# Emotional functioning of parents of children with cancer: the first five years of continuous remission after the end of treatment

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## Abstract

*Objectives*: The aim of this study is to investigate parental emotional functioning during the first five years of continuous remission after the end of their child's treatment and to identify predictors of parental emotional functioning.

*Methods*: Psychological distress and situation-specific emotional reactions were assessed in 122 mothers and 109 fathers from 130 families. Longitudinal mixed model analyses were performed to investigate to what extent generic and disease-related coping, family functioning and social support were predictive of parental emotional functioning over time.

*Results*: Initial elevated levels of distress, disease-related feelings of uncertainty and helplessness returned to normal levels during the first two years after the end of treatment. Being more optimistic about the further course of the child's disease (*predictive control*) was correlated with lower psychological distress and less negative disease-related feelings, while more passive reaction patterns were correlated with higher psychological distress and more negative disease-related feelings.

*Conclusions*: Although in general the parents of children with successfully treated cancer showed adequate emotional resilience, support for these parents should not stop when treatment ends. Parents in need of help can be identified on the basis of their coping abilities. Copyright © 2007 John Wiley & Sons, Ltd.

Keywords: childhood cancer; parental emotional adjustment; psycho-oncology

## Introduction

As a result of advances in the treatment of childhood cancer, the number of successfully treated patients has increased enormously in the last decades. The overall 5-year survival rate for children diagnosed with cancer in Europe is currently more than 70% compared with 30% in the 1960s [1,2]. Childhood cancer is a dramatic event that affects the daily life and emotional well-being of all family members. The results of longitudinal studies showed that, although many parents adjust well to the paediatric cancer experience, a considerable percentage of parents continue to suffer problems such as psychological distress, anxiety and post-traumatic-stress symptoms after termination of their child's cancer treatment [3–7]. More affirmative reactions to the cancer experience have also been observed, however. Cognitive strategies used to cope with stressful events can generate a positive effect; for instance, labelling ordinary events with a positive meaning and appraising stressful situations as challenges rather than burdens, which can generate feelings of mastery and control [8,9].

The authors recently found that parents experienced considerably more emotional distress than the general Dutch population two months after the end of their child's successful treatment for cancer [9]. This is not surprising, since coming off therapy is one of the major transitions in care in the practice of paediatric oncology [10]. It is a very difficult and anxious time for both patients and parents [11,12], while there is a tendency for social and emotional support to decrease when treatment ends [13], even though the family is just starting to come to terms with what has happened.

In the present study, a process-oriented approach was applied to investigate parental emotional adjustment to childhood cancer. We presume that parental emotional functioning is the outcome of a process over time that is influenced by situational characteristics, such as demographic and medical variables, and by psychosocial factors such as coping, social support and family functioning. These psychosocial factors

are important because those can be approached in intervention.

Previous research revealed highly diverse demographic and medical variables that were predictive of parental maladjustment to the cancer experience [14], among others: being a mother, low socio-economic status, recurrence of the disease in the child and worse health status of the child [5,15–18].

The ways in which parents cope with the consequences of childhood cancer can be regarded as an important factor in their adaptation to the cancer experience. According to the model of stress and coping developed by Lazarus and Folkman [19], coping consists of actions, behaviours and thoughts aimed at dealing with the demands of events and situations that are appraised as stressful. In the context of coping with cancer, Grootenhuis *et al.* [20] found the following cognitive control strategies to be relevant in the medical setting: expectations of the further course of the disease (predictive coping), reliance on powerful others such as doctors (vicarious control), attempts to influence the chance-determined outcome, such as hoping for a miracle or wishful thinking (illusory control) and searching for information (interpretative control). Previous research revealed that parents who were optimistic about the further course of their child's disease reported fewer emotional problems [16]. Others reported that higher scores for the 'social support-seeking' generic coping style resulted in lower distress levels one year after the diagnosis of cancer [17].

Several studies indicate that social support can protect parents from the stress caused by their child's disease and treatment [13,21–23]. In families of a child with cancer, higher scores on family cohesion and adaptability were found to be correlated with lower parental anxiety and fewer parental post-traumatic-stress symptoms [22]. In addition, Sloper *et al.* [24] found that stronger family cohesion was associated with lower distress levels in mothers.

The first few years following the end of treatment are considered as an important phase in the adjustment to the cancer experience. A longitudinal study was designed in order to gain insight into the process of parental adjustment during this phase. The main research questions were: (1) How do mothers and fathers adjust emotionally during the first five years of continuous remission following the completion of treatment for childhood cancer? (2) To what extent are coping, family functioning and social support associated with parental emotional functioning during the first five years of continuous remission following the completion of treatment for childhood cancer? The associations between the psychosocial variables (independent variables) and parental emotional outcomes (dependent variables) were controlled for demographic and medical variables.

## Methods

### Procedure

The results presented here concern the longitudinal VOLG study (*Vragenlijsten kinderOncologie Latere Gevolgen* = questionnaires on childhood cancer late sequelae), a Dutch study on the late psychosocial consequences of cancer in childhood, which started in 2000 and ended in 2006. Patients and their parents were recruited from two Dutch university hospitals, the Emma Children's Hospital at the Academic Medical Center in Amsterdam (from March 2000 until the end of 2002) and the Radboud University Nijmegen Medical Center (from June 2002 until the end of 2002). The Medical Ethics Committee of the hospitals have approved the study protocol.

All consecutive patients who met the inclusion criteria during these periods were invited to participate in the VOLG study. The inclusion criteria were: (1) age of the patients 1–18 years; (2) complete remission; (3) end of successful treatment at most two months before; and (4) being able to complete Dutch questionnaires.

Parents were informed about the VOLG study by letter. Once informed consent had been obtained, the parents were telephoned and an appointment was made to fill in the questionnaires at the hospital or at home. The parents completed the questionnaires, four to six times, depending on the year of inclusion. The assessments took place approximately two months (M1), one year (M2), two years (M3), three years (M4), four years (M5) and five years (M6) after the end of successful treatment. The data for parents whose child relapsed were excluded from analysis from the moment of the relapse.

### Measures

#### Dependent variables: parental emotional outcomes

*Parental psychological distress* was measured using the *General Health Questionnaire*-30 (*GHQ*-30) [25,26]. The raw total scale score can be used as an overall index of psychological distress, where higher scores indicate greater distress. According to Goldberg and Williams [25], scores of 5 or more indicate clinically elevated levels of psychological distress. The validity of the 30-item version is well documented and its internal consistency is highly satisfactory [25,26].

*Parental situation-specific emotional reactions* were assessed using the *Situation-Specific Emotional Reaction Questionnaire* (SSERQ) developed at the Psychosocial Department of the Emma Children's Hospital/AMC [27]. It consists of four scales, which describe feelings that can be considered situation-specific for parents of children with cancer, during and after treatment [27],

namely disease-related feelings of loneliness, helplessness, uncertainty and positive feelings. The higher the scores, the more often parents experienced the emotional reactions in question. The validity and internal consistency are satisfactory [27,28].

### Independent variables: situational characteristics

*Medical data* were obtained from the patient's medical record. The prognosis was based on the survival chances at diagnosis as rated by each patient's oncologist, namely <25, 25–75 or >75%. After the end of treatment (M1), the parents were asked to rate their perception of the intensiveness of their child's treatment on a Visual Analogue Scale, from 'totally non-intensive' (0, left end of line) to 'very intensive' (10, right end of line). They were also asked to assess the visible consequences of the disease. Their answers were dichotomized to 'presence' or 'absence' of visible consequences.

*Important family events* during the past year were scored by the parents on a list of 19 such events, including the birth of a child, parental divorce, moving, death of a family member or friend, decline in financial means, change of school and change of job. The total score of important family events was dichotomized to 'less than two' and 'two or more'.

### Independent variables: psychosocial factors

*Generic coping* was measured with the *Utrecht Coping List* (UCL) [29], a questionnaire about coping with stressful or problematic situations. The UCL covers seven coping styles: active problem focusing, palliative reaction pattern, avoidance behaviour, seeking social support, passive reaction pattern, expression of emotions and comforting cognitions. A higher scale score means more use of the coping style. The internal consistency and validity are satisfactory [29,30].

*Disease-related cognitive coping* was assessed using the *Cognitive Control Strategies Scale* for Parents (CCSS-PF). The instrument, based on the model of Rothbaum *et al.* [31], was developed at the Psychosocial Department of the Emma Children's Hospital/AMC [20]. It assesses the extent to which respondents try to gain sense of control over the illness by using four cognitive coping strategies: predictive control (being optimistic about the course of the disease), vicarious control (attributing power to medical-care givers and treatment), interpretative control (searching for information in order to better understand emotional reactions and to gain insight into the situation) and illusory control (attempts to influence the chance-determined outcome). Higher scores represent a stronger reliance upon the control strategy in question. The questionnaire proved to be useful, valid and

reliable in the context of cancer and Inflammatory Bowel Disease [20,28,32–34].

*Family functioning* was measured with the Dutch version of the *Family Adaptability and Cohesion Evaluation Scales* (FACES) [35], developed by Olson and colleagues [36–38]. The Adaptability Scale indicates the extent to which a family adapts its power structure, role definitions and rules to meet internal and external demands. The Cohesion Scale indicates the degree of mutual connectedness between family members. Higher scale scores mean greater adaptability and cohesion. The validity and internal consistency of the Dutch version are good [35].

The amount of *social support* the respondent indicated that he received from the social network was assessed using the *Social Support Questionnaire for Transactions* (SSQT), developed by Suurmeijer and colleagues [39–41]. The SSQT measures the frequency of supportive interactions on seven different scales, together with a total score. The psychometric properties of the SSQT have proved to be good [39,41]. The SSQT total score was used in the present study.

### Statistical analyses

The Statistical Package for Social Sciences (SPSS), Windows version 12.0, was used for all analyses. Missing values were handled according to the guidelines given in the manuals for the relevant questionnaires and, after that, through the expectation-maximization estimation method [42]. Analyses were carried out for mothers and fathers separately.

In order to describe parental emotional functioning over time, the mean scores of psychological distress (*GHQ*-30) and situation-specific emotional reactions (*SSERQ*) are depicted in Figures 1 and 2. For each outcome, we fitted a linear mixed model with a random intercept representing the baseline at the first measurement occasion and fixed slopes representing the deviations from baseline at the other measurement occasions. In addition, parental levels of psychological distress were compared with norm data of the general Dutch population at each measurement occasion, using one-sample *t*-tests and binomial tests at each measurement occasion.

Linear mixed model analysis was further carried out to examine to what extent psychosocial factors over time were predictive of parental emotional functioning over time, while controlling for demographic and medical characteristics. Measurement occasions were treated as nested within respondents. The major advantage of this method is that all available data are incorporated into the analysis, including data from parents who missed one or more measurement occasions. Efficient estimates can be obtained through maximum likelihood estimation procedures if dropout is random (conditionally on the non-missing data) [43]. Hence,
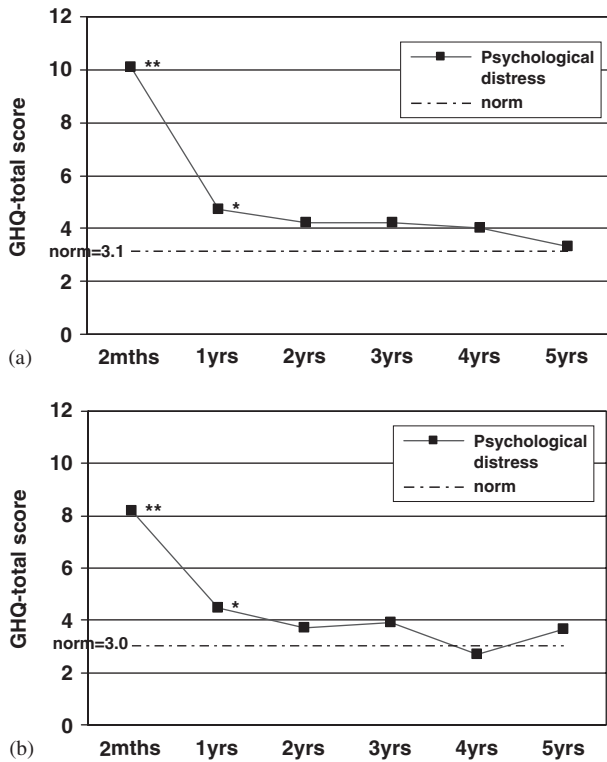
**Figure 1.** Psychological distress (GHQ-total scores) in (a) *mothers* and (b) *fathers* over measurements occasions, compared with the norm (general Dutch population (37)) [*]GHQ-total score differed significantly from the norm at 0.05 and [**]GHQ-total score differed significantly from the norm at 0.001.
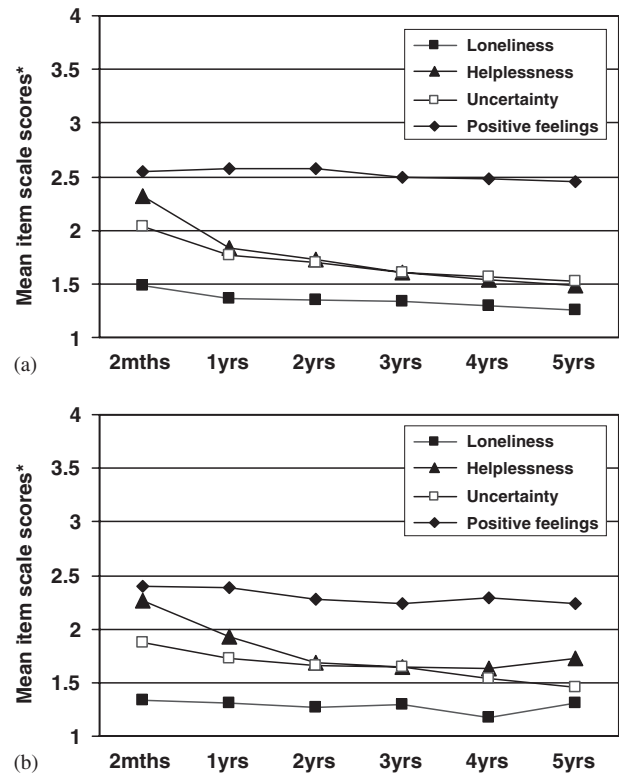


**Figure 2.** Situation-specific emotional reactions (SSERQ) in (a) *mothers* and (b) *fathers* over measurements occasions. [*]1 = never; 2 = sometimes; 3 = often; and 4 = almost all the time.

changes in the numbers of subjects from occasion to occasion do not harm the analysis, other than that the statistical power to find deviations from baseline decreases with higher attrition.

To facilitate interpretation of regression coefficients, all continuous scores on dependent (outcome) variables and independent (predictor) variables were transformed into standard normal scores, expressing deviations from the mean at M1. We followed Cohen [44] in considering standardized regression coefficients of 0.1 as small, 0.3 as medium and 0.5 as large. For binary-coded variables, regression coefficients of 0.2 can be considered small, 0.5 medium and 0.8 large.

Measurement occasions were treated as fixed because growth curve models did not fit the data. The intercept was considered random with its mean fixed at the standardized mean outcome at M1, thus taking the outcome at M1 as the reference point. In this way, parameter estimates for M2–M6 can be interpreted as deviations from baseline (M1). The deviations were treated as fixed parameters as indicated by Akaike's information criterion.

Models were fitted for each of the five outcomes (parental psychological distress and the four situation-specific emotional reactions). Because of the large number of predictor variables, preselection was necessary. The initial model consisted of the random intercept (M1) and the fixed parameters for measurement occasions M2–M6. Predictor variables were subsequently entered in four steps into the initial model, if significant at least at 0.20: (1) demographic and medical variables, (2) disease-related coping, (3) generic coping, (4) family functioning and social support. Once selected, the variables remained in the model, even if they turned out to be non-significant in later steps. The final models thus consisted of the random intercept (M1) and the fixed regression coefficients for M2–M6, completed with the predictor variables that were selected in the stepwise procedure. For one outcome, positive feelings, there appeared to be no intercept variance. The final model for this outcome, both for mothers and fathers, was therefore fitted with a fixed intercept. Percentages of total explained variance were calculated after each step.

For each model, we checked whether the longitudinal covariance structure was best described by compound symmetry or by an autoregressive structure, with reference to Akaike's information criterion. Compound symmetry appeared to give the best fit for all models except that for parental psychological distress, where an autoregressive structure was more appropriate.

We checked whether first-order interaction effects of measurement occasion with medical variables, disease-related coping and social support

should be added to the model. To prevent too many findings occurring by chance, these tests were carried out at a Bonferroni adjusted level of significance. The conclusion under these conditions was that none of the first-order interaction effects considered needed to be added to the model.

## Results

### Participants

A total of 164 consecutive childhood cancer patients who completed treatment successfully at most two months before, and their parents, were invited to participate in the longitudinal part of the VOLG study: 150 patients from The Emma Children's Hospital AMC and 14 patients from the Radboud University Nijmegen Medical Center. The response rate was 81.7% ($N = 134$). The 30 families who did not participate did not differ from participating families with respect to demographic and medical variables ($p < 0.1$ at $t$-tests or $\chi^2$-tests).

Data about emotional functioning of mothers and/or fathers from 130 (of the 134) families were available: 129[1] families at M1 (99.2%), 109 at M2 (83.4%), 105 at M3 (80.8), 97 (74.7%) at M4, 55 at M5 (42.3%) and 25 (19.2%) at M6. Dropout because of non-response was 8.5% ($N = 11$). Furthermore, the data for parents whose children had relapsed were excluded from analysis from the moment of the relapse ($N = 25$, 19.2%). Hence, it depends on the moment of relapse how many measurement occasions were included. Finally, data at M5 and M6 were not available in, respectively, 57 (43.8%) and 91 families (70%) because of the finite follow-up period of the VOLG study.

Only a few significant differences were found between the parents with data until M6 ('complete data') and the parents whose data did not include M6 ('incomplete data'). Firstly, the parents with incomplete data were on average one to two years younger than the parents with complete data. Secondly, incomplete data include fewer parents of patients with a good prognosis (survival chance at diagnosis > 75%) than complete data. This is not surprising because families were excluded from analysis from the moment of a relapse. No other differences in demographic and medical variables were found at a significance level of 0.10. We also compared the mean parental outcomes at M1 for parents with incomplete data with that of parents with complete data and found no difference. The characteristics of patients and parents are presented in Table 1.

### Emotional functioning over time

#### Psychological distress

Two months after the end of treatment, about two-third of the mothers (72%) and fathers (60%) reported clinically elevated levels of psychological distress, which was much higher than the percentage in the general population (24 and 22% for females and males, respectively). One year after the end of treatment, elevated levels were found in 34% of the mothers and 36% of the fathers, still significantly different from the norm. From two years after treatment, mothers and fathers reported normal levels of psychological distress.

Elevated levels of psychological distress were also expressed by the mean GHQ-total scores (Figure 1). Two months after the end of successful treatment, mothers as well as fathers had a significantly higher mean total score than the norm: $T(120) = 10.3$, $p < 0.0001$ and $T(107) = 7.5$, $p < 0.001$, for mothers and fathers, respectively. The distress scores decreased over time. Both mothers and fathers reached a normal level of psychological distress at two years. At one year,

**Table 1.** Characteristics of the patients and their parents at M1 (2 months since end of treatment)

| | N | M | SD | Range |
|---|---|---|---|---|
| Parents | | | | |
| Age mothers (years) | 122 | 37.9 | 5.0 | |
| Age fathers (years) | 109 | 39.7 | 5.1 | |
| Patients | | | | |
| Age at study (years) | 129 | 8.0 | 4.4 | 1.1–18.2 |
| Age at diagnosis (years) | 129 | 6.8 | 4.5 | 0.3–17.2 |
| Time since diagnosis (months) | 129 | 13.7 | 8.4 | 2.0–29.7 |
| Duration of treatment (months) | 129 | 11.5 | 8.4 | 0.6–26.0 |

| | % | N |
|---|---|---|
| Age category (years) | | |
| 1–5 | 41.9 | 54 |
| 6–11 | 34.1 | 44 |
| 12–15 | 20.9 | 27 |
| ⩾16 | 3.1 | 4 |
| Gender | | |
| Female | 41.9 | 54 |
| Male | 58.1 | 75 |
| Diagnosis | | |
| Leukaemia/lymphoma | 48.1 | 62 |
| Solid tumour | 47.3 | 61 |
| Brain tumour | 4.7 | 6 |
| Prognosis | | |
| <25% | 5.4 | 7 |
| 25–75% | 39.5 | 51 |
| >75% | 55.0 | 71 |
| Treatment[a] | | |
| Chemotherapy | 95.3 | 123 |
| Surgery | 46.5 | 60 |
| Radiotherapy | 18.6 | 24 |
| Autologous bone marrow transplantation | 2.3 | 3 |
| Other | 2.3 | 3 |

Number of mothers and fathers at M2–M6: mothers 103 (M2), 98 (M3), 92 (M4), 52 (M5), 24 (M6) and fathers 91 (M2), 87 (M3), 78 (M4), 46 (M5), 19 (M6). [a]More than one answer was possible per patient.

their scores were still higher than the norm: $T \times (102) = 2.5$, $p < 0.05$ and $T(90) = 2.4$, $p < 0.05$, for mothers and fathers, respectively.

### Situation-specific emotional reactions

The frequency of disease-related feelings of loneliness ($p < 0.05$), helplessness ($p < 0.001$) and uncertainty ($p < 0.001$) decreased significantly in mothers during the first year after the end of treatment (M2) and remained low at subsequent measurement occasions. The frequency of disease-related positive feelings was constant over time (Figure 2).

The frequency of feelings of helplessness ($p < 0.001$) and uncertainty ($p < 0.001$) decreased significantly also in fathers during the first year after the end of treatment (M2) and remained low at subsequent measurement occasions. The frequency of positive feelings and feelings of loneliness was stable over time, with the exception of M4, where fathers reported fewer positive feelings than at M1 ($p < 0.05$), and M5, where they reported fewer feelings of loneliness compared with M1 ($p < 0.01$).

## Predictors of emotional functioning

The fixed effects derived from the longitudinal mixed model analyses of parental emotional functioning are shown in Table 2 (mothers) and Table 3 (fathers), as well as the total explained variance after each step in the analysis.

Parental emotional functioning was explained well by the longitudinal mixed models. The total explained variance was more than 40%, except for positive feelings. In general, the amount of variance explained by the medical effects was much smaller than that explained by coping. Disease-related feelings of uncertainty were explained particularly well by coping, 42.9 and 55.1% in mothers and fathers, respectively. It is notable that—apart from the contribution of the time of measurement—parental feelings of helplessness were explained mainly by disease-related coping. Positive feelings, on the other hand, were explained much more by generic coping than by disease-related coping.

Apart from that, most fixed effects were small [44]. Medium-sized effects were found for the effect of predictive control (disease-related coping) and passive reaction pattern (generic coping). The predictors of emotional functioning in both mothers and fathers are presented in greater detail in Tables 2 and 3 and are described below.

### Psychological distress

Both mothers and fathers reported that longer duration of treatment and greater optimism about the further course of the disease (predictive control) were associated with lower levels of psychological distress. Having a more passive reaction pattern

was associated with higher levels of distress. In mothers, we also found that older age and stronger family cohesion was associated with lower levels of distress, while mothers who reported the occurrence of two or more important family events during the past year and mothers who perceived treatment as being more intensive had higher levels of distress. Fathers who had more palliative reaction patterns experienced lower levels of distress.

### Situation-specific emotional reactions

More use of predictive control strategies was correlated with less disease-related *loneliness* in both mothers and fathers, while higher scores on passive reaction patterns and family adaptability were related to more loneliness. In addition, mothers who used more illusory control strategies reported more loneliness, while mothers who reported higher scores on family cohesion experienced less loneliness. Fathers who reported less supportive interactions (social support) experienced more loneliness.

If the children did not exhibit visible consequences of the disease and treatment, their mothers and fathers reported less disease-related feelings of *helplessness*. Predictive control in mothers and fathers was also associated with less helplessness. Higher scores on illusory control, palliative and passive reaction patterns were related to greater feelings of helplessness in both mothers and fathers, as were higher scores on interpretative control, family cohesion and adaptability in fathers.

Mothers and fathers who used more predictive control strategies reported less disease-related feelings of *uncertainty*, while higher scores on palliative and passive reaction patterns and family adaptability were associated with more uncertainty. In mothers, we also found that the use of more illusory control strategies and higher scores on family adaptability were associated with more uncertainty, while older age was associated with less uncertainty. If the children did not show visible consequences of the disease and treatment, the fathers reported less uncertainty. In addition, more active problem focusing was associated with less uncertainty in fathers.

Disease-related *positive feelings* were positively correlated with active problem focusing and comforting cognitions, in both mothers and fathers. In addition, illusory control was positively correlated with positive feelings in mothers, and mothers who had reported more family cohesion and the occurrence of two or more important family events during the past year experienced more positive feelings. Fathers who perceived the treatment as being more intensive, fathers who reported more supportive interactions and fathers who had more palliative reaction patterns experienced more

# Parental emotional functioning after treatment of childhood cancer

**Table 2.** Parameter estimates for longitudinal regression models of emotional functioning in *mothers* predicted by measurement occasion, demographic and medical characteristics, coping, family functioning and social support

| | Psychological distress (GHQ) | Loneliness (SSERQ) | Helplessness (SSERQ) | Uncertainty (SSERQ) | Positive feelings (SSERQ) |
|---|---|---|---|---|---|
| Fixed effects | | | | | |
| *Measurement* | | | | | |
| (deviation from end of treatment; M1) | | | | | |
| One year (M2) | $-0.57^{**}$ | $-0.13$ | $-0.66^{**}$ | $-0.34^{**}$ | 0.13 |
| Two years (M3) | $-0.48^{**}$ | $-0.03$ | $-0.74^{**}$ | $-0.33^{**}$ | $0.19^{*}$ |
| Three years (M4) | $-0.49^{**}$ | $-0.06$ | $-0.95^{**}$ | $-0.48^{**}$ | 0.08 |
| Four years (M5) | $-0.50^{**}$ | $-0.08$ | $-0.99^{**}$ | $-0.52^{**}$ | $0.44^{*}$ |
| Five years (M6) | $-0.36^{*}$ | 0.06 | $-1.08^{**}$ | $-0.42^{**}$ | 0.03 |
| Percentage of explained variance by fixed effects | 12.8 | 2.6 | 21.1 | 11.4 | 0.6 |
| | | | | | |
| *Medical and demographic characteristics* | | | | | |
| Age mother | $-0.12^{*}$ | $-0.11$ | $-0.09$ | $-0.11^{*}$ | $-0.07$ |
| Age patient | | | 0.09 | | |
| Time since end of treatment | | | | | |
| Duration of treatment | $-0.14^{**}$ | | | | |
| Leukaemia or lymphoma | | | | | |
| Radio- and chemotherapy | | | | | |
| Prognosis >75% | | | | | |
| Perceived treatment intensity | $0.10^{*}$ | | | | |
| No visible consequences | | | $-0.26^{**}$ | | |
| Percentage of explained variance by fixed effects | 19.6 | 7.1 | 25.4 | 14.6 | 0.8 |
| | | | | | |
| *Disease-related coping (CCSS)* | | | | | |
| Predictive control | $-0.17^{**}$ | $-0.19^{**}$ | $-0.32^{**}$ | $-0.33^{**}$ | |
| Illusory control | | $0.14^{**}$ | $0.18^{**}$ | $0.10^{**}$ | $0.19^{**}$ |
| Interpretative control | | | 0.06 | | 0.03 |
| Vicarious control | | | | | |
| Percentage of explained variance by fixed effects | 27.2 | 24.6 | 39.6 | 37.8 | 8.6 |
| | | | | | |
| *Generic coping (UCL)* | | | | | |
| Active problem focusing | | | | $-0.06$ | $0.18^{**}$ |
| Expression of emotions | | | $-0.08$ | | $-0.06$ |
| Palliative reaction pattern | | | $0.13^{**}$ | $0.10^{**}$ | |
| Passive reaction pattern | $0.36^{**}$ | $0.37^{**}$ | $0.20^{**}$ | $0.36^{**}$ | |
| Comforting cognitions | | | 0.08 | | $0.21^{**}$ |
| Seeking social support | | $-0.05$ | | | |
| Avoidance behaviour | | 0.07 | | | |
| Percentage of explained variance by fixed effects | 41.0 | 40.3 | 43.4 | 57.5 | 27.5 |
| | | | | | |
| *Family functioning and social support* | | | | | |
| Adaptability (GDS) | | $0.14^{**}$ | | $0.09^{*}$ | |
| Cohesion (GDS) | $-0.10^{*}$ | $-0.09^{*}$ | 0.07 | | $0.09^{*}$ |
| Supportive interactions (SSL) | | $-0.08$ | | | 0.10 |
| ≥2 important family events (last year) | $0.16^{*}$ | | | | $0.15^{*}$ |
| Percentage of explained variance by fixed effects | 42.9 | 42.9 | 44.1 | 58.1 | 30.5 |
| Total number of observations | 421 | 489 | 480 | 489 | 442 |

$^{*}p < 0.05$, $^{**}p < 0.01$.

positive feelings. Finally, expression of emotions was associated with less positive feelings in fathers.

## Discussion

The first five years of continuous remission after the end of treatment were investigated, in view of the importance of this phase for parental adjustment in the run-up to long-term survivorship of their children with cancer. The present study indicates that, on the whole, parents adjust well to the experience of childhood cancer in their family. The findings illustrate that psychosocial variables are stronger indicators of emotional functioning than medical variables.

**Table 3.** Parameter estimates for longitudinal regression models of emotional functioning in *fathers* predicted by measurement occasion, demographic and medical characteristics, coping, family functioning and social support

| | Psychological distress (GHQ) | Loneliness (SSERQ) | Helplessness (SSERQ) | Uncertainty (SSERQ) | Positive feelings (SSERQ) |
|---|---|---|---|---|---|
| Fixed effects | | | | | |
| *Measurement* | | | | | |
| (deviation from end of treatment; M1) | | | | | |
| One year (M2) | −0.38** | 0.13 | −0.36** | −0.11 | 0.12 |
| Two years (M3) | −0.41** | 0.16 | −0.71** | −0.09 | −0.10 |
| Three years (M4) | −0.43** | 0.16 | −0.70** | −0.16 | −0.07 |
| Four years (M5) | −0.48** | 0.03 | −0.69** | −0.21* | −0.08 |
| Five years (M6) | −0.27 | 0.47** | −0.36 | −0.22 | −0.22 |
| Percentage of explained variance by fixed effects | 9.2 | 1.2 | 15.5 | 5.6 | 1.7 |
| | | | | | |
| *Medical and demographic characteristics* | | | | | |
| Age father | | −0.10 | | | |
| Age patient | | | | | |
| Time since end of treatment | | | | | |
| Duration of treatment | −0.12** | | | | |
| Leukaemia or lymphoma | | | | | |
| Radio- and chemotherapy | | | | | 0.35 |
| Prognosis >75% | | | | | 0.30 |
| Perceived treatment intensity | | | 0.01 | | 0.18* |
| No visible consequences | −0.10 | | −0.28** | −0.14* | |
| Percentage of explained variance by fixed effects | 13.0 | 3.8 | 17.6 | 6.7 | 4.9 |
| | | | | | |
| *Disease-related coping (CCSS)* | | | | | |
| Predictive control | −0.11** | −0.14** | −0.34** | −0.30** | |
| Illusory control | 0.06 | | 0.18** | 0.07 | |
| Interpretative control | | | 0.14** | | 0.10 |
| Vicarious control | | | | | 0.06 |
| Percentage of explained variance by fixed effects | 23.3 | 18.4 | 41.1 | 35.2 | 9.0 |
| | | | | | |
| *Generic coping (UCL)* | | | | | |
| Active problem focusing | | | −0.10 | −0.13** | 0.21** |
| Expression of emotions | 0.08 | | | | −0.20** |
| Palliative reaction pattern | −0.08* | | 0.10* | 0.08* | 0.13* |
| Passive reaction pattern | 0.41** | 0.33** | 0.17** | 0.38** | |
| Comforting cognitions | 0.05 | 0.06 | | | 0.12* |
| Seeking social support | | | | | |
| Avoidance behaviour | | 0.03 | | | |
| Percentage of explained variance by fixed effects | 45.7 | 39.4 | 47.6 | 61.8 | 19.4 |
| | | | | | |
| *Family functioning and social support* | | | | | |
| Adaptability (GDS) | | 0.09* | 0.12* | 0.09* | |
| Cohesion (GDS) | | | 0.23** | | |
| Supportive interactions (SSL) | | −0.18** | | | 0.12* |
| ≥2 important family events (last year) | | | | | |
| Percentage of explained variance by fixed effects | 45.7 | 45.0 | 51.3 | 61.8 | 22.5 |
| Total number of observations | 421 | 426 | 353 | 421 | 357 |

*$p < 0.05$, **$p < 0.01$.

Parental levels of distress and disease-related negative feelings returned to normal levels in the first two years after the completion of treatment. This finding is not in line with the results of most other studies, in which elevated levels of distress were found during the first years after the end of treatment [3,4,6,22,27,45–48]. It should be realized that the favourable outcomes presented in the VOLG study could not be extrapolated to the parents of children who suffered a relapse. If these parents are included in the analysis, higher levels of psychological distress and more disease-related

negative feelings are found. In addition, patients with brain tumours were underrepresented in the longitudinal VOLG study for logistical reasons.

While the findings in the present study indicate that, on the whole, parents adjusted well over time to the experience of childhood cancer in their family, early identification of parents who are at the risk of developing adjustment problems is important so that appropriate support can be offered at an early state. The results of our longitudinal mixed model analysis showed that both disease-related and generic coping and family functioning were predictive of emotional functioning, independent of the impact of demographic and medical factors. These results are discussed in greater detail below, followed by discussion of the limitations and clinical implications of the present study.

With respect to *disease-related cognitive coping*, it can be concluded that the more optimistic parents were about the further course of the disease (*predictive* control), the less emotional distress and the fewer disease-related negative feelings they reported. Although the present study does not answer the question of causality, it is plausible to suppose that optimism about the further course of the disease leads to less emotional distress and fewer disease-related feelings of loneliness, uncertainty and helplessness. In other words, being hopeful could protect parents from negative emotions. The protective impact of a positive view on adjustment to stressful events has been previously reported [49,50] and is in line with previous research on the parents of children with cancer [16].

*Illusory* control is found to have a negative association with parental emotional functioning, especially in mothers. This association was also reported by Grootenhuis and Last [16] and studies on care giving for diseased adults showed also that wishful thinking was negatively associated with adjustment. Once again, we cannot establish causality, but it seems plausible that parents who feel lonely, uncertain and helpless about the disease may come to rely on wishful thinking [51,52].

Five out of the seven *generic coping* styles were associated with parental emotional functioning. The passive reaction pattern was the strongest predictor of emotional functioning in both mothers and fathers. Goal-oriented parents who faced the situation calmly (active problem focusing) reported better emotional functioning than parents who coped with stress by taking a passive standpoint and allowing themselves to be totally immersed in the problem (passive reaction pattern) and parents who engaged in distracting activities and tried to relax (palliative reaction pattern). These correlates are not surprising, since passive coping is related to the concept of 'learned helplessness' and active coping to feelings of control over events [53]. The findings in the present study agree with those of

other investigations of parents of children with cancer [53,54] and other life-threatening diseases [55,56].

With regard to *family functioning*, we found a cohesive family structure to be particularly important for mothers. Mothers who reported more family cohesion were less distressed and lonely, and they reported more positive feelings. This finding is not surprising, as many parents mentioned that the experience of childhood cancer led to stronger family bonding [11,57]. Other studies have indicated that stronger family cohesion is related to lower levels of parental distress 18 months post-diagnosis, and to less post-traumatic stress at least one year after the end of successful treatment [3,22].

Higher levels of family adaptability appeared to be associated with stronger feelings of loneliness, helplessness and uncertainty. In the Circumplex model of marital and family systems, the theoretical framework proposed by Olson *et al.* [58], moderate levels of cohesion and adaptability are considered to be related to the most favourable adjustment outcomes in families faced with stress, whereas extreme levels of adaptation ('chaotic' family systems) and cohesion ('enmeshed' family systems) are related to less adaptive functioning. Inspection of our data, however, did not demonstrate extreme high levels of adaptability and cohesion in the families under study. Previous studies showed inconsistent results on this point [46,59–62].

More *supportive interactions* were found to be associated with lower levels of loneliness and more positive feelings. Correlations of social support with psychological distress, helplessness and uncertainty, however, were found in neither fathers nor mothers. Other studies [13,21,22] showed positive correlations between parental emotional functioning and social support, but these findings concern a shorter period after diagnosis than in the present study.

## Limitations and practical implications

Data were as far as possible collected from all parents two months, and one, two and three years after the end of treatment (i.e. at measurement points M1, M2, M3 and M4, respectively). Due to the long inclusion period and the finite follow-up period of the VOLG study, a considerable proportion of the parents did not complete the questionnaires four and five years after treatment (M5 and M6). As a result, sample sizes were relatively small on these two last occasions. The observations at M5 and M6, however, can be used to increase the precision of the parameter estimates that are not specific to M5 and M6. Our investigations showed that parents with incomplete data did not differ from parents with complete data with respect to their scores on the outcome variables at M1, so

that, probably, the incomplete data led to no bias in the results. The small sample sizes at M5 and M6 did, however, result in lower power, and the scores at M5 and M6 carried less weight than those at previous measurement occasions.

Meaningful conclusions cannot be drawn for the parents of children with brain tumours because these children are underrepresented in the longitudinal VOLG study. Furthermore, as mentioned above, the generalizability of the results is—on purpose—limited to parents of children who did not suffer a relapse. This is also a strength of this study.

Another strength of this study is that it includes both mothers and fathers, and a large number of psychosocial variables as possible predictors of parental emotional functioning. A disadvantage of the large number of variables chosen was that it was necessary to pre-select variables for the final analyses, although the sample size was fairly large compared with other studies into paediatric cancer.

Another restriction lies in the variables of the research model chosen. Firstly, the present study focused on a limited number of outcomes, namely psychological distress and situation-specific emotional reactions. Other interesting aspects indicative of parental emotional (mal)adjustment, such as post-traumatic stress symptoms, could also be taken into account [6,22,46–48]. Secondly, we assessed neither previous parental emotional functioning (such as a history of psychiatric problems) nor socio-economic variables (such as income and employment)—factors that have been shown in previous studies to have an impact on parental functioning [14,17]. Intrapersonal factors such as personality and temperament may also affect adjustment [63]. These were partly expressed in the personal coping styles investigated in our study.

In conclusion, despite the overall resilience in parents over time found in this study, there are good reasons why support for parents should not stop when treatment ends. Firstly, continued support for families might relieve psychological distress in the first couple of years after the cessation of treatment and help parents to get back to normal daily life. Secondly, certain subgroups of parents appeared to be at greater risk of worse emotional malfunction. If parents are still experiencing high levels of distress and disease-related negative feelings of helplessness and uncertainty one to two years after the end of treatment, they may suffer adjustment problems. Particular attention should be paid to parents who are less optimistic about the further course of the disease, and parents who have a passive coping style.

Oncologists could play a part in tracing adjustment problems in parents. If they observe that parents continue to report higher levels of distress than called for by the health of the child, psychosocial support may be appropriate. Screening parents in an early stage would make sense, since we did not find any interaction of time with psychosocial predictor variables, which means that the correlations with the outcome are applicable to each measurement occasion. It is important for care givers to understand emotional and behavioural reactions as outcomes of a coping process, so as to be able to respond to them more appropriately. Providing information on the psychosocial consequences of their child's condition and helping them to treat their child as normally as possible could prevent late psychosocial problems by enhancing re-entry into normal life.

The results could yield points of departure for interventions aimed at improving parental emotional functioning. In this perspective, we should consider the question of causality and changeability of the psychosocial factors found to be associated with parental emotional functioning. These questions cannot be answered definitely, but there were some indications that interventions on coping could improve well-being [64]. The results of a pilot study into reducing distress and improving family functioning by means of cognitive behavioural and family therapy for adolescent survivors and their families (the Surviving Cancer Competently Intervention Program—SCCIP) are promising [65,66].

Above all, the aftercare for the survivors should also be directed at the parents, in order to be able to support parents optimally and to trace parents at the risk of adjustment problems. Increasingly, computer-scored individual measurement of HRQoL is used in clinical practice to inform the physician about the patient's HRQoL. Computer-scored measurement of emotional well-being in parents is also recommended.

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## Note

1. Parental data from one family were not available at M1. Data at subsequent measurement occasions were included.

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