Child and parental adaptation to pediatric oncology

Vrijmoet-Wiersma, J.

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Summary and discussion of the results

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Parental reactions to childhood cancer

The diagnosis and treatment of cancer in one's child can cause long-lasting psychological effects in a parent. The review article in Chapter 2 shows that feelings of uncertainty, anxiety, depressive symptoms and posttraumatic stress symptoms (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. This means that most parents are resilient even when confronted with the stressors of long and intensive cancer treatment, possible medical complications and the omnipresent fear of losing one's child. A subgroup of parents reports ongoing stress, even many years post-treatment. 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. The following risk factors for long lasting parental distress have been identified in previous research: pre-existing 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 [1,11]. Certain coping strategies, such as active problem solving, seeking social support and optimism can serve as protective factors [10,20].

One of the problems with pediatric psychology research is the variety of definitions of the core elements of the psychological stress process that are 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 [16]. Furthermore, the existing assessment instruments may fail to assess the specific problems that parents of cancer patients have to deal with [9]. There is a risk of "pathologizing" parental adaptation to childhood illness, which can have negative effects such as increased stigma and a de-emphasis on parents' daily functioning [25].

One way of addressing this problem is to carefully select assessment instruments that capture the broad ray of challenges parents of children with a life-threatening disease are confronted with. The study of the psychometric properties of the Dutch version of an American disease-related measure (Chapter 3) was an effort to describe parental stress in terms of the frequency of illness related events and the perceived difficulty of these events, thereby doing justice to the specificity of the experiences of parents. Parents of 107 children diagnosed less than 18 months ago with cancer in three different academic medical centers were included in this study. Eighty percent of the children were still on treatment. Risk factors for high disease-related stress were female gender, paternal age (older fathers reported significantly more distress than younger fathers) and child age (parents of younger children reported higher stress scores than parents of older children). Furthermore, as was expected, parents of children on treatment had significantly higher stress scores than parents whose children had completed treatment. Parents of children diagnosed more recently reported more stress than parents of children who were diagnosed longer ago. Surprisingly, the perceived difficulty of the stressors did not decline over time. This finding means that although the frequency of disease-related events is diminished with time, parents still perceive these events as difficult.

Alongside the PIP, three other questionnaires were administered, which measured state and trait anxiety, general stress and stress associated with parenting (parenting stress). A high correlation was found between scores on the PIP and on the first two measures, but a low correlation was found with the measure of parenting stress. This means that disease-related stress has an important overlap with anxiety (or worrying about one's child) and stress in general but less with stress associated with disciplining one's child. Disease-related stress however adds to clarify the specific stressors for parents of very ill children, especially during the period of active treatment: frequent hospital visits and admissions, waiting for news from the doctor, watching one's child undergo medical procedures can all add to the stress. These issues are not included in 'generic' questionnaires.

Parental reactions to stem cell transplantation (SCT)

As described in the review study in Chapter 4, the majority of parents of pediatric SCT patients appear to be resilient, 18 months post-SCT and beyond. 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. The most frequently identified risk factor for parental distress in the longer term is the way 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 [4,18]. This finding points to the significance of the state-trait model of anxiety in this context: parents with highest pre-existing anxiety (trait) and parents with the most severe anxiety reactions during the acute phase (state) are most at risk. Certain maternal coping strategies, such as acceptance, humor, putting reason before emotion and having positive cognitive appraisals during the acute phase have been identified as protective factors, e.g. [5].

The longitudinal study (Chapter 5) on parenting stress and child- and parent rated health related quality of life (HRQoL) demonstrated that 31 parents rated their children's HRQoL significantly lower both before and on average ten months after SCT than the 21 children themselves. Total parenting stress levels were significantly higher post-SCT than pre-SCT. An important predictor of proxy-rated HRQoL was found in the

Table 1. Overview of studies presented in this thesis

	Review article Stress reactions of parents of pediatric cancer patients Chapter 2	Cross sectional study PIP: Psychometric qualities, parents of children with cancer Chapter 3	Review article Stress reactions of parents of SCT patients Chapter 4	Longitudinal study Stress of parents and HRQoL of children pre-post-SCT Chapter 5	Cross sectional study Stress reactions of parents 5/10 years post-SCT Chapter 6	Cross sectional study Sequelae in children with LCH Chapter 7
Participants/ Number of articles	n = 67 articles	n = 100 mothers, n = 74 fathers	n = 18 articles	n = 19 mothers n = 12 fathers n = 21 children	n = 38 mothers n = 35 fathers	n = 24 children
Characteristics of the children	Children with cancer	Children diagnosed with cancer 1-18 moths ago (80% on treatment), mean age 9.6	Children who underwent SCT	Children who underwent SCT with heterogeneous diagnoses. Mean age at first assessment 11.0	Children who underwent SCT 5 or 10 years ago with miscellaneous diagnoses. Mean ages 13.4 and 16.6 y	Children with LCH. 21% CNS involvement, 30 % Diabetes Mellitus, 67% chemotherapy. Mean age 11.9
Assessment instruments	*n.a.*	PIP, STAI, PSI-SF, GHQ	*n.a.*	DUX 25 (child and parent form), PSI	PIP-SF, GHQ, CVS	DUX 25, LCH-DUX, WISC III subtests, CBCL/TRF/YSR
Outcome	Most emotional stress reactions were reported around diagnosis, i.e. anxiety, depressive symptoms, PTSS and uncertainty. Most parents seem resilient. A subset of parents reports continuing stress.	The Dutch PIP has adequate (test-retest) reliability and validity. Evidence was found for the four-factor model and the one-factor model.	Highest levels of parental stress are reported in the period preceding SCT and during the acute phase of SCT. Stress subsides in the majority of parents 3-6 months post-SCT.	Diminished HRQoL with regards to home functioning, not on other domains. Higher levels of parenting stress, on average 10 months post-SCT. Parents felt less competent as a parent, post-SCT.	40% mothers 5 years post-SCT reported elevated stress levels. High levels of perceived vulnerability: 94% after 5 years, 76% after 10 years for both mothers and fathers.	Lower physical HRQoL More internalizing problems Miscellaneous cognitive problems. Visual short term memory was affected in many children. Special education: 25%
Risk factors	- High levels of stress at diagnosis - Lower SES - Trait anxiety - Child behavior problems - Less perceived social support - Traumatic life events	- Parents of children on treatment - Less time since diagnosis - Higher paternal age - Lower child age	- High levels of stress during SCT - Avoidant coping - Lack of support - Pre-existing stress - Younger maternal age - Lower SES - ICU transfers - Child mood disturbance	- Older children (lower HRQoL) - High perceived child demandingness (higher parenting stress)	- Parents of children with a malignant disease - Higher paternal age - Being non-Dutch - Higher perceived vulnerability	- Older age - Children with Diabetes Insipidus, other CNS involvement and children who have had chemotherapy

child's demandingness perceived by the parents assessed before admittance and on average 10 months post-SCT. Perceived demandingness is a component of parenting stress. It is operationalized as e.g. 'my child demands more attention from me than I can give'. Post-SCT, significant associations were found between (parent-reported) child demandingness, parental health, role restriction (i.e. the manner in which a parent feels restricted by his or her child) and marital stress on the one hand and proxy-rated HRQoL on the other. Parents felt significantly less competent than parents of healthy children, post-SCT. This may indicate that post-SCT parents are faced with more stress concerning parent-child interaction and marital functioning than pre-SCT. The strain of caring for the child after discharge adds to the already present stressors of parents. Furthermore, the fear of relapse remains and makes parents more vulnerable to stress. This could be reflected in the lower rating of the domain 'home functioning' by parents and children, post-SCT. Ten months post-SCT, most children are back in school, the majority of parents have returned to their work place and visits to the clinic have diminished. However, our findings imply that families are still experiencing serious strain in a period of time when others expect them to pick up their old lives and move on. It may mean that parents lack both professional and social support in this phase.

Scant literature has been published on long-term parental stress post-SCT. The only earlier published report was a qualitative (interview-based) study on parents 4-8 years post-SCT [8]. The study described in Chapter 6 showed that general stress levels seemed to return to normal, 5 and 10 years post-SCT in 38 mothers and 35 fathers. Disease-related stress was relatively high 5 years post SCT, but was lower than the comparison group, 10 years post SCT. However, 5 years post-SCT, the percentage of mothers scoring above the cutoff point of general stress was significantly higher than in the reference group. Another finding of this study was that the majority of parents still perceived their child to be extremely vulnerable, both 5 and 10 years post-SCT. Parents from another cultural background reported higher stress scores than parents who were originally Dutch. Perceived vulnerability was higher in parents of children with a malignant disease, a finding that was expected, since these children are objectively more vulnerable. Risk of relapse, secondary malignancies and late effects are more common in this group of children than in children transplanted for a non-malignant disease and the parents of the first group have had more illness related experiences, which are of direct influence on parental distress [23].

Perceived vulnerability

Perceived vulnerability is an important predictor for disease-related parental stress, as was shown in the study on long-term psychosocial consequences of SCT in parents (Chapter 6). Whether high perceived vulnerability leads to overprotective parenting behavior has not been proven [32], but perceptions of vulnerability do influence child emotional adjustment (i.e. anxiety, depression) negatively [3]. Parental worry, communicated either implicitly or explicitly to a child, may convey that he/she is vulnerable or helpless and thus serves to increase anxiety and/or depression. Whether high perceptions of vulnerability cause psychological or HRQoL related problems in SCT survivors remains an area to be studied.

Assessment of parental stress

Assessment of parental stress reactions shortly after the diagnosis of a life-threatening illness of a child is important to identify those parents or families most in need. Disease-related and disease-specific measures can add important information about parental adaptation to stressful illness related situations. Furthermore, these instruments are more sensitive to change and can help to evaluate the effectiveness of interventions. The assessment measures that were selected for our studies on parental stress were based on availability in the Dutch language, data on reliability and validity and the frequency of use in other (inter)national studies. To study the impact of a life-threatening illness of a child on a parent from different angles, a variety of measures was used, i.e. measures of stress associated with parenting (Parental Stress Index, full and short form), state and trait anxiety (State Trait Anxiety Index), general stress or well-being (General Health Questionnaire) and a measure of perceived child vulnerability (Child Vulnerability Scale).

Because of a lack of disease-related instruments in Dutch, the Pediatric Inventory for Parents (PIP), a disease-related measure of parental stress was translated into Dutch. Results regarding the Dutch version of the PIP were satisfactory [33] and showed that it is possible to make a reliable and valid assessment of the frequency and perceived difficulty of various illness related situations. Reliability scores for the PIP Total scales and three of the four subscales (Medical care, Emotional distress and Role function) were adequate and confirmatory factor analyses showed acceptable fit for the four-factor model. PIP scores correlated strongly with a generic measure of anxiety and general psychological functioning. This means that disease-related distress, although it measures a different construct, can have considerable overlap with general well-being and anxiety. The added value of the PIP, however, is that the instrument assesses parental evaluations of their stress concerning specific disease-related situations, such as '*bringing my child to the hospital*' or '*being in the hospital during weekends and holidays'*. Scores on the PIP could be transformed into an individual 'stress profile', which could be used to tailor psychosocial support.

The low correlation of PIP scores with parenting stress scores suggests that stress resulting from difficulties disciplining and setting limits to one's child (parenting stress) is not the same as stress associated with having a child with a serious illness (parental stress). However, in various studies, e.g. [12], the PSI is used as a measure of parental stress instead of stress associated with parenting. This strategy might result in drawing the wrong conclusions about the stress reactions parents can have as a result of their child's illness.

Children's reactions to SCT and LCH

Children who have undergone SCT can have a compromised HRQoL, due to late effects and ongoing worries about a relapse or other complications. We found that children and adolescents in our study, when assessed on average 10 months after SCT, reported decreased HRQoL scores in the domains 'home functioning' and total HRQoL. Home functioning refers to items like 'A*t home, I often feel…*' or '*The things we do together at home, I find…'*. However, scores on the domains emotional, physical and social functioning were comparable to healthy peers. This finding suggests that children, even if they have not completely recovered physically post-SCT, are resilient and display 'hardiness'. They seem less bothered by the aftereffects of the SCT than their parents, possibly because they tend to live more in the 'here and now' and have the desire to return to their old lives. Adolescents are more at risk for a lowered HRQoL than younger children, a finding that is in line with most research in this area e.g. [6]. Older children might be more aware of the limitations and risks post-SCT. Problems related to a lowered 'home-functioning' could refer to the adolescents' desire to be more autonomous than their parents allow them to be. Perceived vulnerability could be an influencing factor and it would be interesting to study the relation between these concepts.

A phenomenon that needs to be taken into account when measuring HRQoL is 'response shift' [26], which refers to a change in the meaning of one's self-evaluation of a target construct as a result of: (a) a change in the respondent's internal standards of measurement; (b) a change in the respondent's values; or (c) a redefinition of the target construct (i.e. reconceptualization) [27]. Response shift could lead SCT- or cancer survivors to rate their HRQoL higher than expected, because they compare themselves to a period of severe suffering and may conclude that they are enjoying a good HRQoL at present [26].

Children with LCH seem to be affected more by the sequelae of their disease than children who underwent SCT, possibly due to detrimental effects on both cognitive and emotional functioning, resulting in lower HRQoL. Furthermore, LCH is an unpredictable illness in both time and severity, which makes it more difficult to cope with.

Assessment of HRQoL in children

HRQoL in pediatric patients can be assessed with generic and disease-related or disease-specific instruments, just like parental stress. To assess HRQoL in children with

Langerhans Cell Hystiocytosis (LCH), a disease-specific questionnaire was developed and used, the LCH DUX (see Chapter 7). This instrument rendered lower HRQoL scores than the generic HRQoL measure. A possible explanation is that children with a chronic illness, when asked in general how they think they are doing, tend to 'leave out' their illness and report relatively high HRQoL scores. It is unclear if this generic 'not including the illness process' happens unconsciously or results from repressive adaptation, as described in children with cancer [24] or if 'response shift' takes place. When children are approached directly about their illness experiences in a disease-specific questionnaire, they are forced to focus on difficulties they might come across because of their illness. Children with LCH appear to have not only a lowered HRQoL, but also cognitive and educational problems (one quarter of our study group is in special education) and more internalising emotional problems than their healthy peers.

Children and adolescents in our longitudinal study (Chapter 5) reported low HRQoL scores compared to a norm group of healthy peers 10 months post-SCT, especially with relation to functioning at home and physical functioning, but also in the total HRQoL score. In the other HRQoL areas (i.e. social functioning and emotional functioning), scores were comparable to the reference group. Parents rated their children's HRQoL significantly lower both pre- and post-SCT compared to the children themselves and compared to a norm group of healthy peers, a finding that has been reported in several other studies [2,7].

Limitations of the studies

Obtaining a sample size large enough to perform sufficient statistical power is a continuous challenge in pediatric oncology research in the Netherlands. The number of newly diagnosed children in our country is not the main problem, but the fact that the 500 newly diagnosed children per year are spread around the country in seven different academic hospitals. Although the number of children undergoing stem cell transplantation have increased substantially, the numbers undergoing transplant at any one center remain relatively small, a factor that has slowed psychosocial research considerably [22]. Multicenter research is to be preferred, but it is far from easy to organize, as experience taught us while undertaking the study on the psychometric qualities of the Pediatric Inventory for Parents [33]. One of the reasons was that all three Medical Ethical Boards of the participating centers needed to give approval, which was a time consuming process. The other reason was that practical matters were more difficult to tackle from a distance, for example handing the forms to the parents when they were in the hospital or outpatients' clinic was more reliable when the research assistant was present. If all Dutch pediatric cancer patients would be treated in one single center, it would be much easier to conduct large psychosocial studies in this population, in conjunction with medical treatment protocols and during visits to the outpatient's clinics and late effects clinics.

A problem associated with small study groups is the fact that parents of children with different cancer diagnoses often are analyzed together, which was also the case in the studies presented in this thesis. Whether this really is a problem is a matter of debate. As Stein and Jessop have stated, there is more variability within diagnostic groups than between them, hence a non-categorical approach is to be preferred [29,30]. On the other hand, it is conceivable to think that parenting a child with standard risk ALL would be different from parenting a child with a malignant brain tumor or a bone tumor, both during treatment and afterwards, when parents and children have to deal with late effects of treatment. Likewise, parents whose child undergoes SCT once are incomparable to parents whose child needs a second transplantation due to relapse or graft rejection. Parents of children with an underlying malignant disease have different pre-SCT illness experiences than parents of children with a non-malignant disease. These prior illness-related experiences during previous admissions have been found to be predictive of later SCT-related stress [23]. On the other hand, these 'experienced' parents are more used to being in a hospital and dealing with hospital staff and thus might be better equipped to face SCT-related stress during the acute phase, compared to parents without a history of frequent hospital admissions.

Another limitation or challenge is participation. Inviting parents to participate in studies on parental stress while they are still experiencing high levels of distress is not an easy job: parents feel overwhelmed with the burden placed on them by their child's illness and report that any additional request is perceived as 'too much'. Other parents report that they wanted to avoid experiencing intense feelings when confronted with questionnaires about their emotional reactions. This might mean that the parents with highest stress levels did not participate in the study. On the contrary, in studies on long-term psychosocial effects of cancer or stem cell transplantation, parents have answered that they felt 'it was all in the past' and no longer relevant to them. These two phenomena can reflect over- and/or underreporting of stress levels and give different patterns of generalization of the data.

There is a lack of reliable and valid disease-related or disease-specific assessment instruments available to medical and pediatric psychologists [16]. However, using disease-related instruments in clinical practice or research also has its disadvantages, because comparison of results is very difficult, if not impossible. Usually there is no reference group of parents of healthy children available and many instruments have only been used in one illness group. Hence, drawing conclusions based on the results can be difficult and disease-related or disease-specific instruments should always be used together with generic measures. The development of the PIP [31] has helped to identify and assess areas of strain for parents of seriously ill children. A disadvantage of this disease-related measure is that it seems less useful for the assessment of long-term parental stress, because the disease-related events do no longer occur and thus no longer seem relevant to parents. However, we learned that their worries and concerns do not disappear altogether. It is striking that the short form of the PIP (the PIP-SF, which is comprised of 15 items with the highest item-total correlations and the highest clinical relevance), which was used in the study in Chapter 6, contains items that appear to be centered mainly on worrying about the child and its future. Worrying seems to be an ongoing process and has found to be associated with parental perceptions of child vulnerability. The PIP-SF seems a promising screening tool for disease-related parental distress, but needs further studying.

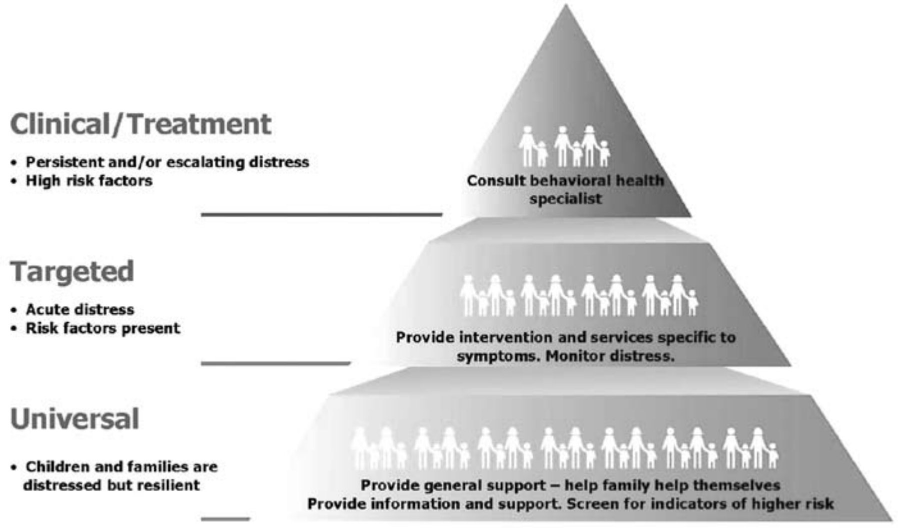
Lastly, data of all of the studies were collected as self-reports, which is the most conventional and convenient method of surveying groups. However, this method of data collection has certain limitations, for example: response style, which may involve either the reluctance to report distress or the tendency to over-report distress. Studies on self-reported distress and parenting stress could contain problems of overlapping concepts and possible underlying personality factors which can contribute to covariation in the assessed variables [19]. Another limitation of this study method has to do with parents from other cultural backgrounds. In all four studies in this thesis, the percentage of non-Dutch parents was low. Language problems seem to be a major cause, but they cannot explain the low rates of participating parents from other cultural backgrounds entirely. It seems that the way most pediatric psychological research is conducted, namely by means of pencil-and-paper self-report questionnaires, does not always match the expectations, preferences or abilities of all eligible parents. It is possible that parents from another cultural background, in which the group is more important than the individual person, perceive questionnaires about how an individual is feeling or coping as less relevant.

Practical implications and directions for future research

Assessment and indication of need

Adequate assessment forms a solid base for both clinical and research purposes. Finding the right assessment instruments to capture the unique experiences of parents of children with a life-threatening disease or children undergoing SCT is a matter of ongoing discussion among pediatric psychologists. All the while, we need to realize that we are dealing with parents who in majority are ‘normal’, psychologically healthy people, but have suddenly ended up in an abnormal situation. An inspiring model, developed by Anne Kazak, is the Pediatric Preventative Health Model (PPPHM), which builds on the assumption that the majority of families with a child with cancer are competent and adaptively organized families, without any elevations in their *a priori* risk (as a group) for psychopathology [15]. The PPPHM model (Figure 1, published with permission of the original author) divides all families into three categories, based on risk factors and needs.

Figure 1. Pediatric Preventative Health Model



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The term *Universal* is used for the largest group of families, who are seen as distressed, but resilient. This category consists of about 50% of all families who are helped with general support, information and preparation to invasive medical procedures by the child life specialist. This universal support is also to prevent more problems in the near future. The term *Targeted* is used to indicate those families at higher risk and in need of services specific to symptoms; about 35% of the families fall into this category. One of the goals of psychosocial services is that these parents do not ‘move up’ in the pyramid because their problems are accumulating. The smallest category on top of the pyramid (about 15% of the families) refers to *Clinical/Treatment*, to highlight those families at highest risk for persisting and escalating stress, who need to be referred to a behavioral health specialist (e.g. a pediatric psychologist, social worker or psychiatrist).

A model like the PPPHM adequately illustrates the fact that the majority of parents are able to cope with a cancer diagnosis and treatment in their child and do not need extensive psychosocial counseling. The model also has an economic advantage: costly and time consuming specialized psychosocial interventions will be offered only to those parents most in need. However, assessing the parents' level of risk and the specific strengths of the family is not an easy job. There is a need for psychometrically sound measures that are appropriate for use with pediatric health populations and for parents of children undergoing SCT or in treatment for cancer. Parents of children who need to undergo SCT could be screened in the weeks before admittance; parents of children newly diagnosed could be screened starting four weeks after diagnosis. The PIP could be used in its short form as one of the screening methods and state and trait anxiety should also be included as ingredients of screening batteries, since both pre-existing (trait) anxiety and acute anxious reactions (state) to diagnosis and treatment have been identified as risk factors for long-term parental stress. In the SCT-setting, the Prior Illness Experience Scale [23] could be used to assess parental and child experiences with cancer treatment before SCT. It has been shown to be predictive of parental stress during and after SCT. Furthermore, known risk factors such as traumatic life events, pre-existing psychopathology and a lack of support (see Figure 1) should be detected in each family early on in treatment by doctors, psychologists, social workers and nurses.

Timing of assessment seems to be a difficult issue, because, as our review study in Chapter 2 showed [34], studies vary considerably in the choice of time points. 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), at the completion of treatment or in the longer term. Pediatric psychology research would benefit from consensus on the optimal points in time to assess emotional reactions in parents following the diagnosis of cancer in their child. Assessment should preferably take place at one, six and 12 months after diagnosis, at the end of treatment and one and/or two years after the cessation of treatment, see Box 1. In this way, the comparison of results from research will be facilitated and patient and parent care will be enhanced. Assessment shortly after diagnosis provides important information on the initial reactions of parents. However, clinical practice has shown that assessment within four weeks after diagnosis is difficult, because parents are often too overwhelmed to take the time to fill in questionnaires. Assessment at six and twelve 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 (post-traumatic) stress levels.

Box 1. Timing of psychological assessment

Proposed time points for assessments
- 1 month post diagnosis
- 6 months post diagnosis
- 12 months post diagnosis
- End of treatment
- One/two years after cessation of treatment

In conclusion, it seems necessary to find (or develop) a reliable, easy to use screening tool for all parents and families of pediatric cancer patients that can be used at different time points and that is focused on known risk- and protective factors. Consensus is needed between all seven child cancer centers in the Netherlands about the content and implementation of such an instrument. A likely candidate for this purpose would be a Dutch version of the Psychosocial Assessment Tool (PAT) [13,21], which is a brief screening tool for psychosocial risk in families of children newly diagnosed with cancer. In addition, one could think about developing a semi-structured interview to capture parental stress and risk- and protective factors.

Intervention

Once the target families have been identified, evidence-based, brief and easy to use interventions need to be implemented in daily practice. Most individual counseling will be aimed at parents in the 'Clinical/Treatment' group, whereas parents in the 'Universal' or 'Targeted' group could also profit from group interventions and psycho-educational programs. The results of the study on long-term parental stress show the importance of early detection of parents at risk to prevent high levels of distress which were observed 5 years after SCT.

Hurdles like enrollment/response rate, prevention of drop out, timing and funding of interventions need to be taken collaboratively. Specifically, parents of children undergoing SCT seem to need more specific information and/or psychoeducation before admission, to reduce pre-transplant stress. Issues such as 'how to prepare for the lengthy and stressful admittance period', 'how to obtain adequate support' and 'what emotional reactions can be expected' need to be addressed. For this purpose, a DVD has been developed at the LUMC for parents of children who need to undergo SCT, with the aim to provide practical information and to serve as a coping model. The DVD was based on an existing film as part of an intervention study for parents of newly diagnosed children with cancer [14,28]. Issues before, during and after SCT are addressed in a group discussion of four families of SCT survivors. Parents receive the DVD approximately 5-6 weeks before admittance and are instructed to watch the DVD at home. They are requested to fill in a short questionnaire on feasibility, acceptability and usefulness of the DVD. For parents in the Universal group of the pyramid, watching the DVD could be sufficient preparation pre-SCT, while parents in the Targeted or Clinical group could perhaps be offered individually tailored sessions pre-SCT. Fragments of the DVD could be watched in these sessions with a psychologist or social worker and discussed with the parents.

During the acute phase of SCT (i.e. Phase II of the Model of Medical Traumatic Stress, see the Introduction Section), support seems to be sufficiently accessible for most of the parents, because they spend most of their time in the clinic and have the access to help and attention from both staff (nurses, physicians, social workers, child life specialists and psychologists) and other parents. The biggest challenge seems to be the period immediately after discharge post-SCT, when parents are facing the burden of caring for their (still ill) child at home and are expected to 'return to normal life'. We need to find a way to be more outreaching to parents in this stage, for example by organizing house visits by child life specialists and/or nurse-practitioners for every family in the first two weeks after discharge post-SCT. More follow-up care should be provided to those parents who suffer from longer term (posttraumatic) stress, i.e. the parents in the upper part of the triangle in Figure 1, by means of referral to local psychosocial health care specialists and/or support groups. The same holds true for parents of children with Langerhans Cell Hystiocytosis, who are largely 'out of sight' of hospital staff once treatment of their child is completed. These parents are burdened with both the care for a child with late effects of the disease and treatment and with their own uncertainty about the future. It would be worthwhile to study the effectiveness of an internet-based intervention and/or peer contact group for these parents.

Considering the adverse effects of childhood cancer treatment, there is a need for disease-specific psychosocial interventions for survivors of childhood cancer. A face-to-face psycho-educational group intervention was developed in the Netherlands, aimed at empowerment of survivors of childhood cancer by teaching disease-related coping skills. The program improved disease-related skills and psychosocial outcomes [17]. E-health developments should be considered as well. E-Health is defined as the delivery of health services and information through the internet and related technologies. It has developed considerably over the past years, with most e-health interventions focussing on adults, and to a lesser extent on children. For children however the use of the computer and internet is part of their daily life. Furthermore, adolescents seem to disclose more problems in online therapies compared to face-to-face interventions. For these reasons, E-Health applications are also of great value for children. E-Health in pediatric psychology is still lagging behind and must address numerous challenges. A first step has been made for childhood cancer survivors. The psycho-educational group intervention previously described has now been reshaped into a chat-group-intervention program and determination of effectiveness is in progress (www.opkoersonline.nl).

Many parents report their children to be vulnerable. Implications of this finding could be that all parents of children with oncological illnesses get psychoeducation about this phenomenon and are coached in setting appropriate limits to and having realistic expectations of their child. This could be done by pediatric psychologists or trained nurses. Parents who are considered to be at risk should be involved in more intensive and individualized intervention programs. Doctors are also important partners, because they can encourage parents to treat their child as normally as possible during visits to the late effects clinics. Psychosocial effects of parental beliefs of vulnerability on children surviving SCT need to be studied and brief routine assessment of both parents and children, even years post-SCT or LCH treatment, is recommended.

In conclusion, results in this thesis show that even though parents of children with cancer or children undergoing SCT as a group are resilient, pediatric psychologists are challenged to develop specific targeted interventions, based on a theoretically sound and easy to use assessment of risk and need. Doctors and nurses should be aware of parents who are at risk for heightened stress, because well-functioning parents are better able to tend to the needs of their children. Following up on parental well-being is important, not only during the active phase of treatment, but also in the long run: out of sight should not be out of mind!

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