Child and parental adaptation to pediatric oncology

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Parental stress before, during, and after pediatric stem cell transplantation: a review article

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Abstract

Goals of work. Pediatric stem cell transplantation (SCT) is a stressful treatment for children with relapsed or high-risk malignancies, immune deficiencies and certain blood diseases. Parents of children undergoing SCT can experience ongoing stress related to the SCT period. The aim of this article was to present a literature review of articles on parental distress and adaptation before, during, and after SCT and to identify risk and protective factors. *Methods.* The review was conducted systematically by using PubMed, Web of Science, PsychInfo and Picarta databases. Eighteen articles met our inclusion criteria: publishing date between January 1, 1990 and January 1, 2009; studies concerning parents of children undergoing SCT; studies examining the psychological adjustment and/or stress reactions of parents as primary outcomes and studies available in English.

Results. Highest levels of parental stress are reported in the period preceding SCT and during the acute phase. Stress levels decrease steadily after discharge in most parents. However, in a subgroup of parents, stress levels still remain elevated post-SCT. Parents most at risk in the longer term display highest levels of stress during the acute phase of the SCT. *Conclusion.* Psychosocial assessment before SCT, during the acute phase and in the longer term, is necessary to identify parents in need for support and follow-up care.

Introduction

Stem cell transplantation (SCT) is an invasive treatment for seriously ill children who have hematological, oncological, or metabolical diseases. Recently, for some high- risk leukemia protocols, SCT has become close to being a first-choice treatment. SCT is a perilous treatment, associated with significant mortality and morbidity [5]. It involves a lengthy hospital admission in an isolated environment to prevent infections and treatment with high doses of chemotherapy and/or radiation, followed by infusions of donor stem cells [22]. During the acute phase of SCT, children report high levels of somatic distress, mood disturbance [30], nausea and pain [7], and fatigue and malaise [29]. In the first 4-6 months post-SCT, children are still susceptible to infections and need to live with restrictions. SCT has a profound impact on the lives of children and parents, both during the acute phase and afterwards. Parents are faced with the need to provide both physical and emotional care for their ill child during a long and stressful period. Furthermore, they have to deal with their own emotions, especially with the realistic fear of losing their child and they have to make complicated decisions about the treatment together with the multidisciplinary team. Some parents are also faced with supporting one of their other children who will be acting as a sibling donor and increasingly, parents are acting as haploidentical donors themselves in the case no appropriate donor has been found.

Despite improved survival rates, SCT remains a high risk procedure. The result of the transplantation depends on several risk factors, including type and status of the underlying disease [5]. After treatment, parents and children are faced with the risk of recurrence, acute or chronic graft-versus-host disease (GVHD), and numerous possible late effects such as pulmonary disease, growth problems, and infertility [4,21]. Even in the longer term, the child's illness and the SCT may influence parents' everyday lives [8]. SCT treatment protocols have changed in the past decades, one of the most important differences between treatment now and in the 1990s is the shortened admission period. On the one hand this is an improvement because parents may have fewer concerns about the practical issues during admission, e.g. being away from home for a long period and dealing with work-related stress. On the other hand, caring for a child at home post-SCT can be a heavy burden on parents and families. In addition, increasing survival rates entail increasing numbers of survivors with possible long-term side effects. Moreover, the fear of losing the child is still realistic. Accordingly, changed treatment protocols may not make any difference for parental stress levels.

The field of parental adaptation to their child's SCT has gained more attention in the past decade; most studies have been conducted in the past 8 years [18,31]. The majority of studies have focused on parental stress and adjustment pre-SCT, during the acute phase, and 3, 6, or 18 months post-SCT, e.g. [5,18]. These time points seem to cover the SCT time frame well: 3 months post-SCT, many children still suffer from the after effects of the SCT, whereas after 6 months, most of the children can return to school and pick up their old lives, even if their health is suboptimal [1]. Twelve to eighteen months post-SCT, the majority of children report to have a health-related quality of life (HRQoL) comparable to healthy peers [2,7]. However, certain aspects of HRQoL seem to be affected in the long-term in many survivors, for example cognitive functions and pain, which has been demonstrated recently by Löf et al. [15].

No review articles have been published in this specific area so far. In related areas, however, review studies have focused on adjustment and coping of parents of children with cancer [9,40]; on the quality of life and/or emotional adjustment of children after SCT [2,38]; on the psychological adjustment of families of adult SCT patients [14] and on the psychosocial impact of SCT of adult patients [11,23]. The aim of our article was to conduct a systematic review of the current literature (1990 to 2008) on parental distress and adaptation to their child's SCT and to identify risk and protective factors.

Methods

Search strategy for identification of studies

Several research engines were used to obtain the studies included in this review: Pubmed, Web of Science, PsychInfo, and Picarta. These databases were searched for one of the words: BMT, bone marrow transplantation, SCT, or stem cell transplantation combined with the following words used in headings, keywords, subjects, or abstracts: pediatric, paediatric, parent, child, mother, father, AND/OR stress, distress, post-traumatic stress disorder (PTSD), Post-traumatic stress symptoms (PTSS), anxiety, depression, parental stress, parental distress, psychological, adjustment, and coping. Subsequently, reference lists of the relevant articles were examined to identify additional papers that met the search criteria and a hand search was conducted in several relevant academic journals.

Criteria for considering studies for the review

Criteria for inclusion were: publishing date between January 1, 1990 and January 1, 2009; studies concerning parents of children undergoing SCT; studies examining the psychological adjustment and/or stress reactions of parents as a primary outcome; and studies available in English. Exclusion criteria were: reviews, guidelines, protocols, commentaries, and other descriptive articles; studies focusing on psychological adjustment of pediatric SCT patients only; studies focusing on other critical illnesses or including other treatments; and studies focusing solely on intervention programs.

Description of the studies

Eighteen studies were selected for this review. Table 1 contains a descriptive summary of the articles. We included the aim of the study, number and characteristics of parents and children, methodological features (e.g. study design, timing of measurement); measures; and main results. The indications for SCT varied among the different studies. Most studies reported the underlying diagnosis, type of transplant and type of donor, transplant risk, and disease risk category [17]. On average 80% of the children undergoing SCT suffered from a malignant disease and around 60% of the children in the studies underwent allogeneic SCT (transplantation with bone marrow from a foreign donor) as opposed to autologous SCT (transplantation with own body material). The studies in our review did not distinguish between the experiences of parents of children undergoing SCT once and parents of children who had multiple SCT experiences, who will undoubtedly have to face a unique set of stressors.

In this review article, we will first discuss the methodological qualities of the studies, next we will summarize the main results of the studies, and lastly we will discuss risk and protective factors of parental adaptation to their child's SCT.

Methodological qualities of the selected studies

Study design and timing of assessment

Three of the included studies used a cross-sectional study design [3,20,27] and one study was descriptive/retrospective [8]. The other 14 studies used a prospective longitudinal design with repeated measures, ranging from two to 13 measurement points. However, only a few of these studies followed a particular aspect of parental distress over time, e.g. [18,32]. Exactly half of the studies included in the review used a multi-centered design, the other nine studies recruited participants from one medical center. Until now, only one intervention study has been published in this area. It included mothers of children undergoing SCT [36] and was based on a stress inoculation model.

In most longitudinal studies, two measurement points were used. The first time point was between 47 to 1 day(s) pre-admission and a few days post-SCT. The time point for the second assessment varied strongly between the studies, ranging from 1 week post-SCT to18 months post-SCT. Phipps et al. [31,32] used up to 13 time points in both studies. Only one of the studies assessed long-term parental distress, 4 to 8 years after stem cell transplantation [8].

Participants

The majority of the studies (13 in total) used only mothers as respondents. Sample size in the studies varied from 11 [36] to 207 parents [18]. Eleven of the studies included more than 90 parents. All studies described the recruitment process. Phipps et al. [31,32]

assessed one of the caregivers, resulting in 90% mothers. In only three studies [3,27,39], both parents were used as respondents. The age range of the children was most often 1 to 20 years of age with a mean age at first assessment between 8 and 9 years. In two studies, the age of the children was not mentioned [8,27].

Outcome measures

The conceptualization of stress or distress varied widely between the studies. The distinction between the assessment of (subclinical) levels of distress on the one hand and clinical psychiatric diagnoses was not always clearly made, which makes comparisons difficult. Anxiety and depression were studied as manifestations of parental distress in nearly all of the studies. Other manifestations of parental distress or psychiatric disorders were disturbed and obsessive-compulsive thinking [3], post-traumatic stress symptoms [16,17], generalized anxiety disorder (GAD), panic disorder (PD), and major depressive disorder (MDD) [20]. Somatic complaints and changes in sleep behavior were added by some researchers [25,34,35] as symptoms of parental distress. Variables influencing parental stress levels were mostly operationalized as 'coping' [18,24,25,32], 'family functioning' [32], 'parenting stress issues' [39] and 'social support', both positively and negatively perceived [19,25,32].

In most studies multiple measures were used, most often self-report questionnaires combined with, or additional to, interviews as a way of collecting data. In the majority of studies standardized questionnaires were used to assess parental distress reactions (e.g. Beck Depression Inventory (BDI), Beck Anxiety Inventory (BAI), State-Trait Anxiety Inventory (STAI), and Parental Stress Index (PSI)), e.g. [17,20]. Disease- or context-specific measures were used less frequently. Phipps et al. [31,32] developed and used the Prior Illness Experiences Scale (PIES) to assess previous parent and child experiences with cancer therapy and inpatient hospitalization. Rini et al. [35] created two items to measure benefit finding in their study and DuHamel and her study group developed a scale to assess maternal fear appraisals [6].

Table 1. Summary of the studies included in the review

No	*Year, Author*	*Aim*	*N parents N and age range/ mean age children*	*Methodological features*	*Measures*	*Results*
1	1990, Dermatis	Determine the nature and prevalence of the psychological symptomatology in parents of children undergoing SCT. Investigate relationship of certain psychosocial factors to parental distress associated with the informed consent process.	46 M, 15 F 61 children Range 1-17 yrs	Single-centered Cross-sectional	BSI, WOC, newly constructed scale on the quality of the physician-parent communication	47% of fathers and 60% of mothers exhibited significant psychological distress of a generalized nature. Mothers reported more severe levels of depression and anxiety than fathers did.
2	1997, Nelson	Examine the stress responses of mothers during their child's hospitalization for SCT; Determine the relationships between mothers' stress responses and the resources for coping and social support.	50 M 50 children Mean age 9.3 yrs	Single-centered Longitudinal, time points: time of admission (T1), second (T2), tenth (T3) and twentieth (T4) day post-SCT	STAI, CES-D, HAS, IES, SSS	Maternal anxiety and depressive scores decreased significantly over time The coping style defined as 'active reviewing of feelings or information associated with the situation' significantly explained variance in scores for anxiety, depressive symptomatology, somatic complaints and sleep behavior.
3	2000, Streisand	Document levels of stress in mothers of children undergoing SCT. Pilot a psychological intervention program.	11 M 11 children Range 2-16 yrs Mean age 8.8 yrs	Single-centered Longitudinal, time points: pre admission to 3 weeks post-SCT	DSI, PSI, SSINT	Most stress was reported pre-admission. Mothers reported using more stress management techniques post-intervention than mothers in the standard care condition. The analyses revealed no significant differences in stress between intervention and control mothers.
4	2001, Manne	Examine anxiety and depressive symptoms among mothers of children undergoing SCT.	115 M 115 children Range 4 months-20 yrs Mean age 9.2 yrs	Multi-centered Cross-sectional: 85% of mothers on day -7 to day -1; 15% of the mothers 10 days post-SCT	BAI, BDI, SCID-NP	20% of mothers were diagnosed with a MDD, a GAD, or a PD. There was evidence of comorbidity between anxiety and depressive disorders. Mothers with lower incomes, who were Caucasian, had received prior psychiatric care and were caring for female SCT patients may be at higher risk for adverse psychological reactions.
5	2002, Manne	Investigate the role of cognitive and social processing in post-traumatic stress symptoms and disorder (PTSD) among mothers of children undergoing SCT.	90 M 90 children Range 9 months-20 yrs Mean age 8.8 yrs	Multi-centered Longitudinal, time points: time of SCT, 3 and 6 months past SCT	SCID-NP-PTSD, PCL-C, BAI, BDI, fear network, CSI, LSCM	Emotional distress, SCT-related fears, and negative responses of family and friends assessed at the time of SCT hospitalization were predictive of later PTSD symptoms. Cognitive processing (the appraisal of threat) at the time of transplantation played the most important role in later PTSD symptoms.
6	2002, Oppenheim	Understand parents' perception of children treated in an SCT unit.	40 pairs of parents No details given	Single-centered Cross-sectional	Interviews	Parents expressed intense distress and disorientation and sometimes difficult relations with their child. Many parents expressed having an ambivalent relation with care providers.

No	*Year, Author*	*Aim*	*N parents* *N and age range/ mean age children*	*Methodological features*	*Measures*	*Results*
7	2003a, Manne	Evaluate the role of maternal coping strategies in depressive symptoms experienced by mothers of children undergoing SCT.	207 M 207 children Mean age 8.3 yrs	Multi-centered Longitudinal, time points: at SCT, 3 and 6 months post-SCT	COPE, BDI, appraisal of fear/ worry medical risk	Acceptance and humor were associated with reductions in maternal depressive symptoms. Planning and alcohol/ substance use were associated with increases in maternal depressive symptoms. Active problem solving and use of instrumental support did not predict changes in depressive symptoms.
8	2003b, Manne	Examine the role of perceived partner criticism and avoidance in anxiety and depressive symptoms of mothers of children undergoing SCT.	148 M 148 children Range 4 months-17 yrs Mean age 8.5 yrs	Multi-centered Longitudinal, time points: at SCT, 3 and 6 months post-SCT	SCID-NP-PTSD, PCL-C, BAI, BDI, Fear Network, CSI, CSI, LSCM	Fear structure, distress, and unsupportive responses by family and friends measured at transplantation were predictive of PTSD symptom severity at 6 months after SCT. Perceived partner criticism was associated with higher average depressive symptoms. ICU transfers and number. of days of hospitalization 6 months post-SCT were risk factors.
9	2003, Nelson	Examine the relationships between maternal anxiety and depressive symptoms and resources during their child's SCT.	23 M 23 children Mean age 8.1 yrs	Single-centered Longitudinal, time points: admission and 10 days post-SCT	STAI, CES-D, SPSI, SSS	The majority of mothers reported moderate to high anxiety levels and were at risk of developing depression. Most of the mothers indicated low or moderate satisfaction with the perceived social support. A relationship was found between a negative problem solving orientation and emotional responses.
10	2004, DuHamel	Investigate the role of cognitive processing in maternal adjustment to a life-threatening pediatric medial procedure.	91 M 91 children Range 9-19 yrs Mean age 8.7 yrs	Multi-centered Longitudinal, time points: 3 days prior to SCT and 3 months post-SCT	Structured interviews Fear network IES, BAI, BDI	Mothers' fear network, intrusions and avoidance played a primary role in their adjustment to their child's transplantation, during and after hospitalization. The article shows a cognitive processing model of psychological distress.
11	2004, Forinder	To get in-depth knowledge of the parents' situation during the SCT-process.	20 pairs of parents 20 children, no details given	Single-centered Longitudinal, time points: 4 to 8 yrs post transplant and 4 yrs after first time point	2 semi-structured qualitative interviews Jalowiec Coping Scale	The child's illness and treatment played an important role in the parents' lives for many years. Those parents who managed to put reason before emotion rated their coping as better. A sense of participation was also a useful coping strategy.
12	2004, Manne	Examine the prevalence and predictors of anxiety, depression and PTSD among mothers of children who underwent HSCT.	111 M 111 children Range 1-18 Mean age 8.2 yrs	Multi-centered Longitudinal, time points: at time of SCT and 18 months post-SCT	BAI, BDI, TSS, ISSB, WOC, COPE 18 month follow up: SCID-NP	Approximately 20% of mothers had clinically significant distress reactions. Mothers who were most at risk were younger and reported anxiety and depressive symptoms at the time of transplantation. The prevalence of depressive disorders declined after 18 months.
13	2004, Phipps	Examine changes in parental distress across the acute phase of SCT. Examine the relationship of parental distress to child distress during the SCT process.	136 M, 9 F, 6 others 136 children Range 1-20 yrs Mean age 8.9 yrs	Single-centered Longitudinal, 13 time points: weekly from week -1 to week +6, after that on a monthly basis through month +6	POMS, PSS, CBS, BASES-P, BASES-C	Parents demonstrated modest, but significant elevations in distress, particularly during the early period from admission through week +3. Parental distress was unrelated to child age, gender, diagnosis, or type of transplant, but was significantly related to parental SES. Moderate correlations were observed between measures of parent and child distress.
14	2004a, Rini	Examine the relation between life stress and basic beliefs about self-worth.	100 M Range 9 months-20 yrs Mean age 8 yrs	Multi-centered Longitudinal, time points: at admission and 1 year post-SCT	WAS, TSS, LES, SF36	Prior trauma and negative events were associated with basic beliefs during hospitalization and with changes in basic beliefs in the subsequent year, with distress mediating some of these relations. Relations were found between basic beliefs and maternal physical and mental functioning.
15	2004b, Rini	Examination of children's medical risk and mother's dispositional optimism and socio-demographic resources as predictors of benefit finding at admission (T1) and 6 months later (T2).	144 M 144 children Range 9 months-20 yrs Mean age 8 yrs	Multi-centered Longitudinal, time points: at admission and 6 months after the first time point	LOF, SF-36 (MHSS), 2 newly created items for benefit finding	Predictors of benefit finding differed systematically across assessments, with optimism and medial risk predicting benefit finding at both time points. Socio demographic resources predicted only T2 benefit finding. T1 benefit finding was positively associated with T2 adaptation only for mothers who scored high in optimism.
16	2005, Phipps	Examine psychosocial predictors of distress in parents of children undergoing SCT.	139 M, 9 F, 3 others 151 children Range 1-20 yrs Mean age 8.9 yrs	Single-centered Longitudinal, time points: weekly basis through week +6 post-SCT, monthly until +6	POMS, PSS, CBS, PIES, CBCL, FES, ISSB, WOC	Significant changes were observed in parental distress across the course of SCT, with relatively high levels of parental distress at admission, slightly increasing and peaking at week +2. Predictors of stress: prior parent and patient illness-related distress, pre-morbid child internalizing behavior problems, the family relationship dimensions of the family environment and parental avoidant coping behaviors.
17	2007, DuHamel	Investigate several potential antecedents of maternal fear appraisals: maternal optimism, recent negative life events, lifetime history of traumatic events, and medical characteristics.	140 M 140 children Range 9 months-19 yrs Mean age 8 yrs	Multi-centered Longitudinal, time points: at admission, 3 and 6 months post-SCT	LOT, LES, TSS, newly created items for fear appraisals	Lower optimism and a greater number of negative life events were independently associated with greater maternal fear appraisals. Lifetime history of trauma was not associated with maternal fear appraisals. Mothers' fear appraisals during their child's hospitalization were associated with their fear appraisals up to 6 months later.
18	2008, Vrijmoet-Wiersma	To assess levels of parenting stress compared to a norm group, to assess differences in parenting stress pre- and post-SCT and to assess the effect of parenting stress on parent-reported HRQoL of the child.	19 M 21 children Range 3-18 yrs Mean age 8 yrs	Single-centered Longitudinal, time points: two weeks before SCT and on average 10 months post-SCT	PSI	Compared to parents of healthy children, parenting stress was higher post-SCT. Post-SCT, parenting stress levels were higher than pre-SCT, both total parenting stress and the perceived demandingness of the child. High levels of parenting stress were predictive of poor parental ratings of child HRQoL post-SCT.

Explanation of abbreviations used

M= mothers; F= fathers; yrs = years
BAI = Beck Anxiety Inventory; BASES-P/C = Behavioral, Affective, and Somatic Experiences Scales – Parent version/Child version; BDI = Beck Depression Inventory; BSI = Brief Symptom Inventory; CBCL = Child Behavior Check List; CBS = Caregiver Burden Scale; CES-D = Center of Epidemiologic Studies Depression Scale; CSI = Cancer Support Inventory; DSI = Daily Stress Inventory; Faces III = Family Adaptability and Cohesion Evaluation Scale; FES = Family Environment Scale; HAS = Health Assessment Scale; IES = Impact of Events Scale; ISSB = Inventory for Socially Supportive Behaviors; LES = Life Experiences Survey; LOF = Life Orientation Test; LSCM = Lepore's Social Constraints Measure; MHSS-SF36 = Mental Health Summary Scale of the Short Form-36; PCL-C = Post-traumatic Symptom Disorder Checklist-Civilian version; PIES = Prior Illness Experience Scale; POQOLS = Pediatric Oncology Quality of Life Scale; POMS = Profile of Mood States; PSI = Parenting Stress Index; PSS = Perceived Stress Scale; SCID-NP = Structured Clinical Interview for DSM-IV, Non-Patient version; SSINT = Semi-structured Interview; SSS= Stress Support Scale; STAI = State-Trait Anxiety Inventory; TSS = Traumatic Stress Schedule; VABS = Vineland Adaptive Behavior Scales; WAS = World Assumptions Scale; WOC = Ways of Coping Checklist.

Results

Parental stress

Feelings of anxiety and depression, post-traumatic stress symptoms, disturbed or obsessive compulsive thinking and somatic complaints are the most common stress reactions parents report before, during and after SCT. Parental stress levels were reported to be higher than norm groups pre-transplant [3,32,36] and during the acute phase of the SCT, peaking at two to three weeks post-SCT [32]. The most common observation was that parental distress levels decreased over time after SCT [5,36], with the sharpest declines between 3-6 months post-transplant [28]. A recent study however showed that parents, on average 10 months post-SCT, reported higher parenting stress levels and, specifically, felt less competent as a parent [39]. In the longer term, 4 to 8 years post-SCT [8], many parents reported that their child's illness and subsequent treatment played an important role in their lives for years, ranging from parents still struggling on a daily basis to parents who put this ordeal more or less behind them. Studies comparing mothers and fathers are few in this area, since most studies included mothers only. Dermatis and Lesko [3] found higher levels of depression and phobic anxiety in mothers than in fathers.

Table 1 depicts the results of the 18 studies included in the review.

Risk and protective factors

Many factors have been identified to influence levels of parental distress. The most frequently identified risk factor for parental distress in the longer term is the manner in which the parent is able to handle stress during the acute phase. We grouped the risk factors into three clusters, based on a manual count of the determinants described in the included studies:

1. Disease factors, i.e. if the child had been transferred to the ICU and if it had had a higher number of hospitalizations 6 months post-SCT, parents reported more anxious and depressive symptoms [19]. Higher transplant risk was also associated with higher parental distress during the child's post hospital stage of recovery [6].
2. Psychological factors and parental coping: mother's appraisal of threat to her child's life [5,16], a greater number of negative life events [6], prior parent and patient experiences of distress associated with the child's illness [32], avoidance and intrusions [18,32], alcohol/substance abuse [18], perceived partner criticism [19], and an unsupportive family environment [32] all added to parental (i.e. mostly maternal) stress levels. Furthermore, mothers experiencing depressive symptoms during the acute SCT phase had a higher probability to be diagnosed with a psychiatric disorder 18 months post-SCT [17]. Parent distress has also been associated with child distress: child mood disturbance at admission was predictive of parent global distress over time [28].
3. Socio-demographic risk factors: younger maternal age [17] and lower social economical status (SES) [31] were associated with higher levels of stress throughout the SCT process.

Factors that did not appear to influence parental stress were: the age of the child, type of diagnosis, type of transplant, or the nature of the germ-free environment in which the child was placed [3,5,32]. Other objective medical aspects of the child's condition (i.e. disease risk, treatment course, and current disease status) than the factors mentioned under the first cluster (see above) were not related to parental stress levels or fear appraisals. It seems that the subjective appraisal of these factors is a better predictor of parental stress than the objective disease characteristics [6].

Protective factors or predictors of good psychological adaptation have been identified in terms of 'benefit finding' [35]: mothers who were optimistic by nature reported most benefit finding both during the acute SCT phase and 6 months later. Benefit finding is defined as 'an attempt to restore positive basic beliefs about the self and the world that have been challenged by a traumatic experience' [35,37]. Acceptance and humor as coping mechanisms were associated with reductions in maternal depressive symptoms [18] and 'putting reason before emotion' was identified as another coping mechanism associated with positive outcomes [8]. Lastly, a supportive family environment was associated with lower distress levels throughout the transplant process [32].

Discussion

Having a child undergo stem cell transplantation is a stressful event for any parent. Feelings of anxiety and depression, post-traumatic stress symptoms, disturbed or obsessive compulsive thinking, and somatic complaints are the most common stress reactions parents report before, during, and after SCT. The process of SCT is comprised of several phases and distress levels seem most elevated in the pre-SCT phase and the acute phase during hospitalization, but can stay elevated after discharge. Most parents return to healthy levels of psychosocial functioning 18 months post-SCT, but a subset of parents reports persistent symptoms years later in terms of anxiety, depressive feelings, and post-traumatic stress symptoms (arousal, avoidance, and recurring memories). Certain maternal coping strategies (e.g. acceptance, humor, putting reason before emotion, and having positive cognitive appraisals) during the acute phase of SCT have been identified as protective factors.

It has been shown that dispositional factors and prior experiences influence the way an individual appraises an event such as SCT and adjusts to it. For example, mothers who were more optimistic by nature reported lower fear appraisals at the time of their child's hospitalization for SCT [6] and later on, post-SCT. Optimism seems to be a more or less stable trait that can serve as a coping mechanism and a buffer [10], like positive reframing [18] and benefit finding [35]. Mothers who have encountered more negative life events in the months before their child's SCT reported more fear appraisals. It is hypothesized that traumatic events can trigger increased arousal, cognitions that one's life is difficult and traumatic [17], and a tendency to interpret potentially harmful new events more negatively [6]. Pre-existing factors should be assessed and used as starting points for psychosocial interventions.

The present review reveals potential areas of improvement in future research. In the 18 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 both the temporal course of a stressor [13] and to identify SCT-specific stressors. Post-traumatic stress reactions imply an existential challenge, but findings suggest that the complex situation of SCT involves several different stressors for the parents. 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 predictor variable and psychological stress as an outcome.

In most of the research in this area, no distinction was made between subclinical manifestations of parental distress versus psychiatric states. This is unfortunate, because in the latter approach, parents tend to be 'pathologized' [33] instead of assuming that the majority of families with a seriously ill child are competent and adaptively organized families, without any elevations in their a priori risk (as a group) for psychopathology [12]. Furthermore, it seems that in multidisciplinary SCT teams often there is no consensus of what is 'normal' distress or 'adequate coping' in this context. For example, young and inexperienced nurses can get worried about a parent in tears whereas an experienced social worker or psychologist may feel that a certain level or manifestation of distress is 'normal'. This issue points to the need for adequate psychosocial screening by pediatric psychologists pre-admission and during the acute phase of SCT, in order to target those parents most in need for psychosocial guidance and intervention.

Family functioning, an area of increasing importance in the pediatric psychology literature, is still understudied in parents of children undergoing SCT. The experience of fathers is another area of neglect. In many studies on parental stress of parents of pediatric cancer patients, higher stress levels have been found for mothers than for fathers [40], but recent research has shown that the experience of the child's illness often is as stressful for fathers as for mothers [26]. This finding points to the need to include fathers in future studies.

Strengths of the studies included in our review are the large number of longitudinal designs and multi-centered studies and the majority of studies with 90 participants or more. We have found only a minority of studies in which disease-or context-specific measures were used and this is unfortunate, because SCT is a highly complex treatment with very specific issues to deal with for parents. A combination of generic and disease-specific instruments could further our understanding of parental distress trajectories during the course of the SCT.

Conclusions

The authors conclude that the majority of parents of pediatric SCT patients are resilient, 18 months post-SCT and beyond. The most frequently identified risk factor for parental distress in the longer term is the manner in which the parent is able to handle stress during the acute phase. Parents (mostly mothers) with the most severe stress reactions and fear appraisals during the acute phase, continue to experience heightened levels of anxiety, depressive symptoms, and PTSS later on.

The next step is to develop and systematically examine feasible, limited, brief interventions for sub-clinical manifestations of psychological distress prior to and during the acute phase of SCT in parents who have been identified as 'risk' group. Follow-up care is needed for parents, especially when their child recovers and when control visits to the hospital become less frequent. Intervention research is a growing area in medical psychology and despite the many methodological challenges, efforts should be made to implement and evaluate existing intervention programs in this parent group. This can only be done through sound –SCT-specific- assessment, well-funded (inter)national cooperation, and well-developed study designs. Lastly, the ethical domain of conceiving designed children as donors is an area of interest that deserves be studied in the future, as well as the issue of stress in parents of children who need to undergo more than one stem cell transplantation or whose children suffer from more serious late effects, such as chronic graft-versus-host disease or other health problems.

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