experience of togetherness in the relationship, which created loneliness within the marriage and sometimes great martial frictions:

> Mother A: "The big difference between you and me was that for a long time you kept looking at things very negatively" Father A: "For me the child was dead already...I found her an iceberg, I couldn't understand it all." Mother A: "And I had no sympathy for people who can let themselves go in that way."...Father A: "At those moments that I reacted quite differently than you did, I was so awfully conscious of being alone. You really want to be together, but is doesn't work..you are super-lonely."

Although parents delineated changes in their marital functioning during treatment, in general these did not seem to persist into their present relationship. Stress in marital functioning as a negative *late* effect was only reported in family G.

Parents of long-term cancer survivors might also be confronted with a third category of losses, namely *losses with respect to the child* as a result of the late physical and/or psychological after-effects:

> Mother G of a child with medical after-effects: "We've been given back a quite different child."
> Father F: "In D. a number of things in his development have been speeded up. We've said from the beginning: he will be getting older earlier, he'll skip his youth partly, and I suppose that has happened" Mother F: "and that's a pity, isn't it, that's bad, that's too bad."

Particularly when these changes were of a permanent, irreversible nature e.g. tooth damage, sterility, and neuropsychological deficits, parents had to live with the fact that the child was cured of cancer, but was not the same healthy child as before. In cases of severe neuropsychological after effects, parents were faced with a child who is healthy with respect to the malignancy, but at the same time is mentally and/or physically handicapped. They had to live with the fact that their child is disabled. Consequently, there can be *loss of the image and reality of a healthy child.*

Perseveration

A second major negative change experienced by parents is the perseveration of uncertainty and anxiety about a recurrence of the disease. In all families this theme emerged as being one of the most important late effects:

> Father A: "There's still the anxiety, you know."
> Father B: "Every cold, sneezing twice, and you blow up."
> Mother D: "Such a child, you are really more anxious about her than about anything else, because you are in fact happy that she is still with you, you know, and this concern stays with you, it certainly remains in the back of you mind."

The frequency and intensity of the occurrence of this theme indicated that even long after the termination of therapy parents were uncertain and anxious about the possibility of recurrence. Parents were not only anxious about relapse but also about the appearance of physical after effects in their child:

> Mother C: "Something happens all the time, and then you think..Now they've begun to keep these growth curves, because these children appear to be behind in growth after all, they have found that out recently, what will crop up next year, you are bearing in mind that something might happen, all these problems coming on top, that doesn't stop when treatment is completed."

In addition to the continuation of uncertainty, there might also be a perseveration of problems. If there were medical side effects, the seriousness and the visibility of these long-term effects made them a constant reminder of the disease. In this way, parents were confronted with the fact that the cancer experience cannot be restored and left behind. Even 'minor' side effects could function as a reminder:

> Mother H: "Because she had to take that medicine, it caught the enamel of her teeth. I didn't like that, you know, the child was cured completely and now there's still something permanent."

Furthermore, extra care from and demands on the parents as a result of the psychological and physical sequelae in the child continued:

> Father G: "Yes, we are often thinking about it, how to deal with J., how to handle him, how to stimulate him, how to go about his reading and writing and arithmetics. Actually, J. is the center of the family all the time, without the rest noticing."

This extra care put a variety of extra demands on the parents. In addition to their own efforts to help the child at home, there might be the necessity for specialized care such as medical treatment, physiotherapy, remedial teaching, and psychosocial support.

As a consequence of the perseveration of uncertainty, anxiety, and problems, parents tended to perceive their child as a *vulnerable child*; vulnerable with respect to both its physical condition and its psychosocial development. They felt that the child, in comparison to siblings and peers, was disadvantaged as a result of the various medical interventions he or she had gone through. The lively and often emotional way in which they recalled the child's events gave an indication of the persistent and ingrained impact of these events on the parents:

> Father A: "That child has known no youth."
> Father F: "Just imagine, he is four...and then suddenly bang!!!, all at once admitted to hospital, jabbed in the back without any preparation or warning, put in bed, you have a drip, you are tied, you have stuff you get sick from...everybody besieges you..."

POSITIVE CHANGES

Incidentally, parents reported positive changes. Some of these concerned their overall *life perspective*, a restructuring of their view on themselves and on the world. They felt more able to put the impact of other events in a proper perspective. Parents also mentioned that they focused on the day-by-day living circumstances instead of looking and planning for the future because they had experienced that suddenly life could take a different turn. They felt a need to live happy now rather than to postpone happiness. This reorientation on life and consequently a shift in standards was illustrated by:

> Mother F: "When you see how easily life may -whoops- go down the drain, I tell you, then it occurs deeply to you how important it is that you live, that you are together and that all is fine, the carpe-diem feeling has got everything to do with it, you live more consciously, you know better how to put things in perspective, when people are on about little things."

Some parents reported personal growth. They were proud of having survived this difficult period psychologically and socially. They felt more able to rely on themselves, which enhanced their feelings of self-esteem.

Positive changes also concerned the *marital relationship*. One family (family A) reported a positive effect here. This effect was strong and it was experienced by both partners. They felt a deeper understanding towards each other and were able to discuss their frictions in a more constructive way, which also could be observed during the interview. Another family (family F) reported a greater sense of closeness within the nuclear family, thereby including the marital relationship.

Finally, positive changes constituting of a *more gratifying relationship with the child* and a change in attitude were found (father A; mother F):

> Father A: "I used to see the child as adding to my own glory. That is different now, I've changed my mind, I mean, the child has gone through all this, and I'm thinking in the way of: what can I do for you that later you won't suffer more than necessary. I'm looking after his interest instead of my own."

SECONDARY GAINS

Very occasionally, gains of negative consequences of the illness for parents were observed, be it indirectly. In one of these families the distress inherent in the disease was cultivated in order to prevent a dysfunctioning of the marital couple. The only emotion they expressed was anger which was especially directed towards the doctors, treatment, and hospital services. For this family, it was important that the illness remained in order to preserve this anger. Parents focussed on detecting any possible long-term consequences in the child which seemed sometimes more of a self-fulfilling prophecy than that they were based on medical facts. By creating a common 'enemy' outside the family, the couple was drawn together:

> Mother C: "When we are not fighting against something, or when there is nothing to fight against, we are fighting with each other."

TABLE 2. Variations in manifestation of pathology

		Psychopathology during treatment	
		No	Yes
Psychopathology after completion of treatment	- No	mother A, father H, families B, C, D, E, F	father A mother H
	- Yes	father G	mother G

Coping

The interviews showed that parents of children with cancer experienced considerable emotional distress and serious problems. However, they put their child's interest first, forcing themselves to keep going and to fulfil their responsibilities and obligations. This finding explains the discrepant results in the literature about parental functioning, supporting the notion that on the emotional level there is serious disturbance, whereas on the level of daily life parents make an all-out effort to function well (22). However, sometimes daily functioning broke down. The different patterns, shown in Table 2, illustrate that not only during treatment serious problems arose, but also after completion of treatment, as a late psychological effect. This is in line with reported late reactions to other stressful life events (23).

Parents used a mixture of coping strategies to survive emotionally the stresses inherent in the disease. From the qualitative analysis, two dimensions relevant to coping processes emerged. The first dimension concerned the confrontation versus avoidance of the stress related to childhood cancer to regulate its emotional impact. Specific strategies were observed by which parents tried to dose the threat to the level they were able to face it. Those elaborated here are the position parents adopt on the survival-death perspective, a strategy distinctive for childhood cancer, and the ability to express one's emotions. The second dimension concerned the attribution of control, the extent to which parents regained control over the situation and were able to remove the stressor.

CONFRONTATION-AVOIDANCE STRATEGIES

Survival - death perspective

From diagnosis onward, one of the dilemmas the parents were confronted with is whether to prepare for death or for survival of the child. Parents ranged on a continuum from no hope (father A), to some hope (mother C), and to extreme hope and denial of the possibility of death (father H):

> Father A: "For me the child was dead already...you are looking after a child you don't have."
> Mother C: "You don't consider it (death) but it's always in the back of your mind."
> Father H: "I've never expected that things would go wrong, never."

Although in the interviews this theme was not often openly discussed, its undercurrent was noticeably present and it became prominent when there was a difference in perspective between parents (families A, G, and H). Furthermore, the perspective might change from diagnosis onward to the present time. Understandably, more parents adopted the survival perspective after the successful termination of therapy than during the disease. However, also denial of an initial experienced death-perspective might have played a role.

Remarkably, directly after the diagnosis, more mothers (mothers A,B,E,F) than fathers (father H) seemed to have taken an intense and strong survival perspective:

> Mother F (reaction on hearing the diagnosis): "The moment this doctor told us..then pop..you suddenly turn the switch around: okay then, so now I know, and now, now we are going to fight..yes it is really true, but we won't allow things to get out of hand, you see, we are just going to handle it all down to the minutest detail so that the child won't die."

Although this sex difference might be a matter of chance, it may be speculated that by having given birth to the child, it is the mother who instinctively feels obliged to show hope. If she would adopt the death perspective or show any doubts about the child's prognosis, she might feel disloyal towards her child. When even *she* does not demonstrate belief in the child's future, who else is going to fight for the child?

At the moment of interviewing, more mothers than fathers were anxious about a recurrence of the disease in spite of their initial survival perspective and more fathers claim that, for them, the illness was over. In particular, fathers, who reported to have *changed* from a death perspective during the disease towards a survival perspective, experienced less anxiety over a recurrence of the disease. Our hypothesis is that the ability to confront oneself early with the worst outcome of the disease, i.e. the possibility of death, has a positive effect on late psychological sequelae in parents.

Expression of emotions

The degree of openness of communication about personal feelings was originally selected as our first dimension of contrast and was therefore most prominent in differentiating between families. Some parents hid their emotions for showing them could be painful and involved risks with respect to one's self-esteem and feelings of control:

> Mother A: "I tell you, I find it awfully gruesome (how you are confronted with yourself by the illness). It scares me to death and I'm glad I have a firm grip on my emotions."
> Mother G: "but I hide everything." Father G: "You are afraid of yourself" Mother G: "Yes, but when I will start screaming and let myself go, then I may go bananas."

Parents developed different strategies to avoid becoming overwhelmed by these personal issues:

> Father A: "How the little one (i.e. the patient) is doing, that is safe, you know it all, then it's you who decide what you are going to talk about, and so you keep out of it yourself."

A variation on this strategy of talking about the child as a way to bypass personal concerns (family D) was that parents did not talk at all about feelings (family B, father H) or talked about their feelings in an indirect way (family C). Other variations were talking about their own feelings in a direct way (families A, G, and mother H), and talking about both the feelings of the child and themselves (families E and F). All these variations represented different positions on the confrontation-avoidance dimension. While the former variations have avoidance characteristics, the latter two strategies have predominantly confrontation characteristics.

Open communication is considered as a positive mode of coping associated with good adjustment (22). In our study, however, no direct positive relation between open communication and good adjustment was observed. Families with the highest level of open communication (families A and G) did not have the highest level of overall adjustment. Families with closed channels of communication about emotions (family B and father H) presented a picture of composed day-to-day functioning, although they were remote from dealing with their emotions. Those who adopted a position towards the middle on the open-closed dimension (families E and F) seemed able to function well emotionally and socially.

ATTRIBUTION OF CONTROL

Childhood cancer is associated with loss of control, defined as the inability to manage or to influence events (24). In this study the attribution of control played an important role in the overall well-being of the parents and in their attitude towards the illness experience. Parents with an extreme internal (father A and mother D) or external control (family C) experienced more difficulties with the disease as a late effect. Parents with extreme internal orientation might be so overcome with a sense of personal responsibility for the occurrence of the disease that they may suffer from anxiety, guilt, and depressive reactions. This was most prominently present in father A. Parents with extreme external control continued to blame their environment, which resulted in feelings of anger and resentment (family C). Parents who appraised their degree of control in a balanced way (families B,E, and F) seemed more accepting and experienced less guilt. In fact, they appraised the situation in a realistic way because there are only limited possibilities to exert direct control. This observation is in line with the hypothesized curvilinear relationship between locus of control orientation and experienced stress, implying that subjects with extreme internal or external control may be especially vulnerable to life stress (25,26). Two areas in which attributional style plays an important role will be discussed: procreative and parenting behavior.

Procreative behavior

We found that variations in progeny can be viewed as strategies to cope with or to repair emotional harm and to regain control over the situation. Some variations occurred in the way parents decided about their future offspring. One variation was that parents felt no change at all in family planning (family C). Some parents indicated that for practical reasons it was impossible to have a baby during the treatment period (families B and F). A third variation was that parents decided to have no more children (family A, mother H):

> Mother A: "I've been thinking about it: one or two, well and he was ill and then I knew it already, so final, isn't it? Absolutely no more children at all, I will never do it again, I found it (cancer and its treatment) so terrible, I don't want it to happen again, not only for me but not of the child either: what hard times he's been going through!"

Family A felt so overwhelmed by the suffering of their child, that they had indirectly caused by giving birth him, that they made the irreversible decision for sterilization. In family H, the mother did not want a second child because she was afraid that she would not be able to care for this child if her daughter might relapse. In addition, she was afraid of not being able to develop the same intense relation with a new baby. All motives to refrain or delay family planning involved elements of avoiding future stress and misfortune.

Still another variation was presented by family B who had a new baby 6 years after completion of treatment. This child differed 11 years in age with the older sibling. After the distress inherent in the disease, this new baby symbolized a positive experience for the whole family. The new child may also be regarded as a psychological replacement child in order to compensate for the losses (a.o. missing a period of the life of their other child(ren)) caused by the illness.

Parenting behavior

Most parents regarded the former patient as a vulnerable child. Consequently, they were extra alert to the child's emotional and physical well-being. Their sense of duty and obligation was increased to overcome the disadvantaged situation of the survivor. These parental strivings were directed to achieve 'normalcy' for the 'non-normal' child, which can be regarded as *the work of undoing*. The extra care of parents concerned both the physical and the psychological well-being of their child. On the physical level, this care took shape in restricting the child's daily activities to prevent possible harm:

> Mother D: "You are more concerned about such a child, about what she is doing. Climbing up a tree for instance, we won't let her do it."

On the psychological level, parents were very concerned about the emotional effects of the illness period on the child. They tried to compensate for the many repugnant events during treatment, and protected their child from more (potentially) unhappy or stressful events. Consequently, parents overindulged the child in material and psychological respect. The latter might result in lowering demands. For example, parents were very reserved in demanding scholastic performance, subordinated their expectations to the happiness of the child, tried not to upset the child unnecessarily, and reduced the number of conflicts by setting less demands on the child. Setting limits on the child turned out to be extremely difficult for parents:

> Father F: "You feel terribly guilty of course when you are just severe on him."
> Mother F: "Guilty because he feels so miserable already and he has to suffer so much and then he gets it in the neck on top."

Thus, on the physical level the demands were constrained, whereas on the psychological level they became more lax. In normal development this situation is the reverse: physical limitations are reduced with increasing age, whereas psychological demands are growing.

The issue of control plays an important role in parenting. It may be hypothesized that parents who attribute the physical and psychological rehabilitation of their child to a large extent to their own efforts (internal locus of control, e.g. father A, mother D), are more persistent in their attempts to support the child and experience more concerns and worries. A salient observation was that, in response to the request whether the child could participate in the study, these parents displayed protective behavior. Father A insisted to be present at the interview in case of emotional upheaval in the child. In family D, parents subdued to the child's wish to play with her friends instead of participating in the study.

Consequences of coping for the other partner

We found that coping behavior used for the emotional adaptation of a parent had implications for the coping behavior of the other parent. One variation was that parents facilitated in each other those strategies that were in line with their own way of coping (families B and C). In other words, they set limits to the coping behaviors of others that jeopardized their own equilibrium. For instance, if a parent suppressed his or her own emotions, he/she might find ways to prevent others from expressing their feelings too. Thus, emotional control was attained by controlling the emotions of other family members as well.

A second variation we found was that partners developed a differentiation in their response to stress. In most families, there was a subtle division in roles, thereby adapting the burden of the illness to the strength or resilience of the other partner. This division of roles was often seen at events that were perceived as extremely stressful by parents, such as lumbar punctions and bone marrow aspirations. Parents often made arrangements about who had to undergo the burden of being present at these procedures. In families A and G this differentiation in roles was most prominent. Because one partner was not able to maintain emotional balance, the other partner was responsible for the family's day-to-day functioning:

> Mother A: "There is only one who is allowed to break down. I've very often had the feeling that I couldn't, because that must not happen. Imagine me doing it, then the whole lot will fall down."

The emotional strength of one partner seemed inversely related to that of the other partner. In family G even an exchange of roles was observed. When after completion of treatment the father in family G became very depressed, the mother grew stronger and was able to care for the family adequately up till the moment the father regained control again.

Both variations may create feelings of loneliness in the other partner, because both imply that the other partner was hindered in expressing emotions. Whereas the first strategy of not showing emotions is used to ward off fear, the second strategy is motivated by not wanting to enhance the burden already experienced by the parent. This protective behavior towards oneself or towards the spouse may reduce the chance of receiving support from each other (27). Therefore the outcome of coping has to be interpreted within the dynamics and context of the marital and family system.

DISCUSSION

Loss and mourning

Upon hearing the potentially fatal diagnosis of cancer, parents' only wish is that their child will be cured. Now that survival seems to have been accomplished, one would expect them to be relieved, even elated. However, the picture that arouse from our interviews was quite different: the illness experience had marked these parents for life. Instead of happiness, perseveration of problems and feelings of loss prevail. We will provide some hypotheses to explain this observed anguish in parents involving processes of mourning over (a) the psychological losses and (b) the anticipated death of the child.

Parents of cancer survivors continue to experience profound losses after treatment ends. As described in the Results section, these experienced losses are unrelated to the actual loss of the child but to psychological losses caused by the illness. In spite of the child's survival, these losses can subsequently induce mourning processes. However, because the child has survived the disease, it is unlikely that parents themselves as well as their social environment will explain their present feelings of unhappiness in terms of mourning. Although these psychological losses are of a less severe nature as compared to the loss in case the child had died, these losses nevertheless do matter for parents. Thus, in considering treatment or death, to have their child treated is in fact only choosing the lesser of two evils.

Apart from mourning about psychological losses, our results shows that mourning over the possible death of the child still plays an important role as well. Three variations will be elaborated on. First, it was observed that during diagnosis and treatment some parents adopted a death perspective. Several authors have pointed to the preparatory and adaptive function of anticipatory mourning in case the child dies (28-31). Since it is increasingly likely that the child survives due to improved treatment, the question is how anticipatory mourning affects parents of a child who survives cancer. Green and Solnit (32) suggest that this anticipatory mourning process may be disruptive in case the child who is expected to die, stays alive. We observed that parents who were unable to switch from a death perspective to a survival one still experienced much anguish and anxiety.

The other extreme, formed by parents who directly from the start take an exclusive and strong survival perspective, is a second variation. Parents can have several reasons for adopting such a perspective: The 'loyal' parent, who is motivated by the best interest of the child, does not deny the possibility of death but directs all energy and attention at survival. The 'denying parent', who is overwhelmed by anxiety, is motivated to adopt this perspective to never allow the possibility of death come into consciousness. In both cases this strategy has adaptive value to ward off vexing uncertainty and to prevent parents from oscillating between contrasting perspectives.

Although parents initially experience little anxiety from adopting a survival perspective, costs may be involved too. A survival perspective blocks anticipatory mourning, preventing parents to go through the process of grieving to a certain point where it can alleviate tension. To go through this process at a later stage in time is extra difficult, because it contradicts the fact that the child is visibly recovering. The time appropriate to grieve over the possible death of the child will not return and becomes more and more remote. At the same time, after successful treatment this remoteness from the possible death of the child may create room for the denying parent to face the possibility of death and for loyal parent to reconsider the life-threatening aspect of the disease once set aside. For both variants, this necessitates to work through a grieving process during an unusual stage of the illness i.e. after treatment ends. Because this mourning process is displaced it is difficult to resolve. This late psychological effect may play a role in parents' current functioning.

From our data, the most beneficial variant seems to be able to change from a death to a survival perspective. Parents, who at first faced the possibility of death and who during the course of treatment changed this perspective into a survival perspective, experienced less stress after stop treatment. They seemed to benefit from having gone through a mourning process. It demonstrates that a combination of coping strategies is more efficacious than only one strategy, particular when timed appropriately (33,34). However, the final adaptive value of these different strategies on psychosocial functioning in the long run needs to be explored.

Dynamics of coping

The long-term sequelae of childhood cancer consist of a complex of stressors, and parents use a differentiated approach to cope with each of its elements. Consequently, dispositional measures of coping may not have predictive value because parents may use different strategies of the same coping dimension dependent on the element involved. In this study it was found that, for instance as to expressing emotions (approach-avoidance dimension), some parents were able to discuss negative changes in personal functioning (approach), but *not* in marital functioning (avoidance). The same observation is made as to attributional style or locus of control. Parents may believe to have little control over the disease outcome (external control), but at the same time experience substantial control over the psychological outcome of the disease in their child (internal control).

In the present study, parents differ in the choice of their strategies. Some used predominantly confrontation, others avoidance, still others used both strategies. In a personality model of coping, Krohne (35) explains these differences between individuals by their dispositional tolerance for uncertainty and for emotional arousal. Furthermore, he considers extreme positions on either confrontation or avoidance to be vulnerable systems of coping. The adaptive cost-hypothesis formulated by Suls and Fletcher (34) points out that avoidance may cost considerable efforts, which, over time, can be debilitating to the individual's psychological and physical resources. But also negative relations with health status are reported for individuals using confrontation strategies (35). A combination of strategies seems more adaptive than using either one intensively (33,34). This is in line with the good outcome in 'balanced' families in our study, which were characterized by the fact that they adopted a middle position on relevant coping dimensions including confrontation - avoidance and attribution of control.

Parenting behavior as a mode of coping

In the literature on childhood cancer, different parenting behaviors have already been observed in parents, but until now no link has been made between parenting and the coping literature. In this study we described a number of parenting behaviors that have a function in reducing negative effects evoked by childhood cancer. In considering parenting behavior from a coping point of view, the issues in parenting (36) become functionally linked to parental coping with childhood cancer. In this way, a new area of research is opened. Insight may be gained into the question which kind of stress induces parents to use a particular parenting strategy. This insight may also lead to intervention programs not directly aimed at altering dysfunctional coping strategies of parents, i.e. a particular type of parenting behavior like overprotection (37), but aimed at altering the negative effects experienced by parents which motivated them to use that particular parenting strategy.

CONCLUDING REMARKS

The qualitative method resulted in an integrated insight into what it is like for parents to live through childhood cancer. On the basis of our analyses, insight is obtained into which late consequences can be expected in parents after cessation of treatment. This information may help health care professionals to develop strategies to prepare parents for the period of survivorship and to help them adapt to this period. Furthermore, it gives researchers insight into the actual issues important for parents after cessation of cancer treatment. The themes and variations emerging from this analysis may initiate new studies to explore a particular topic in more detail.

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CHAPTER 3.2

LATE PSYCHOSOCIAL CONSEQUENCES FOR PARENTS OF CHILDREN WHO SURVIVED CANCER*

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ABSTRACT

Investigated late psychosocial sequelae in 133 parents of children who survived cancer, using questionnaires developed to measure the specific impact of the disease. Childhood cancer had distinct and persistent late psychosocial effects on parents of survivors. Uncertainty and loneliness were the most reported problems. Demographic and situational characteristics such as being a mother, low SES, no religious affiliation, chronic disease in a family member other than the child surviving cancer, and concurrent stresses increased the risk of reporting late problems. Treatment itself had little or no effect on reported problems. However, when treatment led to long-term sequelae in the child, a significant and specific effect on parental problems was observed. No decline of problems over time was found, which has implications for patient care.

INTRODUCTION

As the prognosis of childhood cancer improves, so does concern for the quality of survival. Because cancer and its treatment affect not only the patient but also the family, quality of survival should include the long-term effects for the family as well. Consequently, we need to know the kind of late psychosocial consequences parents experience *after* the child's treatment has been terminated. There are several reasons why parents may experience problems after completion of treatment. Even though treatment has been terminated there is still a possibility that the disease will recur and

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the child might die. Furthermore, parents may experience a setback resulting from the stresses during treatment including the threat of losing their child, the rigors of treatment, and changes in family life (1). In addition, parents can be confronted with new problems evolving from the child's illness, such as long-term medical side effects including growth retardation, sterility, and neuropsychological deficits (2). The first aim of our study was to describe the kind of psychosocial problems parents experience after termination of their child's treatment, and second, to explore which factors are related to the occurrence of these problems.

Empirical studies on late psychosocial consequences of childhood cancer for parents

Studies on the long-term sequelae for parents are sparse. An initial study by Peck (3) revealed that after cessation of treatment, parents most often mentioned anxiety about recurrence of the disease. None of the parents believed their child was cured and, consequently, none considered their child normal. Other ongoing concerns that have been reported are worries about the child's school performance, lack of friends, physical side effects, marriage prospects, and job opportunities (4-8). For most parents, concerns and anxieties over the various aspects of their child's development become an integral part of their daily life (3,5).

In contrast, other studies found no evidence for continuing psychological distress in parents of children who had terminated cancer treatment (9-11). These studies reported an improvement in adjustment over time, using measures of overall functioning. In addition, in comparing parents of cancer survivors and parents of healthy children, no differences in level of parental distress (12-13) or anxiety and depression (14) have been found.

These discrepancies in reports on parental functioning after cessation of treatment can be attributed to the different psychological domains that have been assessed in each of these studies. By focusing on limited areas of functioning, an incomplete picture of parental problems is found. Furthermore, studies on parental concerns are often of a descriptive nature and use nonstandardized instruments; in contrast, studies on general functioning employ standardized but global instruments that conceal the specific issues of concern for parents of cancer survivors. Finally, existing studies have been limited by small samples and the inclusion only of mothers.

Measuring late psychosocial consequences for parents

One of the most common problems in studying the psychosocial aspects of childhood cancer is measuring the specific impact of the cancer experience. Existing standardized instruments allow for reliable judgments but are ill suited to the problem under study. These instruments are inappropriate because they are developed to differentiate between psychopathology and normality. In general, parents of children with cancer are normal individuals showing psychosocial reactions to an extreme stressor (15). Thus, the failure of existing instruments to identify significant findings may be due to their insensitivity in identifying the essential and specific aspects of the cancer experience (16-17).

Therefore, we developed an instrument aimed to assess the specific problems of childhood cancer in parents. This measure was based on the operationalization of stress in a general conceptual framework developed by Van den Borne and Pruyn (18,19) of the impact of a stressor. Although several models (e.g., Thompson et al. (20), Wallander et al. (21)) depict variables influencing the process of adaptation, they do not describe which specific domains of psychosocial functioning should be tapped. The model of Van den Borne and Pruyn (18), however, outlined the domains of psychosocial functioning that are negatively affected by a stressor; as such it provides guidance in operationalizing the relevant concepts needed to measure late psychosocial consequences in parents. According to this model, a stressor is perceived when it causes (a) uncertainty, and/or (b) loss of control, and/or (c) threat to self-esteem, and/or (d) negative feelings. *Uncertainty* is defined as a lack of information about a value system that is important for a person. People strive to experience as little uncertainty as possible in order to predict and influence forthcoming events. This implies that people want to have information when they experience uncertainty. *Loss of control* is defined as the inability to manage or influence events. Loss of control can be experienced in different areas including the individual's daily, physical, emotional, social, and general functioning. *Self-esteem* is related to the image an individual has about oneself. *Negative feelings* include all other feelings by the individual perceived as unpleasant, such as loneliness, depression, and fear. It has been demonstrated that the many psychosocial problems experienced by parents of children with cancer during treatment can be assigned to the problem categories of the model (22). We adapted these four problem categories to the specific problems experienced by parents of cancer survivors.

Factors affecting the experience of parental problems

The second aim of our study was to explore which factors are related to the experience of problems by parents of childhood cancer survivors. The models of Wallander and Thompson and their colleagues (20,21) point to risk and resistance factors that may result in differential adjustment in parents of chronically ill children. Evidence suggests that demographic and situational factors are related to adjustment in parents of cancer patients. Also, we hypothesize that intrapersonal and disease-related variables may affect parental functioning.

Demographic variables. Speechley and Noh (14) found that low socioeconomic status (SES) was negatively associated with parental psychosocial functioning and that gender may also be important as mothers are considered to be more vulnerable in experiencing problems than fathers. Religious belief is perceived by parents as a source of support which may enable them to cope better with the stresses inherent in the disease (23). Furthermore, it was expected that family size would have an impact. Parents of an only child may experience more stress because losing their child also involves the loss of their parenting role.

Situational variables. Kalnins et al. (24) demonstrated that parental problems were negatively affected by concurrent stresses in the family. Furthermore, we expected that the presence of a chronic disease in another family member may add to parental problems.

Intrapersonal variables. Intrapersonal factors can moderate the experience of problems. Locus of control is a variable that influences the person's appraisal of a situation as a stressor and thus may affect whether problems are experienced (25). In addition, Peck (3) noted that parents held back their concerns. Social desirability may have influenced the extent to which parents report problems (26).

Disease-related stressors

Childhood cancer and its treatment involves a variety of disease-related stressors which may add to the risk of emotional upset. Stresses related to the life-threatening nature of cancer include survival chances and the actual recurrence of the disease. Indicative of stressors during treatment may be the duration of treatment, and the modalities of treatment such as chemotherapy, surgery, and irradiation. Long-term sequelae may include specific side effects associated with cancer and its treatment (2).

The present study first describes the problems of uncertainty, loss of control, self-esteem, and negative feelings reported by parents of a child surviving cancer. Next, we investigate whether problems of parents are related to the lapse of time since cessation of treatment, ranging from less than 2 years to more than 5 years. Finally, the relation between demographic, situational, intrapersonal, and disease-related factors and reported problems of uncertainty, loss of control, self-esteem, and negative feelings are explored.

MATERIALS AND METHODS

Subjects

Parents whose native language was Dutch and whose child met the following criteria were approached to participate in the study: (a) the child had successfully terminated cancer treatment, and (b) the child was 8 to 12 years old at the time of the study. All eligible children with leukemia and lymphoma treated at the Sophia Children's

TABLE 1. Demographic, situational, and intrapersonal characteristics of parents of childhood cancer survivors (N=133; fathers n=64, mothers n=69).

Demographic vairables	%	*n*	Situational variables	%	*n*	Intrapersonal variables	%	*n*
Family size			*Concurrent stresses*			*Locus of control*		
-one child	9	13	-yes	29	39	-internal	77	103
-more than one child	91	120	-no	71	94	-external	23	30
Religion			*Chronic disease*			*Social desirability*		
-yes	62	82	-yes	21	28	-yes	38	50
-no	37	49	-no	79	105	-no	62	83
-missing	1	2						
SES:								
-low	59	78						
-medum/high	40	53						
-missing	1	2						

Hospital/University Hospital Rotterdam (one of the four pediatric cancer centers in The Netherlands) and 4 children with a solid tumor, consecutively selected according to the same criteria, were approached. The patients with a tumor resembled the children with lymphoproliferative diseases with respect to prognosis, intensity of chemotherapy, duration of treatment, and type of late medical sequelae. Of the eligible participants 98% consented. In total, 133 parents (69 mothers; 64 fathers) of 70 children surviving cancer participated.

Termination of treatment ranged from 6 months to 7 years 9 months (median 4 years 1 month) at the time of assessment. Table 1 summarizes the demographic characteristics of the parents and children. SES was scored on the basis of the educational level of each parent. Marital status was not included, because only 4 parents were single or divorced.

Procedure

Parents were approached during regular appointments at the (long-term) follow-up outpatient clinic. Written informed consent was obtained 1 week after the initial contact. Parents were interviewed at home in an open, in-depth way about their experiences with their child's disease. After the interview they were requested to complete independently a booklet of questionnaires.

TABLE 2. Psychometric data of the scales uncertainty, loss of control, self-esteem, and negative feelings, and their subscales.

	Number of items	Response format (Likert-scale)	Factor loadings mean (range)	GFI	Item-total correlations mean (range)	Cronbach's alpha
Uncertainty	21	4-point	.71 (.51-.85)	.95	.61 (.46-.77)	.93
Loss of control			.71 (.56-.85)	1	.43 (.38-.48)	.64
- Personal functioning	9	2-point	.77 (.67-.86)	.98	.47 (.37-.56)	.77
- Being a parent	9	2-point	.77 (.63-.87)	.97	.49 (.41-.60)	.80
Self-esteem	4	2-point	.72 (.64-.80)	.99	.52 (.47-.58)	.73
Negative feelings			.69 (.41-.91)	.99	.62 (.41-.77)	.82
- Anxiety	17	4-point	.85 (.70-.99)	.95	.71 (.53-.84)	.95
- Depression	10	4-point	.76 (.61-.93)	.94	.60 (.49-.68)	.87
- Psychological and physical distress	20	4-point	.76 (.62-.89)	.93	.61 (.50-.75)	.93
- Disease-related fear	10	4-point	.73 (.49-.89)	.91	.59 (.40-.67)	.86
- Loneliness	8	4-point	.71 (.63-.81)	.93	.57 (.47-.66)	.84
- Sleep disturbances	5	2-point	.77 (.62-.90)	.99	.46 (.32-.53)	.70

Dependent measures: psychosocial problems

DEVELOPMENT OF THE SCALES

Item pool

To get insight into the specific problems of parents whose child survived cancer, a pilot study was performed with 14 families. Parents were interviewed about their experiences with their child's illness and their present situation. The duration of the interviews ranged from 2.5-4.5 hours and all interviews were audiotaped. From the recorded verbatim protocols and observations during the interview, problems were identified (see also 27). An exhaustive list of items was formulated; items were subsequently assigned to one of the problem categories of the model. All available items from both the pilot study and those developed by Van den Borne and Pruyn (18) for adult cancer patients were included. Then items were selected according to the following criteria: the item (a) was relevant for parents, as measured by the occurrence of the issue in at least two interviews; (b) covered a new issue that was considered important in the context of the model; (c) covered an issue reported in the literature; and (d) was not redundant. In this way, ten *a priori* scales were constructed measuring the four problem categories of the model that were included in the booklet of questionnaires.

Scales measuring the concepts of the model

Number of items and response format of the scales are presented in Table 2. *Uncertainty* was measured by one scale. Items covered the need for information about the child's present condition, long-term emotional and physical sequelae in the child, survival chances, how to handle the child, and parental psychosocial problems. Two subscales were constructed to measure *loss of control* in relation to (a) parents' personal functioning, and (b) his/her parental role. *Self-esteem* was measured by one scale developed by Van den Borne and Pruyn (18). To measure the scale *negative feelings*, six subscales were used: anxiety; disease-related fear; sleep disturbances; loneliness; depression; psychological and physical distress. These feelings are most often reported in the literature on psychosocial functioning of parents of children with cancer during treatment (1).To measure parent's feelings of *anxiety*, we used items 1-5, 7-17, and 15-20 from the state version of the State-Trait Anxiety Inventory (28). *Depression* was measured by an adaptation of the Self-Rating Depression Scale (SDS; 29), (items 1,3,10-12, 14, 15, and 20) and 2 items developed by Van den Borne and Pruyn (18). To measure *psychological and physical distress* we used a 20-item version of the Rotterdam Symptom Checklist (RSCL; 30), containing all items of the psychological distress scale and items 3,5,7,8,15,22-24, and 28-30 from the physical distress scale. The content of items measuring *disease-related fear* was based on the literature on the late physical and emotional consequences in childhood cancer (2,5) and on parental concerns (3). The *loneliness* scale contained general items on

Table 3. Disease-related characteristics of survivors of childhood cancer ($N=70$; male $n=42$, female $n=28$).

Variables related to disease:			Variables related to treatment:			Variables related to long-term sequelae:		
Age:	median	range	*Duration of treatment :*			*Time off treatment:*	%	*n*
age at diagnosis	3y 8mo	1y 3mo-9y 7mo		%	*n*	≤2 year	23	16
age at investigation	10y 10mo	8y 5mo-13y 9mo	- <2 year	16	11	2 year - 5 year	46	32
			- 2-3 year	64	45	≥5 year	31	22
Diagnosis:	%	*n*	- 3-4 year	14	10	*Visible impairment:*		
Leukemia	87	61	- 4-5 year	6	4	- yes	29	20
Non-Hodgkin lymphoma	7	5	*Intensity of chemotherapy protocol:*			- no	71	50
Rhabdomyosarcoma	6	4	- mild	24	17	*Invisible impairment:*		
			- intermediate	52	36	- yes	17	12
Relapse:			- severe	24	17	- no	83	58
-yes	10	7				*Overweight:*		
-no	90	63	*Surgery:*			- yes	44	31
			- yes	13	9	- no	56	39
Survival chances at diagnosis:			- no	87	61	*School problems:*		
			Irradiation:			-regular school	56	39
≤ 50%	51	36	- yes	51	36	-regular school plus school problems	23	16
> 50%	49	34	- no	49	34	-special school	21	15

loneliness developed by Van den Borne and Pruyn (18) and items regarding specific issues of loneliness for parents of childhood cancer survivors. *Sleep disturbances* were measured by a scale developed by Van den Borne and Pruyn (18).

Mean total psychosocial problem scores were computed for each parent by summing the item scores of the uncertainty, loss of control, self-esteem, and negative feelings scales, and dividing this total score by the number of nonmissing items. To investigate the distribution of mean total psychosocial problem scores, parents were classified according to the level of their scores. The level of reported problems on the scales for uncertainty, anxiety, fear, loneliness, depression, and psychological and physical distress, all measured on 4-point scales, was trichotomized into low, medium, and high, if the mean scale score was 1 to 2, 2 to 3, and 3 or higher, respectively. The scales for loss of control, self-esteem, and sleep disturbance, measured by a 2-point scale, were dichotomized: parents with a mean scale score below 1.5 were assigned to the low category, parents with a mean score of 1.5 and above were assigned to the high category.

To explore why some parents reported more problems than others, parents were assigned to a problem group according to their ratings on each of the four main scales. A cutoff point corresponding to the 75th percentile of the distribution of the scale scores on each separate scale was chosen to classify 33 parents as a problem group and 100 parents as a nonproblem group. To examine the validity of cutoff point an external criterion was used: parents rated on a 6-point Likert scale the extent to which they had come to terms with their child's disease.

Method of analysis

To determine whether the scales were unidimensional, we estimated and fitted an one-factor model to the polychoric correlations among the scale items for each *a priori* scale using the LISREL program (31). Polychoric correlations were used because of the categorical nature of the response format. The goodness-of-fit-index (GFI) indicates the degree to which the one-factor model fits the scale scores. If GFI was $> .90$ the one-factor model was accepted as a reasonable approximation of the data. Furthermore, factor loadings $\geq .40$ on the latent factor were considered as an indication that these items measured the defined area and were reliable (32). To explore the reliability of (sub)scale items, Cronbach's alpha and item-total correlations were computed separately for each (sub)scale. Items with an item-total correlation $\geq .30$, considered of medium magnitude (33), were assessed to have an acceptable reliability. Items whose factor loadings and item-total correlations met both criteria were included in the scale. Because loss of control and negative feelings were measured by several questionnaires, first analyses were performed to determine the homogeneity of the separate *a priori* subscales. To investigate whether the two loss of control subscales and the six negative feelings subscales measured one underlying concept, confirmatory factor analysis and reliability analysis were performed on the subscale scores.

Results of the confirmatory factor analysis and reliability analysis revealed satisfactory indices of homogeneity for uncertainty, loss of control, self-esteem, and negative feelings (Table 2).

Independent measures: risk factors

Demographic, situational, and intrapersonal variables of the parents are presented in Table 1. Situational variables (i.e. psychosocial stresses) were measured by two items inquiring about (a) concurrent stresses and (b) a chronic disease or condition in one of the other family members. Intrapersonal variables included locus of control and social desirability. An eight item locus of control scale was developed to measure the control parents perceived over problems (21) Social desirability was assessed by using an eight item version of the Social Desirability Scale (26).

Disease-related stressors are summarized in Table 3. The survivor's statistical survival chances at diagnosis were rated by an oncologist, based on the diagnosis, symptoms at presentation, and the success rates for the treatment protocols at that time. Intensity of chemotherapy was rated by the oncologist according to treatment protocol. Visible impairment was partly measured by the residual physical impairment index (5), consisting of the sum of ratings of current visibility and interference with daily living. Invisible impairment included invisible medical problems such as organ damage and endocrine problems. Overweight was defined as the child's weight at or above the 90th percentile corresponding to Dutch standards (34). School problems were rated present if the child received remedial teaching or was not able to attend a regular school.

RESULTS

Problems reported by parents of childhood cancer survivors

Frequencies of (sub)scale scores are given in Table 4. Parents reported much uncertainty after termination of treatment. Almost 90% of all parents reported uncertainty. Analysis of variance (ANOVA) revealed no difference between fathers' and mothers' mean uncertainty scale scores. The Pearson correlation between scale scores of fathers and mothers was .68 ($p < .001$). Parents were especially uncertain about the late medical and psychosocial sequelae and about the survival chances of their child.

Only 9% of the parents reported much loss of control. ANOVA revealed no difference between fathers' and mothers' mean scale scores of uncertainty. The correlation between both parents' scale scores was .69 ($p < .001$).

Frequencies on the self-esteem scale indicated that most parents (63%) reported high self-esteem. No significant correlation in self-esteem was found between fathers and mothers ($r = .29$, n.s.)

TABLE 4. Percentage of parents ($N = 133$) scoring in different scale categories of uncertainty, loss of control, self-esteem, and negative feelings.

	Scoring		
Scales	Low	Medium	High
Uncertainty	12%	37%	51%
Loss of control	91%	n.a.	9%
-Personal functioning	88%	n.a.	12%
-Being a parent	86%	n.a.	14%
Self-esteem	37%	n.a.	63%
Negative feelings	80%	19%	1%
-Anxiety	58%	32%	10%
-Depression	69%	28%	3%
-Psychological and physical distress	95%	4%	1%
-Disease-related fear	71%	23%	6%
-Loneliness	16%	62%	22%
-Sleep disturbances	86%	n.a.	14%

Note. All items are measured by a 4-point Likert scale, except those of the (sub)scales loss of control, self-esteem, and sleep disturbances. n.a. = not applicable.

Of the parents, 80% reported low levels of negative feelings. No significant differences in negative feelings were found between fathers and mothers. Although mean scores on the negative feelings scale were low, the scores on the separate subscales gave a more differentiated picture. On some subscales, parents indicated a high degree of negative feelings, whereas on others, few negative feelings were reported. The most negative feelings were reported for the loneliness subscale. Intermediate to high levels of anxiety, depression, and disease-related fear were reported by 42, 31, and 29% of the parents, respectively. Sleep disturbances and psychological and physical distress were mentioned the least. The correlations between fathers' and mothers' subscale scores were: anxiety (.51), disease-related fear (.79), loneliness (.68), sleep disturbances (.08), depression (.48), and psychological and physical distress (.23); all were significant ($p < .001$) except for the subscales sleep disturbances and psychological and physical distress.

Interval between time of assessment and termination of treatment

Figure 1A displays the mean scores of the separate scales in relation to time since termination of treatment. For the purpose of comparability across scales, all scores were converted to a 10-point scale (cf. 35). Because negative feelings is a combined score, based on several subscales, the separate subscales over time are also displayed (Figure 1B). Parents in all three groups reported the highest scores for the uncertainty and loneliness (sub)scales.

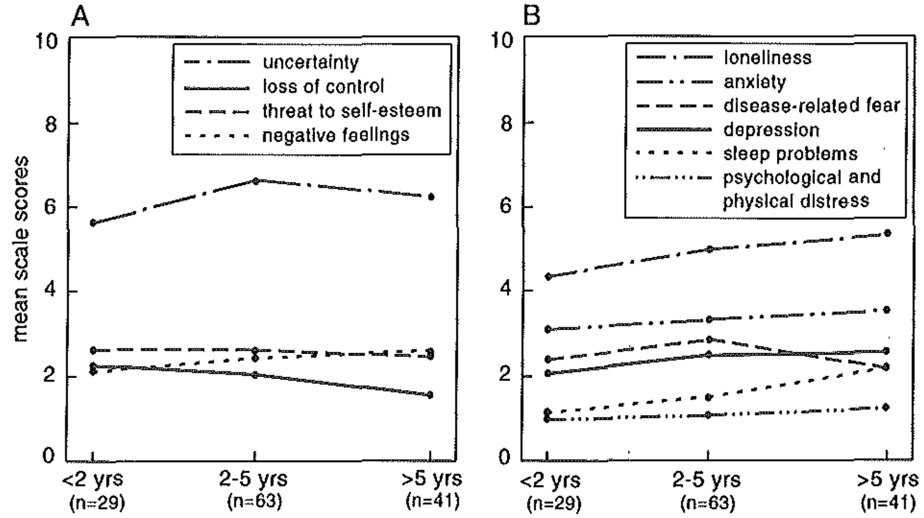

Figure 1. Mean psychosocial problems (A) and mean negative feelings (B) in relation to time since completion of treatment.

ANOVA's were performed to investigate the effect of time and gender. No significant differences were found except for mothers who reported a decrease in loss of control over time (F (1,127)=6.6, p=.011). After Bonferroni correction for three statistical comparisons, this difference failed to reach statistical significance. This implied that there was little variation in scores over time. Because no differences in problem scores were found between fathers and mothers, Figure 1 presents the results for the total sample.

Relation between risk factors and psychosocial problems

Using the cut-off point corresponding to the 75th percentile of the distribution of the scale scores on each separate scale, 50.4% (n=67) of the parents were classified as having no problems, 20.3% (n=27) fell into the problem group on one scale, 12.8% on two scales (n=7), 11.3% on three scales (n=15), and 5.3% (n=7) of the parents on all four scales. A separate problem group was created by combining the two latter groups into a high-risk group (i.e., parents who reported serious problems on 3 scales or more; n=22; 6 fathers and 16 mothers). To investigate the validity of the cutoff points for each of the four problem categories and the high-risk group, parents in the problem groups were compared with parents in the nonproblem groups. ANOVAs showed that parents of the top 25% problem score on uncertainty, loss of control, self-esteem, and negative feelings scales, reported significantly more problems, F (1,131)=126.84, F (1,131)=238.71, F (1,131)=317.55, and F (1,131)=210.97;

respectively, $p < .000$. Furthermore, parental scores on the measure of the extent to which parents had come to terms with their child's disease were compared. Except for the problem group for uncertainty, ANOVA's showed that parents in the loss of control, self-esteem, negative feelings problem groups and the high risk group, were significantly less advanced in this process than parents in the non-problem groups, respectively $F = 13.78$; $F = 25.34$, $F = 12.91$, and $F = 10.72$; for all analyses $df = 1/131$, $p < .000$.

Odds ratios were computed because of our interest in identifying those parents who are most at risk of experiencing problems (36). Odds ratios indicate the relative probability of being assigned to one of the problem groups for parents for whom the risk factor is present, compared to parents for whom this factor is absent. Values smaller than 1.0 indicate a negative relation to psychosocial problems, values greater than 1.0 indicate a positive relation. Odds ratios were computed with two different sets of predictors: the first set included the demographic, situational, and intrapersonal variables (see Table 1), and the second consisted of disease-related variables (see Table 3). Odds ratios were computed separately for each variable and its combination with other variables within the predictor set, provided the number of subjects constituting a risk group was ≥ 10. In Table 5 odds ratios (OR) are reported that differed significantly from 1 (95% confidence interval). This method gives priority to avoiding Type I errors (37). To avoid making Type II errors no corrections for chance findings were made.

In the first set of variables involving demographic, situational, and intrapersonal variables, four risk factors were significantly related to psychosocial problems: low SES, no religion, female gender, and the presence of a chronic disease in a family member other than the child surviving cancer. Low SES and no religion were separately, but also in combination with being a mother and concurrent stresses, positively related to classification in the problem group on negative feelings and threat to self-esteem scales, and the high-risk group. Presence of an additional chronic disease increased chances for a problematic outcome on the loss of control and negative feelings scales and to be classified in the high-risk group. A chronic disease, especially in combination with no religion (OR=6.2) and being a mother (OR=6.5) increased the probability of experiencing serious problems. Intrapersonal factors yielded no significant relation to any of the outcome variables.

The second set of disease-related risk factors showed 10 positive OR that differed significantly from 1 in relation to the loss of control scale. All these risk factors were related to long-term sequelae in the child, except operation. The probability of scoring in the problem group for loss of control was drastically increased for parents whose child suffered invisible impairment in combination with either school problems (OR=8.3) or overweight (OR=9.0). For these parents, the probability to be classified

TABLE 5. Odds ratio of psychosocial problems against disease and demographic and situational factors.

Predictor variables	*n*	Outcome variables				
		Uncertainty	Loss of control	Self-esteem	Negative feelings	High scores on at least 3 scales (*n*=22)
Demographic and situational variables						
-Low SES	78	-	-	2.4	-	-
-No religion	49	-	-	2.2	2.8	2.9
-Chronic disease	28	-	4.5	-	3.0	3.4
-Female	69	-	-	-	-	2.9
-Female x low SES	46	-	-	2.6	-	2.6
-Female x no religion	23	-	-	2.6	2.9	3.4
-Female x concurrent stresses	26	-	-	-	2.8	3.8
-Female x chronic disease	17	-	4.3	-	4.3	6.5
-Low SES x no religion	29	-	-	3.7	3.4	5.2
-Low SES x concurrent stresses	17	-	-	3.9	3.2	4.7
-Low SES x chronic disease	16	-	-	-	-	3.8
-Chronic disease x no religion	10	-	3.5	-	-	6.2
-Chronic disease x concurrent stresses	13	-	-	-	4.2	3.8
Disease related variables						
-Operation	18	-	-	-	2.9	-
-Operation x prognosis ≤ 50%	12	-	-	4.7	5.1	-
-Invisible impairment	23	-	2.9	-	2.0	2.8
-School problems	59	-	2.4	-	-	-
-< 2yrs since stop x school problems	39	-	2.6	-	-	-
-< 2yrs since stop x overweight	35	-	2.8	-	-	-
-Invisible impairment x school problems	15	-	8.3	-	4.3	4.3
-Invisible impairment x overweight	13	-	9.0	-	4.2	5.8
-Visible impairment x school problems	22	-	2.5	-	-	-
-Visible x invisible impairment	13	-	-	-	4.2	3.8

in the problem group on the negative feelings scale and in the high-risk group was increased as well. None of the disease-related variables were risk factors for uncertainty and only one, operation plus poor prognosis, was related to self-esteem.

DISCUSSION

The first aim of our study was to obtain insight into the late psychosocial sequelae of parents of children who survived cancer. Our study demonstrates that after termination of treatment, childhood cancer *has* late psychosocial effects on parents of survivors, and that uncertainty and loneliness are the most frequently reported problems. Because of the high number of parents who reported uncertainty, and because the odds ratios showed that no independent variable specifically affected uncertainty, we conclude that uncertainty in parents is a general characteristic following the cessation of their child's treatment. It is possible that we did not include an independent variable which may have affected uncertainty. However, a large number of demographic, situational, intrapersonal, and disease-related characteristics were included, several of which did have an effect on other problem categories. Comaroff and Maguire (38) argued that uncertainty in parents is the main feature of childhood cancer during treatment. Our results show that this is also the case *after* treatment.

Parents of childhood cancer survivors reported a variety of negative feelings. In general, the incidence of these feelings was not high, except for loneliness which was reported by 84% of the parents. Sleep disturbances and psychological and physical distress were relatively rare. Although disease-related fear, anxiety, and depression were more frequently reported, they were of a moderate intensity. Depression, often reported by parents during treatment, was observed in a relatively small subset of parents, indicating that depression is not a standard long-term outcome of the cancer experience in parents. Speechley and Noh (14) compared parents of cancer survivors to a matched sample of parents with healthy children and found no differences in levels of depression or anxiety between the two groups.

In this study we were able to compare the level of problems reported in different areas. In this way the different findings in the literature about a low level of parental distress (9,12-14) and a high level of lingering concerns (3-5,8) can be linked. Our findings indicate that most parents of childhood cancer survivors continue to be uncertain about the well-being of their children and report lonely feelings, but at the same time these concerns are not accompanied by high levels of distress as measured by anxiety, depression, disease-related fear, sleep disturbances or psychological and physical distress. To interpret the precise meaning of the level of reported problems, comparative data is needed. The measures developed from the model can be used to collect comparative data with other populations.

The impact of childhood cancer on parents seems to persist over time. Although a cross-sectional design was used in the present study, limiting our conclusions about how problems evolve within one family, we found no difference in levels of reported problems among groups of parents. All problem categories remained at the same level, independent of the lapse of time since treatment. With respect to negative feelings there was a slight tendency, although not significant, for these feelings to increase with length of time since treatment. Our findings are in accordance with the study of Speechley and Noh (14) who also used a cross-sectional design but in contrast to the longitudinal findings of Kupst and Schulman (10), who found a decrease in problems 6 years after diagnosis.

The second aim of the study was to get insight into the variables related to late psychosocial outcome. Considering the large number of variables and interaction effects tested, the results are of an exploratory nature and need replication in future studies. An important set of variables influencing the risk of late psychosocial problems are demographic and situational variables. These variables especially had an effect on self-esteem and negative feelings. Low SES, no religious affiliation, and chronic disease in another family member, in combination with being a mother and having other concurrent stresses, increased the risk of late psychosocial sequelae. Although it may be argued that these risk factors per se are associated with low self-esteem and negative feelings, it shows at the same time that, given this situation, an extra stressor as childhood cancer overburdens these parents. Thus, parents who have multiple stressors may have more difficulties in coping with the problems inherent in childhood cancer.

In the literature, a number of variables have been reported to protect the individual from stress. In our study, these protective variables are the *absence* of the above-mentioned demographic characteristics and situational circumstances. A protective variable that emerged from this study is religious belief, particularly for parents with limited resources such as a low SES, and concurrent stresses. In a study on religious belief, depression, and ambulation status in elderly women with broken hips, it was found that religious affiliation was associated with lower levels of depressive symptoms and better ambulation status (39) The present study provides corroborative evidence that religion has a positive effect on negative feelings, including depression, in parents of cancer survivors. It remains unclear why religious belief is associated with fewer problems in parents. Possible explanations are that religious affiliation is associated with social support or that religious affiliation is related to important differences in intrapersonal characteristics. Another possibility is that religion provides extra strategies to cope with childhood cancer. Because childhood cancer is characterized by uncertainty and loneliness, religion may comfort parents and provide a framework to interpret their experiences.

In contrast to demographic and situational variables, intrapersonal characteristics were unrelated to psychosocial outcome. Although intrapersonal characteristics are related to coping strategies, parents may use a differentiated approach to cope with each of the specific problems tapped by each item included in the scale score. By using total scale scores as an outcome variable we did not differentiate in problem-specific responses which may have concealed the relation between intrapersonal variables and psychosocial outcome.

An important issue was whether parents experienced a late setback as a result of the stresses evolving from disease and treatment. Surprisingly, variables related to either the disease itself or the treatment such as the child's prognosis, relapse, length of treatment, intensity of treatment, or irradiation yielded no relation with outcome. This implies that diagnosis and treatment, as such, had little or no effect on late psychosocial problems. Only surgery had an effect, but these children had more invisible (55 vs. 14%) and visible sequelae (88 vs. 24%) in comparison to the survivors without surgery. Long-term sequelae such as invisible medical side effects, including organ damage or sterility, and school problems substantially increased problems in parents. Parents confronted with medical aftereffects reported more loss of control and negative feelings which may be explained by their limited possibilities to actually remedy this unfavorable situation for their child. Parents of a child with school or weight problems in addition to invisible sequelae are especially at risk for later problems. These findings suggest that it is the evolving of new disease-related problems that predict poor parental outcome later, rather than stresses during the acute treatment phase.

Based on the results of this study, there are several implications for patient care. The need for care is underscored by the fact that once problems in parents have emerged, they tend to persist over time. Uncertainty is the main problem for parents of cancer survivors. Information is needed especially on the long-term physical effects of cancer in their child as well as on the impact of the disease on the child's *psychosocial* functioning. However, providing answers may be not sufficient. All parents in our sample regularly visited the (long-term) follow-up clinic. Still, there was a considerable need for information. It seems that families are in need of more information than is available. Therefore, the quest for information can also be an indication of the families' struggle to adjust to the loss of illusion of control and predictability of their lives evolving from the major unexpected trauma of cancer in their child. An opportunity to process these experiences can be helpful by, for instance, exploring parents' individual experiences and way of coping with the disease and discussing its effect on their present way of life and functioning. In applying this approach during interviews as part of this study, parents disclosed that it was the first time they talked about the complete period in an elaborate way, and a number of parents spontaneously reported beneficial effects for themselves as a result of the interview (27).

In conclusion, our study shows that childhood cancer has late psychosocial effects on parents after treatment has ended. Parents especially reported uncertainty about the survival status of their child and about the impact of disease and treatment on the physical and psychosocial well-being of their child. In addition, parents who were confronted with new problems in their child evolving from the illness and treatment, such as medical side effects, reported further problems. These problems, in combination with feelings of loneliness, indicate that many parents suffer in silence, which may continue over the years after completion of treatment. On the basis of these results, routine psychosocial follow-up consultations for parents in medical follow-up programs is recommended.

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CHAPTER 3.3

IMPACT OF CHILDHOOD LEUKEMIA ON FAMILY PLANNING*

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ABSTRACT

Clinical experiences have indicated that family planning is affected by childhood leukemia. To investigate this issue, 130 mothers and fathers of 68 families with a long-term disease-free survivor were studied using interviews and questionnaires concerning the effects of childhood cancer on family planning. In one third of the families, either one or both partners reported that their child's disease affected their reproductive planning in various ways. The most extreme variations included having completed the family before diagnosis but still having another child versus not having completed the family before diagnosis but refraining from further progeny. More than half of the affected families refrained from further offspring, delineating psychological motives for their decision. For parents whose family planning is affected by their experiences with childhood leukemia, the decision-making process is an extra burden. Therefore, we advocate that support for parents of a child with cancer should include counselling on progeny, in which both genetic and psychological information should be provided.

INTRODUCTION

The peak incidence of childhood cancer is under 5 years of age (1,2). This implies that childhood cancer predominantly occurs in families with young children. Several of these families have not yet completed their family. A major life event such as a serious disease in a child can affect the parents' reproductive planning. Studies on the reproductive planning of parents with a genetic risk for a defective or ill child have shown that parents may refrain from further progeny (3). However, life events can also cause families to plan for a new child when they had not done so originally. After the

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death of a child parents may try to substitute the deceased child by having another child. This 'replacement-child' is deliberately planned as a result from unresolved parental grief (4,5).

Childhood cancer is a major life event for parents that affects a great number of parental issues (6). Until now studies on the psychosocial effects of childhood cancer on the parents did not investigate its impact on family planning. Our clinical experience, however, indicates that family planning is also affected by childhood malignancy. During treatment a number of parents were uncertain about postponing or refraining from progeny. They feared negative consequences of a new baby in the family on the ill child indicating that care for the ill child prevails. Sometimes future pregnancies are postponed until treatment for the ill child has been completed. In case of a relapse or advanced maternal age, the issue of having another child became most pressing. This suggests that the child's illness has an impact within the parental subsystem on the spousal and parental roles (7). Parents whose decision is affected by childhood leukemia have to cope with this problem in addition to the other stresses inherent in disease and treatment.

This study explores the extent to which family planning is affected by childhood cancer in mothers as well as fathers. To study the issue, families with a child who had completed treatment for leukemia or lymphoma were approached in order to investigate the possible changes in family planning and its actual outcome. This latter aspect gives insight into the variations of how parents cope with this problem.

MATERIALS AND METHODS

Subjects and procedure

The subjects of this study were parents of 8-12-year-old children with leukemia or lymphoma who were in continuous remission and had completed chemotherapy. Patients were Dutch and represented all eligible children at the Sophia Children's Hospital/University Hospital Rotterdam. Seventy families were approached to participate in the study. Two families refused to participate because they feared a disturbance in their mental balance. One hundred-and-thirty parents of 68 families provided information concerning their procreative behavior by interview and questionnaire ($n=124$) or by interview ($n=5$) and questionnaire ($n=1$) only. The latter parents were for practical and/or emotional reasons unable to give data by both methods. They were included in the sample to avoid selection within the sample. In 6 families information of only the mother was available because she was single ($n=4$), the father refused to participate ($n=1$), or the father was unable to participate due to physical handicap ($n=1$). Table 1 summarizes the characteristics of the sample.

TABLE 1: Demographic and disease-related characteristics.

Children ($n=68$)		Parents ($n=130$)	
Sex:		*Sex:*	
-Male:	60% ($n=41$)	-Male:	48% ($n=62$)
-Female:	40% ($n=27$)	-Female:	52% ($n=68$)
Age: median (range)		*Age: median (range)*	
-At diagnosis:	3y 7m (1y 3m-9y 7m)	-At diagnosis: fathers:	34y (24y-54y)
-At investigation:	11y 3m (8y 5m-14y 1m)	mothers:	32y (23y-43y)
		-At investigation: fathers:	40y (32y-58y)
Diagnosis:		mothers:	39y (31y-55y)
-Leukemia :	93% ($n=63$)		
-Non-Hodgkin lymphoma:	7% ($n=5$)	*Marital status:*	
		-Married:	97% ($n=126$)
Survival chances at diagnosis[1]*:*		-Divorced or single:	3% ($n=4$)
≤ 50%:	51% ($n=35$)		
> 50%:	49% ($n=33$)	*Religion:*	
		-Catholic:	26% ($n=34$)
Time off treatment (median range):		-Protestant:	27% ($n=35$)
4y 1m (range 6m - 7y 9m)		-Other:	9% ($n=12$)
		-None:	36% ($n=47$)
		-Missing:	2% (n=2)

[1] Survival chances were based on the success rates of the treatment protocols at that time.

Parents were interviewed at their homes about their experiences with the disease including its impact on family planning. If both parents participated, fathers and mothers were interviewed together. After the interview, each parent received a booklet questionnaires and was requested to mail the completed forms to the investigator. The use of a retrospective design has inherent limitations as the lapse of time may influenced the recall of past experiences. The fact that we used different methods i.e. interviews and questionnaires to investigate family planning may enhance the reliability of the responses. In addition, interviewing both partners may have given some corrections to possible distortions because parents frequently discussed and commented on the information provided by their partner.

Measures

Two psychologists independently rated all audiotaped interviews regarding the effect of their child's disease on procreative behavior. Raters indicated whether family planning: (a) was affected and parents refrained from further offspring (b) was affected and parents wanted another baby, (c) was not affected by the disease or (d) that it was undecided/unknown from the interview. The interrater-reliability was $k=.84$. The questionnaire was specifically designed to get insight into a change in family planning, its actual outcome, and the accompanying motives. If parental responses from the

TABLE 2. Changes in procreative behavior of parents (*N*=130) upon confrontation with childhood leukemia/lymphoma.

Stage of family planning before diagnosis	Family planning affected by diagnosis and/or disease course	Kind of change	Present status (at the time of the study)	Sterilization
Completed	No 91% (n=62)			
52% (n=68)	Yes 9% (n=6):	-Want to have another child (n=1)	.Unsettled (n=1)	No
		-Unsettled (n=5)	.Had another child (n=3) .No child (n=2)	No No
Unsettled	No 41% (n=13)			
25% (n=32)	Yes 59% (n=19):	-Want to have another child (n=3)	.Had another child (n=3)	No
		-Do not want to have another child (n=12)	.No child (n=12)	Yes (n=7)*
		-Unsettled (n=3)	.Had another child (n=2) .No child (n=1)	No No
		-Abortion (n=1)	.Unsettled (n=1)	No
Not completed	No 73% (n=22)			
23% (n=30)	Yes 27% (n=8):	-Do not want to have another child (n=1)	.No child (n=1)	No
		-Delayed family planning (n=7)	.Had another child (n=5) .No child (n=2)	No No

*Sterilization during or after treatment of childhood leukemia/lymphoma.

interview and the questionnaire differed ($n=5$), findings from the interview were used for further analysis.

Psychosocial functioning of parents was assessed by using questionnaires measuring four problem categories parents experience when coping with childhood cancer as described by a conceptual model on coping with stress (8). These problems categories are: (a) uncertainty (b) loss of control (c) threat to the self-esteem, and (d) negative feelings, including anxiety, fear, sleep disturbances, depression, loneliness, and psychological and physical distress. The reliability of these scales was adequate as measured by Cronbach's alpha (9) of respectively, .93, .64, .73, .95, .86., 84, .70, .87, and .93 (Chapter 2.3). Disease-related variables included prognostic outlook of the child at the moment of diagnosis and late medical sequelae. Late medical sequelae were measured by an adaptation of the residual physical impairment index (10) consisting of a sum of ratings of current visibility of physical residua, interference with daily living, invisible impairment, and school performance.

RESULTS

Seventy-five percent ($n=97$) of parents reported no change in their family planning (Table 2). However, one or both parents ($n=33$; 25%) in one third of the families ($n=22$), reported that their procreative planning had changed as a consequence of the diagnosis and their experiences with the illness. Interviews revealed that if family planning was affected, it was an important and emotional issue for parents.

Three main stages of family planning before diagnosis were identified: completed ($n=68$), unsettled ($n=32$), and not completed ($n=30$). In each stage of family planning, changes were observed; the ensuing variations are presented in Table 2. Fifty-two percent of the parents indicated that they viewed their family as complete before diagnosis. A small proportion of this group (9%, $n=6$) had second thoughts about wanting a new baby and three of them actually had another child. These children were born several years after cancer therapy had been completed.

Parents, who before diagnosis did not resolve the issue whether or not to have another child, were most affected in their family planning. In this group, 59% ($n=19$) of the parents reported being affected, most of them ($n=12$) refraining from further progeny. Seven of these parents decided for sterilization. Although most them did not regret this decision, one parent had serious regrets at the time of the study. Another parent with an unsettled family planning before diagnosis got pregnant during the treatment period, but became so ambivalent towards a new child that she underwent an abortion, and is still uncertain how to proceed with progeny.

In parents who had not yet completed their family before diagnosis, 27% ($n=8$) described a change in family planning. Three parents refrained from further progeny.

TABLE 3. Reasons for change in family planning by parents (n=33).

Reasons	n
Reasons for refraining from progeny (n=24)[*]	
- care of the ill child prevails (including practical limitations during treatment and feelings of guilt towards the ill child)	15
- to prevent a possible suffering from a serious disease in another child	14
- a new child cannot replace the ill child	13
- to avoid the risk of being confronted with a comparable ordeal again	12
- were advised against a new child by lay people	3
- were advised against a new child by professionals	2
Reasons in favor of having a new baby (n=12)[*]	
- to enjoy a child under untroubled circumstance	11
- to consciously experience the development of a child	9
- a new child would help to divert oneself from their child's disease	6
- the family would be too small in case the child would die	6
- it would facilitate coming to term with the child's disease	3
Reasons for refraining from progeny in spite of the wish of having a new baby (n=5)	
- feels emotionally unstable to have a new child	5
- a new non-healthy baby would overburden the family	4
- did not dare after all	3
- partner did not share the same wish	1
- had no partner	1
- had received advice to refrain from further progeny by professionals	0

[*] Some parents displayed in different groups motives for a change in family planning.

Two of these parents belonged to the group of seven parents who postponed childbearing.

Parents with affected family planning (n=33)

Twenty-one mothers and twelve fathers reported that their family planning was affected. In eleven families both partners reported a change in family planning whereas in the other eleven families one partner (ten mothers and one father) reported a change. Of the parents whose family planning was affected by diagnosis and/or course of disease, 39% (n=13) had another child, 6% (n=2) were still unsettled about further progeny, and 55% (n=18) refrained from further progeny. Motives for a change in family planning are presented in Table 3. There was no relation between time since completion of treatment and change in family planning.

Parents with affected family planning were compared with parents who did not report a change in family planning. Because of the sample size of parents whose family planning was affected, no separate analyses of variance (ANOVAs) were performed on the different subgroups within the sample of affected parents. ANOVAs indicated that parents who reported a change in family planning did not differ from other parents with respect to feelings of uncertainty and self-esteem. However, they experienced significantly more loss of control ($F=9.451$; $df=1.128$; $p<.001$) and negative feelings ($F=7.345$; $df=1.128$; $p<.001$). Binomial tests showed that significantly more mothers than fathers ($p<.001$) reported an alteration in their reproductive planning. Religious affiliation was not related to a change in family planning. With respect to disease-related variables, parents of children who had a worse prognosis at diagnosis were more affected in their family planning than parents whose child had a better prognosis at diagnosis ($p<.04$), however this result failed to reach statistical significance after Bonferroni correction for the number of statistical analyses was applied. Parents whose child had school problems reported a change in family planning more often ($p<.001$). Other late effects showed no relation with a change in family planning.

DISCUSSION

Our study shows that the experience of childhood leukemia affects family planning in a substantial number of families. Parents with unsettled or uncompleted family planning prior to diagnosis had a high probability of changing their plans. The affected parents in this study explicitly indicated that this change was due to their experiences with their child's leukemia or lymphoma. However, changes about family planning may occur in every family due to various reasons such as personal maturation, sex of children, and other life experiences. After an extensive (Med-line and Psychlit) literature search we could not find normative data on the 'natural history' of family planning. Nevertheless, an affected rate of 59% in the unsettled group shows that the likelihood of a change in family planning is high when confronted with childhood leukemia/lymphoma.

Furthermore our study gave insight into the various ways in which family planning was affected. Of those parents who reported a change in family planning, more than half refrained from having any more children, even in families who perceived their family as incomplete before the diagnosis of childhood leukemia. In seven families this decisiveness is shown by sterilization. Parents argued that they wished to spare a new child the possibility of suffering and that they themselves did not want to undergo such an ordeal again. Another argument mentioned frequently was that the burden of another nonhealthy child would be too heavy. Similarly, in studies on parents with a genetic risk, the experienced burden with the disorder is a dominant factor in family planning (3,11). Our study indicated that the stress of a serious disease in a child is an important motive to refrain from further progeny and all arguments for not having another child involved elements of decreasing or avoiding future stress and misfortune. In this way the reproductive decision and birth control measures are a way of influencing events and thus can be considered as way of coping to regain control over their situation. That procreative behavior can be regarded as a coping strategy is underlined by our observation that there was a difference in feelings of control between parents who felt that their family planning had been affected and those who had not. Parents who reported that their family planning was affected experienced less control which is in line with a study (12) to the perceived controllability of mothers to prevent a birth of a high risk infant. Mothers who perceived less control in preventing this condition were less positive towards future childbearing. Apparently, because parents of a child with cancer can take no real preventive measures to avoid the occurrence of cancer in a new child, most parents refrain from further offspring.

The desire to have children is also an important factor in the reproductive decision, regardless the kind of disorder (3,11-13). This may explain why parents who have not completed their family and thus have a strong desire for children are less affected in their family planning than parents who were unsettled prior to the diagnosis. A number of parents who wanted to have another child postponed childbearing for practical reasons. These parents explained their decision by saying that the care for the ill child prevails and/or that they wish to avoid the risk that the ill child will experience negative consequences as a result of a new child in the family. Procreative behavior can also be viewed as a coping strategy to ward off feelings of guilt towards the ill child, which results in postponing future pregnancies until treatment has been completed.

Another reason for a change in family planning is that parents may want to replace a child which is expected to die (3,4). We therefore investigated whether the prognostic outlook at diagnosis affected family planning. The reproductive decision proved to be unrelated to survival chances. However, the potential fatal prognosis which is inherent in the diagnosis of leukemia had an effect on parents; some indicated that their family size would be too small in case the child should die and expressed a wish for another child. In contrast, a number of parents refraining from further offspring, argued that their ill child could not be substituted by a new child. Thus our study presents evidence that parents can also have decisive arguments *against* replacing a child.

Surprisingly, some parents of a child surviving leukemia who viewed their family as complete before diagnosis, expressed a wish to have another child. In view of the literature about replacement children (4,5), this seems to be a paradox, since there is no need to substitute a physical loss. However, as a result of their child's disease parents can experience definitive, negative changes in their outlook on life and in their relationship with their partner and/or child, which can be regarded as psychological losses (14). To cope with these losses, a new baby represents a positive experience that can compensate for or substitute the negative feelings experienced by parents in the affected group. In this way a new baby as a result of the cancer experience in parents of long-term survivors may represent a 'psychological replacement child'.

More mothers than fathers indicated that their family planning was affected. In half of the affected families there was concordance between the spouses, whereas in the other half mostly mothers reported a change in family planning. The roles of fathers and mothers seem to differ in families with fathers as being primarily involved with financial concerns and mothers as taking primarily the caretaking roles (7). Because an ill child directly pertains to the mother's role they may be more motivated to ameliorate this stress in the future. The fact that this coping strategy from a biologically point of view is directly under the mother's control may have played a part as well.

The decision-making process on progeny is a complex operation which involves conscious as well as unconscious motives (3,15). Parents indicated to be appropriately informed by physicians about the (often) negligible genetic risk for future children and that they could have another child from a medical point of view. Although information about genetic risk is an essential ingredient in the decision-making process, our study demonstrates that parents of a child with leukemia based their decision about further offspring primarily on emotional grounds. Even in parents with a genetic risk, emotional factors were viewed as more important than the magnitude of this risk (15). Because family planning can be viewed as a coping strategy to master, reduce, or tolerate the emotional impact evolving from the child's illness, the results of our study may be generalized for families with children with other malignancies. Our preliminary data on family planning in parents of children with solid tumors supports this notion. In the same way, these findings may also have implications for parents with a child with other serious diseases or disorders with no or an unknown genetic risk.

Our study reveals important emotional factors influencing the complex decision-making process on progeny in parents of a child with leukemia/lymphoma. These include the perceived burden of the illness, the feeling that the care of the ill child prevails, and the fear not being able to cope with another affected child. Because there is often no medical obstacle to refrain from having another child, these emotional factors remain concealed in the communication between physician and parents. Thus, in addition to provide medical information, the physician needs to give attention to the worries and concerns of parents with respect to their reproductive decisions. By embedding the issue of family planning in a family-based oncology service, the physician provides support by conveying an understanding of this difficult dilemma for parents of a child with cancer.

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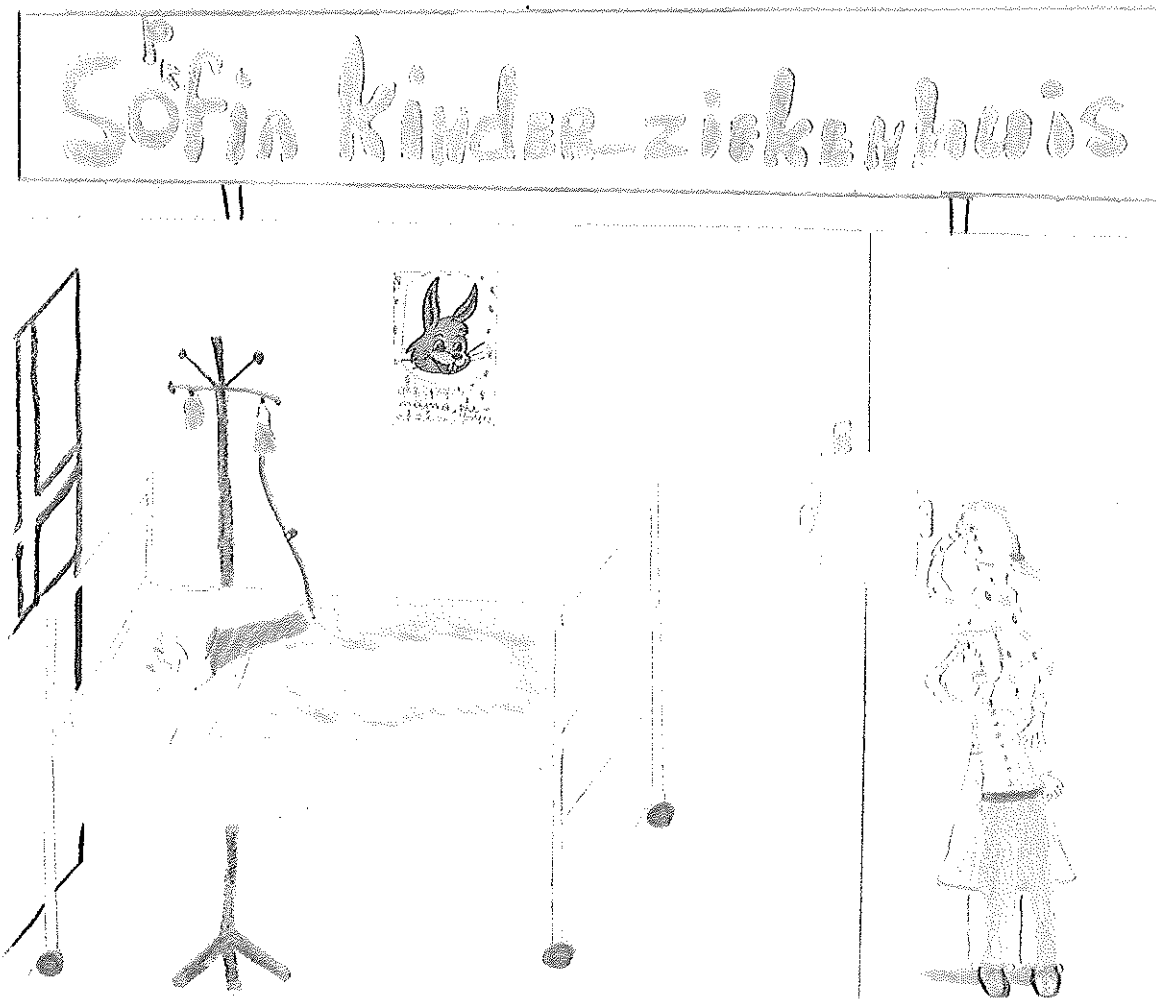
Chapter

4



Joyce, 12 yrs (sibling of a childhood cancer survivor)

Chapter 4 : Surviving childhood cancer: Impact on patients and siblings

4.1 High prevalence of learning problems in children treated for leukemia and lymphoma is exclusively related to cranial irradiation: A comparative study
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Accepted for publication in: Eur J Cancer. 231

CHAPTER 4.1

HIGH PREVALENCE OF LEARNING PROBLEMS IN CHILDREN TREATED FOR LEUKEMIA AND LYMPHOMA IS EXCLUSIVELY RELATED TO CRANIAL IRRADIATION: A COMPARATIVE STUDY*

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ABSTRACT

Prophylactic treatment of the central nervous system (CNS) in childhood leukemia and lymphoma is associated with negative effects on intelligence. We investigated the clinical significance of these findings by comparing the effect of different types of CNS prophylaxis on the survivor's learning capabilities. Over 80% of children who received cranial irradiation had learning problems, which is in sharp contrast to the 16% of children with intrathecal chemotherapy only. Similar low frequencies of learning problems were found in childhood cancer survivors without any form of CNS prophylaxis as well as in matched controls. These data underline the need to eliminate cranial irradiation from CNS prophylaxis in children with leukemia and lymphoma.

INTRODUCTION

Prophylactic treatment for early eradication of occult central nervous system (CNS) involvement in children with leukemia and lymphoma has contributed significantly to improved survival rates (1). Most initial treatment protocols with effective CNS prophylaxis contained cranial irradiation (2400 cGy or later 1800 cGy) in combination with intrathecal chemotherapy. During the last decade, cranial irradiation has been replaced in several treatment protocols by intravenous high dose methotrexate (MTX) plus intrathecal chemotherapy. This replacement was based on reports that intellectual

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and neuropsychological dysfunction was observed in survivors of childhood leukemia following CNS prophylaxis with cranial irradiation (2,3). Other reports presented, however, conflicting data which made it difficult to draw definitive conclusions about the adverse role of cranial irradiation (reviewed in 3). Apart from methodological shortcomings (reviewed in 3 and 4), most studies were based on small patient groups and only recently a large study demonstrated that leukemia patients who received cranial irradiation had lower IQ scores than children treated with intrathecal chemotherapy alone (4).

The clinical significance of lower IQ scores for the child's learning capabilities and school achievements is not clear, because IQ scores of survivors treated with cranial irradiation were still within the normal range (2-4). In addition, CNS prophylaxis with chemotherapy potentially may also have neurotoxic effects (5,6). Finally, the effect of each type of prophylactic CNS treatment on the occurrence of learning problems has not been investigated (3). Therefore, we investigated the contribution of different types of prophylactic CNS treatment to the child's learning problems and compared these results with other childhood cancer survivors who did not receive any form of CNS prophylaxis as well as with matched controls.

MATERIALS AND METHODS

Childhood cancer survivors (excluding patients with brain tumors) treated at the Sophia Children's Hospital, who were in symptom-free remission and attended grade 3-6 in primary school, were consecutively selected when they visited the outpatient clinic (compliance 96%). The cancer survivors (*N*=100) were divided into three groups: 31 children diagnosed with solid tumors treated without CNS prophylaxis, 38 children with leukemia and lymphoma treated with CNS prophylaxis consisting of MTX intrathecally and intravenously, and 31 children with leukemia and lymphoma treated with CNS prophylaxis including 1800 cGy or 2400 cGy cranial irradiation. The three groups were matched with respect to gender, age at diagnosis (median 4 years and 6 months) and time since cessation of treatment (median 3 years and 9 months), because girls and younger children are more at risk of neuropsychological sequelae, which may only become apparent several years after CNS treatment (3-5). There were no significant differences between the groups with respect to race and age at assessment. Children surviving cancer were compared with a control group (*N*=265), matched for race, gender, age, and social economic status (SES).

Mild learning problems were ascertained, if children attending regular school had learning problems as reported by parents and teachers and if these problems were confirmed by psychological assessment and/or the necessity to receive individual tutoring or special classes. Not included in this category were school problems associated with emotional or behavioral difficulties and repeating grades due to missing classes during treatment or revalidation.

Serious learning problems were ascertained, if children were not able to benefit from regular education, even with extra help, and were referred to special schools for the learning disabled. In The Netherlands, children are only admitted to these special schools after extensive psychological and didactic selection procedure (including IQ testing). The severity of their learning problems has to be validated by a committee of educational specialists.

RESULTS AND DISCUSSION

The group of childhood cancer survivors treated with CNS prophylaxis had significantly more learning problems than those treated without CNS prophylaxis (45% versus 16%; $p<.001$; z-test two-tailed probability). This high rate of learning problems in children treated with CNS prophylaxis is similar to that reported in other studies (3,6-9), in which educational problems were detected in 33% to 50% of patients receiving CNS prophylaxis. However, these studies did not evaluate the consequences of different types of CNS prophylaxis. Our results show that the high proportion of learning problems was *exclusively* associated with cranial irradiation. Over 80% of children with cranial irradiation had learning problems against 16% of children without cranial irradiation ($p<.000$; z-test two-tailed probability), indicating that cranial irradiation results in a 5-fold increase of learning problems as compared to CNS prophylaxis without cranial irradiation. This increase concerned both mild learning problems and serious learning problems (Table 1).

Remarkably, children treated for leukemia or lymphoma receiving CNS prophylaxis without cranial irradiation did not show more learning problems than childhood cancer survivors without any form of CNS prophylaxis. Furthermore, both patient groups did not differ from the matched control group. Compared to the matched control group, childhood cancer survivors with cranial irradiation had a 10-fold increase of serious learning problems (Table 1). These findings provide extra evidence that the negative effects of CNS prophylaxis on the child's learning abilities must be attributed to cranial irradiation.

It has been shown that CNS prophylaxis without cranial irradiation has equivalent or even better survival rates than those with cranial irradiation (1,10). Therefore, the dramatically increased prevalence of learning problems in children treated for leukemia and lymphoma with cranial irradiation emphasizes once again the necessity to eliminate prophylactic cranial irradiation from current treatment protocols. One should even consider to eliminate cranial irradiation from treatment protocols for leukemic children with initial CNS disease or CNS relapse where cranial irradiation still is the mainstay of treatment. Therefore we initiated a study to treat young children with initial CNS involvement and children with isolated CNS relapse with intraventricular chemotherapy only. The first experiences with this type of CNS treatment seem to be promising.

TABLE 1. Relative frequencies of learning problems in childhood cancer survivors and control groups.

	Childhood cancer survivors (*N*=100)				Controls	
	With CNS prophylaxis[a]			Without CNS prophylaxis[b] (*n*=31)	Matched control group[c] *N*=265	Dutch population[d]
	Total group (*n*=69)	Cranial irradiation (*n*=31)	No cranial irradiation (*n*=38)			
Serious learning problems	22% *(n=15)*	39% *(n=12)*	8% *(n=3)*	10% *(n=3)*	4% *(n= 11)*	5%
Mild learning problems	23% *(n=16)*	42% *(n=13)*	8% *(n=3)*	6% *(n=2)*	13% *(n=35)*	NA
Total learning problems	45% *(n=31)*	81% *(n=25)*	16% *(n=6)*	16% *(n=5)*	17% *(n=46)*	NA

Abbreviations used: CNS: central nervous sustem; NA: not available.

[a] Leukemias and lymphomas

[b] Solid tumors excluding lymphomas and brain tumors

[c] Matched controls with respect to race, sex, age, and socioeconomic status (SES).

[d] Data 1994/1995 from the Centraal Bureau voor de Statistiek (Dutch Central Office for Statistics), The Hague, The Netherlands.

Our data clearly demonstrate the deleterious effects of CNS irradiation on the child's learning abilities. We hope that this report will stimulate the development and clinical evaluation of alternative treatment modalities to improve the child's quality of life after cessation of therapy.

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CHAPTER 4.2

PSYCHOSOCIAL FUNCTIONING OF CHILDREN SURVIVING CANCER DURING MIDDLE CHILDHOOD*

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ABSTRACT

Psychosocial functioning of 95 8- to 12-year-old children who survived cancer was compared with that of healthy peers using parent and child reports. The total group of children who survived cancer showed more social problems and internalizing problems than matched peers. Although the majority of cancer survivors adjust well, a considerable increase in serious adjustment problems found, especially in boys: 27% of male survivors had serious problems as compared to 10% of healthy peers. For female survivors a non-significant increase in serious adjustment problems was found. Our results show that childhood cancer increases the risk of serious adjustment problems. The current view that childhood cancer survivors are not different from healthy peers needs to be reconsidered.

INTRODUCTION

Surviving childhood cancer is a relatively new phenomenon. As a result of the improvement in medical treatment, at this moment between 60% and 70% of all children with a malignancy will survive. The ultimate goal of current cancer treatment is not only to save the child's life, but also to save the quality of this survival (1). For several reasons the child surviving cancer may be at risk for developing psychosocial problems. Firstly, even though treatment has been terminated there still is the threat that the illness will recur and that the child may die of cancer. This chronic uncertainty about relapse and survival has been labeled *The Damocles Syndrome* (2). Secondly, the nature and duration of stresses during treatment such as physical discomfort, bodily

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changes, hospitalizations, and changes in family functioning may result in posttraumatic stress reactions (3). Thirdly, the child will be confronted with new problems associated with the re-entry into the 'pre-illness' lifestyle (4). This transition from the patient role to the 'healthy status' may be difficult, because the child may be treated differently in the family and society. Finally, physical sequelae evolving from cancer and its treatment, including growth retardation, neuropsychological deficits, sterility, and bodily disfigurements (5) may affect psychosocial functioning after treatment is terminated.

Studies concerning the long-term psychosocial consequences of childhood cancer have yielded mixed results. Early epidemiological surveys found few emotional problems among the survivors and concluded that they made excellent adjustment even surpassing their premorbid expectations (6,7). These studies have been criticized, because mental health was not assessed directly but inferred from global sociological measures, such as level of education, employment, and marital status (8). One of the first in-depth studies on the psychosocial functioning of a large number of survivors was conducted by Koocher and O'Malley (2,9). They found that 59% of their former patients had at least mild psychiatric symptoms, such as depression, anxiety, problems of self-esteem, and fluctuations in mood. However, Koocher and O'Malley (2,9) did neither provide normative data nor psychometric data for the psychiatric interview they used to classify the adjustment of long-term survivors.

Since then most studies employed standardized assessment instruments to measure the impact of childhood cancer on the survivors' psychosocial functioning. A part of these studies reported overall good adjustment and few differences between cancer survivors and controls or normative samples were found (10-17). Because of these findings and on the basis of their own study, Kazak and Meadows (17) concluded that there is sufficient evidence to accept the null hypothesis that children surviving cancer are not different from healthy children. In contrast, others have documented emotional and adjustment difficulties in childhood cancer survivors including depression, poor self-esteem, problems in social competence, and an increased rate of somatic concerns and learning difficulties (18-23). These conflicting findings started a debate about the nature and the severity of psychosocial consequences for childhood cancer survivors.

Unfortunately, almost all existing studies on the late effects of childhood cancer have methodological shortcomings (3,10,17,24,25), such as heterogeneity of the sample by including children both on and off treatment and including subjects across a wide age range, nonrepresentative samples (low compliance or not reported compliance), small sample size, lack of adequate comparison groups, lack of multiple informants to assess psychosocial functioning, poor psychometric properties of the instruments, and flaws in statistical analysis. In addition, none of the studies has adequately evaluated the severity of psychosocial problems using standardized instruments, i.e. whether problems of cancer survivors are normal variations or clinically significant problems.

Consequently, it is not clear whether childhood cancer survivors are at risk for psychosocial maladjustment.

Furthermore, it is important to identify factors related to late psychosocial consequences of childhood cancer. The risk of psychosocial problems has been found to increase by factors related to the physical outcome of treatment, such as functional impairment (20) and severity of late physical sequelae (12). However, the role of different characteristics related to the disease (onset, diagnosis, and course), types of treatment (chemotherapy, surgery, radiotherapy), and intensity of treatment has received little attention (26,27). Kazak (26) and Armstrong (27) advocate to test the specific characteristics of disease and treatment in long-term survivors to elucidate their role in long-term adjustment.

Therefore, we designed an optimal study to investigate the nature of late consequences of childhood cancer, to identify the factors related to these late consequences, and to determine the severity of the late consequences. Firstly, a large, representative, and homogeneous sample of 95 children was recruited. All children were off treatment and were within a particular developmental stage, i.e. middle childhood. This stage was chosen, because of its neglect in past research, which focused predominantly on adolescents and adults surviving childhood cancer. Secondly, the psychosocial functioning of children surviving cancer was compared with a matched comparison group of 95 healthy peers. Thirdly, both parents and children were used as informants, because each type of informant may have a different but valid view in assessing psychosocial functioning (28,29). Fourthly, we explored the relation between risk factors and adjustment to childhood cancer. For this purpose we adapted the conceptual framework by Wallander et al. (30) that classifies two major clusters of risk factors, namely disease related variables and psychosocial stressors. Finally, we wished to investigate the severity of problems and used an assessment instrument with documented satisfactory reliability and validity for this purpose (31,32).

MATERIALS AND METHODS

Subjects

Children with a malignancy and meeting the following criteria were included in the present study: a) successful termination of cancer treatment, b) 8 to 12 years old (5 children who were slightly older than 12 but attended elementary school were included), and c) whose native language was Dutch. They were all treated at the Sophia Children's Hospital, one of the four pediatric cancer centers in the Netherlands. All eligible children with leukemia and lymphoma were approached. Children with a solid tumor were consecutively selected according to the same criteria. These latter patients resembled the children with lymphoproliferative diseases with respect to prognosis, treatment protocol, difficulty of treatment, length of treatment, and late

TABLE 1: Disease-related characteristics of long-term survivors of childhood cancer (N=95; male n=59, female n=36).

Variables related to disease:			Variables related to treatment:			Variables related to long-term sequelae:		
Age	median	range	*Intensity of treatment protocol :*			*Time off treatment:*	%	*n*
- age at diagnosis	4y 8mo	0y 8mo-12y 0mo		%	*n*	≤2 year	32	30
			- mild	21	20	2 year - 5 year	42	40
			- intermediate	43	41	≥5 year	26	25
			- severe	36	34	*Invisible impairment:*		
Diagnosis:	%	*n*				- yes	24	23
- Leukemias and lymphomas	77	73	*Duration of treatment:*			- no	76	72
- Solid tumors	23	22	- <2 year	39	37	*Visible impairment:*		
			- 2-3 year	46	44	- yes	40	38
Relapse:			- ≥ 3 year	15	14	- no	60	57
- yes	7	7				*Overweight:*		
- no	93	88	*Surgery:*			- yes	33	31
			- yes	35	33	- no	67	64
			- no	65	62	*School problems:*		
Survival chances at diagnosis:			*Cranial irradiation:*			-no problems (regular school)	63	60
≤ 50%	51	48	- yes	37	35	-mild problems (regular school plus school problems)	18	17
> 50%	49	47	- no	63	60	-severe problems (special school)	19	18

medical sequelae. Parents of 99 children were asked to participate and to give written informed consent. Four families (4%) did not participate because parents of three children feared that the study would be emotionally upsetting to the child and one mother insisted to be present at the child's assessment. The sample consisted of 62% (n=59) boys and 38% (n=36) girls with a median age of 10 years and 3 months (range: 8 years and 4 months to 13 years and 9 months). The mean social economic status (SES) of the parents, rated according to Dutch standards (33) on a six step-scale of occupational level (highest occupational level=6), was 3.26 (missing information on 2 families).

To compare the parents' report of children surviving cancer with healthy peers, a subsample of 95 children were randomly selected from a normative sample (n=2033) of the general population (see for sample selection and data collection procedure 34) matched for gender, age, and SES. To compare the child reports, a sample of 90 children was randomly selected from healthy children (n=300) from two local elementary schools and matched for gender and age (for sample selection and data collection procedure, see 35).

Disease-related and psychosocial risk factors

Disease-related variables are presented in Table 1. Survival chances were based on the oncologist's rating of the child's statistical survival chances based on the diagnosis, symptoms at presentation, and the success rates of the treatment protocols at that time. Intensity of therapy protocol was rated according to the treatment protocol. Invisible impairment included invisible medical problems such as organ damage and endocrine problems. Visible impairment was partly measured by the residual physical impairment index (2), consisting of the sum of ratings of current visibility and interference with daily living. Overweight was defined as the child's weight at or above the 90th percentile corresponding to Dutch standards (36). School problems were rated as present if the child received remedial teaching or was not able to attend a regular school. Psychosocial stressors included concurrent stresses in the family unrelated to childhood cancer and a chronic disease or chronic condition in one of the other family members. These stresses were present in 29 and 18 families out of 70 families, respectively (missing information on 25 families).

Procedure

All instruments were individually administered to survivors. Parents were asked to complete a questionnaire while their child was being evaluated. Although five survivors did not participate personally (three were mentally retarded and two refused), their parents' reports were included.

Instruments

Child Behavior Checklist (CBCL) (31). The CBCL for ages 4-18 is a standardized procedure to obtain parents' reports of their child's behavioral and emotional problems. The CBCL has been translated into Dutch, and its good reliability and discriminative validity were replicated in Dutch studies (29,34). The problem section of the CBCL consists of 120 items. Parents are requested to circle a 0 if the problem item was not true for the child, and a 1 or a 2 if it was sometimes true or very true, respectively. Total problem scores were computed by the scores on each item. High scores reflect a high degree of problems. Achenbach (31) empirically constructed eight syndromes and their generalizability for Dutch children has been demonstrated (37). Five syndromes could be grouped under two broad band scales: Internalizing and Externalizing. Internalizing problems reflect internal distress, whereas externalizing problems reflect conflicts with other people and their expectations of the child. The internalizing group consists of the following syndromes: Anxious/Depressed, Somatic Complaints and Withdrawn. The externalizing group consists of Aggressive Behavior and Delinquent Behavior.

Self-Perception Profile for Children (SPPC) (38). The SPPC is a 36 item questionnaire measuring self-concept or perceived competence in 8- to 12-year old children. The SPPC contains six subscales, tapping a) scholastic competence, b) social acceptance, c) athletic performance, d) physical appearance, e) behavioral conduct, and f) general self-worth. Each subscale contains six items on which the child is asked to rate its competence on a 4 point Likert scale. Subscale scores range from 6 to 24 and a higher score is associated with higher perceived competence. The Dutch version of the SPPC had alpha coefficients and test-retest reliabilities for the six subscales ranging from .65 to .81 (mean .76) and from .66 to .83 (mean .76), respectively. Internal validity of SPPC was shown by the replication of the multi-factorial structure by exploratory and confirmatory factor analysis (35).

Amsterdam Biographic Questionnaire for Children (ABV-K) (39). The ABV-K is a forced choice (yes/no) personality questionnaire for 9- to 17-year old children measuring a) Neurotic Complaints (30 items), c) Somatization (20 items) d) Extroversion (20 items), and d) Testing-attitude (16 items). The Neurotic Complaints-scale measures psychoneurotic complaints with descriptive statements such as "I worry a lot" and "Deep down I am scared for a lot of things"; the Somatization-scale measures somatization of neurotic complaints, with items such as "I often have a headache" and "My belly often aches"; the Extroversion-scale measures the child's position on the dimension introversion-extroversion with descriptive statements such as "I feel self-conscious" and "When being with other children, I stay in the background"; and the Testing-attitude-scale measures social desirability with items such as: "I lie sometimes", "I only have good thoughts about other people". The total scores on each subscale reflect's the individuals position on a continuum ranging from 0-100. For the four scales Cronbach's alpha were respectively .82, .72, .75, .68 for boys and .83,

.76, .74, .73 for girls and test-retest reliabilities ranged from .74-.80 (mean .77) for boys and from .76-80 (mean .78) for girls (39). Correlations between the subscales of the ABV-K and the Eysenk Personality Questionnaire-Revised-Junior were: .70 between both Neurotic Complaints subscales, .36 between both Extroversion subscales, and .60 between the Testing-attitude scale and Lie subscale. For the Somatization-scale a correlation of .70 was found with the Neurotic Complaints scale of the ABV-K (40).

Children's Depression Scale (CDS) (41). The CDS is a 63 item self-report scale to asses depression in 9- to 15-year old children. Children sort cards with descriptive statements on a five-point Likert scale. Scores range from 63-315 and high scores indicate a higher level of depression. Rotundo and Hensley (42) found a Cronbach's coefficient alpha of .98 for the CDS and a correlation of .84 between the CDS and the Children's Depression Inventory.

Data analysis

The impact of cancer was studied by comparing the mean CBCL, SPPC, ABV-K, and CDS total and scale scores of the patient and the comparison groups using a series of one-way analyses of variance (ANOVA). Data analyses were performed for the total group and for each gender separately. To determine differences in the seriousness of problems between the patient groups and the comparison groups, binomial tests were performed to compare the percentages of children in each group scoring above the 90th percentile of scale-scores on the CBCL, the SPPC, the CDS, and the ABV-K. To investigate the effects of risk factors on reported psychosocial problems, we performed two types of analyses. First, we used ANOVAs to compare the mean problem scores of survivors with a particular risk factor with those survivors without that risk factor. Second, we separated the problematic from the non-problematic survivors using the 90th percentile cutoff point of scale scores, and investigated differences in the presence of risk factors between the two groups.

Because of the explorative nature of our study, a significance level of $p < .05$ was applied. Using this value, priority is still given to avoiding type I errors rather than to avoid type II errors (43,44). To avoid a further increase in the chance of failing to find phenomena that exist (type II errors), no corrections for chance findings were made.

RESULTS

Nature of problems

Comparisons of psychosocial problem scores of survivors and their healthy comparisons are presented in Table 2. There were significant differences between the total sample cancer survivors and their healthy peers on nine problem scales. Parents scored cancer survivors as having significantly more problems (mean Total Problems

TABLE 2. Mean scale scores for children surviving cancer (N=95) and their healthy controls (N=95) and results of tests of differences in mean scores in ANOVAs.

	Total sample				Boys				Girls			
Scale	Cancer survivors	Healthy children	*p*	Effect size[a]	Cancer survivors	Healthy children	*p*	Effect size[a]	Cancer survivors	Healthy children	*p*	Effect size[a]
CBCL (parent)	N=95	N=95			n=59	n=59			n=36	n=36		
-Withdrawn	2.9	1.8	.005	4.7	3.2	2.0	.05	5.3	2.3	1.6		
-Somatic Complaints	1.2	0.7	.05	3.1	1.4	0.7	.05	3.5	1.0	0.6		
-Anxious/Depressed	2.9	2.5			3.1	2.9			2.6	1.8		
-Social Problems	2.7	1.5	.001	6.0	3.1	1.6	.005	8.2	2.0	1.4		
-Thought Problems	0.5	0.2	.01	3.5	0.5	0.2	.05	4.0	0.4	0.2		
-Attention Problems	4.0	3.5			4.6	3.9			3.1	2.8		
-Delinquent Behavior	1.4	1.0			1.8	1.2			0.8	0.7		
-Aggressive Behavior	6.2	5.7			7.7	6.8			3.6	4.1		
-Internalizing	7.0	4.9	.05	3.1	7.6	5.4	.05	3.1	5.9	3.9		
-Externalizing	7.6	6.7			9.5	7.9			4.4	4.8		
-Total Problems	24.5	19.4	.05	2.2	28.4	21.9	.05	3.3	17.9	15.4		

(continued) →

TABLE 2 (continued).

Scale	Total sample				Boys				Girls			
	Cancer survivors	Healthy children	*p*	Effect size[a]	Cancer survivors	Healthy chidden	*p*	Effect size[a]	Cancer survivors	Healthy children	*p*	Effect size[a]
SPPC (child)	*N*=90	*N*=90			*n*=58	*n*=58			*n*=32	*n*=32		
-Scholastic Competence	17.1	17.1			17.6	18.2			16.0	15.0		
-Social Acceptance	17.6	18.6			17.4	19.4	.05	4.4	18.0	17.5		
-Athletic Competence	16.9	18.5	.005	4.4	17.5	19.7	.001	10.2	15.7	16.4		
-Physical Appearance	20.2	18.8	.05	2.4	20.6	19.9			19.6	16.7	.01	9.5
-Behavioral Conduct	17.9	17.6			17.5	17.7			18.5	16.9	.05	6.4
-General Self-worth	20.4	19.8			20.4	20.6			20.3	18.5		
ABV-K (child)	*N*=90	*N*=90			*n*=58	*n*=58			*n*=32	*n*=32		
-Neurotic Complaints	47.3	48.1			50.0	46.1			43.2	51.7		
-Somatization	53.8	56.3			57.4	54.4			47.4	60.0		
-Extroversion	48.8	63.9	.001	6.1	46.3	67.7	.001	11.6	53.1	57.2		
-Social Desirability	53.5	51.8			53.7	54.1			52.3	47.9		
CDS (child)	*N*=90	*N*=30										
-Total Score	151.9	147.5										

[a] Percentage of explained variance.

TABLE 3. Influence of risk factors on mean scale scores of psychosocial functioning of boys surviving cancer (n=59).

		Parent report						Child report		
Risk-factors	*n*	With-drawn	Somatic Complaints	Social Problems	Thought Problems	Internal-izing	Total Problems	Social Acceptance	Athletic Competence	Extro-version
Diagnosis: leukemia/solid tumor	46/13	3.4/2.5	1.4/1.2	3.2/3.0	0.6/0.3	7.8/7.0	28.5/28.1	16.7/19.6	17.4/18.2*	44.3/50.9
Age at diagnosis: <4yrs/≥4yrs	19/40	2.3/3.6	0.8/1.6	3.4/3.0	0.4/0.6	5.5/8.6	23.6/31.0	15.5/18.2*	16.9/17.8	38.2/49.0
Prognosis at diagnosis: ≤50%/>50%	30/29	3.2/3.2	1.3/1.4	4.2/2.0***	0.5/0.6	7.4/7.9	30.8/26.0	16.4/18.2	17.0/18.1	44.8/44.9
Severe treatment: yes/no	20/39	3.0/3.3	0.9/1.6	3.0/3.2	0.6/0.5	7.6/7.6	29.7/27.8	18.7/16.8	18.3/17.2	53.4/42.0
Duration of treatment: <3 yrs/≥3yrs	52/7	3.1/3.7	1.3/1.7	2.8/5.3*	0.5/0.7	7.7/7.4	28.0/31.7	17.8/13.5*	17.9/15.2*	47.8/29.
Surgery: yes/no	21/38	2.9/3.4	1.1/1.5	2.8/3.3	0.5/0.6	7.3/7.8	29.1/28.1	18.2/16.8	17.6/17.5	48.5/44.3
Cranial irradiation: yes/no	21/38	3.3/3.1	1.2/1.4	4.4/2.4*	0.6/0.5	6.7/8.1	27.6/28.9	15.3/18.5*	16.9/18.0	33.6/52.5*
Time off treatment: < 5yrs/≥5yrs	47/12	3.3/2.6	1.5/1.0	2.7/4.6	0.5/0.6	7.9/6.3	28.9/26.7	17.9/15.0*	17.9/16.3	48.5/35.7
Invisible impairment: yes/no	14/45	3.4/3.1	1.1/1.4	2.6/3.3	0.3/0.6	7.1/7.8	25.6/29.3	17.3/17.4	16.9/17.8	39.3/47.8
Visible impairment: yes/no	22/37	2.5/3.6	1.1/1.5	3.3/3.0	0.4/0.6	6.3/8.4	28.1/28.6	17.9/17.0	17.1/17.9	43.9/47.0
Overweight: yes/no	18/41	4.2/2.8	1.4/1.3	4.0/2.7	0.6/0.5	9.2/7.0	30.3/27.6	15.8/18.0	17.4/17.7	39.4/48.3
School problems: yes/no	21/38	3.1/3.2	1.1/1.5	4.6/2.3***	0.5/0.6	6.9/8.0	30.6/27.3	16.1/18.0	18.0/17.4	43.3/47.1
Age at investigation: <11yrs/≥11yrs	32/27	3.6/2.7	1.1/1.6	2.3/4.0*	0.5/0.5	8.0/7.1	27.6/29.4	17.8/16.8	18.5/16.5*	42.1/50.0
SES: low (≤3)/high (>3)	31/27	3.5/2.9	1.2/1.5	3.2/3.1	0.6/0.5	8.0/7.4	30.3/26.5	17.5/17.2	18.1/16.9	47.5/43.8
Concurrent stresses: yes/no	20/22	3.7/2.6	2.1/1.0	4.0/2.7	1.0/0.27**	10.1/5.8	34.3/22.9*	15.8/17.4	17.3/17.6	40.1/52.1
Chronic disease: yes/no	10/32	4.1/2.9	2.3/1.3	3.8/3.2	1.3/0.4***	11.6/6.7	39.6/24.8*	14.9/17.2	16.9/17.6	46.0/46.5

Note: Table entries indicate mean scale scores; on the left side the mean scale score corresponds with the risk factor on the left, on the right side the mean scale scale score corresponds with the risk factor on the right.
* .05; ** .01; *** $p < .005$

was 24.5 versus 19.4; $p<.05$). Survivors were rated as having more problems than peers on the CBCL scales Withdrawn ($p<.005$), Somatic Complaints ($p<.05$), Social Problems ($p<.001$), Thought Problems ($p<.01$), and Internalizing ($p<.05$). On the SPPC, cancer survivors scored significantly lower on Athletic Competence ($p<.005$), and higher on the Physical Appearance ($p<.05$) scale than their healthy peers. On the ABV-K Extroversion subscale their scores were significantly lower ($p<.001$). Separate analysis for gender showed that on most scales boys surviving cancer had more psychosocial problems than comparable healthy boys, but girls did not differ significantly from girls in the comparison groups (Table 2). Parents scored male survivors as having more problems than healthy boys on the CBCL syndromes: Withdrawn ($p<.05$), Somatic Complaints ($p<.05$), Social Problems ($p<.005$), Thought Problems ($p<.05$), Internalizing ($p<.05$) and on Total Problems ($p<.05$). Boys surviving cancer scored themselves on the SPPC as less Socially Accepted ($p<.05$) and less Athletic Competent ($p<.001$). On the ABV-K their scores on the Extroversion scale were significantly lower ($p<.001$) than healthy boys. Parents scored girls surviving cancer as having slightly more problems, but differences were not significant. On the SPPC, girls surviving cancer scored themselves significantly higher on the Physical Appearance scale ($p<.01$), and on the Behavioral Conduct scale ($p<.05$) than girls in the comparison group.

To explore whether these marked differences between boys and girls could be attributed to differences in sample characteristics, chi-square tests ($df=1$) were performed to compare male and female survivors on demographic, psychosocial, and disease-related characteristics and long term sequelae. The only significant differences we found, were that more boys than girls were older than 5 years at diagnosis ($p<.01$), had a higher SES ($p<.01$), and were longer off treatment ($p<.05$).

Relation between risk factors and psychosocial adjustment

To determine which risk factors were associated with significantly elevated problem scores for boys and girls, separate ANOVAs were performed for male and female cancer survivors, in which we compared survivors with each other using demographic, psychosocial, disease-related characteristics, and long-term sequelae as independent variables and the deviating psychosocial problem scores as dependent variables. Consequently, for boys we tested the risk-factors in relation to the CBCL scales Withdrawn, Somatic Complaints, Social Problems, Thought Problems, Attention problems, Internalizing Behavior and Total Problems, the ABV-K Extroversion scale and the SPPC Social Acceptance and Athletic Competence scales. For girls, the dependent variables were the SPPC Physical Appearance and Behavioral Conduct scales.

Table 3 shows that for boys, these independent variables had an effect on six areas of psychological functioning. Parents reported significantly higher scores on the CBCL Social Problems scale for boys who had a survival chance at diagnosis $\leq$ 50% ($p<$

TABLE 4. Influence of risk factors on mean scale scores of psychosocial functioning of girls surviving cancer ($n = 32$).

		Child report	
Riskfactors	n	Physical Appearance	Behavioral Conduct
Diagnosis: leukemia/solid tumor	24/8	19.0/21.4	18.4/18.8
Age at diagnosis: <4yrs/≥4yrs	20/12	19.5/19.8	18.3/18.9
Prognosis: ≤50%/>50%	15/17	19.3/19.8	19.3/17.8
Severe treatment: yes/no	13/19	18.3/20.5	17.5/19.2
Duration of treatment: < 3 yrs/ ≥3 yrs	26/6	20.2/17.2	18.4/19.0
Surgery: yes/no	11/21	19.6/19.5	17.6/19.0
Cranial irradiation: yes/no	11/21	18.4/20.2	20.0/17.7
Time off treatment: <5yrs/≥5yrs	20/12	18.7/21.1	18.2/19.1
Invisible impairment: yes/no	8/24	19.4/19.7	20.0/18.0
Visible impairtment: yes/no	15/17	20.1/19.2	19.4/17.7
Overweight: yes/no	11/21	16.4/21.3***	17.6/19.0
School problems: yes/no	16/16	18.1/20.4	19.7/17.9
Age at investigation: <11yrs/≥11yrs	16/16	19.7/19.5	17.4/19.6
SES: low (≤3)/high (>3)	22/9	19.0/20.8	18.3/18.4
Concurrent stresses: yes/no	9/15	17.8/20.1	18.0/19.1
Chronic disease: yes/no	6/18	22.0/18.3	20.3/18.2

Note: Table entries indicate mean scale scores; on the left side the mean scale score corresponds with the risk factor on the left, on the right side the mean scale scale score corresponds with the risk factor on the right.
***$p < .005$

.005), had received treatment ≥ 3 years ($p < .05$), were cranially irradiated ($p < .05$), had school problems ($p < .005$) and whose age at investigation was ≥ 11 years ($p < .05$). Parents reported higher scores on the CBCL Thought Problems scale when there were concurrent stresses ($p < .01$) or a chronic disease in one of the other family members ($p < .005$). A mean elevation in CBCL Total Problem scores of 11 and 15 points was found for boys who came from families with respectively concurrent stresses ($p < .05$) and chronic disease ($p < .05$). Boys reported themselves as being less Socially Accepted when their age at diagnosis was < 4 years ($p < .05$), when they were treated ≥3 year ($p < .05$), were cranial irradiated ($p < .05$), or when they were more than five years off treatment ($p < .05$). We found less favorable Athletic Competence scores for boys who were diagnosed with leukemia, were treated ≥ 3 year ($p < .05$) or were ≥ 11 years at investigation ($p < .05$). Furthermore, boys who were cranial irradiated had lower ABV-K Extroversion scores than non-irradiated survivors.

For girls we found only one risk factor which was significant related to psychosocial problems, namely overweight (Table 4). Girls who had weight problems scored themselves lower on the SPPC scale Physical Appearance ($p < .005$).

Risk factors were combined into three major risk groups: (a) demographic and psychosocial factors (including age ≥ 4 at diagnosis, age ≥ 11 at investigation, low SES, concurrent stresses and another chronic disease in the family); (b) disease-related factors (including prognosis ≤ 50%, severe treatment, treatment ≥ 3 years, surgery and irradiation); and (c) long-term sequelae (including <5 years from treatment, invisible impairment, visible impairment, school problems, and overweight). We computed sum scores for the three major risk groups and a total risk score and tested if these four risk scores were more accumulated in male cancer survivors than in female cancer survivors. No significant differences in mean total risk score between boys and girls were found. Next, we compared problematic and non-problematic boys on the four risk sum scores. We found that all mean risk scores, except demographic and psychosocial related factors, were higher for boys having deviant scores on the CBCL Social Problems scale (respectively $p < .05$, $p < .05$, $p < .001$ and variance explained 7.0%, 10.0% and 19.6%). Demographic and psychosocial risk factors were more accumulated for boys scoring above the cut-off point on the CBCL syndromes: Somatic Complaints ($p < .005$; explained variance 13.7%), Thought Problems ($p < .001$; explained variance 17.1%), Attention Problems ($p < .05$; explained variance 7.4%), and the Total Problem score ($p < .05$; explained variance 10.7%). The total risk score was significantly higher for boys scoring in the problematic range on the CBCL scale Attention Problems ($p < .01$; explained variance 12.4%) and for the Total problem score ($p < .05$; explained variance 10.7%). For girls scoring above the cut-off point on the CBCL scales Withdrawn, risk-factors related to long-term sequelae were more accumulated ($p < .01$; variance explained 17.9%).

TABLE 5. Percentage of boys (*n*=59) and girls (*n*=36) surviving cancer and their healthy controls exceeding cut-off scores.

	Boys > Cut-off (%)			Girls > Cut-off (%)		
Scale	Cancer survivors	Healthy chidren	Binomial test P	Cancer survivors	Healthy children	Binomial test P
CBCL (parent)	*n*=59	*n*=59		*n*=36	*n*=36	
-Withdrawn	16.9	10.2		25.0	2.8	.001
-Somatic Complaints	22.0	10.2	.01	25.0	11.1	.05
-Anxious/Depressed	10.2	10.2		16.7	11.1	
-Social problems	27.1	10.2	.001	19.4	11.1	
-Thought problems	15.3	5.1	.005	22.2	11.1	
-Attention problems	20.3	10.2	.05	16.7	13.9	
-Delinquent behavior	18.6	10.2		5.6	2.8	
-Aggressive behavior	15.3	8.5		11.1	8.3	
-Internalizing	16.9	10.2		27.8	11.1	.005
-Externalizing	18.6	6.8	.001	13.9	11.1	
-Total Problems	27.1	10.2	.001	13.9	11.1	
SPPC (child)	*n*=58	*n*=58		*n*=32	*n*=32	
-Scholastic Competence	8.9	12.1		3.1	6.3	
-Social Acceptance	17.9	8.6	.05	3.1	9.4	
-Athletic Competence	17.9	6.9	.005	0	9.4	
-Physical Appearance	5.4	10.3		6.3	9.4	
-Behavioral Conduct	5.4	8.6		3.1	9.4	
-General Self-worth	14.3	10.3		3.1	9.4	
ABV-K (child)	*n*=58	*n*=58		*n*=32	*n*=32	
-Neurotic Complaints	12.5	6.9		3.1	6.3	
-Somatization	7.1	8.6		6.3	9.4	
-Extroversion	12.1	8.6		10.8	12.1	
-Social Desirability	12.5	10.3		8.8	9.4	
CDS (child)	*n*=58	*n*=30		*n*=32	*n*=30	
-Total Score	5.4	10.0		5.9	10.0	

Abbreviations used: CBCL=Child Behavior Checklist; SPPC=Self Perception Profile for Children; ABV-K=Amsterdam Biographic Questionnaire for Children; CDS=Children's Depression Scale.

Severity of problems

To determine the proportion of children in the patient and in the comparison group scoring in the deviant range, the 90th percentile of the cumulative frequency distribution of the scores of the comparison groups on the different instruments was chosen as the cut-off point (for the SPPC the 10th percentile was chosen). Table 5 shows the proportions of children scoring above this cut-off point in the patient and comparison groups. These proportions were compared using binomial tests of difference (*p*=two tailed probability). More boys surviving cancer had deviant scores than boys in the comparison group on five CBCL scales, respectively Somatic Complaints (22.0% versus 10.2%), Social Problems (27.1% versus 10.2%), Thought Problems (15.3% versus 5.1%), Attention Problems (20.3% versus 10.3%), and Externalizing (18.6% versus 6.8%), as well as on Total Problems (27.1% versus 10.2%). On the SPPC Social Acceptance and Athletic Competence scales we found that 17.9% of the male survivors had deviant scores versus respectively 8.6% and 6.9% of the control boys. For girls, we found significant higher percentages of problems on three CBCL scales, respectively Withdrawn (25.0% versus 2.8%), Somatic Complaints (25.0% versus 11.1%), and Internalizing (27.8% versus 11.1%).

DISCUSSION

Our extensive study on 95 children surviving cancer during middle childhood (compliance of 96%) and 95 matched controls allowed us to identify the nature of late psychosocial problems, to get insight into the factors that affect the child's adaptation, and to determine the severity of late psychosocial problems.

Nature of problems

Children surviving cancer were more withdrawn and displayed more introvert behavior, had more somatic complaints, and showed more problems with social competence as compared to matched healthy children. Parent and child reports coincided on social problems as well as on internalizing behavior and these problems corroborate consistently with those reported in other studies (18-23). The status of social and internalizing problems as a late consequence of childhood cancer is firmly established both in our study and in the literature, but the nature of other problems such as negative feelings (neurotic complaints, depression) and somatic complaints is less clear. Discrepancies in parent and child reports on these problems found in our study give more insight into the nature of these problems.

With respect to negative feelings, the increased Total Problem scores as reported by parents was not confirmed by elevated scores of children on the ABV-K Neurotic Complaints and Depression scales. Sanger et al. (22) and Olson et al. (18) also found that children surviving cancer reported less problems than their parents. The lack of depression in children with cancer during or after treatment is frequently found (10-12,19,45-47). It has been suggested that children with cancer are repressing negative information (47,48). Social desirability, measured in our study by the ABV-K, can be viewed as an indication of repression, but was found not to be different from peers. An alternative explanation might be that in rating everyday life stresses, children surviving cancer assess the problem items on the depression scale and ABV-K differently than their healthy peers. As a result from their experience with cancer there may be a change in children's conceptualization of problems. Consequently, a change in the child's internal standard affects the perception of his of her level of functioning on the dimension being measured. As a result of this 'response shift', problems are being underreported. This 'response shift' has also been described in adults with cancer (49).

Another discrepancy between parent and child reports was that the elevated parental scores on the Somatic Complaints scale were not accompanied by elevated ABV-K Somatization scores of the children themselves. This finding seems to reject the suggestion that cancer survivors have hypochondriacal tendencies (20). The discrepancy between parent and child may be attributed to the underreporting of problems by the child due to the above described 'response shift'. However, the instrument and the parent as informant may also affect the results. With respect to the instrument, the value of the Somatic Complaints scale of the CBCL has been questioned in applying it to chronically ill children, because somatic complaints may be confounded with symptoms related to the disease (50). In principle, this critique does not apply to survivors of childhood cancer who are assumed to be disease free. Thus, increased somatic complaints as reported by parents on the CBCL are probably valid but may be explained by a heightened parental concern about the potential recurrence of the disease which cause them to be more alert to physical symptoms in the child.

Gender and adjustment to childhood cancer

Although differences in psychosocial adjustment between children surviving cancer and healthy children were demonstrated, analysis for each gender separately revealed that these differences can be attributed mainly to a poorer adjustment in male survivors. The problems in boys surviving cancer focussed mainly on internalizing behavior and social competence. Parents rated their son on the CBCL as having more internalizing behavior and more social problems. On self-report measures, boys reported to be less socially accepted by peers, less athletically competent, and less extrovert than healthy peers. The observation by Chang et al. (19) that cancer survivors exhibit passive and non-assertive behavior corroborates with our findings. Boys seem to keep themselves distant from social interaction, and feel socially and physically less competent. The picture that emerges is that these boys assert themselves less, both socially and physically. For boys, physical affirmation is considered to be important in establishing an optimal position in the social order (51). It seems that the cancer experience handicaps the male cancer survivor in the race of establishing and maintaining dominance, which leads to inequities and problems in interpersonal relationships. The fact that physical sequelae in boys increase the likelihood of social problems supports this notion.

For girls surviving cancer less psychosocial sequelae were reported than for their male counterparts. They even reported better Behavioral Conduct on the SPPC than peers. In addition, female survivors perceived themselves as more attractive than peers. This is a remarkable finding, because physical appearance is the most important factor in determining girl's sense of self-worth (35). No risk factors were found to affect the girl's self-concept except for weight problems. High self-esteem is considered a characteristic of resilient children (52) and might explain why girls show lesser problems than boys.

The question remains why the cancer experience has more psychosocial late-effects on boys than on girls. This differential sensitivity to childhood cancer is in line with the literature that before adolescence boys are more vulnerable to the exposure of stress than girls (53-54). However, little is known about the underlying mechanisms of gender differences in adaption to stress. Various suggestions related to genetic, personal, and environmental factors have been put forward (52,55). It is suggested that boys use different coping styles than girls, which may prove to be less effective in dealing with stressors (56). Another possibility is that parents employ different socialization behaviors towards boys versus girls (57). Childhood cancer may affect these parenting styles in such a way that it has a larger impact on boys than on girls. In addition, differential cultural stereotypes and norms for boys' and girls' behavior may affect the assessment of problems by parents. A study on adult perceptions of children indicates that adults have biased expectations towards children with cancer (58). Boys with leukemia were rated as adjusting less well to the future than healthy boys, whereas no such difference was found for girls. These perceptions might affect the response of adults to children with cancer, because a self-fulfilling prophecy process might be initiated by the label cancer. Such processes might affect male cancer survivors, although it has to be demonstrated whether this perceptual bias has an impact on adult behavior.

Other factors related to psychosocial adjustment

Insight into factors which may affect adjustment in children revealed relatively few individual factors that increased the risk of psychosocial problems. Except for children who received cranial irradiation, had school problems, or had a long (≥ 3 yrs) treatment course, factors related to the disease itself (diagnosis, prognosis, age at onset) and type and severity of treatment had little effect on psychosocial adjustment. Psychosocial stressors especially affected the level of general adjustment. Most effect on psychosocial functioning was found when all risk factors (see Tables 4 and 5) and those related to long-term sequelae were combined. This suggests that risk factors may be cumulative which is in line with a meta-analysis on studies of factors related to adjustment in children with physical disorders (25,59). It was shown that disease, family, and child characteristics all contribute to the child's adjustment, except for SES (59). The overall effect of risk factors on psychosocial sequelae in our study was, however, modest. This finding is interpreted that, besides gender, the cancer experience per se is the most dominating factor in explaining late problems.

Severity of problems

Another aim of our study was to investigate whether differences between cancer survivors and healthy children can be regarded as normal variations or as clinically significant problems. Our study demonstrated a considerable increase in serious adjustment problems in children surviving cancer, especially for boys. This statement is made on the basis of parental information, because they are considered to be generally the most knowledgeable observers of their child's behavior, and because we used an instrument that has demonstrated its reliability and validity for this purpose (46). For the total group, 21.1% of the survivors had serious adjustment problems, reflecting a level of problems that is typically for referral to mental health services. Interestingly, this proportion of children with adjustment problems corroborates with the Koocher and O'Malley study (2), in which 21.4% of survivors displayed moderate to marked impairment in their overall functioning.

Traditionally, it has been assumed that psychopathology is a common response to chronic illness in childhood, but this could not be substantiated by empirical research (60). Probably as a reaction to this assumption, the current view (over)emphasizes that there are few differences between children surviving cancer and healthy peers (10-17,61). Although we also found that the majority of survivors adjust well, thereby underscoring the resilience of children, a substantial proportion of children had serious problems and thus refutes the opinion that childhood cancer causes no or only a slightly increased risk for psychopathology. Several studies (32,25,59) have lucidly outlined how methodological problems may lead to failure in finding differences in adjustment between survivors and comparisons. The design of our study (i.e. unbiased sample, large sample size, limited age range, all children off treatment, carefully matched controls), our instruments (reliable, valid), and our analyses (separate for gender) may explain why we found differences between cancer survivors and healthy peers.

CONCLUSION

Our study demonstrates that children surviving cancer differ in psychosocial functioning from that of matched peers. Children surviving cancer have more social problems and internalizing problems than healthy children. Gender and the cancer experiences per se are the most dominating factors in explaining late psychosocial problems. Although the majority of cancer survivors adjust well, childhood cancer substantially increases the risk of serious late adjustment problems. These findings have important implications for patient care. Whereas the current view emphasizes the similarities between cancer survivors and healthy children, we found that surviving childhood cancer has to be considered as an decisive factor in late psychosocial problems of survivors. Consequently, this information will facilitate a correct diagnosis and intervention for the individual survivor with psychosocial problems.

At this time we do not know what the long-term consequences are of poor or good adaptation during middle childhood for the survivor's adaptation during later developmental stages. In healthy children, poor adaptation in middle childhood is a good predictor of vulnerability in adolescence (62). However, the continuity of problems over time is not clear. In contrast to the higher prevalence rates of psychopathology in boys during childhood, in adulthood, females exceed males in certain disorders (50). Adaptation is a process and poor adaptation at one stage of development does not necessarily preclude an improvement during subsequent developmental stages and visa versa (63). However, the finding that older children (≥ 11 yrs) and those who were longer off treatment (≥ 5 yrs) had more problems, presents a serious concern for future adjustment of cancer survivors. In different developmental stages, different risk and protective factors may play a role in hampering or facilitating adaptation. Our study significantly contributes to knowledge on the long-term consequences of cancer in middle childhood, but we advocate follow-up studies of this population of childhood cancer survivors to obtain insight into the *process of adaptation* across developmental stages.

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CHAPTER 4.3

SIBLINGS OF CHILDHOOD CANCER SURVIVORS: HOW DOES THIS FORGOTTEN GROUP OF CHILDREN ADJUST AFTER CESSATION OF SUCCESSFUL CANCER TREATMENT?*

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ABSTRACT

Siblings of childhood cancer patients are labelled the 'forgotten children' because they experience significant psychosocial distress and are isolated from support systems inside and outside the family. This study investigates the late consequences of the cancer experience for siblings. Sixty siblings of cancer survivors were compared with control subjects on measures of psychosocial adjustment. No differences were found between siblings of cancer survivors and controls on emotional and behavioral problems and competence, suggesting than siblings adjust well to the period of survivorship. The effect of demographic, family, and disease-related characteristics on the siblings' psychosocial adjustment was limited. Whereas during treatment many psychosocial problems for siblings have been reported, this does not result in a heightened risk for psychological disturbance for siblings as a late effect. Implications for patient care are discussed.

INTRODUCTION

Progress in the field of pediatric oncology has drastically improved the rate of survival in childhood malignancies. As more children survive their disease, the focus is now on the quality of this survival. Studies on the long-term consequences of cure should include the late effects for patients, parents, and siblings because cancer and its treatment not only affect the patient but also the other members of the family.

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Information on the long-term sequelae for patients is accumulating, and to a lesser extent this is also the case for parents, but studies on siblings are sparse. Some studies investigated siblings' retrospective experience with the illness (1-3). However, to date no studies exist on the current psychosocial functioning of siblings of childhood cancer survivors.

Siblings of childhood cancer patients are considered to be vulnerable for psychosocial problems. Before diagnosis the prevalence of psychosocial problems among siblings was similar to the general population, however, after diagnosis siblings experienced significantly more emotional and behavioral distress than controls (4). Problems frequently noticed by parents are feelings of jealousy and guilt, academic underachievement, somatic problems, and acting out behavior (5,6). It has been found that during treatment there are striking similarities in the emotional distress between sibling and the pediatric cancer patient, including anxiety, social isolation, vulnerability to illness, and feelings of low self-esteem (7): In some areas, the siblings showed even more signs of distress that the patient (7). The major theme in discussing the sibling's concerns is the disruption they experience in interpersonal relationships, especially with their parents (8,9). Siblings of leukemic children more frequently felt a lack of close relationship with their parents than siblings of other chronically ill children. It was shown that within the family, the needs of the sibling are met least of all (10). Simultaneously, they felt isolated from supportive systems outside the family (11). As a result siblings of cancer patients have been labelled as the 'forgotten children' (11).

Summarizing these findings, the question evolves what the long-term consequences are for healthy siblings of being exposed to prolonged periods of family disequilibrium. The first aim of the study was to investigate the psychosocial functioning of siblings of childhood cancer survivors by comparing their functioning to that of healthy peers. Studies on siblings of childhood cancer patients have been criticized for the use of non-standardized instruments and for the lack of multiple informants because most previous work is based on parents' reports only (2,12). Therefore, we used parents and siblings reports on standardized measures.

The second aim of our study was to investigate which factors affect the siblings' psychosocial adjustment. It has been observed that individual differences and family constellation variables, such as age of the sibling, gender, birth order, family size, contribute to adjustment, but these variables have rarely been examined in relation to the sibling's psychosocial functioning (13,14). Also, disease-related characteristics, which are a possible indication of the disease-related stress, are seldomly considered. Consequently, the impact of demographic, family, and disease-related characteristics on the sibling's psychosocial adjustment is explored in the present study.

MATERIALS AND METHODS

Subjects

Siblings of childhood cancer survivors between 4 and 16 years of age were selected from families with a childhood cancer survivor. The childhood cancer survivor had to meet the following criteria: (a) the child was diagnosed with leukemia, non-Hodgkin lymphoma (NHL) or rhabdomyosarcoma (RMS) (b) the child had successfully terminated cancer treatment, (c) used Dutch as native language, and (d) was 8 to 12 years old at the time of the study. Childhood cancer survivors meeting these criteria were diagnosed between November 4, 1976 - December 23, 1987. During this time period, 473 patients were diagnosed with a malignancy at the Sophia Children's Hospital/University Hospital Rotterdam (one of the four pediatric cancer centers in The Netherlands). According to the patient selection criteria, 231 patients were eligible with respect to diagnosis. Of this group, 53 patients died of their disease. Of the remaining 178 patients, 10 patients were excluded because Dutch was not their native language. In addition, 90 patients were excluded because their age was outside the requested range. In total, 78 survivors were eligible according to all selection criteria. Of this group 73 children were approached to participate in the study, representing all children with leukemia and NHL, and 5 children with RMS. These latter patients resembled the children with lymphoproliferative diseases with respect to prognosis, treatment protocol, difficulty of treatment, length of treatment, and late medical sequelae.

Of the 73 families approached, three families refused to participate because they felt that taking part in the study was too distressing for themselves ($n=2$) or the sibling ($n=1$). In seven families, the patient was the only child. In three families, the sibling fell outside the age range. From each family, one sibling was selected to control for patient and family characteristics. Preference was given to the sibling closest in age to the former patient. In total, 60 siblings were enrolled in the study.

Demographic, family, and disease-related variables

Demographic, family, and disease-related characteristics are presented in Table 1. Socioeconomic status (SES) was rated according to Dutch standards on a six-step scale of occupational level (highest level=6) (15). Family stressors unrelated to the disease were rated as concurrent stresses. Chronic disease was scored if one of the family members other than the former patient had a chronic disease or a chronic condition.

Characteristics related to disease, treatment, and late sequelae are also summarized in Table 1. The survivor's statistical survival chance at diagnosis was rated by an oncologist, based on the diagnosis, symptoms at presentation, and the success rates for the treatment protocols at that time. Intensity of chemotherapy was rated by the

Table 1. Demographic, family, and disease-related characteristics of the sample ($N=60$).

Demographic characteristics of siblings			**Disease characteristics of survivors**		
	n	%		*n*	%
Sex			*Diagnosis*		
Boys	34	57	Leukemia	50	83
Girls	26	43	Non-Hodgkin lymphoma	5	8
			Rhabdomyosarcoma	5	8
Age at investigation					
Median 10 year 6 months			*Survival chance*		
Range 5 to 16 year			≤ 50%	30	50
			> 50%	30	50
Age at diagnosis					
Median 4 year and 1 month			*Intensity of treatment*		
Range 0 to 11 year			Less severe	43	72
Unborn 4 siblings			Very severe	17	28
Family characteristics			*Duration of treatment*		
			< 3 years	50	83
Socioeconomic Status			≥ 3 years	10	17
Unskilled employees	5	8			
Skilled manual employees	20	33	*Surgery*		
Minor professionals	9	15	Yes	10	17
Owners small business	7	12	No	50	83
Professional	9	15			
Major professional	7	12	*Radiation*		
Housewife	1	2	Yes	29	48
Missing	2	3	No	31	52
Number of siblings			*Time off treatment*		
One sibling	36	60	< 5 years	45	75
More than one sibling	24	40	≥ 5 years	15	25
Birth order			*Visible impairment*		
Older than survivor	28	47	Yes	16	27
Younger than survivor	32	53	No	44	73
Concurrent stresses			*Invisible Impairment*		
Yes	25	42	Yes	12	20
No	35	58	No	48	80
Chronic Disease			*Weight problems*		
Yes	15	25	< 90th percentile	37	62
No	45	75	≥ 90th percentile	23	38
			School problems		
			Yes	25	42
			No	35	58

oncologist according to treatment protocol. Visible impairment was partly measured by the residual physical impairment index (3) consisting of the sum of ratings of current visibility and interference with daily living. Invisible impairment included invisible medical problems such as organ damage and endocrine problems. Weight problems were defined as the survivor's weight was at or above the 90th percentile corresponding to Dutch standards (16). School problems were rated present if the survivor received remedial teaching or was not able to attend a regular school.

Comparison groups

Parent reports on the sibling's behavior were compared with that of children matched for sex, age, and SES from a representative sample of the general population (*N*= 2033; see reference 17 for sample selection and data collection procedure). Child reports were compared with randomly selected healthy children (*N*=300) from two local elementary schools and matched for sex (see reference 18 for sample characteristics and data collection procedure).

Procedure

Parents were approached during regular appointments at the follow-up outpatient clinic. They were asked to inform the sibling about the study and to explore the sibling's willingness to participate. If the sibling agreed, written informed consent was requested. Siblings were interviewed at home in an open, in-depth way about their experiences with the illness. After the interview the instruments were administered. Instruments were only administered if they were appropriate for the sibling's age. Mothers were asked to complete a questionnaire while their child was being evaluated. Although two siblings refused to participate, their parents' reports were included. In one family the mother did not complete the questionnaire because of clinical considerations, but information from the sibling was available.

Measures

Child Behavior Checklist (CBCL) (19). The CBCL for ages 4-18 is a standardized procedure to obtain parents' reports of their child's behavioral and emotional problems. The CBCL has excellent psychometric properties, which have been replicated in Dutch children (17). The problem section of the CBCL consists of 120 items. Parents are requested to circle a 0 if the problem item was not true for the child, and a 1 or a 2 if it was sometimes true or very true, respectively. High scores reflect a high degree of problems. Total problem scores are computed by summing the scores on each item. Achenbach (19) empirically constructed eight syndromes: Withdrawn, Somatic complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Delinquent Problems, and Aggressive Behavior. Five of these syndromes could be grouped under two broad band scales: internalizing and externalizing. Internalizing problems reflect internal distress, whereas externalizing problems reflect conflicts with other people and their expectations of the child. The internalizing group consists of the following syndromes: Anxious/Depressed, Somatic Complaints, and Withdrawn. The externalizing group consists of Aggressive Behavior and Delinquent behavior.

Self-Perception Profile for Children (SPPC) (20). The SPPC is a 36-item questionnaire measuring self-concept or perceived competence in 8- to 12-year old children. The SPPC contains six subscales, tapping a) Scholastic Competence, b) Social Acceptance, c) Athletic Performance, d) Physical Appearance, d) Behavioral Conduct, and f) General Self-worth. Each subscale contains six items on which the child is asked to rate its competence on a 4-point Likert scale. Subscale scores range from 6 to 24 and a higher score is associated with higher perceived competence. Reliability and validity of the SPPC has been established for Dutch children (18).

Amsterdam Biographic Questionnaire for Children (ABV-K) (21). The ABV-K is a forced choice (yes/no) personality questionnaire for 9- to 17-year old children measuring a) Neurotic complaints b) Somatization c) Extroversion, and d) Testing-attitude. The Neurotic complaints-scale measures psychoneurotic complaints with descriptive statements such as "I worry a lot" and "Deep down I am scared for many things"; the Somatization-scale measures somatization of neurotic complaints, with items such as "I often have a headache" and "My belly often aches"; The Extroversion scale measures the child's position on the dimension introversion-extroversion with descriptive statements such as "I feel self-conscious" and "When being with other children, I stay in the background"; and the T-scale measures social desirability with items such as: "I lie sometimes" , "I only have good thoughts about other people". The total scores on each subscale reflect the individual's position on a continuum ranging from 0-100. Satisfactory internal consistency and test-retest reliabilities are reported (21,22).

Children's Depression Scale (CDS) (23). The CDS is a well-established measure of the severity and the presence of depressed mood in 9- to 15-year old children. Children sort 63 cards with descriptive statements on a five-point Likert scale. Scores range from 63-315. High scores indicate a higher degree of depression. Reliability and concurrent validity have been established (24).

Statistical analysis

To test differences between siblings and comparison groups, analyses of covariance (ANCOVA) were performed with age at investigation as covariate. The total group of siblings and boys and girls separately were compared with children from the general population. The impact of demographic, family, and disease characteristics (see Table 1) on psychosocial problems was assessed by comparing the mean CBCL, ABV-K, SPPC, and CDS scale scores of siblings with and without a characteristic present in a series of one way analysis of variance (ANOVA). Non-dichotomized variables were categorized as follows: age sibling at investigation (<11 years old, ≥ 11 years old); age sibling at diagnosis (<4 years old, ≥ 4 years old); socioeconomic status (skilled manual employee or lower, minor professional or higher).

The sample size of this study was not large, limiting the statistical power of the tests. To get insight into the exact power of each analysis to detect differences that do exist but fail to reach statistical significance ($p<.05$), and thus avoid type II errors, we performed post hoc estimations of power and effect size. According to Cohen (25) group differences of around .20, .50, and $>.80$ standard deviation are considered a small, medium, and large effect size, respectively. The sample size in this study was not large enough to detect small effects (power ranges from .09 to .20). Estimation of power showed that with an α of .05 , there was a 80% to 100% chance of finding large effects in the total group of siblings and for each sex separately, and 73% to 85% chance of finding medium effects in the total group of siblings. Thus, for these analyses the power is adequate ($>.70$) to excellent ($>.90$). However, in comparing male and female siblings to their controls, the power to detect medium effects was inadequate (range: .33 - .61). Differences that showed a medium effect size, but failed to reach statistical significance due to low power, will be reported in the results section, but should be interpreted with caution.

RESULTS

Differences between siblings and comparison groups

Results of the comparison between siblings and children from the general population are presented in Table 2. Comparison between the total group of siblings and the comparison groups yielded 22 non-significant differences and one significant difference: on the ABV-K Somatization scale siblings of cancer survivors ($M=44.8$) reported less problems than controls ($M=58.2$) at $p<.05$. Analyses for each sex separately showed one significant difference for the boys and two for the girls. Male siblings had a lower mean score on the ABV-K Extroversion scale ($M=50.0$ versus $M=71.9$ at $p<.05$) and female siblings had a lower mean score on the ABV-K Somatization scale ($M=38.4$ versus $M=61.1$) and a higher mean score on the SPPC Social Acceptance scale ($M=19.4$ versus $M=16.2$) both at $p<.05$. In addition, two medium effects for girls were found that did not reach statistical significance due to low power. Female siblings had higher scores on the SPPC Behavioral Conduct scale and on the SPPC Total Score. Finally, no significant differences for the CBCL or for the CDS were found.

Effect of demographic, family, and disease-related variables on adjustment

Significant differences at $p<.05$ and percentages of explained variance are presented in Table 3. Sex and age at diagnosis of the sibling had an effect on four self-report

TABLE 2. Mean for age adjusted scale scores for sibling and control groups[1]

Scale	Total			Boys			Girls		
	Sibling	Control		Sibling	Control		Sibling	Control	
CBCL (parent)	*n*=59	*n*=59		*n*=34	*n*=34		*n*=25	*n*=25	
Withdrawn	1.9	1.8		2.0	2.2		1.6	1.3	
Somatic Complaints	0.9	0.8		1.0	0.8		0.8	0.7	
Anxious/Depressed	2.3	2.5		2.4	2.9		2.0	2.0	
Social Problems	1.4	1.6		1.5	2.1		1.1	0.8	
Thought Problems	0.5	0.2		0.6	0.2		0.3	0.2	
Attention Problems	3.1	3.2		3.7	3.8		2.3	2.4	
Delinquent Behavior	1.0	1.4		1.0	1.5		1.0	1.2	
Aggressive Behavior	5.6	6.6		6.7	8.4		4.1	4.2	
Internalizing Behavior	5.0	5.0		5.4	5.7		4.5	4.0	
Externalizing Behavior	6.6	8.0		7.7	9.9		5.2	5.4	
Total Problems	18.6	21.1		21.1	25.0		15.1	15.7	
SPPC (child)	*n*=46	*n*=46		*n*=26	*n*=26		*n*=20	*n*=20	
Scholastic Competence	17.5	16.1		18.0	17.1		16.7	14.7	
Social Acceptance	18.1	17.8		17.1	19.0		19.4	16.2	***12.4***
Athletic Competence	17.1	18.0		17.6	19.3		16.3	16.6	
Physical Appearance	18.3	17.0		19.4	18.5		17.0	14.8	
Behavioral Conduct	16.3	16.3		15.8	16.8		17.1	15.4	
General Self-worth	19.6	18.7		20.2	19.7		18.8	17.5	
Total Score	106.9	103.8		108.2	110.5		105.3	95.2	
ABV-K (child)	*n*=46	*n*=46		*n*=25	*n*=25		*n*=21	*n*=21	
Neurotic complaints	47.2	47.5		47.1	41.3		47.8	54.4	
Somatization	44.8	58.2	***4.9***	50.1	55.7		38.4	61.1	***14.5***
Extroversion	59.0	63.5		50.0	71.9	***12.5***	68.3	54.8	
Testing Attitude	48.4	45.6		47.3	50.7		49.7	39.4	
CDS (child)	*n*=44	*n*=30							
Total score	151.2	147.1							

Abbreviations used: CBCL=Child Behavior Checklist; SPPC=Self Perception Profile for Children; ABV-K=Amsterdam Biographic Questionnaire for Children; CDS=Children's Depression Scale.

[1] Percentage of explained variance is indicated in bold italics.

TABLE 3. Percentages of variance accounted for by significant ($p < .05$) effects of demographic, family, and disease-related characteristics on psychosocial problems[1]

	Characteristics						
	Demographic		Family		Disease		
	Sex	Age at diagnosis	Chronic disease	SES	Radiation	Time since treatment	School problems
CBCL (parent)							
Withdrawn							
Somatic Complaints						11.6[g]	
Anxious/Depressed			7.4[d]				
Social Problems			11.6[d]				
Thought Problems							
Attention Problems							
Delinquent Behavior			7.0[d]				
Aggressive Behavior							
Internalizing							
Externalizing							
Total Problems			10.8[d]				
SPPC (child)							
Scholastic Competence		9.6[c]					
Social Acceptance	8.9[a]			10.5[e]			
Athletic Competence							
Physical Appearance	10.1[b]						
Behavioral Conduct							
General Self-worth					11.1[f]		
Total Score							
ABV-K (child)							
Neurotic complaints							
Somatization				8.9[e]			
Extroversion	12.0[a]						
Testing Attitude					8.9[f]		
CDS (child)							
Total score					18.2[f]		10.4[f]

[1] Only those characteristics with at least one significant result are listed. *Abbreviations used:* CBCL= Child Behavior Checklist; SPPC=Self Perception Profile for Children; ABV-K=Amsterdam Biographic Questionnaire for Children; CDS=Children's Depression Scale; SES=socioeconomic status. [a] higher problem scores for male siblings; [b] higher problem scores for female siblings; [c] higher problem scores for older siblings (≥4 yrs at diagnosis); [d] higher problem scores for siblings with characteristic present; [e] higher problem scores for siblings from lower SES; [f] lower problem scores for siblings with characteristic present; [g] higher problem scores for siblings of survivors ≥5 years off treatment.

scales. Male siblings had a lower ABV-K Extroversion score (M=50.0) than girls (M=68.3) and a lower SPPC Social Acceptance score (M=17.1 versus M=19.4). Female siblings had a lower SPPC-Physical Appearance score than male siblings (M=17.0 versus M=19.4). The mean SPPC Scholastic Competence score was lower for siblings who were older than four years when the disease was diagnosed than for siblings who were younger (M=16.2 versus M=19.3). With respect to family characteristics, we found that parents of siblings coping with a chronic disease in another family member reported more psychosocial problems. The mean CBCL Total Problem score was 28.3 for siblings from these families versus 15.3 for siblings from other families. The mean CBCL syndrome scores that were elevated were respectively Anxious/Depressed (M=3.6 versus M=1.8), Social Problems (M=2.7 versus M=0.91), and Delinquent Behavior (M=1.9 versus M=0.8). Furthermore, siblings from lower SES-families had higher ABV-K Somatization scores (M=54.0 versus M=36.0) and reported lower Social Acceptance on the SPPC (M=17.0 versus M=19.6) than sibling from higher SES families.

The impact of disease-related factors on the siblings was restricted to an effect of radiation and time since cessation of treatment and if the survivor had school problems. Siblings of survivors who were radiated during treatment, scored themselves higher on SPPC General Self-worth (M=20.5 versus M=17.9), higher on the ABV-K Testing Attitude scale (M=55.0 versus M=37.0), and lower depression scores on the CDS (M =142.0 versus M=162.0). Siblings whose brother or sister had school problems as a late effect reported also lower depression scores on the CDS (M=142.0 versus M= 157.4). For siblings whose brother or sister was more than five years off treatment, parents reported more Somatic Complaints (M=1.4 versus M=0.5).

DISCUSSION

The aim of the study was to investigate how siblings of cancer survivors adjust after cessation of treatment. The results of the present study showed that the psychosocial functioning of siblings of childhood cancer survivors is comparable to that of children in the comparison groups. Although small differences between groups may exist, which could not be detected due to limited statistical power, no major differences in psychosocial functioning was found. On all measures of emotional and behavioral problems and competence, the total group of siblings fared the same or even better than those to whom they were compared, showing that childhood cancer does not result in a heightened risk for psychological disturbance for siblings as a late effect. Depending on the informant, estimates of the frequency of siblings' adjustment problems may vary. The fact that in our study parents and siblings reports coincided, underscores that the stresses associated with cancer and its treatment have no serious late psychosocial sequelae for the sibling of the survivor.

Although clinical experience and research demonstrates that siblings during diagnosis and treatment have serious problems (4-11, 26-28), after termination of treatment siblings adjust well to the period of survivorship. The absence of long-term sequelae in siblings stands in sharp contrast to the late effects of the cancer experience on the survivors themselves and their parents. In comparing childhood survivors and siblings to their matched controls on the CBCL, siblings showed significantly fewer problems than the survivors (JEWM Van Dongen-Melman, unpublished data). In the survivor group, 21.1% of the children had serious adjustment problems versus 6.8% in the siblings ($p = .01$; z-test two-tailed probability). Moreover, childhood cancer has distinct and persistent late effects on parents that even tend to increase over time (29). Whereas it has been reported that during treatment siblings were less adjusted than other family members (10), after termination of treatment this situation seems to be reversed. It might be hypothesized that siblings after termination of treatment are able to distance oneself from the cancer experience, whereas for survivors and parents the confrontation with the disease, the concerns and anxieties over the recurrence of the disease, and its long-term medical consequences continues.

Few demographic, family, and disease-related factors were found to affect the siblings' adjustment. This finding can be attributed to the sibling's general well functioning because these factors have to discriminate between well adapted and better adapted siblings. In the literature, several factors are considered important to explain the differential adaption in siblings of chronically ill children (13,14,30). The situation for siblings of childhood cancer survivors differs, however, from siblings of children with cancer undergoing cancer treatment. After termination of cancer treatment the direct medical stresses subside whereas siblings of children with cancer during treatment are continuously confronted with the illness. The fact that a chronic disease in another family member did increase adjustment problems for siblings may lend support to this supposition.

Interestingly, siblings of a survivor who received cranial radiation or who had school problems showed significantly lower depression scores and higher scores on general perceived competence. Simultaneously, they gave more socially desirable answers that might explain these favorable scores. Cranial radiation is associated with neuropsychological deficits that negatively affect academic achievement. Because one would expect that long-term sequelae in a survivor would increase problems, the question is why do these siblings need to present themselves more favorably. A possible explanation might be that they did not want to show their concerns or problems which might enhance their parents' burden or worries (28). Siblings may feel the need to excel to compensate for the survivor's problems and thus make a valuable contribution to family life. Alternatively, these siblings may feel jealous towards the survivor and thus like to overemphasize their adequate adjustment in contrast to that of the survivors.

The fact that childhood cancer does not result in an increased risk for major psychological disturbance does not, however, signify that siblings may not have undergone alterations in psychosocial functioning or do not experience distress. The individual sibling may have lingering concerns and problems that do require attention. From the interviews with children we found that if there were lingering concerns, they focussed on the relationship with the parents and their position within in the family. Short-term counseling-sessions were developed to help parents to initiate communication about these issues with their children. Parents reported back that addressing the disadvantaged situation of the sibling, was very effective to alleviate these problems. Recent data suggest that siblings who reported more communication about the illness indeed perceived their lives to be less affected by the cancer experience (2). Even though siblings fare well, their quality of life can be enhanced by well attuned communication.

Our study shows no major differences in psychosocial functioning between siblings of cancer survivors and controls, suggesting that childhood cancer does not result in a heightened risk for psychological disturbance for siblings after cessation of treatment. Although potential reassurance can be given to parents about the long-term outcome for siblings, simultaneously efforts to facilitate communication between parents and siblings should be made. Our study provides information to develop adequate support strategies for families of childhood cancer survivors. It may help the health care team to focus care on those family members who are in need of support.

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Chapter

5

Johan, 11 yrs (cancer survivor)

CHAPTER 5

GENERAL DISCUSSION

SURVIVING CHILDHOOD CANCER: ALL IS WELL THAT ENDS WELL?*

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INTRODUCTION

Progress in medical treatment has drastically raised the survival rate in childhood malignancies. Three decades ago, childhood cancer had an almost invariably fatal prognosis. When cancer was diagnosed, the child often died after a short disease course. At this moment childhood cancer has a much more favorable prognosis. The child has not only an extended life expectancy, but also a real chance of cure (Figure 1). Along with this progress in medical therapy, the psychological dilemma changed from one of adapting to the imminence of death to one of coping with uncertain survival. Paradoxically, with an increased chance of cure, the uncertainty over the ultimate outcome increased.

With successful treatment the costs of cure appeared. Medical side effects related to disease and treatment were documented, including disorders in growth and puberty, obesity, neuropsychological deficits, musculoskeletal abnormalities, amputation, cardiopulmonary dysfunction, and the development of second malignancies (reviewed in Chapter 1.1). The understanding that disease-free survival is not synonymous with cure, led to a new conceptualization of cure. In his book *'The truly cured child'* (1), Van Eys explained that biological cure must extend to the quality of survival. Thus, in addition to the medical consequences, the psychosocial consequences of childhood cancer should be evaluated as well.

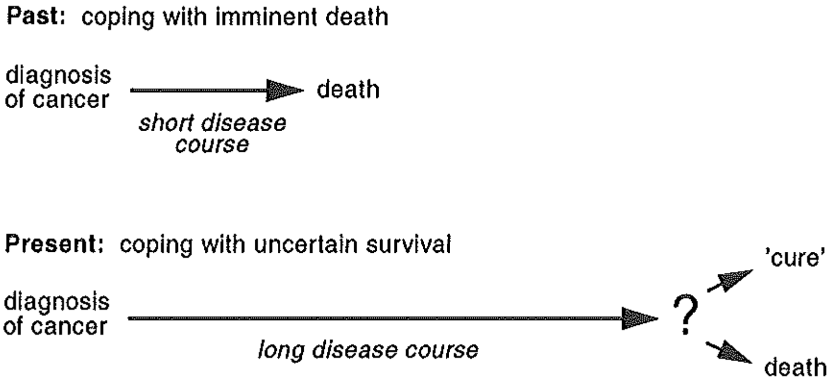

Figure 1: Changes in medical outlook of childhood cancer and ensuing coping tasks.

Although information is accumulating on the medical side effects of cancer and its treatment, considerably less is known about the late psychosocial consequences for the child, in particular about the late consequences for the child's family. The child and its family can be at risk for psychosocial problems, because of the nature and duration of stresses during treatment (summarized in Chapter 1.2) and because of the confrontation with new problems evolving from the illness and its treatment. We investigated the late psychosocial consequences for patients, parents, and siblings, and will discuss our findings in the context of the literature. Based on this study, we provide recommendations for patient care.

LATE PSYCHOSOCIAL CONSEQUENCES FOR THE PATIENT

Learning problems

Learning problems for children treated for cancer have often been reported (2-11). In our sample of 95 survivors, 35% had learning problems (Chapter 4.2). This sample consisted of childhood leukemia/lymphoma survivors and survivors treated for solid tumors. For children surviving leukemia or lymphoma it has been reported that central nervous system (CNS) prophylaxis with cranial irradiation to prevent CNS relapse is associated with neuropsychological impairment (12-16). Many studies found a decline in intelligence and problems with attention, memory, and visual-motor functions (12-20). In addition, anatomical abnormalities have been found on computer tomography (CT) scans, such as white matter damage, calcifications, and atrophy (4,21,22).

Although neuropsychological dysfunction and neurological abnormalities may be associated with poorer academic performance, there is no straightforward relationship between these measures and the child's learning capabilities. Firstly, there is a poor correlation between neurological and (neuro)psychological measures (4,21,22). Children with impaired neuropsychological functioning may have normal CT scans and vice versa. Secondly, children receiving CNS prophylaxis had a decline in IQ, but their IQ scores were still within the normal range (12,18,23). Finally, despite normal IQ scores and normal CT scans, children with leukemia were found to have learning problems associated with their disease (7). These findings imply that neurological and psychological measures are not sufficiently refined to assess all possible sequelae and to predict their effect on the child's daily life. Therefore, the clinical significance of these findings, i.e. their effect on the child's learning capabilities, should be assessed. Although neuropsychological sequelae have most often been associated with cranial irradiation, it is not clear whether learning problems are an effect of treatment, the disease itself, or both (6,7,12,13). Furthermore, it is suggested that other types of CNS prophylaxis, consisting of chemotherapy only, also have neurotoxic effects (4,21,24).

No study has been performed to isolate the effects of cranial irradiation from the effects of CNS prophylaxis with chemotherapy only, and from other treatment and disease variables. We performed a comparative study in which children with different types of CNS prophylaxis (leukemia or lymphoma patients) were compared with children who received systemic chemotherapy but no CNS treatment (patients with solid tumors) and with matched healthy controls (Chapter 4.1). We found an unexpectedly high prevalence (80%) of learning problems in children who received cranial irradiation as CNS prophylaxis. These children had a ten fold increase in serious problems as compared to healthy children: almost 40% had serious learning problems that required special education. A similar percentage of serious learning problems associated with cranial irradiation was reported in another Dutch study (4). Furthermore, our study showed that CNS prophylaxis with chemotherapy only was not associated with an increase in learning problems (16%) as compared to cancer survivors without any form of CNS treatment (16%) and matched healthy children (17%). With this study we demonstrated that learning problems are exclusively related to cranial irradiation (Chapter 4.1). Our data underline the need to eliminate cranial irradiation from CNS prophylaxis in children with leukemia and lymphoma and to consider alternative treatment regimens for children with initial CNS disease where cranial irradiation is still the mainstay of treatment.

Social problems and internalizing problems

Many studies report problems in social adjustment for children surviving cancer (6,8,11,25-27). We also found more social problems and problems with social acceptance as compared to matched healthy children. The consistent report of problems in social adjustment, using different assessment methods and informants, make it a core issue for the child surviving cancer.

We found several interesting correlates with social adjustment. Children were more withdrawn and showed more introvert behavior. The child's responses on the extroversion scale indicated that they distance themselves and derive less pleasure from social interaction with their peers. This cluster of social problems and internalizing behavior fits into the description of socially withdrawn children (28). Stewart and Rubin (28) showed that withdrawn children experience a relative high degree of social rebuke and failure in the company of peers. When these children try to gain peer compliance, their approaches are made with caution and are mostly ineffective. It has been noted that male survivors indeed assert themselves less, socially as well as physically (24; Chapter 4.2). The subsequent failures of their nonassertive behavior in social interactions are likely to result in social withdrawal, which will lead to avoidance of these children by peers (28) and may explain social isolation reported for children surviving cancer (26,29-31).

Furthermore, school problems and cranial irradiation were related with social adjustment in boys but not in girls. Children who received cranial irradiation had more social problems and exhibited more introvert behavior, showing that cranial irradiation has an effect on both the child's learning capabilities and its behavior. A study on socially adaptive behavior of cancer patients confirms the relationship between cranial irradiation, learning deficits, and social adaptation (32). It can be hypothesized that cranial irradiation causing cognitive deficits may further hamper the male survivor in developing social skills.

Gender differences in adjustment and risk factors affecting adjustment

Gender differences in psychosocial adjustment of children surviving cancer have seldomly been reported, because most studies did not perform separate analysis by gender. We found that male survivors were more vulnerable to psychosocial problems than female survivors. This differential sensitivity to childhood cancer is in line with the literature that before adolescence boys are more vulnerable to exposure to stress than girls (33-35). Various suggestions related to genetic, personal, and environmental factors have been put forward (35-38). For instance, boys may use different coping styles than girls, which may be less effective in dealing with stressors (37). In addition, observer bias, different cultural stereotypes, and different norms for boys' and girls' behavior may have an effect. However, still little is known about the underlying mechanisms for explaining gender differences in adaptation to stress.

The differential sensitivity of the sexes to stress is illustrated by the effect of risk factors on psychosocial functioning (Table 1). The risk factors measured in our study consisted of psychosocial stressors as well as disease-related stressors (i.e. related to disease itself, treatment, and long-term sequelae). Whereas several psychosocial and disease-related stressors affected psychosocial adjustment in boys, only one factor had an effect on girls (overweight). Effect of individual disease-related stressors on psychosocial adjustment was limited (Table 1). However, when all risk factors related

TABLE 1. Risk factors affecting psychosocial functioning of boys and girls surviving cancer.[1]

Type of stressor	Boys		Girls	
	Stressor	Effect[2]	Stressor	Effect[2]
Psychosocial stressors	- concurrent stresses - chronic disease in family	2 2		
Disease-related stressors	- long treatment course (≥ 3 yrs) - cranial irradiation - school problems - poor prognosis (≤ 50%) - age at diagnosis (≤ 4 yrs) - older age of investigation (≥ 11 yrs) - longer from treatment (≥ 5 yrs) - diagnosis of leukemia	3 3 3 1 1 1 1 1	- overweight	1

[1] No effect was found for the following disease-related stressors: severity of chemotherapy treatment, surgery, invisible impairment, visible impairment, and socioeconomic status.

[2] Number of psychosocial outcome variables affected by risk factor.

to long-term sequelae were combined, children with multiple side effects were most affected in their psychosocial functioning. Other studies confirm this relationship (6,39). In addition, we found that all risk factors combined had the largest effect on psychosocial adjustment, but the overall effect was modest. Also other studies found weak or no association between demographic variables, disease-related variables, and long-term adjustment (6,39-42). We conclude that gender and the cancer experience per se are the most dominating factors in explaining late problems.

Severity of problems

Studies concerning the severity of long-term psychosocial consequences of childhood cancer have yielded mixed results. One of the first in-depth studies on psychosocial functioning of a large number of survivors was conducted by Koocher and O'Malley (43,44). They found that 59% of their former patients had at least mild psychiatric symptoms, such as depression, anxiety, problems of self-esteem, and fluctuations in mood. However, Koocher and O'Malley (43) neither provided normative data nor psychometric data for the psychiatric interview they used to classify (mal)adjustment. Since then, most studies employed standardized assessment instruments, but failed to replicate Koocher and O'Malley's findings. Instead a number of these studies reported overall good adjustment and few differences between cancer survivors and controls or normative samples (9,29,31,39,40,45-50). Other studies have documented emotional and adjustment difficulties in childhood cancer survivors (6,8,11,25,27,29,42,51-53), but they did not adequately evaluate whether these differences were normal variations or clinically significant problems. Consequently, the severity of psychosocial consequences for survivors remained unresolved.

In our study we tested directly whether the cancer survivors were at risk of developing serious problems. Although the majority of cancer survivors adjust well, a substantial increase in serious problems for the total group of survivors was found: 21.4% versus 10.8% of controls. Especially boys appeared to be vulnerable to developing serious problems. One out of every four male cancer survivors has serious problems, whereas among healthy boys, one out of every ten boys has serious problems. Consequently, the current opinion that childhood cancer causes no or only a slightly increased risk for psychopathology needs to be reconsidered.

Difficulties in detecting adjustment problems in childhood cancer survivors

Why did our study show differences in problems between survivors and healthy children whereas other studies did not? A number of methodological differences between our study and those reported in the literature may be responsible for this fact: sample size, heterogeneity of the sample, representativeness of the sample, type of informant, separate analysis for gender, and study approach (6,8,9,11,25-27,29-31,39,40,42-78; summarized in Table 2).

Heterogeneity of the sample

Subject characteristics vary widely across studies with respect to age and treatment phase. As indicated in Chapter 2.1, the age range of subjects varies widely and is considered one of the most significant methodological concerns (79). In some studies the age range of the subjects is ≥ 15 years (25,40,43,49), but our study has a very narrow age range (only 5 yrs). In addition, our sample included only children after termination of treatment.

Representativeness of the sample

Many studies do not explain how the selection of subjects from the total pool of eligible patients was established (Table 2). This information is essential to evaluate if any bias entered into the selection of patients for evaluation. Fritz et al. (40) clearly explained that in their study part of the subject group could not be reached. These subjects were thought to experience more psychosocial disturbance than those that could be reached for their study (40).

Compliance rates vary widely among studies: some are as low as 15%, others claim to have a rate of 100%, and sometimes the compliance rate is not reported (Table 2). One should be aware that sample bias might result in a high compliance rate.

The importance of an unbiased sample is illustrated by a study of Kazak and Meadows (46), who found no differences between survivors of childhood cancer and a comparison group. They claimed that the so-called 'null hypothesis' needs to be accepted, stating that cancer survivors are not different from healthy children, although their compliance rate was only 74.5%. In contrast, our study with a compliance rate of 96% revealed that 21.4% of survivors have serious adjustment problems. It is striking to note that studies reporting no adjustment problems do not mention biased patient selection or low compliance as a possible explanation for their results (45,46). Our experience indicates that cancer survivors and families who have difficulties in coping with the disease are reluctant to participate. Thus, it is no surprise to find good adjustment in a biased sample of cancer survivors.

Type of informant

Response shift? Discrepancies between child reports and other informants have been found in our study as well as in several other studies (6,11,26,29,42). Thus, the source of information may be a critical factor in finding serious problems. Our study shows that children reported less problems than parents. Interestingly, in the general population, differences between parent and child reports of problem behavior point to the opposite direction: children reported more problems than their parents and this discrepancy increased with age (80). This supports the suggestion that cancer survivors perceive problems differently (Chapter 4.2). As a result of the cancer experience, they use a different internal standard to evaluate problems. Day-to-day stressors and problems are viewed from a different perspective against the experiences of having had a life-threatening disease. It may reflect the often noticed 'lay' observation by adults that children surviving cancer have grown 'wise' beyond their age. This 'response shift' may be responsible for underreporting problems by survivors themselves. If children with cancer indeed use different internal standards to evaluate their problems, it has important and far reaching implications, methodologically *and* clinically, for evaluating the child's mental health. Assessment measures normed on healthy children might not be appropriate for children having (had) a chronic disease. A child scoring within the normal range on such instruments would in fact receive higher scores if controlled for the illness experience. Therefore, further studies are needed to investigate the existence and nature of a 'response shift' in children with cancer.

Observer bias. In our study, the seriousness of problems was based on parental information. Parents are considered most knowledgeable about their children's behavior (81). However, parental perception of the child may be influenced by their own functioning and their own coping strategies (82,83). Our clinical experience indicates that parents are more reluctant than eager to report problems in their child, which lends extra support to the severity of problems reported by parents. To control for bias in parental report, independent raters were used to evaluate the patient's general psychosocial functioning on the Children's Global Assessment Scale (CGAS) (84,85). The mean CGAS score for children surviving cancer was 70.6 (SD=15.6). Thirty-one percent of patients (18 boys and 3 girls) had a CGAS score <61 and were classified as having moderate to marked adjustment problems. These results support the parent's view about the seriousness of problems in survivors and show that childhood cancer can cause serious adjustment problems.

TABLE 2. Methodological characteristics of studies on psychosocial adjustment of children surviving cancer.[1]

Study (reference)	Sample				Representativeness of sample		Control group		Informants	Analysis by gender	Psychometric properties of instruments[2]
	n	Age (yrs)	Age range (yrs)	On/off treatment	Un biased selection[3]	Compliance	Present	Pair-wise comparison			
Fergusson, 1976 (51)	18	10.9	8-15	off	-	NR	-	NA	P,C	NT	-
Obetz et al., 1980 (48)	18	NR	NR	off	-	NR	-	NA	C	NT	-
Koocher et al., 1981(43)[4]	117	18.4	5-32	off	-	96%	+	-	C	NT	-
Schuler et al., 1981 (52)	48	9.5	5-16	off	-	NR	-	NA	C	NT	-
Sawyer et al., 1986 (8)[5]	42	10.4	NR	off	+	95%	+	+	P,T	NT	+
Chang et al., 1987 (25)	42	17.2	11-25	off	+	96%	-	NA	P,C	T	?
Kaplan et al., 1987 (50)	38	mixed[6]	7-18	mixed	+	93%	-	NA	C	NT	+
Fritz et al., 1988 (40)	52	15.9	7-21	off	-	88%	-	NA	P,C	NT	-
Fritz et al., 1988 (53)	41	17.3	NR	off	-	NR	-	NA	C	NT	-
Greenberg et al., 1989 (39)	138	12.5	8-16	off	+	100%	+	-	P,C	NT	+
Kazak et al., 1989 (46)	35	12.2	10-15	off	+	75%	+	-	P,C	NT	+
Mulhern et al., 1989 (6)	183	12.2	7-15	off	+	100%	-	NA	P	NT	+
Sawyer et al., 1989 (9)[5]	32	13.4	NR	off	+	93%	+	+	P,T	NT	+
Noll et al., 1990 (26)[7]	24	12.3	8-18	mixed	-	89%	+	+	T	NT	+
Spirito et al., 1990 (31)	56	9.2	5-12	off	-	96%	+	-	P,C,T	NT	+
Noll et al., 1991 (29)[7]	24	12.3	8-18	mixed	-	89%	+	+	C,Cl	NT	+

(continued) →

TABLE 2 (continued):

Study (reference)	Sample				Representativeness of sample		Control group		Informants	Analysis by gender	Psychometric properties of instruments[2]
	n	Age (yrs)	Age range (yrs)	On/off treatment	Un biased selection[3]	Compliance	Present	Pair-wise comparison			
Sanger et al., 1991 (27)	48	12.5	4-17	mixed	-	95%	-	NA	P,T	T	+
Brown et al., 1992 (47)	55	7.6	4-10	mixed	+	90%	NA[8]	NA	P,C,T	T	+
Olson et al., 1992 (11)	20	9.7	6-16	off	-	NR	+	+	P,C,T	NT	+
Rait et al., 1992 (54)	88	15.6	12-19	off	+	90%	NA[8]	NA	C	T	+
Noll et al., 1993 (30)[7]	19	14	11-18	mixed	-	85%	+	+	C,T,Cl	NT	+
Madan-Swain et al., 1994 (45)	25	15.7	12-18	off	+	15%	+	-	C,T	NT	+
Sloper et al., 1994 (42)	31	12.3	9-18	off	+	81%	+	+	C,P,T	T	+
Kanabar et al., 1995 (49)	30	mixed[9]	3-26	off	+	NR	-	-	mixed[10]	NT	-
Van Dongen-Melman et al., (Chapter 4.2)	95	10.3	8-13	off	+	96%	+	+	P,C	T	+

Abbreviations used: NA=not applicable; NR=not reported; NT=not tested; C=Child, P=Parent; T=Teacher; Cl=Classmate/peer.

[1] Studies were included, if the mean age of the sample was ≤ 18 yrs; this means exclusion of refs. 59-76. No accessibility to original source: refs. 57 and 58.

[2] When information was reported on reliability and validity of instruments used, this item was scored positive.

[3] When no information was reported on total number of eligible patients or on selection procedure, this item was scored negative.

[4] Also published in refs. 44,55,56.

[5] The sample of Sawyer et al., 1986 was retested in 1989.

[6] Sample consisted of two subsamples: mean age of 9.7 yrs (*n*=21) and mean age of 15.4 yrs (*n*=17).

[7] In Noll et al., 1990 and 1991 the same sample was used which was retested in Noll et al., 1993

[8] Control group is not applicable because a within-group design was used.

[9] Sample consisted of two subsamples: mean age of 8 yrs (*n*=15) and mean age of 19 yrs (*n*=15).

[10] Either parent report or child report, sometimes both were used.

Statistical analysis by gender

Often no separate analysis by gender has been made, which may explain the failure to find differences between cancer survivors and healthy children. Especially when the sexes are equally distributed in the sample, the better adjustment of girls may compensate the poorer adjustment of boys.

Study approach and view on the child surviving cancer

As outlined in Chapter 2.1 it is essential to view a childhood cancer survivor as a normal individual under extreme stress. Many researchers in pediatric psychology have adopted this view. In line with this view, the literature focusses on adaptation and coping in children and families with chronic disease including childhood cancer. Traditionally, the child's adaptation was discussed in psychiatric terms and it was assumed that deficiency was an inevitable outcome to chronic disease (86). Probably as a reaction that a negative outcome is expected, the current opinion overemphasizes similarities in mental health between healthy and ill children. This view resulted in a growing resistance to use instruments that assess serious impairment or psychopathology, because they are directly associated with the traditional 'deficit' approach. However, these instruments are designed to discriminate between clinical symptoms and normal variations and thus are helpful to give insight into the child's functioning on a continuum of mental health. The Child Behavior Checklist (CBCL; 87), which was used in our study, is not designed to assess all variations in mental health. However, it is a valid and reliable instrument for detecting problems at the extreme end of the mental health continuum. Instead of providing a 'golden standard' (88) to which the behavior of children with chronic illness can be compared, the CBCL is a valuable tool to assess the stressfulness of the disease for the child.

Reflections on our study

While we were able to evaluate the child's psychosocial functioning, the instruments in our study gave no insight into the specific problems for children surviving cancer. Even though the majority of children experience good overall adaptation, cancer may result in specific problems on various life domains. Interviews can complement this weakness of general instruments, as outlined in Chapter 2.1. Interview studies on adolescents and young adults give insight into how they perceive the effects of illness on their daily existence (43,77). Analysis of our child interviews may clarify how younger children during middle childhood relate about their experiences of surviving childhood cancer.

Our study delineates the effects of childhood cancer on middle childhood in detail. However, further studies are needed to elucidate whether these findings can be generalized to other developmental stages. In addition, we do not know whether poor or good adaptation during this developmental stage predicts vulnerability during later stages. Adaptation is a process and poor adaptation at one stage of development does not necessarily preclude an improvement during subsequent developmental stages and vice versa (89). However, the finding that older children and those who are longer off treatment have more problems, presents a serious concern for future adjustment of cancer survivors. Therefore, follow-up of this population of childhood cancer survivors is important to get more insight into the process of adaptation across developmental stages.

LATE PSYCHOSOCIAL CONSEQUENCES FOR PARENTS

Changes in life

Studies on the impact of cancer after cessation of treatment on the parents' life are sparse. Existing interview studies have focussed on their retrospective experiences with the illness, on lingering concerns (in particular the fear of relapse), and on changes in the marital relationship (39,43,46,90-99; Table 3). How cancer affects the parents' personal life has received little attention. From the qualitative analysis of our in-depth interviews, a number of changes in their personal life was identified (Chapter 3.1). Parents indicated that cancer in their child was the most overwhelming experience in their life. By comparing their life before and after the treatment period, parents reported a number of significant changes. These changes were not incidental, but were of a definitive, long-lasting nature. The far majority of these changes was labelled as negative and most of them could be categorized as losses. For instance, their outlook of life changed drastically, faith in life vanished, and with that the purpose of life, as is illustrated by a parent: *"You are disappointed in life, pur sang in the things you believe in."* Other parents indicated that they can never be the same relaxed person again and experienced a loss of joyfulness. Parents reported that they feel more vulnerable for traumatic events. When confronted with new stresses they reported to be more easily disturbed in their psychological equilibrium and less able to cope with these stresses. Other parents reported a loss of part of their life-time, because of the energy and time consuming treatment of their child. They could have spent this large amount of energy and time differently, if their child had not been ill. In addition, losses within the marital relationship and losses with respect to the health of their child were experienced. These negative changes were so profound that they led to changes in the parents family planning (see Chapter 3.3).

The finding of losses seems paradoxical, because the child survived cancer. In the literature, losses have only been discussed in relation to the child dying from cancer. However, we demonstrate that losses also occur when the child survives, but these are of a psychological nature. These losses induced mourning processes in spite of the child's survival. In addition, we focussed on the implications of the potentially fatal prognosis of cancer on parental functioning. The potential loss of the child and the psychological losses experienced by parents and the subsequent mourning processes are essential in describing the life situation of parents of a child surviving cancer. In this way, we integrated the study approaches of chronically ill and fatally ill children, as is advocated in Chapter 1.2.

It is often emphasized that studies on the impact of cancer should not only focus on the negative aspects but also on its positive aspects. Positive changes, such as personal growth and a more gratifying relationship with the spouse and the child, were indeed reported (Chapter 3.1). However, the number of positive changes and the few families who reported them obliges one to consider the status of these gains and to examine which meaning they have for the individual. Assigning a positive meaning to childhood cancer, may be a way of giving meaning to an extremely stressful period in life. In this way, it may be a style of coping, enhancing one's feelings of self-esteem and control (Chapter 2.2). In our interviews, parents made clear that the positive changes did not compensate the losses resulting from the illness. The emphasis in the literature on positive gains seems to lack this subtlety. It may be speculated that the emphasis on positive changes in the literature, reflects the researcher's own need to find a meaning for such traumatic experiences. We doubt whether gains influence the subject's happiness. Therefore, it should be further investigated in what way positive changes are related to psychosocial well-being.

Uncertainties and worries

Interview studies on the life situation of parents found many lingering concerns about the long-term effects of treatment and uncertainties associated with the well-being of childhood cancer survivors (43,90,92,95). Parents experienced a perseveration of uncertainty and anxiety related to the potential recurrence of the disease, as is illustrated by a father: *"We've always had that uncertainty: will it come back or won't it. That's what worries you"*. Of all problems measured (Chapter 3.2), uncertainty was most often reported by parents: almost 90% of parents reported uncertainty with 51% to a high degree. Parents felt uncertain about long-term survival chances in their child and the child's current condition, about the physical and emotional after-effects on the child, and how to support the child in coping with disease (Table 4). Whereas Comaroff and Maguire (100) argued that uncertainty in parents is the main feature of childhood cancer *during* treatment, we showed that uncertainty is also a general characteristic *after* cessation of treatment.

A number of lingering parental concerns and worries have been mentioned such as the fear of relapse, the child's ability to form relationships, the child's marriage prospects, and the child's ability to have children (Table 3). Table 5 presents the frequencies on the disease-related fear scale, which was used in our study to measure parental worries. Relatively low frequencies of parental concerns were reported. Peck (90) also found low frequencies of parental concerns by questionnaire. She indicated that these frequencies did not reflect the anxieties of parents as perceived during the interview. Especially the fear of relapse often mentioned in the interview data (43,90,92,95) was

TABLE 3. Summary of studies on psychosocial functioning of parents of childhood cancer survivors.[1]

Study (reference)	Purpose of the study	Sample	Instruments	Major findings
Peck, 1979 (90)	Identification of problems in families	24 families Compliance: NR	Interview; Questionnaire	Parents were anxious about recurrence of the disease and had lingering concerns about the survivor.
Koocher et al., 1981 (43)	Investigation of current and retrospective experiences with the illness	173 parents (98 mothers, 75 fathers) Compliance: NR	Interview	Parents expressed concerns about long-term effects of treatment, relapse, fertility, and overall physical condition of the survivor. Parents' marriage was stable over time.
Morrow et al., 1981 (91)	Comparison of adjustment of parents of pediatric cancer patients	48 parents with a child on treatment, 22 parents with a child off treatment, 37 parents of a deceased child; Compliance: NR	Psychosocial Adjustment to Illness Scale	Parents whose child was on treatment or had completed treatment showed better adjustment than parents whose child had died.
Lewis & LaBarbera, 1983 (92)	Investigation of parental experiences	20 parents (19 mothers, 1 father) Compliance: 100%	Interview	Anxiety and fear are normal emotional responses at cessation of treatment.
Wallace et al., 1987 (93)	Investigation of parental needs and concerns	93 parents Compliance: 61%	Questionnaire	Major concerns were related to marriage prospects, fertility, relapse, and emotional development of the survivor. Parents need ongoing support.
Kupst & Schulman, 1988 (94)	Determination of long-term coping	43 families, including families whose child relapsed or died; Compliance: 88%	Interview; Current Adjustment Rating scale	Improvement in adjustment since diagnosis (almost 7 yrs ago) were reported. No differences between bereaved parents and parents of a long-term survivor.

(continued) →

TABLE 3 (continued)

Study (reference)	Purpose of the study	Sample	Instruments	Major findings
Spinetta et al., 1988 (96)	Longitudinal study of families' adjustment	51 families including families whose child died Compliance: 94%	Interview	Parents whose child died had more adjustment difficulties. Families who functioned well during treatment continued to do so after cessation of treatment.
Greenberg et al., 1989 (39)	Assessment of parental functioning	118 mothers Compliance: NR Comparison group: n=98	Derogatis Stress Profile	Levels of psychosocial distress did not differ from levels in comparison parents.
Kazak & Meadows, 1989 (46)	Assessment of parental functioning	35 families Compliance 75% Comparison group: n=13	Langer Symptom checklist	Levels of psychosocial distress, i.e. depression and anxiety, did not differ from levels in comparison parents.
Greenberg & Meadows, 1991 (97)	Description of positive and negative experiences resulting from illness	120 parents Compliance: NR	Interview	76% statements were positive concerning intact support systems, changed values, and marital adjustment. 79% statements were negative concerning recurrence of cancer, physical sequelae, and marital relationship.
Speechley & Noh, 1992 (98)	Assessment of psychological distress	112 parents (63 mothers, 49 fathers) Compliance: 82% Comparison group: n=112 (64 mothers, 62 fathers)	Depression-scale, Anxiety scale (STAI)	Levels of depression and anxiety in parents of a long-term survivor did not differ from comparison parents. Parents with social support experience less psychological distress.

(continued) →

TABLE 3 (continued)

Study (reference)	Purpose of the study	Sample	Instruments	Major findings
Van Dongen-Melman et al., (Chapter 4.1)	Qualitative analysis of the impact of illness on their personal and current functioning and ways of coping	16 parents (8 mothers and 8 fathers selected from 162 parents) Compliance: 98%	In-depth interview	Parents reported psychological losses resulting from illness in personal functioning and in relation to partner and child. Mourning processes prevailed in spite of child's survival. Parents reported preservation of anxiety and problems and few positive changes. Specific coping strategies observed were expression of emotions, outlook on child's survival, parenting, and family planning.
Van Dongen-Melman et al (Chapter 4.2)	Assessment of psychosocial problems and risk factors affecting these problems	133 parents (69 mothers, 64 fathers) Compliance: 98%	Questionnaire measuring uncertainty, loss of control, self-esteem, and negative feelings (depression, anxiety, disease-related fear, psychological and physical distress, sleep disturbances, loneliness; see Chapter 2.3).	Uncertainty and loneliness were the most reported problems: parents 'suffer in silence'. No decline in problems over time was found. Risk factors for late problems included: female sex, parents without religious affiliation, low SES, and family stressors unrelated to cancer.
Van Dongen-Melman et al., (Chapter 4.3)	Investigation of impact of childhood cancer on family planning	130 parents (68 mothers, 62 fathers) Compliance: 96%	Interview; Questionnaire	In one third of families family planning was affected. Most affected families refrained from further offspring. Psychological reasons for their decisions were given.

[1] Studies with no separate analysis for disease stage were excluded: refs. 95 and 99.

TABLE 4: Frequencies on the uncertainty scale by parents ($N=133$).

Description and instruction of items of the uncertainty scale	%	*n*
Parents of children who have been ill are left with a number of questions after treatment has been completed, questions they would like to know more about. To what extent do you like to know more about:		
1. the cause of the disease	81	108
2. the present physical condition of your child	81	108
3. the long-term chances of survival of your child	80	106
4. possible side effects of the treatment in the long run	88	117
5. the extent to which your child has been physically impaired by the treatment of the disease	84	110
6. the purpose of the follow-up examinations	68	90
7. when your child is considered cured	50	66
8. how to talk or deal with a doctor	50	66
9. which people you come in contact with, you have to inform about the disease	50	67
10. where to go with questions and problems around the disease of your child	56	75
11. where to obtain good information material or literature about the period after completion of treatment	61	81
12. the extent to which the illness left its marks on the child's emotions	84	112
13. what is going on in your child's mind in connection with the disease	86	114
14. how you can help your child in getting over the disease and treatment	81	106
15. whether you should talk about the disease with your child or not	53	71
16. in how far you should adapt your child's upbringing to what he or she experienced because of the disease	46	61
17. which demands you may make as to your child's learning at school	57	75
18. how you can help your child to mix with peers	51	67
19. what questions other parents have about their child who has been ill	39	52
20. how other parents try to solve these questions	37	49
21. how other parents cope with the disease of their child personally	37	48

not revealed by questionnaire (93). In our study only 25% ($n=33$) of the parents indicated by questionnaire to worry about a potential recurrence of the disease. Thus, discrepancies may be explained by differences in assessment methods (interview versus questionnaires), but also by differences in conceptualization. For example, although relatively few parents reported fear of relapse, 80% of parents reported to be *uncertain* about the survival chances of their child (Table 4). Furthermore, age of the survivor may play a role in reporting concerns. For instance, Wallace et al. (93) found that the most common concern of parents was related to the child's ability to have children. The limited worries of parents of 8- to 12-year old children in our sample about issues as fertility, marriage prospects, and jobs, may become more prominent during adolescence and young adulthood.

TABLE 5. Frequencies on the disease-related fear subscale by parents (N=133).

Description of instruction and items of the disease-related fear subscale	%	*n*
Below you find a number of concerns expressed by parents with a child who has been ill. Could you indicate to what extend you are worried:		
1. about the late after-effects of treatment	42	42
2. about the growth in height of your child	17	22
3. about his/her sexual development	20	27
4. about his/her (later) fertility	20	27
5. about the school performance of your child	24	32
6. about your child's possibility of finding a good job in the future	28	37
7. about the way your child associates with other children	13	17
8. about the emotions of your child	14	18
9. about the chances for your child to find a partner in life	12	16
10. about a complete decline of your child	8	9

Overall psychosocial functioning

We were able to compare the levels of problems reported in different areas. In this way the different findings in the literature about low levels of parental distress (39,46,47,98) and high levels of lingering concerns (40,43,90,97) can be linked. Our findings indicate that most parents of childhood cancer survivors continue to feel uncertain about the well-being of their children (90%) and report feelings of loneliness (84%), but at the same time these concerns are not accompanied by high levels of distress as measured by anxiety, depression, disease-related fear, sleep disturbances or psychological and physical distress.

To interpret the precise meaning of the level of reported problems, comparative data would be useful. In our study no comparison group was included, but more insight in the level of problems is provided by rating parental functioning on the Global Assessment Scale (GAS) (101). A mean GAS score of 73.5 (SD=12.6) was found for mothers and a mean score of 75.6 (SD=9.9) for fathers. Moderate to marked impairment was found in 11% of the mothers and in 6% of the fathers. These scores confirm that mothers experience more problems than fathers (Chapter 3.2). Furthermore, findings on the GAS support the hypothesis that parents may experience stresses, but than these stresses do not lead to serious problems in day-to-day functioning. This idea is in line with several studies who reported that anxiety and depression in parents of childhood cancer survivors did not differ from comparison groups (39,46,98).

Perseveration of problems

The impact of childhood cancer on parents does not decrease over time (Chapter 3.2; 98). All problem categories remained at the same level, independent of the lapse of time since treatment. There was a slight tendency for negative feelings to increase with length of time since treatment. This is in contrast to the longitudinal findings in the Kupst and Schulman study (94), who found improvement in adjustment 6 years after diagnosis in comparison to the time under treatment. However, attrition of the sample and poor psychometric properties of the instrument measuring adjustment may have affected their results. We hypothesize that although there may be a decrease in level of problems as compared to the time at diagnosis, once treatment has been terminated, existing problems will decrease no more.

Risk factors affecting psychosocial problems

Confrontation with long-term physical sequelae in the child

Many parents are uncertain and worry about possible late-effects in their child (Table 4), but when parents actually are confronted with late physical sequelae, it has a substantial effect on late psychosocial problems (39; Chapter 3.2). Particularly when these changes are of a permanent and irreversible nature (e.g. tooth damage, sterility, and neuropsychological deficits), parents have to live with the fact that the child is cured of cancer, but is not the same healthy child as before. Even minor side effects are a constant reminder of the fact that the cancer experience cannot be restored and left behind. Long-term sequelae substantially increased loss of control and negative feelings in parents. The significance of long-term sequelae on parental psychosocial problems is further illustrated by the fact that other variables, related to either the disease itself or the treatment (e.g. the child's prognosis, relapse, length of treatment, intensity of treatment, or irradiation), yielded no relation with outcome. This suggests that it is the present confrontation with stresses rather than stresses in the past (i.e. during the acute treatment phase) that predicts poor psychosocial outcome in parents (Chapter 3.2).

Other factors affecting psychosocial functioning of parents

An important set of variables influencing the risk of late psychosocial problems were demographic variables and psychosocial stressors. These variables especially had an effect on self-esteem and negative feelings. Being a mother, low SES, no religious affiliation, and a chronic disease in another family member increased the risk of late psychological sequelae. Although it may be argued that these risk factors per se are associated with low self-esteem and negative feelings, it also shows that an extra stressor such as childhood cancer overburdens these parents. Thus, parents who experience multiple stressors have more difficulties in coping with the problems inherent to childhood cancer. Marital status may also have an effect (6), but we could not investigate this risk factor because there were only four single-parent families.

In the literature, a number of variables have been reported that were found to protect the individual from stress. A protective variable that emerged from this study is religious belief, particularly for parents with limited resources such as having low SES and concurrent stresses. In Western industrialized society where the role of religion has rapidly declined, it was surprising to find religion strongly related to lower levels of psychosocial problems. The supportive role of religion may be explained by providing extra strategies to cope with childhood cancer. Apparently, religion is a powerful tool that gives meaning to suffering. Furthermore, religion may provide an extra source of support and comfort, especially when people are actively involved in the religious community.

Marital relationship

During treatment heightened marital discord has been reported (102-106). In our sample, parents reported considerable strain on their marriage. When a child falls ill, parents may expect that they go through this ordeal together and share the same feelings. But, although it is the same child who is seriously ill, partners may experience the illness differently. In addition, they may cope with it in different ways. They are confronted with the fact that the illness is not a shared experience. Partners can become aware that there is a maximum to their togetherness and that one can feel very lonely, despite the marital relationship. This sense of loneliness within the marriage can cause great marital frictions, as is illustrated by a parent: *"Apart from the risk of losing your kid, you are also facing the risk of losing each other"*. It may be hypothesized that the larger the difference between the partners in experience and coping, the more loneliness is experienced. It has been shown that parallels between partners' coping styles are related to the marital quality and support from the spouse (102).

Although parents delineated changes in their marital functioning during treatment, in general these did not seem to persist into their present relationship. Divorce rate is an indication of the instability of the relationship. Of the 71 families who participated in our study, only two couples had divorced since the diagnosis. This divorce rate is lower, although not significantly, than in the general population for the comparable age-range as our parents (3% versus 6%; two-tailed z-test; $p=0.12$; data of the general Dutch population derived from the Centraal Bureau voor de Statistiek [Dutch Central Office for Statistics], The Hague, The Netherlands). Also in other investigations a low divorce rate in parents of cancer survivors was noted (39,43). Therefore, we conclude that the cancer experience does not increase divorce rate.

These consistent findings on the stability of parents' marriages might be explained by the notion that traumatic experiences bring couples closer together. Some parents

TABLE 6. Summary of studies on siblings of childhood cancer survivors.[1]

Study (reference)	Purpose of the study	Sample	Instruments	Informant	Major findings
Gogan et al., 1977 (131)	Investigation of current and retrospective experiences with the illness	13 siblings Median age: 17 yrs (range: 8-28 yrs) Compliance: NR	Interview	C	Siblings reported no major changes in life resulting from the illness.
Peck, 1979 (90)	Identification of problems in families	20 siblings Age: NR Compliance: NR	Interview	P	In 40% of families siblings had unresolved problems.
Gogan et al., 1981[2] (132)	Investigation of current and retrospective experiences with the illness	101 siblings Mean age: 17.8 yrs (range: 7-34 yrs) Compliance: NR	Interview; Death Anxiety Questionnaire	C	Illness made profound impact on their life: feelings of jealousy, resentment, isolation, and fear for own health were relatively common during treatment. Most siblings had resolved these feelings at the time of the study.
Van Dongen-Melman et al., (Chapter 4.3)	Investigation of current psychosocial adjustment	60 siblings Median age: 10.6 yrs (range: 5-16 yrs) Pair-wise matched controls Compliance: 95%	CBCL; Self-Perception Profile for Children; Children's Depression Scale; ABV-K: Neurotic Complaints, Somatization, Extroversion, Social Desirability	P,C	No major differences in emotional and behavioral problems and competence between siblings and comparison group were found. Siblings are not at risk for adjustment problems.

Abbreviations used: NR=not reported; C=Child; P=Parent; CBCL=Child Behavior Checklist; ABV-K=Amsterdam Autobiographic Questionnaire for Children.

[1] Studies on siblings of patients on and off treatment were excluded, because no separate analysis for disease stage was made: refs. 111 and 123.

[2] The study by Gogan et al. (1981) probably includes the sample of their previous study (1977).

reported that their relationship had improved. The number of parents reporting positive changes in marital functioning was small and may not account fully for the stable marriage rate. Marital distress may not only have adverse effects. Sabeth and Leventhal point to the adaptive functioning of conflict in families of chronically ill children (107). Other factors may also contribute to a stable relationship. Firstly, spouses have been reported to be the most important source of social support during the illness (102). Secondly, parents may learn that unhappiness caused by divorce is something they are able to control themselves in contrast to the limited control they have over the outcome of their child's disease. Thirdly, conflicts troubling other couples may seem trivial to these parents in comparison to childhood cancer. Fourthly, parents are very concerned about the survivor and may not want to jeopardize his development further by divorce. Further studies are needed to elucidate the dynamics affecting quality and stability of the marital relationship.

LATE PSYCHOSOCIAL CONSEQUENCES FOR SIBLINGS

Childhood cancer is one of the few chronic diseases of which the patient can become completely disease-free. At cessation of treatment, the sibling makes a transition from being a sibling of a chronically ill child to one of a healthy child. Late medical effects, however, may cause the sibling to become a sibling of a physically or mentally handicapped child.

Most studies report that during treatment, siblings of cancer patients experience more emotional problems (108-111). Others studies, however, focusing on adaptation and coping, indicated that siblings cope well (112,113) and do not experience significant behavioral problems (114). Several reviews (115-117) lucidly describe the problems in comparing and interpreting studies on psychosocial functioning of siblings of chronically ill children. These studies encounter comparable methodological problems as described for studies on childhood cancer survivors. Although results on adjustment and outcome variables differ, there is much consensus about the subjective distress for siblings of cancer patients (118-127). The major theme in discussing the siblings' concerns is the disruption they experience in the relationship with their parents (127,128). They are treated differently and feel isolated from support systems inside and outside the family (129,130). As a result they have been labelled as the 'forgotten children' (130). The question is what the consequences are of being exposed to a prolonged period of family disequilibrium.

Studies on siblings after cessation of treatment are sparse (90,111,123,131,132; summarized in Table 6). Although well-designed, reported studies were not planned to assess the current psychosocial functioning of the siblings in-depth. Therefore, we performed a study on the psychosocial functioning of siblings by standardized measures (Chapter 4.3). Our results show that siblings of childhood cancer survivors did not differ in psychosocial functioning from children in comparison groups. In fact, on all measures of emotional and behavioral problems and competence, the total group of siblings fared the same or even better than their comparisons. Although clinical experience and research demonstrates that during treatment siblings have emotional problems (108-111,118-130), our data indicate that childhood cancer does not result in a heightened risk for psychological disturbance for siblings as a late effect. The fact that parent and sibling reports coincided, underscores this finding.

Although parent and sibling reports coincided on psychosocial adjustment, uncertainty remains about the validity of these findings. Siblings may have underwent a similar 'response shift' as patients and therefore they may underreport problems. In addition, parental reports may be biased. In Chapter 2.3 it was found that parental adaptation was more related to the survivors' functioning than to the siblings' functioning. Despite attempts to focus on the impact of the disease on siblings during the interview with parents, this topic was generally addressed briefly. It may be that parents attempt to disaffirm the impact of childhood cancer for the siblings as a coping device. Consequently, a favorable score by the parents on siblings' functioning may have resulted from limited attention to the siblings or from strategies to cope with the disease. However, non-familial assessment of the siblings' adjustment validated siblings' and parental perception. A mean of 80.1 (SD=12.5) for siblings on the CGAS was found. Only 9% of the siblings had moderate to marked adjustment problems, whereas 85% fell into the category of showing adequate to very good adjustment. These findings underline that siblings adjust well to the period of survivorship.

We found few demographic, family, and disease-related factors that influenced siblings' adjustment. This finding can be attributed to the sibling's overall good functioning, because these factors have to discriminate between well adapted and better adapted siblings. In the literature, several factors are considered important to explain the differential adjustment in siblings of chronically ill children (115-117). The situation for siblings of childhood cancer survivors differs, however, from that of siblings of children undergoing cancer treatment. During treatment, siblings of childhood cancer patients are continuously confronted with the illness, whereas after termination of cancer treatment the direct medical stresses subside. The fact that the permanent presence of a chronic disease in another family member increases psychosocial problems for siblings may lend support to this supposition.

LATE PSYCHOSOCIAL CONSEQUENCES FOR THE FAMILY

Twenty-five years ago, Binger et al. (133) wrote an influential article on the emotional sequelae on the family of a child who died of leukemia. Binger et al. (133) found in more than 50% of the families a severe problem in one of the family members. At that time childhood leukemia had an almost invariably fatal prognosis. As a result from improved medical treatment, we are now able to describe the emotional consequences on

TABLE 7. Psychosocial adjustment of childhood cancer survivors and their families (N=71).

	Children		Parents	
(C)GAS category[1] (score)	Patients (n=67)	Siblings (n=56)	Fathers (n=63)	Mothers (n=71)
Moderate to marked adjustment problems (31-60)	31% (n=21)	9% (n=5)	6% (n=4)	11% (n=8)
Mild adjustment problems (61-70)	16% (n=11)	5% (n=3)	24% (n=15)	28% (n=20)
No adjustment problems (71-100)	52% (n=35)	86% (n=48)	70% (n=44)	61% (n=43)

[1] Adjustment of patients and siblings was measured by the Child General Assessment Scale (CGAS) (84) and adjustment of parents was measured by the General Assessment Scale (GAS) (101).

the family of a child who survived cancer. Although using different criteria as Binger et al. (133), we found that in 42% of families at least one member had a score on the GAS or CGAS that fell in the category of moderate to marked adjustment problems. Even though the outcome of medical treatment is different, childhood cancer remains a major crisis for the family with long-lasting aftereffects.

In Table 7, the adjustment problems for all family members are summarized. Although the childrens' scores on the CGAS and the parents' scores on the GAS can be compared, comparisons between childrens' and parental scores need to be cautiously interpreted. As indicated in Chapter 2.1, the cut-off points for the GAS are less well validated. Therefore we tentatively compared the scores between parents and children. Table 7 shows that family members are differentially affected by the cancer experience. The cancer survivors are most seriously affected, followed by mothers, fathers, and siblings. These findings in combination with the results of Chapters 3 and 4 can be interpreted that in patients cancer results in significant changes in psychosocial adjustment. Mothers and fathers experience permanent changes in their perspective on life and experience a continuation of uncertainties and problems with respect to the survivor, but the impact of these changes on adjustment have a milder effect on psychosocial adjustment. In siblings, childhood cancer has minor long-term consequences for adjustment. The absence of sequelae in siblings stand in sharp contrast to the late effects of the cancer experience on survivors and their parents. It might be hypothesized that as the direct stressors associated with the illness subside, the family restructures towards a pre-illness stage, which will alleviate the major stressor for the siblings. Thus for siblings, the stressors associated with childhood cancer decrease, whereas for survivors and parents the confrontation with the disease, the concerns and anxieties over the recurrence of the disease, and its long-term medical consequences continue. This suggestion is supported by the fact that long-term sequelae in the survivor affect patients and parents, but not siblings (see Table 8).

TABLE 8. Effects of risk factors on psychosocial functioning of patients, parents, and siblings.

Risk-factors	Patients	Parents	Siblings
Demographic factors			
.gender	- -[1]	- -[2]	-[1,2]
.age at investigation	-[3]	0	0
.low SES	0	- -	- -
.no religion	NT	- -	NT
Intrapersonal factors			
.locus of control	NT	0	NT
.social desirability	NT	0	NT
Family-related factors			
.family size	NT	0	0
.birth order	NT	NT	0
.concurrent stresses	- -	-	0
.chronic disease	- -	- -	- -
Disease-related factors			
Diagnosis			
.diagnosis leukemia	-	0	0
.age at diagnosis	-[4]	0	-[3]
.prognosis at diagnosis ≤ 50%	-	0	0
.relapse	NT	0	NT
Treatment			
.severe treatment	0	0	0
.duration of treatment ≥ 3yrs	- -	0	0
.surgery	0	-	0
.cranial irradiation	- -	0	+ +
Long-term sequelae			
.time off treatment ≥ 5yrs	-	0	-
.invisible impairment	0	- -	0
.visible impairment	0	-	0
.overweight	-	-	0
.school problems	-	- -	+

Abreviations used: SES=socioeconomic status; NT= not tested.

--: strong negative effect (in children: risk-factor has effect on at least two outcome variables; in parents: individual risk-factor has effect on reported problems) -: negative effect, 0: no effect, +: positive effect, ++: strong positive effect (risk-factor has effect on at least two outcome variables),

[1] male; [2] female; [3] older age; [4] younger age

Table 8 summarizes the effects of risk factors on psychosocial functioning of family members that were assessed in this study. Two risk factors had an effect on the whole family: gender and the presence of a chronic disease in another family member. The limited influence of risk factors in children may be due to the nature of risk factors tested. In addition, it has been hypothesized that family resources, parental behavior, social support, intrapersonal factors, and patterns of coping can moderate the effects of risk factors on adaptation (134). Open communication, psychosocial adjustment of child and parent, patterns of coping, and the availability of support enhance adjustment (40,56). These factors point to the primary importance of the family in the adaptation of family members to childhood cancer.

Family dynamics

Cancer in a child causes a change in the life pattern of the family (128). In terms of family-system theory, a change in a part of the system affects other parts and the system as a whole. The family members interact in a balanced way to maintain homeostasis (Figure 2), but is brought into disequilibrium by the disease (135). The illness requires changes in family structures that can threaten the integrity of the family. Consequently, the family will search for a new equilibrium (136). In contrast to an acute illness, a chronic disease forces the family to make long-term adjustments. Each family member has to cope with the changes in family interaction in addition to the individual emotional adaptation to the illness. Logically, parents will direct most time and attention to the ill child. This implies that there is less interaction with the other children and also less time and energy for the partner. The ill child becomes the most important family member, because the emotional atmosphere in the family is dependent on his or her physical and emotional well-being (Figure 2).

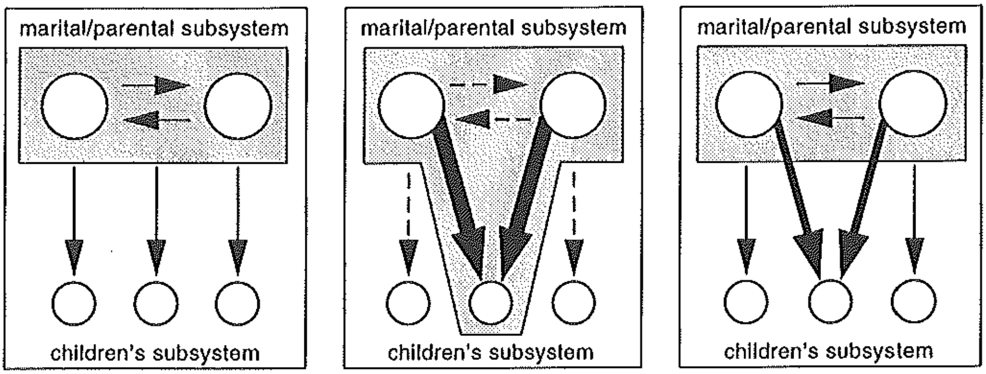

Figure 2: Hypothetical functioning of the family system according to disease stage: pre-illness period (left panel), during treatment (middle panel), and after cessation of treatment (right panel).

After termination of treatment, the family has to adapt once again to a new situation. Will the family system return to its former structure or will a new structure develop? After termination of treatment the direct stressors associated with the disease decrease. The family will reorganize to adapt to the new needs and demands of living with a cancer survivor. We found that parents, as a result from the perseveration of uncertainty, anxiety, and the confrontation with late sequelae, tend to perceive their child as a vulnerable child; vulnerable with respect to its physical condition and its psychosocial development. They may feel that the child is disadvantaged in comparison to siblings and peers. This disadvantaged position is a major reason for parents to improve the child's emotional and physical well-being. They respond with ardor to fulfill their responsibilities in order to overcome this situation and they strive to achieve 'normalcy' for the 'non-normal' child (Chapter 3.1). In this way, parental actions will influence the parent-child interaction. Therefore, we expect that the family moves towards a new equilibrium different from the pre-illness period (Figure 2). In has been observed that families with a survivor differ in family structure, i.e. in cohesion and adaptability, from comparison groups and normative samples (39,46,54).

Parental actions towards the child can be considered as a mode of reducing stress. By improving the situation with respect to the child, parental stress is alleviated. In this way parenting can be viewed as a coping strategy (Chapter 3.1). More insight is needed into what kind of parenting behavior is used towards the survivor and its effect on the child's adaptation. In addition, differential treatment can affect the siblings' adaption (137-140) and therefore the effect of parenting on the siblings needs to be studied as well. Currently, we are conducting such an investigation.

In Figure 2, changes in family structure over the three disease stages are depicted for one particular type of family system. However, each family typology (see for instance 141-144) may adapt differently to each new phase of the disease. Longitudinal studies are needed to elucidate how different family typologies proceed through the various stages of the disease.

IMPLICATIONS FOR PATIENT CARE

It is a general belief that when the child has been cured of cancer, all is well. A related assumption is that as the period of remission lengthens, the stress associated with the child's cancer dissipates. We have demonstrated that both beliefs lack scientific basis. The two major findings of our study are (a) that after termination of treatment, childhood cancer *has* late psychosocial effects for the family and (b) that these consequences persist over time. While the latter finding underscores the need for care, the former gives implications for the type of care needed.

Care for survivors

Our study shows that learning problems is a major issue for cancer survivors. Especially children who receive cranial irradiation, including children with leukemia, lymphoma, and brain tumors, need to be monitored (neuro)psychologically over a prolonged period of time. Psychological assessment should start as soon as possible after diagnosis and should continue after cessation of treatment. In this way changes in intellectual development are detected early and unnecessary delay in appropriate management of learning problems can be prevented. It is important to inform children, parents, and school about the child's potential capacities and about the possibilities and limitations to improve the learning difficulties by training programs. Especially when no improvement can be reached, alternative strategies to cope with their deficits need to be developed to help them adapt to their capabilities.

Our results show that children surviving cancer have an increased risk of problems as a result from the stressfulness of the disease. This finding has important clinical implications, because if a child after cessation of treatment has serious problems, they may be the result from the stresses associated with childhood cancer. Without the realization that childhood cancer may cause serious problems, problems of these children may not be adequately diagnosed. Our study clearly showed that childhood cancer needs to be considered as a decisive factor in the symptomatology and type of psychosocial problems of children treated for childhood cancer. The importance of a correct diagnosis relates directly to the type of intervention. We hope that this information will result in an optimal and specialized intervention for the child who is in need of psychosocial care.

The nature of late consequences found in our studies also gives direction which type of psychosocial sequelae can be prevented. Social adjustment, introvert behavior, and interactions with peers need special attention. Recently special intervention programs have been developed, which may help to prevent late sequelae in this area (145,146). These programs start during treatment and provide extra coping strategies for the child how to deal with the diagnosis, visible effects of treatment, and other treatment-related issues. Children are stimulated and modelled with alternative behavioral strategies to cope with these cancer-related issues in social situations. Children with cancer may also profit from intervention programs which have been developed for children with dysfunctional styles of interaction. Such programs can be useful to develop a broader repertoire of social problem-solving skills. Special intervention programs are needed for socially withdrawn children to strengthen their sense of social competence and to develop assertiveness in peer-interaction (28,147).

Care for parents and survivors

Uncertainty is a main problem for parents of cancer survivors. Especially information is needed on the long-term consequences of cancer in their child. With respect to physical sequelae, since 1978 the Division of Hematology/Oncology provides medical follow-up services, which have been instigated by Dr. K. Hählen and Dr. G.E. Van Zanen, pediatric oncologists. The need for such consultations have recently also been acknowledged by others (148,149) and in an increasing number of clinics this kind of care for long-term survivors has now been introduced. This long-term follow-up will not only have beneficial effects for the survivors, but can also provide information on the physical sequelae for their parents.

In addition, parents are not only uncertain about the medical consequences, but they are equally uncertain about the impact of the disease on the child's psychosocial functioning. Therefore psychosocial follow-up services are needed, where parents and children can discuss their concerns and which are informative about current issues and developments in psychosocial aspects of childhood cancer. When the child completes treatment, support systems are either not offered or become less available to parents over time (46,93,97). For instance, the hospital staff is becoming less available. In addition, to discuss worries and concerns outside the family is not easy, because ignorance and preconceptions about the late consequences are an obstacle. Consequently, family and friends may not understand or anticipate needs. If parents try to talk about their worries, they often get uninviting responses and consequently, do not discuss the subject again. When available support systems decline, psychosocial follow-up is an effective support system for parents. Parents indicated that the mere fact of knowing that such a service exists, were they can turn to if needed, would be helpful.

Another way to provide information to parents is by written material. Wallace et al. (93) reported in her study that the majority of parents designated written material to be read at home as a means for help. In our study more than 60% of the parents indicated that it was difficult to get good reading material or literature about the period after completion of treatment. As a result of this study, a booklet is being written in association with the Dutch Society "Parents, Children, and Cancer" (cf. Candlelighters in the U.S.A.) to inform parents about the late psychosocial consequences of childhood cancer for the whole family.

However, providing information and answers is not sufficient. All parents in our sample regularly visit the (long-term) follow-up service. Still, there is a considerable need for information. Their quest for information is probably also a reflection of the families' struggle to adjust to the psychological losses evolving from a major unexpected trauma. Our experiences from the research project give corroborative evidence that many parents need an opportunity to discuss their own feelings with a psychologist specialized in childhood cancer. The high compliance rate in our study underscores this need, because participating in the study, in fact, meant an opportunity to receive care. By the in-depth interviews, parents were able to put their experiences into perspective and to process them.

The absence of serious problems is no reason to refrain from patient care. Most patients, parents, and siblings, adjust reasonably well on their own. One cannot have but great respect for the way they ensure their own emotional survival. However, the nature and seriousness of the consequences they are confronted with can be significant. Questions, uncertainties, worries, and late problems, can be dealt with more easily and effectively, if psychosocial support is available. Furthermore, there may be difficulties, albeit non-pathological, which can be solved or improved relatively easily and which make a dramatic difference on the quality of life of the individual family member. The fact that parents *spontaneously* reported beneficial effects from the interviews is characteristic. Ten years after the diagnosis of leukemia in his son, a father reported during the second interview: *"The first time you visited us, I said it was wonderful that you came along, because my wife needed an opportunity to talk. She's still so full of emotions. But the days after the interview, I noticed a change myself. It felt as if a stone had been lifted from my stomach, which I hadn't even noticed it was there".*

Another important task is to help children and parents cope when confronted with medical and psychosocial sequelae. After cessation of treatment, they can be faced with the fact that their child is not the same healthy child as before. Rehabilitation may be offered, but in a number of children residual sequelae remain. The literature on parents' adaptation to the birth of a mentally or physically handicapped child points to the stresses involved to accept this new reality. For parents of cancer survivors, this process starts later in the child's life. Psychosocial follow-up services need to support parents and children in their struggle to *accept* this situation.

Care for siblings

As discussed above, the fact that childhood cancer does not result in an increased risk for major psychological disturbance in siblings, does not mean that they may not have undergone alterations in psychosocial functioning or do not experience distress. The individual sibling may have lingering concerns and problems that do require attention. From the interviews with children we found that if there were lingering concerns, they focussed on the relationship with the parents and their position within in the family. We developed short counseling-sessions to help parents to initiate communication about these issues with their children. Afterwards parents reported that addressing the disadvantaged situation of the sibling was very effective to alleviate these problems. Even though siblings fare well, their quality of life can be enhanced by well attuned communication.

Obstacles in providing care and ways to overcome them

Parents may be reluctant to seek help for several reasons. They may feel stigmatized by referral to mental health professionals. In addition, parents said that they did not want to burden the staff with their non-medical problems. Parents felt that their worries as compared to parents with children under treatment were of lesser concern and consequently did not want to take up the staff's time. In addition, they are thankful that their child is alive and did not want to disappoint the oncologist or to look ungrateful to him/her by indicating that not all is well.

Our clinical experiences from the present research project show that inviting parents routinely to participate can alleviate these obstacles. We strongly advocate to provide psychosocial follow-up consultations on a routine basis. To reduce practical obstacles as transportation time and costs, we propose that these psychosocial follow-up consultations are combined with the medical follow-up consultations.

CONCLUSION

We conclude that surviving childhood cancer has late psychosocial effects for patients, parents, and siblings. These consequences do not dissipate over time. This underscores the need for psychosocial follow-up care for families after cessation of treatment. In psychosocial consultations, knowledge about the late effects of cancer in children and their families, and possibilities and limitations for rehabilitation should be integrated. The type of care should comprise a variety of approaches such as emotional and cognitive diagnostic procedures, counseling, interventions, and adequate referral for problems not primarily associated with cancer. This form of individualized family care should be fully integrated in the medical-follow up as a routine part of the care system. Optimal integration of medical and psychological knowledge about late effects is needed; therefore follow-up care should be provided in the cancer center where the child has been treated. With combined medical and psychosocial follow-up services, one can strive towards a situation where all is well that ends well.

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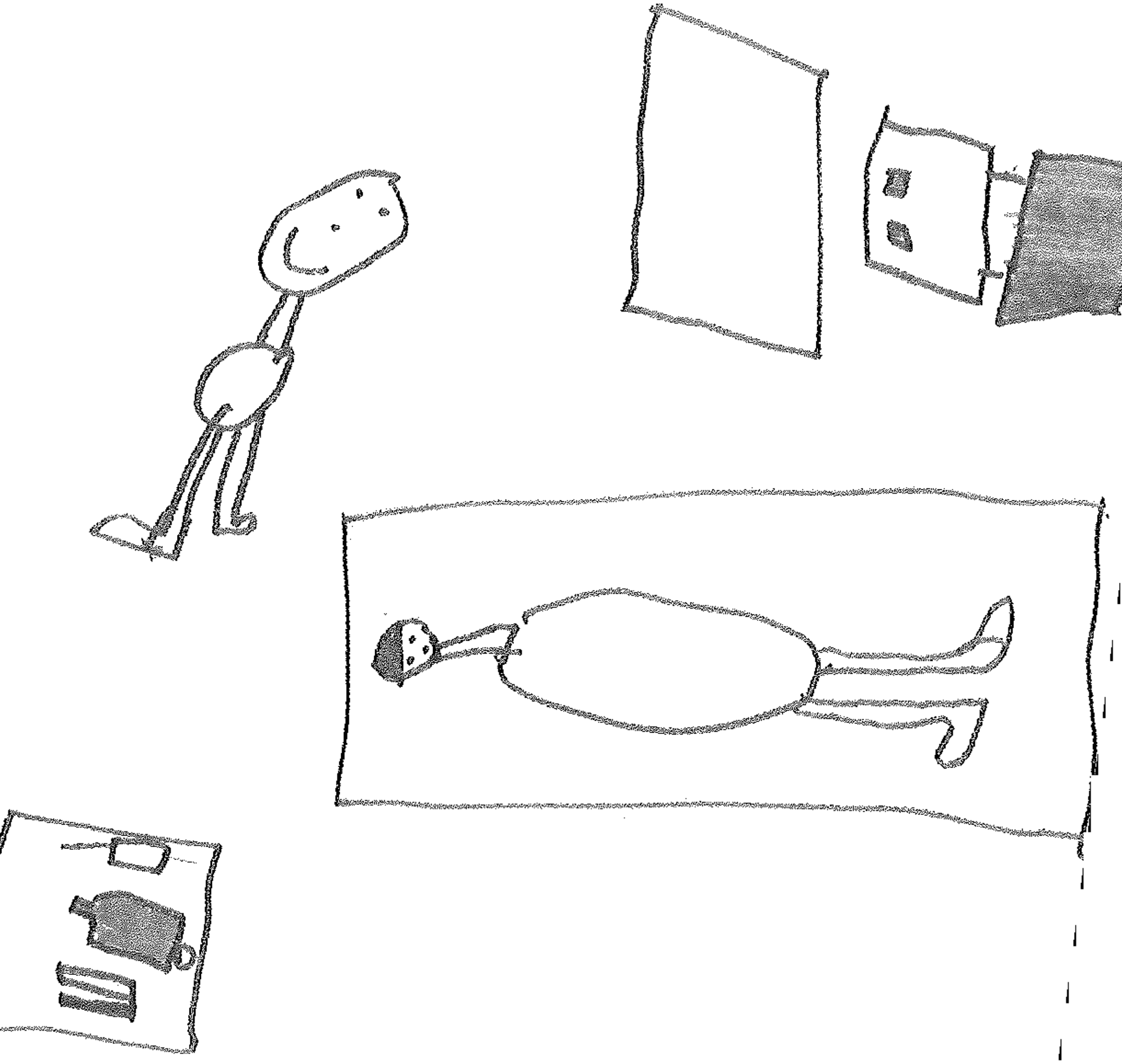
Summary

Jack, 8 yrs (cancer surviv



SUMMARY

Childhood cancer is a rare disease which yearly affects approximately 380 children in The Netherlands. Progress in medical treatment has drastically raised survival rates for childhood cancer patients. Whereas three decades ago, cancer in a child had an almost invariably fatal prognosis, at this moment childhood cancer has a much more favorable outlook. The child not only has an extended life expectancy but also a real chance of cure. At this moment about 70% of children with cancer survive their disease. However, childhood cancer treatment is long and intensive and the modern treatment regimens are not without side effects. Several side effects are reversible, such as physical discomfort, nausea and vomiting, weight loss, and loss of hair; other side effects are of a permanent nature, including growth retardation, skeletal abnormalities, overweight, sterility, and a decrease in IQ. Some of these late side effects only become apparent several years after cessation of treatment. With increased survival rates the costs of cure become apparent (Chapter 1.1).

In pediatric oncology, it is emphasized that disease-free survival is not synonymous with cure. Successful treatment of cancer must aim at 'truly' curing the child, reflecting that biological cure must extend to the quality of survival and also must ensure psychological and social well-being. Therefore, after cessation of treatment the main concerns are: (a) how the stresses during cancer and its treatment (Chapter 1.2) affect late psychosocial functioning and (b) which new problems evolve from the illness and treatment.

It has been estimated that by the year 2000, one out of 1000 young adults has suffered from childhood cancer. As more and more children survive their disease, insight into the long-term consequences of disease and treatment become increasingly important. Although information is accumulating on the medical side effects of cancer and its treatment, considerably less is known about the late psychosocial consequences for the child. There is especially a lack of knowledge about the late consequences for the child's family. Therefore, the purpose of this thesis was to obtain insight into the late psychosocial consequences on the family: patients, parents, and siblings. A related aim was to identify which children and families are most 'at risk' of developing late psychosocial sequelae.

Chapter 2 describes the materials and methods of the study. Studying the psychosocial consequences of childhood cancer is complex due to a number of methodological problems. These problems are not unique for childhood cancer, but pertain to chronic illness in childhood in general. Some of these methodological problems concern the sample, including sample size, subject selection, and compliance; other problems are related to the type of informants and assessment methods. Chapter 2.1 describes how we approached these issues in our study. With respect to sample, the importance of an unbiased, homogenous, and substantial sample size is underlined and our procedure to reach these standards is described. Furthermore, the need for different informants

reporting on psychosocial functioning is explained and we describe how they were introduced into the study design.

In studying the psychosocial responses to cancer the choice of instruments is a common problem. Already existing, highly standardized instruments allow for reliable judgement, but they often do not measure the problems specific for childhood cancer. Moreover, illness-specific instruments are sparse and their psychometric properties are often insufficiently studied. At this moment there are no instruments to assess the specific problems for parents after cessation of treatment. We developed such an instrument (questionnaire) for parents from a conceptual framework of coping with cancer. This framework describes the impact of a stressor on the individual and we show in Chapter 2.2 that this framework is applicable to describe the problems associated with childhood cancer. The use of a conceptual framework in developing an instrument is essential, because it indicates which aspects of psychosocial functioning need to be measured when parents are confronted with childhood cancer. In Chapter 2.3 this questionnaire, measuring uncertainty, loss of control, threat to the self-esteem, and negative feelings, is described. We extensively investigated the psychometric properties of the questionnaire. The results showed that the homogeneity of the scales was satisfactory, so that the questionnaire could be used in a reliable way to measure the problems for parents.

Because there is an emphasis to view the child as a normal individual adapting to a stressful situation, there is a need for instruments that can detect variations in psychosocial functioning within the normal range. The Self-Perception Profile for Children (SPPC), measuring self-concept, is such an instrument which is developed in the U.S.A. Before the SPPC could be used in Dutch childhood cancer survivors and their siblings, an adaptation of the instrument had to be made and had be to tested in Dutch children. In Chapter 2.4 we report satisfactory indices of reliability and validity demonstrating that the instrument holds cross-cultural generalizability.

In Chapter 3, the impact of childhood cancer on the parents is described. Chapter 3.1. describes our extensive qualitative analysis of in-depth interviews with parents to obtain insight into what it is like to live through childhood cancer. Parents indicated that having a child suffering from cancer was the most overwhelming experience in their life. By comparing their life before and after the treatment period, parents reported a number of significant changes. These changes were not incidental, but were of a definitive, long-lasting nature. The far majority of these changes was labelled as negative. Most of these negative changes could be categorized as losses. Although the child has survived cancer, the finding of losses seemed paradoxical. However, the losses are not related to the physical loss of the child, but are related to psychological losses. Parents experienced losses in the outlook of life, losses within the marital relationship, and losses with respect to the health of their child. As a result mourning processes prevailed in spite of the child's survival. In addition, parents experienced a perseveration of uncertainty, anxiety, and problems related to the recurrence of the disease, the occurrence of late medical and psychosocial side effects, and rehabilitation efforts. The losses experienced with respect to the child and the perseveration of problems made parents perceive their child as a vulnerable child. Positive changes such as personal growth, and a more gratifying relationship with the spouse and child, were incidentally reported by parents. A third category of secondary gains was added to describe negative consequences which are secondarily reinforcing to parents. Parents may use late sequela to conceal other problems such as marital problems. These gains were least found and only indirectly observed.

The use of our illness-specific instrument confirmed that childhood cancer had late effects on parents (Chapter 3.2). Uncertainty and loneliness were the most frequently reported problems by parents. Almost 90% of parents reported uncertainty and 84% reported feelings of loneliness. Furthermore, the relation between psychosocial problems and variables related to demographic, disease-related, and personality characteristics were investigated to identify parents 'at risk'. Parents with low socioeconomic status, who had no religious affiliation, who had additional stressors, and were a mother, had an increased risk of experiencing late problems. Treatment by itself had little or no effect on reported problems. However, when treatment led to long-term medical sequelae, a significant and distinct effect on parental problems was observed. This suggests that it is the present confrontation with stresses rather than stresses in the past (i.e. during the acute treatment phase) that predicts poor psychosocial outcome in parents. Surprisingly, no decline of problems over time was found. These results indicate that many parents 'suffer in silence', which may continue over the years after treatment has ended.

How parents cope with these issues and create their own psychological survival can be explained by delineating the major coping dimensions (Chapter 3.1). Specific coping strategies which originate from these coping dimensions are described. The first coping dimension concerns approaching or avoiding stress related to childhood cancer to regulate its emotional impact. The following strategies with this function were found: the ability to express one's emotions and the perspective one adopts concerning their child's survival. The latter strategy, taking a 'life' or 'death' perspective, is distinctive for childhood cancer. The second dimension concerns the attribution of control i.e. the extent to which parents regained control over the situation and/or were able to remove the stressor. A strategy in which attribution of control plays an important role is parenting behavior. Parents strive to minimize the disadvantaged position of the survivor. They try to achieve 'normalcy' for the 'non-normal' child. Another coping strategy on this dimension, is the way how parents proceed with family planning (Chapter 3.3). It was found that in one third of the families, either one or both parents reported that their reproductive planning was affected in various ways. The most extreme variations included having completed the family and still having another child versus not having completed the family prior to diagnosis but refraining from further progeny. Families who were unsettled concerning their family planning were most affected. In those who refrained from progeny, this strategy was used to regain control by preventing future stress or misfortune. In those who deliberately wanted another child, the new baby represented a compensation or a substitution for the negative feelings as a result from childhood cancer.

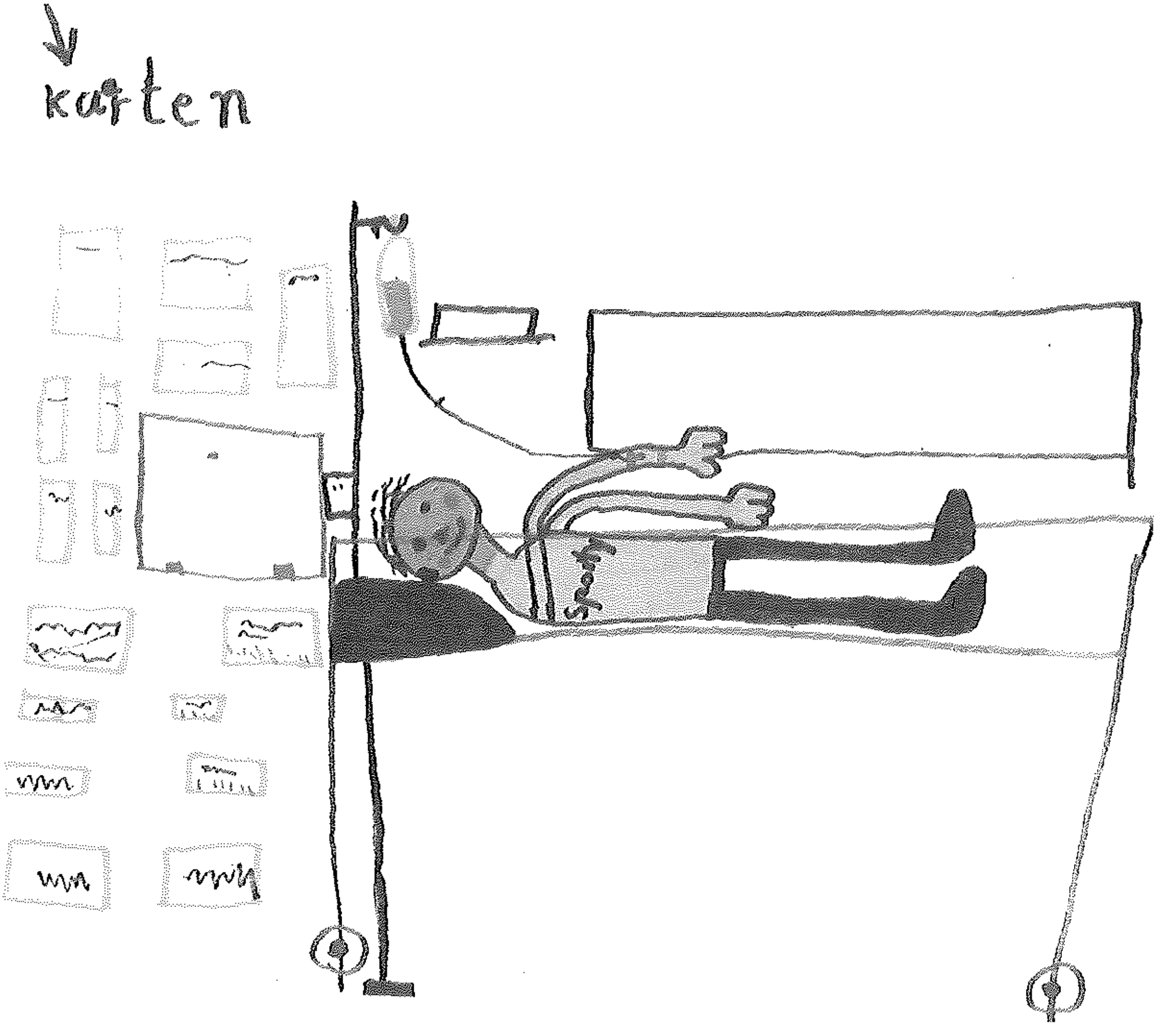
In Chapter 4 the late consequences for children surviving cancer and their siblings are described. In a study on the survivors' academic functioning, we found a dramatic increase in learning problems for children treated for leukemia and lymphoma with cranial irradiation to prevent involvement of the central nervous system (CNS). Over 80% of children treated with cranial irradiation had learning problems with half of these problems of such serious nature that special education was required. However, CNS preventive treatment with chemotherapy (methotrexate) was not associated with an increase in learning problems as compared to a group of survivors without any form of CNS treatment and matched healthy controls. These results underline once again the necessity to remove cranial irradiation from CNS preventive treatment. In addition, the results showed that other forms of CNS prophylaxis without cranial irradiation are not associated with an increase in learning problems (Chapter 4.1). Our data underline the need to eliminate cranial irradiation from CNS prophylaxis in children with leukemia and lymphoma and to consider alternative treatment regimens for children with initial CNS disease where cranial irradiation still is the mainstay of treatment.

Chapter 4.2 describes the nature and severity of late psychosocial effects on children surviving cancer. As compared to healthy peers, children surviving cancer showed more social problems and internalizing problems. Furthermore, our aim was to investigate whether differences between cancer survivors and healthy children can be regarded as variations in normal development or as clinically significant problems. Although the majority of the survivors adjusted well, underscoring the resilience of children, a substantial proportion of children had serious adjustment problems, especially boys: 27% of male survivors had serious problems as compared to 10% of their healthy peers. For girls, a slight, non-significant, increase in serious problems was found. Our study is the first to demonstrate an increased risk of psychosocial sequelae for cancer survivors by standardized measures. It refutes the current view that surviving childhood cancer causes no or only a slightly increased risk of psychopathology. Few individual factors related to disease and psychosocial stressors were found to increase the risk of late psychosocial problems. All risk factors combined showed a modest effect on psychosocial sequelae. It appears that gender and the cancer experience per se are the most dominating factors in developing late problems.

Little is known about the psychosocial adjustment of siblings of cancer survivors. Therefore, we investigated how this forgotten group of children adjusted after cessation of treatment (Chapter 4.3). Siblings of childhood cancer survivors were compared to control subjects on measures of general adjustment, perceived competence, neurotic complaints, somatization, extroversion, social desirability, and depression. On all measures of emotional and behavioral problems and competence, the total group of siblings functioned equally or even better than their comparisons, indicating that childhood cancer does not result in a heightened risk of psychological disturbance for siblings as a late effect. The fact that childhood cancer does not result in an increased risk for major psychological disturbance does not imply that siblings may not have undergone alterations in psychosocial functioning or do not experience distress. The individual sibling may have lingering concerns and problems that do require attention. From the interviews with siblings we found that if their were lingering concerns, they focussed on the relationship with the parents and their position within in the family.

Chapter 5 summarizes the results of our study on surviving childhood cancer, discussing the late psychosocial consequences for patients, parents, and siblings in the context of the literature. We conclude that surviving childhood cancer has different late psychosocial effects for patients, parents, and siblings. The child who survives cancer is most seriously affected, followed by mothers, fathers, and siblings. The complexity, seriousness, and duration of these consequences underline the need for psychosocial follow-up services. On the basis of our results we recommend routine psychosocial follow-up consultations integrated in medical follow-up programs to improve the quality of life of all family members and the family as a whole after treatment has ended.

Samenvatting



Peter, 9 yrs (cancer survivor)

SAMENVATTING

Kanker is een zeldzame ziekte bij kinderen. In Nederland krijgen per jaar ongeveer 380 kinderen kanker. Door de nieuwe behandelingsmethoden zijn de overlevingskansen de afgelopen decennia sterk toegenomen. Dertig jaar geleden had een kind met kanker meestal nog maar korte tijd te leven, maar momenteel bereikt ongeveer 70% van deze kinderen een 5-jaars overleving. De tegenwoordige behandeling biedt het kind met kanker niet alleen een langere levensverwachting, maar ook een reële kans op genezing. De behandeling van kanker is echter zeer intensief en langdurig en gaat gepaard met vele bijwerkingen. Sommige bijwerkingen zijn reversibel, zoals misselijkheid, braken, gewichtsverlies en kaalheid; andere bijwerkingen hebben een permanent karakter, zoals groeiachterstand, overgewicht, onvruchtbaarheid en een daling van de intelligentie. Deze effecten kunnen soms pas jaren na het beëindigen van de behandeling duidelijk worden. Doordat steeds meer kinderen hun ziekte overleven wordt de prijs van hun genezing duidelijker zichtbaar (Hoofdstuk 1.1).

In de kinderoncologie wordt benadrukt dat overleving niet hetzelfde is als genezing. Genezing houdt een volledig herstel in van de mogelijkheden van het kind zoals deze vóór de ziekte aanwezig waren. Dit houdt in dat naast biologische genezing ook gestreefd moet worden naar psychologische en sociale 'genezing'. Er is pas sprake van een volledige genezing, wanneer ook de kwaliteit van overleven bevredigend is. Vandaar dat na het beëindigen van de behandeling de volgende vragen uiterst relevant zijn: (a) in welke mate wordt het huidig functioneren van de ex-patiënt beïnvloed door de belasting tijdens ziekte en behandeling (samengevat in Hoofdstuk 1.2) en (b) met welke nieuwe problemen, voortkomend uit de ziekte en behandeling, wordt het kind later geconfronteerd.

Steeds meer kinderen overleven kanker. Recente schattingen geven aan dat in het jaar 2000, 1 op de 1000 jong volwassenen in zijn/haar jeugd kanker gehad zal hebben. Daarom wordt het in toenemende mate belangrijk om inzicht te krijgen in de lange-termijn gevolgen van ziekte en behandeling. Terwijl op medisch gebied veel onderzoek wordt gedaan naar de late lichamelijke gevolgen van kanker, is er nog weinig bekend over de late psychosociale gevolgen voor het kind. Nog minder is bekend over de lange-termijn effecten op het gezin. Het doel van dit promotie-onderzoek was om inzicht te krijgen in de late psychosociale gevolgen van kinderkanker op het hele gezin: het ex-patiëntje, de ouders, en de broers en zusjes. Tevens werd onderzocht welke kinderen en gezinnen extra risico lopen op het ontstaan van late psychosociale problematiek (Hoofdstuk 1.3).

De opzet van het onderzoek wordt besproken in Hoofdstuk 2. Eerst volgt een uiteenzetting over de methodologische problemen die het psychosociaal onderzoek naar de gevolgen van kanker bemoeilijken. Deze zijn niet uniek voor het onderzoek naar kinderkanker, maar gelden in het algemeen voor psychosociaal onderzoek bij kinderen met een chronische ziekte. Een deel van de methodologische problemen betreft de steekproef, zoals de grootte van de steekproef, selectie criteria en het percentage mensen dat toestemming geeft om aan het onderzoek deel te nemen. Daarnaast wordt het onderzoek gecompliceerd door de keuze van informanten en meetmethoden om het psychosociaal functioneren te onderzoeken. In Hoofdstuk 2.1 wordt onze benadering van de hierboven genoemde methodologische problemen beschreven en wordt aangegeven hoe wij deze problemen in ons onderzoek hebben gehanteerd. Het belang van een steekproef, die representatief, homogeen en van voldoende grootte is, wordt uiteengezet. Er wordt een onderzoeksprocedure beschreven om een dergelijke steekproef te verkrijgen. Verder wordt ingegaan op het belang om verschillende informanten bij het onderzoek te betrekken en hoe wij deze informanten in onze onderzoeksopzet hebben ingepast.

Een van de steeds terugkerende problemen in psychosociaal onderzoek is de keuze van meetinstrumenten om het psychosociaal functioneren in kaart te brengen. Reeds bestaande, goed gestandaardiseerde instrumenten geven weliswaar betrouwbare gegevens, maar kunnen niet de specifieke problemen meten die samenhangen met kinderkanker. Instrumenten die ziekte-specifieke problemen meten zijn echter schaars en hun psychometrische eigenschappen zijn vaak onvoldoende onderzocht. Zo bestaan er geen instrumenten die de specifieke problematiek kunnen meten bij ouders met een kind dat aan kanker heeft geleden. Voor dit onderzoek werd daarom een instrument (vragenlijst) ontwikkeld om de problematiek bij ouders in kaart te brengen. Hierbij werd gebruik gemaakt van een theoretisch model dat in Hoofdstuk 2.2 wordt beschreven. Dit model beschrijft welke problemen een individu ervaart, wanneer het geconfronteerd wordt met een stressor. Er wordt aangetoond dat dit model ook van toepassing is om de problemen die samenhangen met kinderkanker systematisch te beschrijven. Het gebruik van een theoretisch model bij het ontwikkelen van een vragenlijst is van essentieel belang, omdat het model richting geeft aan welke aspecten van het psychosociale functioneren gemeten moeten worden wanneer ouders geconfronteerd worden met kinderkanker. De ontwikkeling van het meetinstrument vanuit dit model en de beschrijving van het instrument wordt gegeven in Hoofdstuk 2.3. Vervolgens werd de vragenlijst uitgebreid onderzocht op zijn psychometrische eigenschappen. Uit de resultaten bleek dat de homogeniteit van de schalen goed was, zodat het instrument kon worden gebruikt om op een betrouwbare manier de problemen van ouders te meten.

Bij het meten van de invloed van kinderkanker is het belangrijk om de betrokkenen als normaal functionerende individuen te beschouwen, die moeten omgaan met een extreem stressvolle situatie. Er is daarom behoefte aan instrumenten die variaties in het normale gebied van het psychosociaal functioneren kunnen meten. Een dergelijk instrument is de 'Self-Perception Profile for Children' (SPPC), een zelf-concept vragenlijst die in Amerika is ontwikkeld. Voordat deze vragenlijst gebruikt kon worden in ons onderzoek moest deze eerst bewerkt en uitgetest worden bij Nederlandse kinderen. In Hoofdstuk 2.4 wordt beschreven dat onze Nederlandstalige SPPC cross-culturele validiteit heeft en voldoende betrouwbaar en valide is om bij Nederlandse ex-kanker patiëntjes en hun broers en zusjes te worden gebruikt.

In Hoofdstuk 3 worden de effecten van kinderkanker op de ouders beschreven. Via kwalitatieve analyse van *diepte-interviews*, werd inzicht verkregen in wat het voor ouders betekent een kind te hebben dat aan kanker heeft geleden en hoe dit hun huidige leven en functioneren beïnvloedt. Uit het onderzoek bleek dat ouders de ziekte en behandeling van hun kind als zeer traumatisch hebben ervaren. Voor veel ouders was de ziekte van hun kind een keerpunt in hun leven. In vergelijking met de periode vóór de diagnose was hun leven ingrijpend en blijvend veranderd. De meerderheid van deze veranderingen kon worden gecategoriseerd als negatief. Veel van deze negatieve veranderingen konden als verliezen worden beschouwd. Gezien het feit dat het kind is blijven leven, lijkt deze bevinding paradoxaal. Echter, de ervaren verliezen hadden geen betrekking op het daadwerkelijk verlies van het kind, maar op psychologische verliezen. Zo hadden veel ouders ervaren dat zij anders ten opzichte van het leven kwamen te staan. Zij ervoeren verlies van onkwetsbaarheid, teleurstelling in het leven, verlies van een toekomst, verlies van onbezorgdheid en verlies van levensvreugde. Ook in de relatie met hun partner en met betrekking tot de gezondheid van hun kind werden verliezen ervaren. Tevens beleefden ouders onzekerheid en angst, vooral met betrekking tot de mogelijke terugkeer van de ziekte en het ontstaan van mogelijke lichamelijke en psychologische bijwerkingen. Wanneer ouders werden geconfronteerd met blijvende lichamelijke beschadigingen bij het kind was dit vaak een extra zorg, ook in praktisch opzicht. Door deze blijvende zorgen en verliezen ten aanzien van hun kind was in de ogen van de ouder het ex-kanker patiëntje een kwetsbaar kind geworden. Het verwerken van deze verliezen houdt in dat rouwprocessen plaatsvinden, ondanks dat het kind is blijven leven. Naast negatieve veranderingen werden ook positieve veranderingen gerapporteerd, zoals een persoonlijke groei en een meer bevredigende relatie met partner en/of kind. Zowel het aantal positieve veranderingen als het aantal ouders dat deze rapporteerde was klein. Een laatste categorie om de ervaren veranderingen van ouders te beschrijven was secondaire ziektewinst. Deze categorie werd toegevoegd, omdat bleek dat enkele ouders uit de negatieve veranderingen die de ziekte met zich meebracht indirect voordeel wisten te halen. Zo werden negatieve veranderingen als gevolg van de ziekte in stand gehouden om andere problemen in het gezin te maskeren, zoals bijvoorbeeld relatieproblemen.

Ook de resultaten van de *vragenlijst* die ontwikkeld was om de specifieke problemen van ouders te meten na het staken van behandeling lieten zien dat kinderkanker specifieke lange-termijn gevolgen heeft voor ouders (Hoofdstuk 3.2). Ouders gaven hierop aan dat ze met name onzekerheid en eenzaamheid ervaren na het stoppen van de behandeling. Bijna 90% van de ouders gaf aan onzekerheid te ervaren en 84% rapporteerde gevoelens van eenzaamheid.

Tevens werd onderzocht welke ouders meer kans hebben op late psychosociale problematiek. Demografische, persoonlijkheids- en ziekte-relateerde karakteristieken werden gebruikt om het optreden van late psychosociale problematiek te voorspellen. Van de demografische- en persoonlijkheidskenmerken vergrootten de volgende vier kenmerken het optreden van late problematiek aanzienlijk: een lagere sociaal economische status, het ontbreken van een religieuze achtergrond, het geslacht van de ouder (moeders) en de aanwezigheid van extra stressoren in het gezin, zoals een chronische ziekte bij een ander gezinslid dan het kind dat aan kanker heeft geleden. Variabelen gerelateerd aan ziekte en behandeling zoals prognose, behandelingsverloop, type behandeling, en zwaarte en duur van de behandeling, hadden nauwelijks effect op late problematiek bij de ouders. Echter wanneer de behandeling bij het kind resulteerde in een blijvende beschadiging, leidde dit tot significant meer problemen bij de ouders. Dit betekent dat late problematiek bij ouders niet zozeer wordt veroorzaakt door de stress tijdens de behandeling, maar dat late problematiek voornamelijk een gevolg is van problemen die na het stoppen van de behandeling optreden. Een onverwachte bevinding was dat de problemen van ouders niet afnamen in de loop van de tijd. Deze bevinding, tezamen met het feit dat ouders vooral onzekerheid en eenzaamheid ervaren, kan geïnterpreteerd worden dat ouders 'in stilte lijden' (Hoofdstuk 3.2).

Verder werd inzicht verkregen in de wijze waarop ouders omgaan met de problemen die voortvloeien uit de ziekte (Hoofdstuk 3.1 en Hoofdstuk 3.3). Met andere woorden, hoe bewerkstelligen ouders hun eigen psychologische overleving. Twee belangrijke coping dimensies worden beschreven: (1) toenadering-vermijding van de stressor en (2) attributie van controle. In Hoofdstuk 3 wordt aangegeven op welke manier deze coping dimensies vorm krijgen in het gedrag van de ouders. De toenadering-vermijding coping dimensie, die een belangrijke rol speelt in het reguleren van emoties, komt tot uiting in een strategie die specifiek is voor kanker, namelijk de mate waarin de ouders een levens- of doodsverwachting hebben ten aanzien van de afloop van de ziekte (dood-leven spoor). Een andere strategie waarin deze dimensie tot uiting komt, is de mate waarin ouders hun emoties bespreekbaar kunnen maken. De tweede coping dimensie, attributie van controle, speelt een rol in het verkrijgen van controle over de situatie en het ongedaan maken van de stressor. Dit werd terug gevonden in het opvoedingsgedrag van de ouders. Ouders streefden er naar om de psychische en lichamelijke belasting die het ex-patiëntje heeft ondergaan op te heffen. Verder is ook gezinsplanning een middel om controle over de situatie te krijgen of om emotionele schade te verminderen. Deze laatste coping strategie wordt uitgebreid beschreven in Hoofdstuk 3.3. In een derde van de gezinnen gaven ouders aan dat hun gezinsplanning beïnvloed werd door de ziekte van hun kind. Er waren ouders die vóór de diagnose nog geen compleet gezin hadden, maar door de ziekte van verdere gezinsuitbreiding afzagen en er waren ouders die vóór de diagnose een compleet gezin hadden, maar als gevolg van de ziekte toch nog een baby wilden en deze ook kregen. Ouders die vóór de diagnose niet zeker waren of hun gezin al dan niet compleet was, rapporteerden het vaakst een verandering in hun gezinsplanning. Meer dan 60% van deze ouders zag af van verdere gezinsuitbreiding. Het motief voor ouders die na de diagnose geen kinderen meer wilden was dat zij hiermee mogelijk leed voor een nieuw kind wilden voorkomen. Voor de ouders die bewust nog een kind wilden betekende een nieuwe baby een vorm van compensatie voor de negatieve gevolgen van kinderkanker.

In Hoofdstuk 4 worden de late gevolgen van kanker voor de kinderen in het gezin beschreven: het ex-patiëntje en zijn broers en zusjes. In Hoofdstuk 4.1 worden de late gevolgen op het schoolfunctioneren van het ex-kankerpatiëntje beschreven. Er werd een sterke toename in leerproblemen gevonden bij kinderen die behandeld werden voor leukemie of non-Hodgkin lymfoom. Bij meer dan 80% van de kinderen die behandeld werden met schedelbestraling ter preventie van centraal zenuwstelsel (CZS) leukemie werd leerproblemen geconstateerd. Bij de helft van deze kinderen waren deze problemen dermate ernstig van aard, dat ze speciaal onderwijs moesten volgen. Bij kinderen met leukemie die een preventieve CZS behandeling met chemotherapie hadden ondergaan, werd geen stijging in leerproblemen gevonden, zowel in vergelijking met ex-patiënten die helemaal geen CZS behandeling hadden ondergaan als in vergelijking met gezonde kinderen. Deze bevindingen geven aan dat schedelbestraling als onderdeel van preventieve CZS behandeling volledig vervangen moet worden door cytostatische behandeling. Ook voor de behandeling van CZS leukemie, waarbij bestraling nog steeds de eerste optie van behandeling is, dient gezocht te worden naar alternatieve vormen van behandeling.

De aard en ernst van psychosociale problemen bij kinderen die aan kanker hebben geleden worden besproken in Hoofdstuk 4.2. In vergelijking met leeftijdgenoten hebben ex-kanker patiëntjes significant meer sociale problemen en laten zij meer internaliserend gedrag zien. Verder werd onderzocht of deze problemen beschouwd kunnen worden als een normale variatie in het psychosociaal functioneren of dat er sprake is van klinische problematiek. Alhoewel een ruime meerderheid van kinderen die aan kanker hebben geleden goed functioneerden, werden bij een substantieel aantal kinderen ernstige problemen gesignaleerd, met name bij jongens: 27% van de jongens had ernstige problemen. Dit was bijna 3x zoveel als bij leeftijdgenoten. Bij meisjes werd een lichte, niet significante stijging in probleemgedrag waargenomen. In ons onderzoek werd voor het eerst met gestandaardiseerde meetinstrumenten aangetoond dat kinderen met kanker een verhoogd risico hebben op ernstige late psychosociale problematiek. De huidige opvatting dat kinderkanker niet of nauwelijks invloed heeft op het psychosociale functioneren wordt hiermee weerlegd. Verder werden er weinig factoren gevonden die het probleemgedrag beïnvloeden. Dit betekent dat het geleden hebben aan kanker en het geslacht van het kind het meest bepalend zijn voor het optreden van lange-termijn problematiek.

Over de broers en zusjes van de ex-kanker patiëntjes is momenteel het minst bekend. In Hoofdstuk 4.3 wordt het onderzoek besproken naar het psychosociaal functioneren van deze 'vergeten' groep kinderen. Zestig broers en zusjes werden vergeleken met leeftijdgenoten op een groot aantal meetinstrumenten, die door de kinderen zelf en hun ouders werden ingevuld. Er werden geen verschillen gevonden in psychosociaal functioneren tussen beide groepen. Het bleek zelfs dat op sommige instrumenten broers en zusjes zichzelf competenter vonden en minder problemen ervoeren dan leeftijdgenoten. Geconcludeerd mag worden dat kanker bij een kind niet leidt tot een verhoogd risico op het ontstaan van late psychosociale problematiek bij de andere kinderen in het gezin. De resultaten houden echter niet in dat broers en zusjes geen veranderingen in psychosociaal functioneren hebben ondergaan of dat zij geen problemen ervaren. Uit de interviews met de broers en zusjes bleek dat zij zich zorgen maakten en onzeker waren over hun plaats in het gezin en de relatie met hun ouders. Deze gevoelens behoeven terdege aandacht en hebben implicaties voor de patiëntenzorg.

In Hoofdstuk 5 worden de late gevolgen van kinderkanker op het ex-patiëntje, ouders, en broers en zusjes in de context van de literatuur samengevat. De resultaten van alle gezinsleden werden geïntegreerd, zodat een totaalbeeld ontstond over de lange termijn gevolgen van kinderkanker voor het gehele gezin. We concluderen dat kinderkanker late psychosociale gevolgen heeft voor het gehele gezin, maar dat de ernst van de gevolgen voor elk gezinslid verschillend is. Het kind dat aan kanker heeft geleden ervaart de meeste problemen na het stoppen van de behandeling, gevolgd door moeder, vader en broers en zusjes. Op basis van het onderzoek worden aanbevelingen gegeven voor de nazorg aan ex-patiëntjes en hun gezinsleden. Gezien de complexiteit, ernst en duur van de late psychosociale gevolgen is het van belang om routinematig een psychosociaal follow-up spreekuur te houden. Dit spreekuur zal het best bij de behoefte van ouders en kinderen aansluiten, wanneer het wordt ingebed in het medisch follow-up programma.

ABBREVIATIONS

ABV-K : Amsterdam Autobiograhic Questionnaire for Children
AGFI : adjusted-goodness-of-fit index
ALL : acute lymphoblastic leukemia
AML : acute myeloid leukemia
ANCOVA : analysis of covariance
ANLL : acute non-lymphoblastic leukemia
ANOVA : analysis of variance
BFM : Berlin-Münster-Frankfurt study group
BMT : bone marrow transplantation
CBCL : Child Behavior Checklist
CDS : Children's Depression Scale
CGAS : Children's General Adjustment Scale
CNS : central nervous system
CT : computer tomography
DCLSG : Dutch Childhood Leukemia Study Group
df : degree(s) of freedom
GAS : General Adjustment Scale
G-CSF : granulocyte colony stimulating factor
GFI : goodness-of-fit index
ICC : intra-class correlation
IQ : intelligence quotient
M : mean
MTX : methotrexate
N : number of total sample
n : number of subsample
NA : not available/not applicable
NR : not reported
ns : not significant
NT : not tested
OR : odds ratio
p : probability
r : correlation
RMS : rhabdomyosarcoma
SES : socioeconomic status
SPPC : Self-Perception Profile for Children
SD : standard deviation

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Jeanette

Nieuwerkerk aan den IJssel,
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6. Van Dongen-Melman JEWM, Koot HM, Verhulst FC. Cross-validation of Harter's Self-Perception Scale for Children in a Dutch sample. Educational and Psychological Measurement 1993;53:739-753.

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15. Van Dongen-Melman JEWM, Hählen K, De Groot A, Van Dongen JJM, Verhulst FC. High prevalence of learning problems in children treated for leukemia and lymphoma is exclusively related to cranial irradiation: A comparative study. Submitted for publication.

16. Van der Reijden-Lakeman IEA, Slijper FME, Van Dongen-Melman JEWM, Verhulst FC. The effect of two years of growth hormone treatment on self-concept in intrauterine growth retarded children. Submitted for publication.

17. Van Dongen-Melman JEWM, De Groot A, Hählen K, Leenders-Van Loo I, Niesse-Lockefeer J, Verhulst FC. Late consequences of childhood cancer on parent-child interaction. In preparation.

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18. Van Dongen-Melman JEWM, Sanders-Woudstra JAR. Leerproblemen bij kinderen met acute lymfatische leukemie na CSZ-profylaxe. IKR-Bulletin 1985(1);9:20-21.

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