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Disease-related Distress in Parents of Children with Cancer at Various Stages after the Time of Diagnosis

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This study evaluates and describes disease-related distress in parents, with particular focus on the association between the time elapsed since the child's cancer diagnosis and a number of indicators of distress. In a cross-sectional design, 264 mothers and fathers of children with various malignancies completed a multidimensional questionnaire focusing on 11 illness-specific and general indicators of distress. Parents were assessed from 4 weeks to 14 years after the child's diagnosis, and age of children at onset of illness ranged from newly born to 21 years (mean ~ 6 years). The levels of distress related to loss of control, self-esteem, anxiety, depression, sleep disturbances, and psychological and physical distress were lower among parents for whom a longer period of time had elapsed from the time of diagnosis. However, the time elapsed could not explain all of the variation in these stress reactions, or any of the variation in uncertainty, disease-related fear and loneliness. The child's age at diagnosis and treatment situation at assessment were surpassed by time elapsed since diagnosis as predictors of variance in parental distress. The pattern observed indicates the presence of disease-related distress even years after the completion of medical treatment. The findings point to the need for research to identify parents at particular risk of suffering long-term harmful consequences from the prolonged stress of parenting a child with cancer. The necessity of longitudinal studies to evaluate the proportion of acute stress in relation to chronic or cumulative parental stress is emphasized.

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Marked progress in the detection and treatment of childhood cancer has been made during the past few decades. During this period, there has also been an enhanced understanding of the necessity of dealing not only with the psychosocial needs of the sick child, but also those of the entire family, as part of the total care concept in paediatric oncology. The psychological and psychosocial well-being of the family, and in particular of the parents, is strongly affected by the uncertainty about the initial outcome of treatment, the immediate experience of seeing one's child suffer, worries about invasive treatment procedures and the uncertainty about the future and the child's long-term physical and psychological health and development (1, 2). Together, these factors constitute potential sources of immediate, as well as continuous and cumulative stress—in other words, an extraordinarily complex stress situation. A deeper understanding of how parental distress may or may not be expected to change with time provides invaluable guidance concerning who should be offered psychological support, and in what form, during the course of the child's illness and treatment.

The psychological consequences of parenting a child with cancer have been studied from different perspectives and with a variety of conceptual targets in focus, such as psychological or emotional adjustment, mental health, marital and family dynamics and the overall psychosocial impact on the life situation. The literature shows that these areas have been investigated with the focus on a variety of distress indicators, e.g. psychological symptoms like anxiety (3–6) and depression (5, 6), mental health (7–9), psychosomatic symptoms (10), symptoms of stress (11), posttraumatic stress (12–14), marital distress (3, 4) and quality of life (15).

Previous studies indicate that, immediately after the information about the diagnosis is given, various indicators of parental distress and psychological problems are found to be elevated when compared to normative data and data from parents of healthy children (3, 7, 11, 16). At this early phase parents have described how they experienced tremendous stress (17). After a period of time, however, lower levels of distress are usually found (4, 7). Indeed, according to single studies, several years after diagnosis, distress has been found to appear at lower levels corresponding to those of the general population (7), although other data suggest that a number of parents still experience increased distress and exhibit stress symptoms even after this long period of

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time (5, 9, 11, 16). In addition, certain types of anxiety appear to persist for decades after the initial diagnosis (1, 12, 18).

The findings reported in connection with previous investigations have been inconsistent, and occasionally the study designs used have had various shortcomings. The number of subjects examined has often been relatively small, making it difficult to draw definitive conclusions and to arrive at general principles on the basis of the trends observed. Several studies have restricted their focus to one or two dimensions of psychological distress only, and those assessment instruments that have been used have often failed to cover illness-specific measures of the unique problems relevant for this population. Furthermore, many studies have addressed mothers only, instead of both parents.

In the present study we seek to circumvent these problems, at least in part. The design, although cross-sectional, encompasses a large sample, and allows analyses of large subgroups of parents at different periods of time after diagnosis, thus providing insight into the variations in the level of parental distress at different points in time. By considering a number of various indicators of disease-related distress, the study supplements previous studies, which have focused primarily on the phenomena of anxiety and depression, or on more general concepts such as overall adjustment or mental health. Finally, in contrast to several earlier studies, in which only mothers were the subjects, the explicit aim here is to understand the reactions of both the mothers and fathers of the children with cancer.

The following specific questions were addressed:

1) What are the levels of the various studied indicators of disease-related distress among parents of children with cancer?
2) Do the studied indicators of distress in parents vary in a predictable manner following the time of diagnosis?
3) Is there a relationship between disease-related distress and the prognosis, the course of treatment and the age of the child?

MATERIAL AND METHODS

Procedures

Parents where consecutively included, when they attended the Paediatric Oncology Unit at Karolinska Hospital in Stockholm for treatment of their hospitalized child, or visited the outpatient clinic for their child's treatment or medical follow-up after therapy had been completed. Altogether, the study group comprised a total of 264 parents of 150 paediatric cancer patients.

Parents visiting the inpatient clinic were informed about the investigation during their children's hospitalization, while the parents of children attending the outpatient unit were informed by telephone. After the parents had received the initial information, an informative written letter including the questionnaires was handed over or sent by mail. If at the initial contact, parents had not already declared that they agreed to be included, a member of the study team contacted the families a second time about participation after they had viewed the written information and the questionnaire booklet. The shortest period that elapsed between the child's diagnosis and receiving an invitation to participate in the study was 4 weeks. Both parents received the questionnaire along with a return envelope, and the instructions were that each parent was to complete the questionnaire independently, without consulting the other parent.

Subjects

Parents, whose child had a known poor prognosis, i.e., would probably die of their disease, were excluded from the study, although exclusion a priori on the basis of the type of malignancy was done only in the case of pontine glioma. Parents of patients undergoing palliative treatment and parents who had lost their child were not approached. Insufficient fluency in the Swedish language (in which the instructions and questionnaires were written) was an additional criterion for exclusion. The language skills of parents were assessed by members of the clinical staff who had been in close contact with them. Of the 345 parents invited, 268 completed and returned the questionnaires. Four of the questionnaires received were excluded because of incomplete data, giving an overall participation rate of 77%. The final study group consisted of 264 parents (146 mothers and 118 fathers). Although not based on a participant attrition analysis, the impression was that attrition was more often than not associated with parents experiencing a more distressing life situation than 'normal'. One participant was a stepmother, and another a stepfather, while the remainder were the biological parents of the patients. All of the children were either undergoing treatment or being subject to routine medical follow-up for paediatric cancer or Langerhans' cell histiocytosis (LCH). Table 1 documents the distribution of diagnoses among the 150 patients whose parents took part in the study. The age of the patients at the time of diagnosis varied from immediately after birth to 21 years, with a mean value of 5 years, 10 months (SD 5 years).

The children were at different stages of treatment or had even completed treatment. The period that had elapsed from the time of diagnosis to the evaluations described below ranged from 4 weeks to 14 years 5 months, the mean value being 34 months (median 27 months, SD 34 months). The children of 103 parents (39%) were receiving active or maintenance treatment for their disease, while for the

Table 1

Distribution of the parents and their children according to the diagnoses of children, survival rates, and mean age of children at diagnosis in different diagnostic subgroups

	Children				Parents	
Diagnosis	Prognosis[a]	n	%	Mean age[d]	n	%
Leukaemia	0.72	68	45.3	5.5	118	44.7
Lymphoma	0.82	11	7.3	8.0	19	7.2
CNS tumours	0.69	15	10.0	5.4	28	10.6
Neuroblastoma	0.53	12	8.0	1.6	22	8.3
Renal tumours	0.88	10	6.7	4.5	20	7.6
Bone tumours	0.57	10	6.7	12.8	17	6.4
Soft tissue sarcoma	0.70	7	4.7	7.4	12	4.6
Germ-cell neoplasms	0.82	7	5.3	9.0	13	4.9
Langerhans cell histiocytocis	0.90[b]	8	5.3	5.21	12	4.6
Other/unspecified	[c]	2	0.7	0.8	3	1.1
Total		150			264	

[a] Retrospective 12-year survival probability for diagnostic groups in the Nordic countries (21).
[b] 5-year survival.
[c] Survival data not available.
[d] Age at diagnosis, years.

children of 161 parents (61%) treatment had been completed.

Evaluation of distress

A multidimensional questionnaire was used to evaluate indicators of disease-related distress. The assessment instrument focused on Parental Psychological Distress in Childhood Cancer, and will hereafter be referred to as PPD-C. This instrument was originally developed by van Dongen-Melman (19). The conceptual framework of the assessment model is based on theoretical modelling, the literature and in-depth interviews with parents of childhood cancer patients. A detailed account of the development of the instrument and the conceptual model behind it is presented elsewhere (6, 19, 20). The three subscales used to assess state anxiety, depression and psychological and physical distress were originally adapted for the childhood cancer parent population from three commonly used psychological scales, i.e., the State-Trait Anxiety Inventory, The Zung Self-Rating Depression Scale and the Rotterdam Symptom Checklist.

The remaining eight scales employed were constructed on the basis of the stress theory, scientific literature on emotional consequences of childhood cancer and in-depth interviews with parents of paediatric cancer patients aimed at identifying problems specific for this population. In the present study, we used a version of this questionnaire translated into Swedish which consisted of 11 subscales designated to evaluate the following variables: degree of uncertainty; loss of control by the parents as regards their functioning as individuals; loss of control as regards their parenting the sick child; loss of control as regards their parenting the sibling(s); level of self-esteem; anxiety; disease-related fear; feelings of loneliness; sleep disturbances; depression; and psychological and physical distress. The 125 items of the questionnaire were answered according to 2-, 3-, or 4-point Likert scales (Table 2). The in-depth interviewing of parents, which was part of the construction process of the original questionnaire, ensures the construct validity of the scales (19). Comprehensive validity and reliability evaluations will be documented in a forthcoming article focusing especially on the psychometric properties and on presenting normative data for the Swedish version of the assessment instrument.

Handling of the data and statistical analysis

For each subscale of the assessment instrument, a score was calculated for the respondents by dividing the total of all the individual values by the total number of answered items. In this way each parent had a mean value for each of the 11 scales. Respondents who had not completed at least 75% of the items at a given subscale were not included in the analyses of that particular scale.

In order to examine the distribution of scores and levels of distress (research question 1), parents were classified into three categories defined a priori, i.e. low, intermediate or high distress. In the case of 4-point scales, low, intermediate and high mean values were defined as <2, $2-3$ and >3, respectively. For 2- and 3-point scales only low (value <1.5 and <2, respectively) and high (value ≥ 1.5 and ≥ 2, respectively) categories of distress were employed. This classification strategy was chosen to be in concordance with the one used by Van Dongen and co-workers (6), and to facilitate comparisons with the Dutch study. The categorization, together with a supplementary statistical

Table 2

Measurement of psychometric variables by the PPD-C scale, and basic outcome data for the study group

Variable	No. of items	Response format (Likert scale)	Disease-related or general[a]	Mean value for all parents	SD	Cronbach's alpha
Uncertainty	21	4-point	DR	2.75	0.72	0.95
Loss of control – personal functioning	10	2-point	DR	1.39	0.30	0.82
Loss of control – being a parent	9	2-point	DR	1.39	0.29	0.80
Loss of control – the sibling(s)	10	2-point	DR	1.38	0.29	0.84
Self-esteem	4	3-point	G	1.76	0.55	0.76
Anxiety	17	4-point	G	2.43	0.68	0.95
Disease-related fear	11	4-point	DR	1.95	0.64	0.91
Loneliness	8	4-point	DR	2.08	0.69	0.91
Sleep disturbances	5	2-point	DR	1.47	0.34	0.77
Depression	10	4-point	G	2.08	0.59	0.86
Psychological and physical distress	20	4-point	G	1.72	0.56	0.93

Abbreviations: [a] DR = disease-related; G = general.

approach, was also used to delineate the level for clinically significant distress.

The length of time that had passed between when the parents found out that their child had cancer and when they answered the questionnaires was treated as a continuous variable. The situation regarding treatment of the child was considered as a dichotomous variable, with patients receiving active or maintenance treatment being assigned the value of 1, and those off treatment the value of 0. The value assigned for prognosis was based on retrospective statistical data for the Nordic countries concerning the probability (in %) of 12-year survival with the given diagnosis (21).

An explorative analysis of the interrelationships between the background variables was done by calculating the Pearson correlation coefficients, and the relationships between distress and background variables (research question 3) were analysed by performing a univariate regression analysis. Finally, a series of multiple regression analyses (MRAs) were executed in order to examine the absolute and relative abilities of the background variables—time from diagnosis in particular (research question 2)—to predict disease-related distress. In the first phase of these MRAs, the variables age of the child at the time of diagnosis, prognosis and situation with regards to treatment were considered. In the second phase, the variable of primary interest, i.e. the amount of time that had elapsed since the parents were told that their child had cancer, was entered, in order to assess the unique influence of this parameter alone.

RESULTS

Basic findings

Basic outcome data for the parents' responses to the questionnaire are presented in Table 2. All 11 scales exhibited acceptable-to-high internal reliability, with Cronbach's a values ranging from 0.76 to 0.95 (Table 2).

For *uncertainty*, the group mean (2.75 for this 4-point scale) was within the higher half of the severity span, corresponding to almost two-thirds of the parents responding at the higher end of the severity span. This means that they reported a fair amount or a considerable amount of uncertainty concerning, for example, the degree to which the illness had permanently affected the child, possible late effects of treatment and/or whether their child had suffered permanent damage because of the treatment. For *anxiety*, the group mean (2.43 for this 4-point scale) indicated a similar degree of severity, corresponding to about 45% of the parents feeling fairly or very afraid, worried, and/or brooding about unpleasant things.

The group mean for *sleep disturbances* (1.47 for this 2-point scale) indicated that about 46% of the parents reported that they suffered from difficulty in sleeping, early wakening, and/or during sleep re-experiencing situations associated with the child's illness. For *loss of control* items the group means (1.38–1.39 for these three 2-point scales) indicated that 26–37% of the parents reported experiencing these problems; for example they felt they were less in control of their emotions, had difficulty in feeling that they understood what was going on in their child's mind, and/or felt that their relationship with the patient's sibling(s) was less easy than before.

Less severity was noted for the other factors, with *psychological and physical distress* being the distress cluster with the least severity reported. Here, the mean (1.72 for this 4-point scale) corresponded to about 88% of the parents reporting problems in the lower half of the severity span. That is, they reported little, or no, suffering from, for example, headaches, tiredness, lack of energy and/or feelings of despair when thinking about the future.

Categorization into distress level groups

Table 3 documents the categorization of the scores of the parents with respect to the different indicators into low, intermediate and high levels of distress. As can be seen, *uncertainty* was an indicator for which high levels were reported by most of the parents, with about only 17% of the parents expressing low levels of uncertainty. Seventy percent of the parents expressed intermediate or high levels of anxiety, while 28% of the parents in the entire group reported psychological and physical distress at intermediate or high levels.

Based on the distress level group categorization, an effort was made to delineate the amount of clinically significant distress. With regard to the 4-point scales (categorized as 'low', 'intermediate' and 'high'), clinically significant distress was considered to be present if the parent had been assigned to the intermediate or high distress group. For 2-point and 3-point scales (categorized as 'low' and 'high' only), clinically significant distress was considered to be present if the parent had been assigned to the high distress group. Parents were then grouped into three subgroups of different distance in time from their child's diagnosis: 0–1 year; > 1–4 years; > 4 years. Analysis revealed that in all three parent groups the mean score for *uncertainty*, *loneliness* and *anxiety* exceeded the cut-off limit for clinically significant distress based on distress level group membership, indicating the presence of clinically significant parental distress in these areas close to time of diagnosis as well as in the long-term. Supplementary analyses using a general cut-off corresponding to the 75th percentile of the distribution of the scores on each separate scale, without reference to the high, intermediate and low distress group categorization, revealed that 6%–30% of parents in the group most distant from diagnosis presented scores at, or above, the cut-off. Within this most distant group parents scored at, or above, the cut-off most frequently at *loneliness* (30.3%), *uncertainty* (23.4%) and *control–the sibling* (19.7%). The lowest proportions of high scorers were found for *anxiety* (6.1%), *depression* (7.6%), and *physical and psychological distress* (12.1%), while the percentage for the rest of the distress variables ranged from 15.2 to 17.2.

Table 3

Distribution of the scores for the different variables assessed into low, intermediate and high levels of disease-related distress

Variable	Distress		
	Low %	Intermediate[a] %	High %
Uncertainty	16.5	41.7	41.7
Loss of control—personal functioning	57.8	[a]	42.2
Loss of control—being a parent	60.3	[a]	39.7
Loss of control—the sibling(s)	66.1	[a]	33.9
Self-esteem	59.0	[a]	41.0
Anxiety	29.9	46.2	23.9
Disease-related fear	60.2	31.8	8.0
Loneliness	42.7	46.2	11.1
Sleep disturbances	52.9	[a]	47.1
Depression	44.7	46.2	9.2
Psychological and physical distress	72.0	24.5	3.4

[a] 2- and 3-point response scales, for which only the categories 'low' and 'high' were employed.

Regression analysis

Regression analyses were preceded by the exploration of the relationships between the studied background variables. As might be expected, there was a marked correlation between the child's treatment situation and the time that had elapsed since the parents were informed about the diagnosis ($r = 0.56$, $p < 0.001$). The age of the child at the time of diagnosis was weakly associated with the time that had elapsed since the parents were informed about the diagnosis ($r = 0.16$, $p < 0.05$). No other associations between background variables were found.

Univariate regression analyses revealed that the parameters *time elapsed since diagnosis*, *treatment situation* and *age of the child at the time of diagnosis* were associated with some aspects of disease-related stress (Table 4).

Time elapsed explained a certain amount of the variance in all the variables examined, with the exceptions of *uncertainty*, *disease-related fear* and *loneliness*. The R^2 values obtained indicate that variation in time elapsed could explain 2.2%–13.9% of the variations in the different aspects of distress. The longer the period since the parents had been informed about the diagnosis, the lower were the levels of disease-related stress, as shown by the negative β-coefficient values. The regression lines for those three distress factors for which a marked decrease with time from diagnosis was indicated, together with three factors for which persistence over time was indicated are presented in Fig. 1.

The univariate regression analyses indicated that the other background variables examined were less strongly associated with distress. The treatment situation explained part of the variations in seven of the distress variables, while the age of the child at the time of diagnosis predicted a small degree of the level of four of the distress variables assessed. The prognosis failed to explain to any significant degree any variation in any of the outcome variables (not shown).

The MRA was performed in order to determine the relative significance of the time elapsed when the other background factors were controlled for (Table 5). Variation in time elapsed could predict 2.6%–5.5% of the total variance in the outcome concerning loss of control—personal functioning, loss of control—being a parent, self-esteem, anxiety, sleep disturbances, depression, and

Table 4

Univariate regression analyses of associations between background and distress variables

Variable	Time elapsed			Treatment situation			Age of child at time of diagnosis		
	R^2	β	F	R^2	β	F	R^2	β	F
Uncertainty			n.s.			n.s.	0.023	0.150	5.8*
Loss of control—personal functioning	0.120	−0.346	34.9***	0.097	0.312	27.6***			n.s.
Loss of control—being a parent	0.139	−0.372	41.1***	0.127	0.356	37.0***	0.018	0.134	4.6*
Loss of control—the sibling(s)	0.022	−0.150	5.2*	0.019	0.138	4.4*			n.s.
Self-esteem	0.101	−0.318	29.1***	0.039	0.196	10.4***	0.016	0.127	4.3*
Anxiety	0.133	−0.364	40.0***	0.084	0.289	23.9***	0.020	0.142	5.4*
Disease-related fear			n.s.			n.s.			n.s.
Loneliness			n.s.			n.s.			n.s.
Sleep disturbances	0.048	−0.219	13.2***	0.025	0.158	6.7**			n.s.
Depression	0.094	−0.306	26.8***	0.049	0.221	13.3***			n.s.
Psychological and physical distress	0.063	−0.250	17.3***			n.s.			n.s.

*** $p < 0.001$; ** $p < 0.01$; * $p < 0.05$; n.s. = not significant.

psychological and physical distress. In accordance with the outcome of the univariate regression analysis, level of distress was predicted to decrease as the time elapsed since information about the diagnosis increased.

In summary, certain symptoms of distress tended to be somewhat less frequent in parents whose children were in the post-treatment follow-up stage, whereas the child's age at the time of diagnosis and prognosis apparently did not contribute systematically to variation in distress. For most of the distress factors examined, lower scores were observed for parents who a longer time ago had been informed that their child had cancer although this association appeared to be weak.

DISCUSSION

The study was designed to investigate the level of disease-related distress, and to examine the time elapsed since diagnosis as possible predictors of psychological disease-related stress among parents of children with cancer. Seven

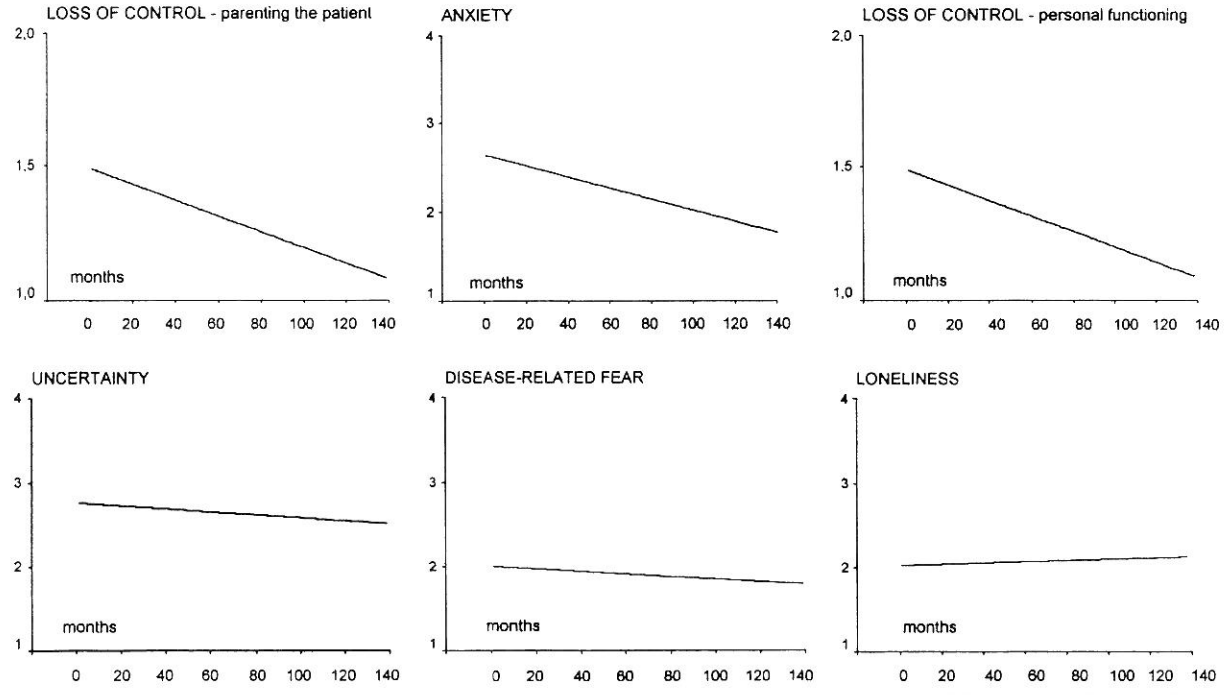

Fig. 1. Regression lines for distress indicators among parents of children with cancer associated with a decrease over time (upper row), and persistence over time from diagnosis (lower row). Distress represented by subscale scores (y axis), and time shown as months from diagnosis (x axis).

Table 5

Multiple regression analyses of predictors of disease-related distress in parents of children with cancer

Variable	Step 1				Step 2				
	R^2	β^a	β^b	β^c	Change in R^2	β^a	β^b	β^c	β^d
Uncertainty	n.s.				n.s.				
Loss of control—personal functioning	0.109***	n.s.	n.s.	0.319***	0.050***	n.s.	−0.140*	0.167*	−0.275***
Loss of control—being a parent	0.148***	n.s.	n.s.	0.355***	0.034***	n.s.	n.s.	0.229***	−0.227***
Loss of control—the sibling(s)	n.s.				n.s.				
Self-esteem	0.053**	n.s.	n.s.	0.191**	0.055***	n.s.	n.s.	n.s.	−0.288***
Anxiety	0.100***	n.s.	0.130*	0.283***	0.053***	n.s.	n.s.	n.s.	−0.282***
Disease-related fear	n.s.				n.s.				
Loneliness	n.s.				n.s.				
Sleep disturbances	0.039*	n.s.	n.s.	0.150*	0.026**	n.s.	n.s.	n.s.	−0.200**
Depression	0.054**	n.s.	n.s.	0.221***	0.044***	n.s.	n.s.	n.s.	−0.258***
Psychological and physical distress	n.s.				0.048***	n.s.	n.s.	n.s.	−0.269***

*** $p < 0.001$; ** $p < 0.01$; * $p < 0.05$; n.s. = not significant.
[a] Prognosis.
[b] Child's age at time of diagnosis.
[c] Treatment situation.
[d] Time elapsed since being informed about the diagnosis.

of the 11 manifestations of distress assessed demonstrated a negative linear relationship with the time elapsed, as might be expected. It is remarkable, however, that four of the symptoms of distress examined were not affected by the time in this manner and, furthermore, that the time elapsed explained only a minor portion of the variation in the other seven symptoms. It can be surmised that differences in the distress levels of parents with children who have cancer are influenced more strongly by other factors.

Certain study limitations should be considered. Although the presented analyses indicate that the stress symptoms assessed are sustained over time, the design of the study prevents us from drawing definitive conclusions regarding the amount of psychological discomfort experienced by these parents in comparison with parents of healthy children. The PPD-C questionnaire used consists largely of disease-specific scales that address the unique problems of this parent population and thus cannot as such be used to evaluate control parents. Consequently, these disease-specific scales only allow for within-group comparisons of parents presenting with various amounts of distress. However, facing a potentially fatal disease of one's child constitutes an indisputable threat. It is, therefore, reasonable to assume increased levels of stress in parents of children with cancer compared to what would be seen in parents of healthy children. And it is, of course, essential to characterize the particular difficulties experienced by parents whose children suffer from life-threatening illness, and who may often have to depend on years of invasive, and physically and mentally demanding treatment. Guided by how parents in the entire group reported severe distress along the severity span, in an attempt to ease the interpretation and understanding of the findings, we came to the conclusion that, with regard to some of the indicators studied, clinically significant distress was still experienced by a substantial number of parents, even distant in time from the child's diagnosis and treatment.

Classification of the parents into groups exhibiting low, intermediate and high levels of distress was designed as a measure of how strongly and frequently the various symptoms were expressed. Uncertainty was reported most noticeably, while psychological and physical distress was experienced less often. In the case of the other symptoms assessed, the distribution of low and high scores was more similar. Using the same questionnaire in a Dutch sample, Van Dongen-Melman et al. (6) have reported a similar distress profile, although they found lower levels of parental distress at almost all subscales of the instrument. This difference between findings may be explained by differences between the samples. Unlike in our study, all of the children of the parents in the Dutch sample had successfully terminated their cancer treatment at assessment. In our study some of the indicators of distress were represented at a somewhat lower level among parents of children off treatment. The systematic differences in distress outcome between these two studies may also be an effect of cultural differences; for instance, how care is organized and how psychosocial demands are met in the two national medical care systems may influence the findings. In line with part of the results presented here, the Dutch study did not find any significant variation in distress over time within the group of parents studied. The lack of such a finding for any of the distress factors may partly depend on the Dutch group being more homogeneous with regard to amount of time that had passed from diagnosis to assessment.

Some of the subscales of the questionnaire used assess general psychological symptoms and some of these are derived from scales commonly used in clinical psychological

practice and research. However, these scales were employed here in abbreviated form and the findings can thus not be directly compared with existing normative data for the complete versions. In a forthcoming report, the psychometric properties of the combination of subscales included in the PPD-C will be described comprehensively. That study will also include data concerning a sample of parents of healthy children, i.e., normative data for the general subscales of the PPD-C.

The study design employed here was cross-sectional, i.e. the parents were asked to respond on one single occasion, at which point different lengths of time had elapsed since the diagnosis of their child. A longitudinal design, in which individual parents are followed over a period of time, would provide more reliable indications of the nature of the changes occurring with time. However, since our study focused on the stress process over a period of a decade, a corresponding longitudinal investigation could not have been completed for many years to come. A problem that is indeed pertinent to this study, but which would remain unresolved even in a longitudinal follow-up design, concerns the rapid alterations and the improvement in the treatment of these patients. It is unavoidable that the way in which the parents experience their child's cancer will always be influenced by the standards of treatment and care at the time.

It has been shown that it is not unusual for parents of a child with cancer to worry about the possibility of a relapse years after successful completion of treatment, even when the patient has reached adulthood (1, 12, 18). When item content is examined, the closely disease-related items of some subscales in our questionnaire provide more detailed information about the specific worries of parents. A kind of concern, which was not linearly related to the time elapsed since diagnosis, is the feeling of uncertainty, with respect to which 40% of the parents experienced levels of distress categorized as high. Those parents expressed a strong or very strong desire to know more about the majority of the 21 disease-related issues covered by the items of the subscale. One such concern is the uncertainty about 'what is going on in (the) child's mind because of his/her disease', with respect to which 80% of the parents exhibited great worry. Eighty-four percent reported a need to learn more about the possible long-term side effects of the treatment. With regard to *disease-related fear*, 7.5% of the parents experienced a high level of distress, declaring that they were to some extent or extremely worried about most of the issues referred to in this subscale. In addition, almost two-thirds (63%) of the parents were extremely worried that their child's illness would reappear in the future, making this the most frequently reported concern. Approximately half of the parents (47%) were equally concerned about possible late effects of treatment. In contrast, only 12% were extremely worried about how their child's illness might affect his/her relationship with other children.

In our study, the time elapsed was to some extent associated with the level of *psychological and physical distress*, a subscale which covers a number of common physical symptoms of distress. However, this was the cluster where problems were experienced to the lowest degree by our subjects, almost three-quarters of whom reporting little or no suffering from the majority of the symptoms. This finding suggests that general physical stress symptoms do not truly reveal the core concerns of these parents.

In general, parents' experience being told that their child has cancer as a trauma, and anxiety and dissociative symptoms, indicative of psychological crisis, appear to occur immediately after such a diagnosis (16, 17). The most significant reduction in anxiety is usually observed during the first month following diagnosis (e.g. 4). In this study, the shortest period between diagnosis and assessment of parents was 4 weeks. Primarily owing to ethical considerations, parents were not approached earlier. In addition, the focus here was on distress in a long-term perspective. It is likely, however, that the immediate reactions caused by the crisis had declined in strength during this first month.

Our findings indicate a continued reduction in the level of many distress symptoms with time, although the decline is not common to all the types of distress studied, and the suggested reduction is relatively small for many of them. Experiences of anxiety and loss of control could be the consequence of chronic stress, and, indeed, these indicators of distress were still prevalent among some of the parents for whom the longest priod of time had elapsed sine the child's diagnosis. On the other hand, experiences of anxiety and loss of control could also be the consequence of chronic stress, and, indeed, these indicators of distress are still prevalent among some of the parents for whom the longest period of time had elapsed since the child's diagnosis.

In addition to the time elapsed, three other independent variables were analysed as possible predictors of distress, i.e., prognosis, treatment situation and the age of the child at diagnosis. When evaluating our findings, with respect to the prognosis, it must be remembered that the longer the time-lapse since diagnosis, the greater the proportion of children who have been cured. In other words, among parents who have received the diagnosis more recently, there is a greater proportion of their children who will later die. Therefore, a higher level of distress among these parents could be expected. However, the medical prognosis appeared not to be associated with the level of distress. This highly interesting finding implies that the statistically based, diagnosis-specific estimate of survival is relatively far removed from the core sources triggering the traumatic crisis of parents. Dockerty et al. obtained a similar outcome (9) when they found no associations between prognosis (as

perceived by the parent) and the level of parental distress. Thus, the child's prognosis—as based on statistics or perceived by the parent—does not appear significantly to influence parental distress, regardless of whether the child is presently being treated or not. It can, nevertheless, be assumed that the worsening of the prognosis associated with a relapse is an additional cause of distress. Grootenhuis & Last (5) have demonstrated that the recurrence of the child's cancer is associated with an intensification of the parents' feelings of helplessness and uncertainty. In our study, parents of children being treated for relapse were not analysed separately. The reactions of parents to a relapse in their child's illness are an important issue, which has received little attention to date. The increased emotional vulnerability of such parents is of obvious psychosocial interest and deserves further investigation.

For some distress factors, parents of children undergoing treatment reported higher levels of distress than parents of children off treatment. To establish the influence of the treatment situation per se is difficult, however, as it interacts strongly with time elapsed since diagnosis in particular. The effect of the treatment situation was reduced or even eliminated when the time factor was taken into consideration. For age of the child at the time of diagnosis, only weak linear associations with a few of the distress subscales were detected. In the multivariate analysis the effect of child's age disappeared for all subscales except for uncertainty, indicating a minor importance of the variable for distress outcome in comparison to time elapsed since diagnosis.

The psychological distress observed among parents long after the diagnosis and treatment of their child have been given indicates that the parents have to adjust not only to a new situation, but also to an ongoing, long-term process that is characterized by recurrent and chronic causes of stress. On the other hand, remaining symptoms of distress may also originate from traumatic disease-related early experiences that have not been successfully coped with or worked through. Although usually the most upsetting individual moment, hearing the diagnosis is not the only traumatic stress factor and may not even be the most crucial one for all parents (14). A large number of factors, situational as well as others, may potentially influence the amount of distress experienced by parents of a child with cancer, and the present study has focused on only a few of them. Having to witness invasive treatment of one's child, and the subsequent experience of being unable to protect the child from suffering can create additional uncertainty regarding the parental role, and this can become an additional source of distress (22, 23). Furthermore, the psychological status of the parents has, not surprisingly, been shown to be related to the level of disease-related distress; especially the presence of trait anxiety (12, 16). Other factors likely to affect the level of distress include the parents' life experiences prior to the child's illness (9, 16), socioeconomic factors (9, 10), perceived social support (8, 9, 13), and the quality of the relationship between the parents themselves (10, 15). With the exception of the distressing treatment procedures, all these factors are of the kind that they may hypothetically affect parental distress at all stages in time, and far beyond the time of diagnosis and treatment. Thus, if a relationship between time and distress does exist, it would not necessarily need to be systematically affected by any of those factors.

Contrary, perhaps, to what might be expected, the relationships detected in this study between time and disease-related stress were relatively weak, or, for some indicators, could not be demonstrated whatsoever. As the study did not include a control group, we cannot be sure of how the suffering of these parents compares to data from parents in the community. However, our attempt to delineate the amount of clinically significant levels of distress among the parents, and the fact that the data demonstrate a considerable dispersion with regard to individual distress scores at different points in time, together demonstrate that a substantial number of parents experience significant distress all along the time span studied.

CONCLUSIONS

The results demonstrate the presence of significant disease-related distress in a substantial number of parents, even years after their child's cancer diagnosis. These findings suggest that psychosocial attention is required, not only at the early stages of onset and treatment of the child's illness, but even several years after treatment has been completed. Longitudinally designed research would further reveal the nature of the stress process following the exceptional life situation of parents of children with cancer.

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REFERENCES

1. Van Dongen-Melman JE, S-WJ. A. Psychosocial aspects of childhood cancer: a review of the literature. J Child Psychol Psychiatr 1986; 27: 145–80.
2. Grootenhuis MA, Last BF. Adjustment and coping by parents of children with cancer: a review of the literature. Support Care Cancer 1997; 5: 466–84.
3. Dahlquist LM, Czyzewski DI, Copeland KG, Jones CL, Taub E, Vaughan JK. Parents of children newly diagnosed with cancer: anxiety, coping, and marital distress. J Pediatr Psychol 1993; 18: 365–76.

4. Fife B, Norton J, Groom G. The family's adaptation to childhood leukemia. Soc Sci Med 1987; 24: 159–68.
5. Grootenhuis M, Last B. Predictors of parental emotional adjustment to childhood cancer. Psycho-Oncology 1997; 6: 115–28.
6. Van Dongen-Melman JE, Pruyn JFADG, Koot HM, Hahlen K, Verhulst FC. Late psychosocial consequences for parents of children who survived cancer. J Pediatr Psychol 1995; 20: 567–86.
7. Sawyer M, Antoniou G, Toogood I, Rice M, Baghurst P. Childhood cancer: a 4-year prospective study of the psychological adjustment of children and parents. J Pediatr Hematol Oncol 2000; 22: 214–20.
8. Hoekstra-Weebers JE, Jaspers JP, Kamps WA, Klip EC. Psychological adaptation and social support of parents of pediatric cancer patients: a prospective longitudinal study. J Pediatr Psychol 2001; 26: 225–35.
9. Dockerty JD, Williams SM, McGee R, Skegg DC. Impact of childhood cancer on the mental health of parents. Med Pediatr Oncol 2000; 35: 475–83.
10. Sloper P. Predictors of distress in parents of children with cancer: a prospective study. J Pediatr Psychol 2000; 25: 79–91.
11. Lähteenmäki PM, Salmi TT, Salo MA. Children with malignant disorders: the health and life situation of their parents examined over a 7-year interval. Acta Pædiatr 1996; 85: 70–5.
12. Kazak AE, Stuber ML, Barakat LP, Meeske K, Guthrie D, Meadows AT. Predicting posttraumatic stress symptoms in mothers and fathers of survivors of childhood cancers. J Am Acad Child Adolesc Psychiatry 1998; 37: 823–31.
13. Manne S, Duhamel K, Redd WH. Association of psychological vulnerability factors to post-traumatic stress symptomatology in mothers of pediatric cancer survivors. Psycho-Oncology 2000; 9: 372–84.
14. Stuber ML, Christakis DA, Houskamp B, Kazak AE. Posttrauma symptoms in childhood leukemia survivors and their parents. Psychosomatics 1996; 37: 354–261.
15. Goldbeck L. Parental coping with the diagnosis of childhood cancer: gender effects, dissimilarity within couples, and quality of life. Psycho-Oncology 2001; 10: 325–35.
16. Hoekstra-Weebers JE, Jaspers JP, Kamps WA, Klip EC. Risk factors for psychological maladjustment of parents of children with cancer. J Am Acad Child Adolesc Psychiatry 1999; 38: 1526–35.
17. Levi RB, Marsick R, Drotar D, Kodish ED. Diagnosis, disclosure, and informed consent: learning from parents of children with cancer. J Pediatr Hematol Oncol 2000; 22: 3–12.
18. Leventhal-Belfer L, Bakker AM, Russo CL. Parents of childhood cancer survivors: a descriptive look at their concerns and needs. J Psychosoc Oncol 1993; 11: 19–41.
19. Van Dongen-Melman JEWM. Psychometric properties of an instrument measuring psychosocial functioning of parents of children who survived cancer. In: On surviving childhood cancer. Late psychosocial consequences for patients, parents, and siblings. Rotterdam: Department of Child and Adolescent Psychiatry, Sophia Children's Hospital/University Hospital Rotterdam, Erasmus University Rotterdam; 1995 (Dissertation).
20. Van Dongen-Melman JEWM, Pruyn JFA, Van Zanen GE, Sanders-Woudstra JAR. Coping with childhood cancer: a conceptual view. J Psychosoc Oncol 1986; 4: 147–61.
21. Childhood cancer in the Nordic countries. Report on epidemiologic and therapeutic results from registries and working groups, Aalborg: Nordic Society of Pediatric Haematology and Oncology, 2000.
22. LaMontagne LL, Wells N, Hepworth JT, Johnson BD, Manes R. Parent coping and child distress behaviors during invasive procedures for childhood cancer. J Pediatr Oncol Nurs 1999; 16: 3–12.
23. Ljungman G, Gordh T, Sorensen S, Kreuger A. Pain in paediatric oncology: interviews with children, adolescents and their parents. Acta Paediatr 1999; 88: 623–30.