Assessment of Parental Psychological Stress in Pediatric Cancer: A Review

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Objectives We present an overview of the literature between 1997 and 2007 on parental stress reactions following the diagnosis of childhood cancer and we evaluate methodological strengths and weaknesses of the studies. Methods PubMed, PsychInfo, and Cinahl databases were used. Sixty-seven were included in the review. Results The conceptualization of parental stress and timing of assessment varies considerably between the studies, which makes comparison difficult. Most emotional stress reactions are seen around the time of diagnosis, with mothers reporting more symptoms than fathers. As a group, parents seem relatively resilient, although a subset of parents reports continuing stress even up to 5 years or more postdiagnosis. Conclusions The authors recommend clear definitions of parental stress, fixed points in time to assess parental stress, and an approach that highlights both parental strengths and weaknesses. Improved assessment can contribute to tailoring psychological care to those parents most in need.

Key words assessment; parent; pediatric cancer; psychology; stress reaction.

The diagnosis of childhood cancer is one of the most intense, disruptive, and enduring experiences that parents can have. The often unexpected and life-threatening diagnosis and the initiation of invasive medical treatment and its sequelae interfere with the entire family’s normal activities and routines for a long period of time and impose stressors of varying duration, predictability, and impact (Grootenhuis & Last, 1997a; Kazak et al., 2001; Kazak, Simms, & Rourke, 2002; Lindahl-Norberg, 2004; Stuber, Kazak, Meeske, & Barakat, 1998). Since substantial progress has been made in cancer treatment and coordination of care, types of cancers that were once regarded as fatal are presently curable with treatment and have instead become chronic life-threatening diseases (Eiser, Hill, & Vance, 2000; Stewart & Mishel, 2000). Nowadays, three out of four children diagnosed with a malignancy will survive their disease and treatment (Greenlee, Murray, Bolden, & Wingo, 2000).

When parents are confronted with a diagnosis of cancer in their child a process starts, referred to as psychological stress (Glover & Poland, 2002; Lindahl-Norberg, 2004; Young, Dixon-Woods, & Heney, 2002). In the literature, definitions of core elements of psychological stress vary considerably, often depending on the underlying theory (Pai, Drotar, Zebracki, Moore, & Youngstrom, 2006). Much research has been focused on stress reactions observed in emotional manifestations of strain (anxiety, depressive symptoms) and more situation-specific emotional manifestations of strain (uncertainty, helplessness, loneliness, and disease-related worry concerning future health and recurrence of the disease) (Grootenhuis & Last, 1997a; Lindahl-Norberg, 2004; Stewart & Mishel, 2000). Furthermore, a growing body of research has suggested that the impact of childhood cancer on the parents can well be conceptualized in terms of trauma-related symptoms or posttraumatic stress symptoms (PTSS) (Kazak, Alderfer, Rourke, et al., 2004; Kazak, Alderfer, Streisand, et al., 2004; Kazak, Alderfer, Streisand, et al., 2004; Stuber et al., 1998). The outcome of the psychological stress process is generally referred to as adjustment (Grootenhuis & Last, 1997a; Lindahl-Norberg, 2004). The current review is based on the theory on stress and coping by Lazarus and Springer (1984): when parents are confronted with a cancer diagnosis in their child (i.e., the stressor), a process starts,
involving the appraisal of the stressor, followed by strain, (i.e., pressure or demand), and stress reactions, or the manifestations of strain, which become manifest as uncertainty, anxiety, depressive symptoms, and PTSS.

The aims of the present review are, first, to evaluate the methodological strengths and weaknesses of studies on the emotional manifestations of strain in parents of children who have been diagnosed with cancer. Second, we will describe the prevalence and nature of parental strain according to disease phase (diagnosis, active treatment, maintenance, and long-term survival), gender differences, and risk and protective factors. Throughout the review article, we will group parental stress reactions into four main diagnostic categories, namely uncertainty, anxiety, depressive symptoms, and posttraumatic stress (PTS). Recommendations will be made for future research.

Method

The following sources of published reviews have been consulted: PubMed, PsychInfo, Cinahl, The Cochrane Library, and Web of Science. We prepared search filters and consulted databases to be accessed. The computer databases PubMed, PsychInfo, and Cinahl were used for a search with the key words: parent, mother, father, stress reaction, psychological stress, adaptation, coping strategy, neoplasm/psychology, and pediatric cancer. Next, all reference lists of identified papers were examined and then a hand search for identified relevant studies was conducted.

The following criteria for inclusion were applied: firstly, year of publication: studies were published between January 1, 1997 and May 31, 2007; second, language: English language studies; third, method: standardized measures of well-documented psychometric quality and the conduct of statistical tests; and lastly, aim: assessment of parental strain, parental stress reactions, and the adaptation related to caring for a child with cancer. The following exclusion criteria were applied: case studies, qualitative studies, book chapters, guidelines, commentaries, and dissertations. Reference Manager Version 10 for Windows was used as the bibliographic software package to organize the relevant references.

Results

We found four other review articles on parental stress, adjustment, and coping, first of all the extensive review by Grootenhuis and Last (1997a) on articles published between 1980 and 1997. A recent review article with a more theoretical character (Klassen et al., 2007) presents an overview of existing literature on the factors influencing parental health and well-being and a review by Bruce (2006) has focused on PTS in both childhood cancer survivors and their parents. Lastly, Cant Peterson and Drotar (2006) published a review article on the family impact of neurodevelopmental late effects in pediatric cancer survivors. Although there are overlapping issues discussed in our review article and the abovementioned reviews, we also see differences between the articles concerning aims and scope. The present review could be seen as a follow-up on the work done by Grootenhuis and Last, concerning articles published in the last 10 years, that is between January 1, 1997 and May 31, 2007.

We found 79 articles with our search strategy, of which 67 articles met the inclusion criteria. Selected studies are summarized, in chronological order, in Supplementary Table I (Note: the table is available at http://jpepsy.oxfordjournals.org). Studies referring to the same sample are described together. The studies reported in this review are difficult to compare, because they do not only differ in design, but also in sample (both size and heterogeneity), inclusion of control groups, time of assessment, definition of core elements of psychological stress, and measurements.

Methodological Issues

Terms used to describe the core elements of parental psychological stress vary considerably between the studies: from emotional strain or psychosocial difficulties to care-giving demands, from affective responses and psychological symptoms to uncertainty, anxiety, depression and posttraumatic stress disorder (PTSD), and from distress, well-being, and mental health to psychosocial functioning and adjustment.

One time, cross-sectional surveys were employed in the majority of studies. Although these designs are not appropriate to assess the effect of time since diagnosis, they have been used very frequently to assess parental strain in relation to disease phase. Sixteen studies (23% of the total) employed longitudinal designs, in order to assess parental manifestations of strain in relation to disease phase. Six intervention studies were included, one of which employed a case control design (Kazak et al., 1999) and five were randomized controlled trials (RCT) (Hoekstra-Weebers, Heuvel, Jaspers, Kamps, & Klip, 1998; Kazak, Alderfer, Streisand, et al., 2004; Kazak et al., 2005; Sahler et al., 2002, 2005). The intervention studies will not be further discussed in this review article.
because this has been done in a recent meta-analysis by Pai and colleagues (2006).

Sample sizes ranged considerably from 15 to 544 parents in cross-sectional studies, from 21 to 164 parents in longitudinal studies and from 18 to 252 parents in the intervention studies. While the majority of studies included both mothers and fathers, 12 studies focused solely on the mothers and one study (Mu, Ma, Hwang, & Chao, 2002) included fathers only. Results were compared with control groups, norm groups of the measures used, and groups of parents of children with other illnesses.

The majority of studies used heterogeneous samples, that is, parents of children with mixed cancer diagnoses. Among the various cancer diagnoses, treatment course varies considerably, with an ensuing risk for complications such as required hospitalizations for chemotherapy, unanticipated hospitalizations for fever and/or neutropenia, and varying foci for radiotherapy treatment. These treatment-related events can have a different impact on parental stress. A number of studies did focus exclusively on parents of children with leukemia (Best, Streisand, Catania, & Kazak, 2001; Iqbal & Siddique, 2002; Kazak, 1997; Lou, 2006) or a brain tumor (Bonner et al., 2006; Fuemmeler, Mullins, & Marx, 2001).

In 26 studies, parents of children who had recently been diagnosed with of cancer were included, ranging from 1 week to 6 months postdiagnosis. Furthermore, 24 studies assessed parents of children in active and/or maintenance treatment, 24 studies assessed parents of children both in- and off-treatment, and 26 studies solely included parents of children off active cancer therapy, that is parents of survivors. The definition of survivorship varied considerably between studies. Some researchers considered the number of months and/or years since completion of cancer treatment to be indicative of survivorhood, while others used the number of months and/or years since diagnosis to indicate survivorhood. Survivors ranged from 6 months to 10 years since completion of cancer treatment and from 15 months to 13 years since the diagnosis of cancer.

Although it is often concluded that traditional measures of psychopathology may be ineffective and/or insufficiently sensitive in the assessment of psychologically “healthy” parents in an abnormal crisis situation (Barrera et al., 2004), a substantial body of research still depends on these instruments. This is also true for the studies included in this review.

However, a number of pediatric psychologists have developed and used promising disease-related measures (Derogatis, 1986; Goldbeck & Storck, 2001; Grootenhuis & Last, 1997b; Moore & Mosher, 1997; Streisand, Kazak, & Tercyak, 2003) and disease-specific measures (Bonner et al., 2006; Kazak 2001; Kazak et al., 2003; McCubbin & Svavarsdottir, 1999) to assess parental stress reactions related to childhood illness or specifically, childhood cancer. In the majority of studies, these newly developed instruments were used alongside traditional measures on anxiety, depression, PTSS, and uncertainty.

Time of assessment of parents of children with cancer ranged considerably between studies. Timing in the cross-sectional studies ranged from diagnosis to >7 years posttreatment. In the longitudinal studies, first assessment of parents ranged from 1 week postcancer diagnosis to >3 years postcancer diagnosis.

### Emotional Manifestations of Strain According to Disease Phase

Several salient themes appear when examining emotional strain by phase of disease; these include the proportion of parents reporting strain, the correlates of stress reactions, and the evolution of these reactions in time. Phases that are distinguished are the diagnostic or consolidation phase, the initial treatment phase, the active treatment phase, the maintenance phase, and survivorhood. We will discuss these phases for each diagnostic category.

### Uncertainty

Broadly defined, parental uncertainty in childhood cancer pertains to both acute and ongoing or pervasive fear of possible disease consequences like relapse or death (Stewart & Mishel, 2000). In six studies, all cross-sectional, the construct of uncertainty in childhood cancer was investigated (Boman, Lindahl, & Björk, 2003; Fuemmeler et al., 2001; Grootenhuis & Last, 1997b; Mu, Ma, Hwang, & Chao, 2002; Santacroce, 2002). Uncertainty in parents of children with cancer has not been compared to uncertainty levels in parents of healthy children.

Compared to parents 1–5 years posttreatment, parents of children immediately after completion of treatment reported the most feelings of uncertainty (Stam, Grootenhuis, Brons, Caron, & Last, 2006). Between 66% and 90% of parents reported feelings of uncertainty after termination of treatment (Boman et al., 2003).

Some parents of childhood cancer survivors may continue to be uncertain about the well-being of their children many years after the cessation of treatment (Grootenhuis & Last, 1997b). In the short term, high levels of uncertainty may interfere with making health decisions. In the longer term, when parental uncertainty
becomes chronic, pervading the disease trajectory, it can lead to the development of PTSS (Mishel, 1999).

**Anxiety**

Anxiety refers to a complex combination of emotions that include fear, apprehension, and worry. Since anxiety entails an expectation of diffuse and uncertain threat, it plays an obvious role in the experience of parents when confronted with the life-threatening diagnosis of cancer in their child. Approximately 22 studies included in this review investigated the construct of anxiety, of which 13 studies employed a cross-sectional design, five studies a longitudinal or prospective design, and four studies a RCT or case control design (Supplementary Table I).

Anxiety occurs most frequently around the time of diagnosis and decreases over time. Parents of children newly diagnosed or in active cancer therapy reported higher levels of anxiety than parents of children off active cancer therapy, in remission, or parents whose child has relapsed (Moore & Mosher, 1997; Yeh, 2002; Santacroce, 2002). In turn, parents of children with a relapse reported higher anxiety levels than parents of surviving or deceased children (Wijnberg-Williams, Kamps, Klip, & Hoekstra-Weebers, 2006a).

Longitudinal designs show that anxiety levels at diagnosis decrease across time to (near) normal levels 5 years postdiagnosis (Wijnberg-Williams, Kamps, Klip, & Hoekstra-Weebers, 2006a,b). Yet, symptoms of anxiety seem more common among parents of children with cancer, compared to parents of healthy children, even up to 5 years postdiagnosis. This suggests that feelings of anxiety are maintained over time with a subset of parents continuing to be anxious. Prospective longitudinal research has shown that highly anxious parents are at risk for the development of PTSS (Best et al., 2001; Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1999). Psychosocial functioning at 6 months after diagnosis seemed to predict later psychosocial functioning best (Wijnberg-Williams et al., 2006b).

**Depressive Symptoms**

Parents may react to the diagnosis of cancer in their children with depressive symptoms (Barrera et al., 2004; Lindahl-Norberg, Lindblad, & Boman, 2005b). Depressive symptoms include, but are not limited to, a persistent sad, anxious or empty mood, feelings of hopelessness or pessimism, feelings of guilt or helplessness, decreased energy, difficulty concentrating or making decisions, restlessness, and insomnia or oversleeping. Eighteen studies included in this review investigated the construct of depression, of which 11 studies employed a cross-sectional design, five studies a longitudinal or prospective design, and two studies a RCT (Supplementary Table I).

High levels of depressive symptoms are reported shortly after diagnosis (Allen, Newman, & Souhami, 1997; Yeh, 2002). Mothers of children newly diagnosed, in active cancer therapy and 1 year postdiagnosis reported more depressive symptoms than mothers of children off active cancer therapy (Von Essen, Sjödén, & Mattsson, 2004). Compared to parents of healthy children, parents of children with cancer showed higher levels of depressive symptoms at multiple points from the time since diagnosis (Dockerty, Williams, McGee, & Skegg, 2000; Lindahl-Norberg, Lindblad, & Boman, 2005b).

In mothers and fathers for whom a longer period of time had elapsed from the time of diagnosis, depressive symptoms were less common (Boman et al., 2003) but in another study parents consistently reported higher depression scores than the norm group of the questionnaire under study (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1998).

Longitudinal studies suggest that depressive symptoms may be maintained over time, especially when parents initially react with moderate to severe levels of depressive symptoms. However, one cannot automatically conclude that the child’s diagnosis is the cause of depressive symptoms in parents (Manne et al., 1996). Other events, such as marital or financial problems, may also result in depressive symptoms and should be assessed simultaneously. Furthermore, because it is not possible to assess parents prior to the child’s cancer diagnosis, the possibility that the depressive symptoms represent a preexisting state cannot be ruled out (Manne et al., 1996). Depressive symptoms of the parent may interfere with, for example, health decisions, frequent clinic appointments, and the parent–child relationship and communication.

**PTSS**

Learning that one’s child has a life-threatening disease is a qualifying event for PTSD or PTSS (American Psychiatric Organization [APA], 1994). PTS acknowledges the life threat inherent in childhood cancer, while also providing a framework in which ongoing symptoms such as intrusive thoughts, arousal, and avoidance may be conceptualized and treated (Kazak et al., 2004). Twenty studies included in this review investigated PTSS or PTSD, of which 13 studies employed a cross-sectional design, three studies a longitudinal or prospective design, and four studies a RCT or case control design (Supplementary Table I).
Approximately 68% of mothers and 57% of fathers of children currently in treatment report PTSS in the moderate to severe range (Kazak, Boeving, Alderfer, Hwang, & Reilly, 2005). Sub-clinical PTSS such as intrusive thoughts about cancer, physiological arousal at reminders, and avoidance of treatment-related events have been found to be even more prominent (Alderfer, Cnaan, Annunziato, & Kazak, 2005). For parents of childhood cancer survivors, the rates of PTSS have been found to range from ~10% (Kazak et al., 1997) to 42% (Fuemmeler et al., 2001).

Parents of children recently diagnosed or currently in treatment report higher rates of PTSS and current PTSD compared to parents of childhood cancer survivors (Kazak et al., 2004, 2005; Lindahl-Norberg, Lindblad, & Boman, 2005b; Phipps, Long, Hudson, & Rai, 2005; Santacroce, 2002). Mothers and fathers of childhood cancer survivors show significantly higher levels of PTSS and lifetime PTSD than parents of healthy children (Barakat et al., 1997; Brown, Madan-Swain, & Lambert, 2003; Kazak et al., 1997; Pelcovitz et al., 1996) but lower than symptom levels for other stressed and traumatized groups (Kazak et al., 1997). An extensive review article on PTSS and PTSD in childhood cancer survivors and their parents has been written by Bruce (2006). He summarized the following risk factors associated with PTSS and PTSD: female gender, greater physical late effects, increased number of prior stressful life events, perceived severity of cancer and treatment, family conflict, poor social support, and emotion-focused coping.

It remains a matter of debate whether traumatic stress is a relevant model to describe the emotional reactions of parents of children with cancer (Phipps, Larson, Long, & Rai, 2006; Stuber et al., 1998). However, symptoms of PTS in parents are a concern and may be an appropriate target for intervention, particularly in the period following diagnosis (Phipps et al., 2005). Early signs and symptoms of PTS require early assessment and intervention since the disruptive symptoms may linger over time in a subset of parents (Barakat et al., 2000; Stuber, 2006).

Emotional Manifestations of Strain and Gender of the Parent

Stress reactions can take different forms in fathers and mothers and it may be relevant to identify these differences in order to deliver specific interventions. Twenty-three studies included in this review compared emotional manifestations of strain in mothers and fathers of children with cancer, of which 13 studies employed a cross-sectional design, nine studies a longitudinal or prospective design, and one study employed a RCT (Supplementary Table I).

Gender Differences in Uncertainty, Anxiety, Depression, and PTSS

Evidence for gender differences in parental uncertainty in childhood cancer has not been well established. In one study, mothers of children in remission or with a relapse reported higher levels of uncertainty than fathers (Grootenhuis & Last, 1997b).

Mothers of children newly diagnosed, in remission, relapsed, or off-treatment report higher levels of anxiety and depressive symptoms than fathers of children with cancer (Allen et al., 1997; Dockerty et al., 2000; Von Essen et al., 2004; Iqbal & Siddique, 2002; Lindahl-Norberg, Lindblad, & Boman, 2005a; Wijnberg-Williams et al., 2006a; Yeh, 2002), whereas other researchers found no gender differences (Frank et al., 2001; Hoekstra-Weebers et al., 1998). In one study that focused on fathers who identified themselves as the primary caregiver, elevated rates of depressive symptoms were found more in fathers than mothers (Bonner et al., 2007). Perhaps being the primary medical caregiver adds to the strain instead of the gender of the parent?

With regard to PTSS and PTSD, mothers have been reported to display more symptoms than fathers (Alderfer et al., 2005; Bruce, 2006; Phipps et al., 2005; Yeh, 2002), especially re-experiencing and arousal symptoms. However, other studies show relatively equal levels of PTSS and rates of current PTSD (Kazak et al., 2004; Magal-Vardi et al., 2004; Phipps et al., 2005). Gender differences in the experience of PTSS may be related to the time of evaluation: over time, only the fathers’ symptoms decreased, whereas the mothers’ symptoms remained high (Magal-Vardi et al., 2004).

In agreement with gender studies on the prevalence of psychological problems in the general population, mothers of children with cancer tend to report more and higher levels of symptoms than fathers. However, it is still not clear whether the differences between mothers and fathers in these studies represent different stress reactions to childhood cancer or are related to general population differences between men and women (Sloper, 2000). Women seem more willing to report discomfort than men. Therefore, gender differences may be due to reporting style (Greenberg & Meadows, 1991). Another explanation may be that mothers more often have the main responsibility for the care of the child with cancer and fathers are more peripherally involved in childcare.
The question remains whether it is necessary and possible to tailor interventions to specific needs of mothers and fathers of children with cancer.

**Risk Factors**

Since parents of children with cancer are at risk for the development of disruptive emotional manifestations of strain, which persist over time among a subset of parents, it seems important to obviate risk factors early in order to detect and support parents most at risk for later maladjustment. Several variables have been indicated as risk factors for the development of emotional manifestations of strain.

Risk factors include, but may not be limited to, the following findings: parents who display the most and highest levels of emotional manifestations of strain at diagnosis continue to experience the highest levels of symptoms, even after treatment ends. Certain demographic characteristics have been identified as risk factors: parents of children with cancer who are less educated and parents with lower SES (Iqbal & Siddique, 2002) or parents with a "perceived unsatisfactory financial status" (Lou, 2006) report more depressive symptoms.

Trait anxiety has been identified as a predictor of posttreatment PTSS for mothers (Best et al., 2001) and for both mothers and fathers (Hoekstra-Webers et al., 1999; Kazak et al., 1998; Stuber, 2006). No association with treatment intensity and minimal associations with time since diagnosis have been found (Kazak et al., 2005). Child behavior problems (Barrera et al., 2004) were found to be predictive of parental depressive symptoms. High levels of care giving demands, past traumatic life events, and less perceived social support have also been identified as risk factors for the development and maintenance of emotional manifestations of strain.

Attention should be given to parents with preexisting psychological problems, because they may be less able to deal with the crisis of having a child with a life-threatening disease. Knowledge of risk factors may help identify those parents most in need of psychological care and interventions, preventing these parents from developing disruptive emotional manifestations of strain beyond the "normal" reactions to the life-threatening diagnosis of cancer.

**Protective Factors**

Several studies have focused on protective factors and on parental adjustment rather than parental stress. We will summarize the positive effect that coping strategies, social support, and family relations are shown to have on parental adaptation.

**Coping Strategies**

Because stressors change with the different phases of cancer, studies on parental coping strategies should be classified according to the phase of cancer (Goldbeck, 2001; Trask et al., 2003). Moreover, the adaptive value of a coping strategy is likely to be dependent upon the phase of cancer. Studies addressing changes in coping strategies over the course of childhood cancer are relatively scarce (Trask et al., 2003). Avoidance seems to be functional in the early phase of childhood cancer when parents are overwhelmed with stressors. However, in face of active treatment and maintenance, avoidant behavior of the parent has been related to elevated levels of emotional manifestations of strain, e.g., anxiety and depression (Hoekstra-Webers et al., 1999; Lindahl-Norberg, Lindblad, & Boman, 2005a).

According to Grootenhuis and Last (1997b), low levels of predictive control coping (i.e., finding it difficult to have positive expectations about the course of the disease), were related to higher levels of emotional manifestations of strain in mothers and fathers of children in remission or with a relapse. More frequent use of active problem focused coping strategies (e.g., acting immediately, being goal oriented), and less frequent use of palliative reactions, avoidance behavior, passive reactions, and expressing negative emotions were associated with less depressive symptoms and anxiety in parents of children in active cancer treatment and children that are cancer-free (Lindahl-Norberg, Lindblad, & Boman, 2005a).

We recommend longitudinal studies with repeated measures within the same cohort over time to examine, which coping strategies are likely to be maladaptive during a particular phase of childhood cancer and require early assessment in order to prevent further psychological problems.

**Social Support**

Social support seems to have a moderating effect on the impact of anxiety, depressive symptoms and PTSS (Barakat et al., 1997; Dockerty et al., 2000; Frank et al., 2001; Lindahl-Norberg, Lindblad, & Boman, 2005b; Manne, DuHamel, & Redd, 2000; Sloper, 2000). Higher levels of perceived social support have been associated with less anxiety (Dockerty et al., 2000; Lindahl-Norberg, Lindblad, & Boman, 2005a, 2006; Sloper, 2000), lower PTSS levels (Barakat et al., 1997; Kazak et al., 1997), and better
adjustment to medical disease (Han, 2003). On the other hand, a small network size, more perceived social constraint, and a less perceived sense of belonging have been associated with more PTSS in parents of pediatric cancer survivors (Best et al., 2001; Bruce, 2006; Kazak et al., 1998; Sloper, 2000).

Assessing and evaluating both the parents’ specific needs for support and the availability of support is important to meet those needs throughout the course of childhood cancer (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 2001).

**Family Relations**

The family plays an important role in the psychological functioning of both the parents and the child with cancer (Kazak et al., 1997, 2002; Barakat et al., 1997; Robinson, Gerhardt, Vannatta & Noll, 2007). Good family relations, adequate family coping, and stable family functioning have been reported (Kazak et al., 1997; Lähteenmaki, Sjöblom, Korhonen, & Salmi, 2004; Sawyer, Antoniou, Toogood, & Rice, 1997, 2000) in studies with a systemic focus. However, marital distress (Yeh, 2002), poor family functioning, and poor family relationships have been reported as well (Streisand et al., 2003).

Although in most studies family functioning has been investigated as an outcome variable, some studies consider family functioning as a predictor variable for parental adjustment to childhood cancer (Grootenhuis & Last, 1997a). Less family cohesion, satisfaction, adaptability, and communication have been correlated to parental anxiety and therefore indirectly predicted PTSS (Kazak et al., 1998).

Screening for family functioning, at diagnosis, seems important to identify strengths that can serve as buffers to cope with the stressors to come.

**Discussion**

The diagnosis and treatment of cancer in one’s child can cause long-lasting psychological effects in a parent. Feelings of uncertainty, anxiety, depressive symptoms, and PTSS are most prevalent shortly after the parents are confronted with the diagnosis of childhood cancer. These emotional manifestations of strain decrease to near normal levels over time in the majority of the parents, but have been found to persist in a substantial proportion of the parents, even many years posttreatment. Furthermore, as is often found in the general population, mothers tend to report more and higher levels of symptoms than fathers with respect to anxiety, depression, and PTSS. These differences may well be related to the traditional distribution of care-giving tasks and responsibilities. Also, since women seem more willing to report discomfort than men, gender differences may also be due to reporting style (Greenberg & Meadows, 1991). The question remains whether these gender differences are meaningful and, consequently, whether mothers require specific intervention efforts.

Assessment of parental stress reactions is important to identify those parents most in need. The following risk factors have been indicated: female gender, preexisting psychological problems, high trait anxiety, low social economic status and financial worries, child behavior problems, high perceived care-giving demands, and less perceived social support. Certain coping strategies, such as active problem solving, seeking social support, and optimism can serve as protective factors. Specific strengths of the family should be identified and used. Parents might well benefit from a tailored intervention based on strengths and weaknesses that is targeted to their specific needs with respect to the phase of childhood cancer (Hoekstra-Weebers et al., 1998; Hoekstra-Weebers et al., 1999; Kazak et al., 2004b; Sloper, 2000; Streisand, Braniecki, Tercyak, & Kazak, 2001).

In most of the studies included in this review, parents of children with heterogeneous diagnoses were assessed, making comparisons difficult. Different rates of uncertainty, anxiety, and other stress reactions may be directly associated with the child’s type of cancer (e.g., parenting a child with standard risk ALL vs. a child with a malignant brain tumor). The inclusion of predominantly white parents and the assessment of either mothers alone or parents as a couple causes bias and generalization problems. The inclusion of nonnative speaking parents continues to be a difficulty, although efforts are being made to translate assessment instruments and intervention programs for these groups (Sahler et al., 2005, 2006).

A wide variety of assessment measures to measure parental emotional manifestations is seen across studies. As has been stated by many others, relevant, reliable, and valid assessment tools for parents of children with cancer are critically important in advancing the field of pediatric psychology because they can provide further evidence of the impact of childhood chronic disease on parents, as well as the potential need for and impact of psychological interventions (La Greca & Lemanek, 1996; Grootenhuis & Last, 1997a; Streisand, Braniecki, Tercyak, & Kazak, 2001). However, parents of children with cancer are coping with an abnormal situation and therefore existing instruments may fail to assess their specific problems.
psychological stress as an outcome. Investigators must clear distinction between stress as a primary reaction and reactions of both parents and patients, and to make a improvement in future research. In the 67 studies 

Disease-related and disease-specific measures can provide valuable, additional information when administered together with general measures (Streisand et al., 2003). It would be beneficial to both research and patient care to make use of the strengths of each different type of instrument. Sound psychometric properties of disease-related and disease-specific measures still need to be established and comparison groups are often small. Multicentered research and (inter) national collaboration is needed to obtain larger samples and to validate disease-related and disease-specific questionnaires developed by others or—to better yet—to develop new measures together. The DISABKIDS project (Bullinger, Schmidt, & Petersen, 2002) and the KIDSCREEN project (Ravens-Sieberer et al., 2001) are excellent examples of successful international collaborative projects yielding valid and reliable assessment tools to measure health-related quality of life (Goldbeck, 2006) in children with chronic conditions. Unfortunately, oncology was not incorporated in these projects.

Looking back on the last 10 years in pediatric psycho-oncology research, there is a trend toward larger studies; almost half (32) of the studies included at least 100 parents (in most cases, both mothers and fathers were included). The proportion of longitudinal studies seems to rise somewhat (14% in the Grootenhuis and Last review vs. 23% in the present review), but the majority of designs is still cross-sectional. This seems somewhat surprising, because in almost all articles the necessity of longitudinal designs is argued.

**Recommendations**

The present review study reveals potential areas of improvement in future research. In the 67 studies included in this review, a variety of definitions of the core elements of the psychological stress process have been used, often described together and simply referred to as “stress.” It is important to clarify what is meant by “stress” and to specify the temporal course of a stressor (La Greca & Lemanek, 1996). To facilitate communication and collaboration, it is necessary to be more specific in the terminology used to describe the psychological reactions of both parents and patients, and to make a clear distinction between stress as a primary reaction and psychological stress as an outcome. Investigators must determine whether they are interested in the person’s appraisal of the stressors, or simply in the occurrence of verifiable events. Another issue is the temporal course of the illness or condition itself, since the phase of an illness guides the “timing” of the assessment (Drotar, 1994). These aspects need to be specified before proceeding further with the study design and measurement strategy. In many instances, it matters whether the investigator is interested in processes that occur at the time of disease onset, in the period following initial diagnosis, during the course of treatment, when complications arise (such as a relapse) or in the longer term.

It seems we have no more need of more cross-sectional research in this area, given its limitations. Repeated, ongoing assessment with longer time frames remains necessary to follow parents prospectively through the different phases of illness, treatment, and long-term survival. It is recommended that a consensus be established on the optimal points in time to assess emotional reactions in parents following the diagnosis of cancer in their child. If assessments would take place for example 1, 6, and 12 months after diagnosis, at the end of treatment and 1 and/or 2 years after the cessation of treatment, the comparison of results from research would be facilitated and patient and parent care would be enhanced. Assessment shortly after diagnosis provides important information on the initial reactions of parents. However, clinical practice has shown that assessment within 4 weeks after diagnosis is difficult, because parents are often too overwhelmed to take the time to fill in questionnaires. Assessment at 6 and 12 months post-diagnosis will give insight in parental stress over time according to different disease phases. The end of treatment brings new challenges for parents and longer term follow-up is necessary to keep track of the parents who still report high-stress levels.

After identifying those parents most in need of more intensive psychological care, the next step is to deliver feasible, limited, brief interventions for sub-clinical manifestations of psychological distress. Intervention research is a growing area in pediatric psychology and despite the many methodological challenges; efforts should be made to implement and evaluate existing intervention programs to prove effectiveness. This can only be done through (inter)national cooperation and well-developed study designs.

Furthermore, it is recommended that investigators routinely describe their reasons for using particular assessment tools or questionnaires, which should be embedded in an underlying theoretical model.
Researchers seldom document their arguments for the selection of assessment measures used in their studies. This is unfortunate, because it would give more insight in the underlying theoretical model and it could facilitate discussion and communication among peers. One should also consider that measures could be used for different purposes. Important questions are: What does this measure do best? Is it a screening tool? Is it able to establish a diagnosis or to obtain a detailed picture of the problem? Is suitable for evaluating treatment outcome? (La Greca & Lemanek, 1996). Method and measure should match the study’s purpose. A screening instrument is not intended to analyze a person, but to direct scarce professional time to cases meriting more in-depth study or support (Cronbach, 1990). Development of brief screening instruments is important to identify parents at risk for preexisting, ongoing, and escalating emotional manifestations of strain (Kazak et al., 2001).

Lastly, instead of “pathologizing” parents by classifying them as anxious or depressed (Quittner et al., 1998, in Roberts, 2003 p. 696–709), it would be more helpful to investigate parents’ quality of life. Parental adjustment to childhood illness should be considered as a normative process involving additional daily responsibilities, limitations in major roles, and increased strain in close relationships. What is asked of parents is much more than in a normal parenting situation and acknowledging this would help parents cope better with the difficult and stressful situations with which they are confronted.

Supplementary Data

Supplementary data are available at JPEPSY Online.

Conflicts of interest: None declared.

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References


