Psychological adaptation to childhood cancer
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Chapter 7

General discussion
Resilience, a relatively common phenomenon in pediatric oncology, was taken as the point of departure for this thesis. The intriguing observation that most parents and children are able to either resist (children) or bounce back (parents) from adversity initiated a wish to look into underlying mechanisms.

Guided by the stress-coping model of Lazarus and Folkman [1] this thesis focused on the following concepts: caregiving stress (appraisal), dispositional optimism/pessimism (antecedent variables), goal adjustment and benefit finding (coping) (Figure 1). We were particularly interested in the longitudinal changes in these concepts during the first year after the diagnosis of cancer.

This final chapter reflects on the main findings of the studies described in this thesis and discusses methodological issues. The chapter ends with directions for future research and clinical practice.

Discussion of the main findings

Change in appraisal (caregiving stress)
Chapter 2 addressed the course of caregiving stress during the first year post-diagnosis in mothers of children recently diagnosed with cancer. Two competing models in the caregiving literature offer opposing predictions regarding the experience and impact of caregiving over time: the wear-and-tear model and the adaptation model [2]. The wear and tear model predicts that caregiving burden increases over time and deteriorates the caregiver’s physical and mental health. In contrast, the adaptation model states that caregiving burden is relatively
short-lived because new adaptation capacities will develop during the caregiving trajectory. The findings in Chapter 2 are more in line with the adaptation model than the wear and tear model. Our findings are similar to those observed in the context of adult oncology [3, 4], but they are contrary to evidence for the wear and tear model as found in the context of older adults [5, 6] and parents of children with complex health problems [7]. The fact that the adaptational pattern of caregiving best fitted the data can be explained by the relatively short duration of cancer caregiving. Although intense, the period of cancer caregiving is, compared to those of other populations (e.g., parents of children with a handicap), relatively short. This was especially true for the sample presented in Chapter 2, which consisted mainly of mothers of children with a relatively good prognosis.

In light of these findings, future researchers may want to examine which other factors contribute to the adaptive competence of mothers of children with cancer. Literature suggests that internal- (e.g., dispositional attributes) and external (e.g., social support) resources play a role in caregiving [8]. The examination of the direct and indirect pathways between these resources and outcomes (caregiving stress, parental functioning) and the interaction of these resources and caregiving stress on parental functioning are interesting venues for further research.

**Shifts in antecedent variables: dispositional optimism/pessimism**

The study described in Chapter 3 looked into the levels of dispositional optimism/pessimism in adolescents with cancer. The chapter supports previous research in adult oncology with its finding that children recently diagnosed with cancer report relative high levels of overall optimism. Further analyses showed that high overall optimism did not reflect high optimism, but low pessimism. Two points merit attention. First, our research shows that, in contrast to lay beliefs, that the (adolescent) patients do not see the world through rose colored glasses. In fact, rather than being overly positive, they seem to temper their negative expectations for the future (e.g., by not focusing on things that might go wrong). Second, our study suggests that a severe stressor such as a cancer diagnosis may trigger a shift in a personality trait which is generally assumed to be stable over time. Previous research has yielded similar results. Notable is the study by Phipps et al [9], which reported an increased prevalence of repressive adaptive style (also a disposition), in children recently diagnosed with cancer. Other research provided evidence for short-term reactive changes in dispositional optimism/pessimism in healthy adults confronted with major life events [10]. So our study as well as other studies supports the plasticity of the human system [11], for it seems that all means are mobilized when it comes to a confrontation with a severe stressor.
Our study also found that dispositional optimism and pessimism were related to different aspects of well-being. More specifically, optimism was related to positive and pessimism to negative aspects of well-being. These findings, which are in concordance with the adult oncology literature, provide further support for the bi-dimensionality of dispositional optimism in adolescents with cancer [12]. Future research should consider examining the relationship between dispositional optimism and positive indicators of well-being in more detail, especially since researchers have emphasized the adaptive significance of positive emotions in times of stress. It seems that emotions such as curiosity, confidence, joyfulness, and hope broaden one's scopes of attention and thinking which in turn may improve one's way of coping with stress [13-15].

**The emergence of meaning-focused coping: goal adjustment and benefit finding**

Meaning-focused coping refers to strategies that alleviate distress by either changing one's general orientation system (changing one's global beliefs, values, or goals to make them more congruent with the situation) or one's evaluation of the stressor [16, 17]. Chapters 4-6 of this thesis looked into two aspects of meaning-focused coping: goal adjustment and benefit finding. The studies presented in these chapters, which are the first to examine this topic in adolescents recently diagnosed with cancer, suggest that adolescents use adult-like strategies to cope with the challenges of cancer.

There is a large body of theoretical literature on goal adjustment (bringing one's goals more in line with one's current conditions), in which several goal adjustment strategies have been proposed. However, these strategies have never been empirically tested. In Chapter 4 a method to measure goal adjustment strategies in adolescents with cancer was presented. This method was used to calculate the strategies in a group of adolescents. The results showed that adolescents with cancer used several of the strategies in response to either goal disturbance or changed values. Putting the goal on hold was the most frequently used strategy. In Chapter 5, goals of adolescents with cancer were compared with those of healthy controls. At 3 months post-diagnosis, the adolescents with cancer showed an intrinsic goal orientation (i.e., reported more intrinsic than extrinsic goals, assigned higher ratings of importance to intrinsic goals but lower ratings of importance to extrinsic goals) and rated their goals on a lower level of abstraction than controls. The study described in Chapter 5 also looked into the goals of the adolescents with cancer over time. Findings showed that the goal orientation and goal abstraction level of the adolescents with cancer remained stable throughout the year following the diagnosis.

The findings presented in Chapters 4 and 5 support previous research in healthy adolescents [18], which has suggested that adolescents possess the cognitive skills for intentional self-regulation (i.e., that they are able to use the
flexible structure of the goal system to make their goals more in line with current
conditions and to find other paths to higher order goals when their goals become
blocked). This may have important implications. Directing attention (temporarily)
away from goals that cannot be achieved is important, not only for the preservation
of well-being but also for the conservation of resources (time, energy) that are
needed for the pursuit of more important goals (e.g., to recover from cancer).
However, this does not necessarily mean that other goals have to be abandoned.
Scaling back allows the adolescent to remain involved in the pursuit of a higher-
order goal, but at a slower pace. A goal on hold is still important, but active goal
pursuit is postponed until normal life can be resumed. Such mechanisms are
important, especially for the population studied in this thesis (i.e., adolescents
with a relatively good prognosis).

Treatment for childhood cancer is temporary and adolescents with a good
prognosis have to prepare for their life after the completion of cancer treatment.
Future research may want look into goal processes in survivors in order to
determine what will happen with the aforementioned goal adjustment strategies
in the long-term. Furthermore, since goal adjustment has been identified as one
of the pathways toward resilience [19] future research might focus on goal
adjustment in relation to well-being.

Benefit finding, which refers to people’s belief that adversity has brought them
something good, was the focus of Chapter 6. Benefit finding can be considered
as a meaning-focused coping strategy since it involves a change in the meaning
of a stressful situation (i.e., the appraised meaning of the stressor is transformed
to a more positive one [17]). There is much controversy in the literature regarding
the trajectory of benefit finding (e.g., when it emerges, how it evolves over time).
Based on the thought that benefit finding is more likely to occur with increasing
time since the adverse event [20], most research on this topic has been carried
out in cancer survivors. This thesis showed, however, that benefit finding is also
possible with less time elapsed since diagnosis. This is consistent with findings
of the few studies in adult oncology that examined benefit finding in the period
shortly after diagnosis [21-23].

There is much discussion in the literature about the nature of benefit finding,
i.e., whether it should be regarded as a positive illusion, self-deception, a
reflection of the current circumstances (e.g., getting more attention from family
and friends, receiving perks) or as enduring positive change (i.e., transformation)
[20, 24-26]. This raises the question whether benefit finding proximal to
diagnosis represents the same phenomenon as benefit finding observed later
on. More research is needed to answer this question. Furthermore, despite the
fact that this thesis was unique in that it studied benefit finding over time,
benefit finding is preferably studied over a still longer period of time, particularly
because benefit finding in pediatric populations has been hypothesized to dissipate over time [27]. Researchers have identified benefit finding as one of the potential mechanisms underlying resilience in children with cancer [28]. Therefore the longitudinal relationship between benefit finding and well-being offers a suggestion for future research.

**Temporal dynamics**

One of the conclusions of this thesis is that adaptation and adaptive mechanisms start to emerge relatively early in the cancer trajectory. The results presented in Chapter 2 showed a rapid decrease in caregiving stress together with an improvement of mothers’ functioning early in the disease process (during the first 3 months post-diagnosis). This suggests that, despite the fact that most stressors of caregiving tend to continue over the first year post-diagnosis, mothers of children with cancer seem to get used to the demands of caregiving in a relatively short time. The findings presented in Chapters 3-6 showed that the adaptive mechanisms (shifts in dispositional pessimism, goal adjustment, benefit finding) in the adolescents with cancer occurred relatively early in the disease trajectory (3-6 months post-diagnosis). Thereafter, dispositional pessimism, goal orientation/abstraction level, and benefit finding did not change during the first year. This fits in with other research in oncology which suggested that adaptive mechanisms may appear within a very short interval (days, weeks) following the diagnosis, remaining stable thereafter for some time [29, 30].

**Towards a synthesis**

The studies presented in this thesis raise several questions. How to interpret their findings? Why do dispositions seem to change? Why are goal adjustments made? What brought the adolescents to report positive consequences about their disease while they were still in the midst of adversity? And why does this occur so early in the disease trajectory? The overarching principle of homeostasis may serve as an explanation. This principle forms the basis of the stress-coping model by Lazarus and Folkman (1984) and other theories such as the set-point theory [31, 32], Carver and Scheier’s cybernetic control model [33], subjective well-being homeostasis theory [34], and the meaning making model by Park and Folkman [16]. At the heart of the homeostatic principle lies the idea that human beings strive towards a state of physical and psychological homeostasis (i.e., to keep their physiological and mental state at a steady level or equilibrium [35]). When the homeostasis is disturbed by, for instance cancer-related stressors, attempts are made to restore the homeostasis [34]. Homeostatic stabilizing forces, behavioral or cognitive, often act in conjunction with other (non-conscious) processes, such as attention (e.g., attention to threat, counter-regulation) [36]. Research shows that life-threatening danger suppresses
attention to threat [37, 38], and this may help explain why adolescents who were relatively recently diagnosed with cancer scored so low on dispositional pessimism. Counter-regulation, the mechanism in which attention automatically orients towards stimuli that are opposite in valence to the current affective state [39], may have played a role in benefit finding (i.e., a negative affective state is supposed to increase the salience of positive information which can be used for benefit finding [40]). The findings with regard to dispositional pessimism and benefit finding could be considered as homeostatic restoring processes. Goal adjustment is another mechanism that can help to restore homeostasis. Goal adjustment is based on a perceived discrepancy between a preferred (goal) and the actual state. This sets into motion a discrepancy-reducing process [33, 41, 42], such as the scaling back of goals or reprioritizing.

Homeostatic theory may also explain why the adaptive mechanisms emerged relative early in the cancer trajectory since a disruption of homeostasis will set into motion homeostatic stabilizing forces to restore the homeostatic state.

Methodological considerations
The strengths and limitations of the studies are described in more detail in the separate chapters of the thesis. A short synopsis of the main issues is presented below.

A clear strength of this thesis is its conceptual and longitudinal approach. Despite the fact that concepts (e.g., dispositional optimism, benefit finding) covered in this thesis have been previously identified as key areas for research on resilience in children with cancer [28], the concepts had yet to be explored in the context of pediatric oncology. Moreover, with regard to some of these concepts we adopted novel approaches such as the bidimensional investigation of dispositional optimism and the examination of situational goal adjustment rather than goal adjustment tendencies. Examining the concepts over time in a consecutive sample of children recently diagnosed with cancer makes the thesis rather unique. Another strength concerns the inclusion of a control group. Since we were not able to assess the adolescents before the onset of cancer, data of healthy peers (serving as a proxy for the adolescent’s pre-illness situation) were used to get at least some insight into the potential effect of cancer on aspects of adolescents’ stress-coping process.

Major limitations involved the sample size and timing of measurement. Because of the small number of adolescents the influence of individual difference variables could not be explored. As a consequence, the conclusions may not hold true for all adolescents, for instance adolescents with a poor prognosis). Second, the assessment of some of the concepts in the study started rather late namely at 3-6 months post-diagnosis. Literature suggests, however, that mechanisms underlying adjustment may start to emerge more proximal to diagnosis. This
could be within days or weeks post-diagnosis) [9, 30]. Furthermore, this thesis focused on main effects only, despite evidence of mediation and moderation effects in other contexts [43-46]. Last, this thesis solely investigated unidirectional relationships, while it is likely that reciprocal relations of causality exist [14, 47].

Future research

Suggestions for future work based on the limitations of the present thesis
To overcome one of the methodological limitations described above, the recruitment of larger samples is an important consideration for future research. This may become possible with the advent of the Dutch national Princess Maxima Center for Pediatric Oncology where all research on pediatric oncology in the Netherlands will be put together. Large samples allow for more sophisticated methods of analysis (e.g., latent growth mixture modeling), which are better suited for the examination of (potential) heterogeneity in stress-coping responses.

Another methodological issue that warrants further consideration is the timing of the first assessment. Participation in large scale population based cohort studies (e.g., Lifelines) may help overcome lack of premorbid data on variables of interest (e.g., on dispositional optimism, mental and physical health) which is a common issue in healthcare research. Apart from this, the immediate post-diagnosis period seems to mark the beginning of adaptation. For a better understanding, future research should look into this period in more detail. Ideally the measurement of relevant concepts should start within days of the cancer diagnosis and should be frequently monitored thereafter.

Furthermore, more research is needed to properly address the complex pathways between the elements of the stress-coping process. Interesting avenues for further research include the exploration of pathways from dispositional optimism/pessimism to well-being via coping [48], the association between dispositional optimism and goal engagement via goal prioritization [49] and the relationship between benefit finding and goal adjustment [19]. Furthermore, future research may consider the examination of the bidirectional relationships of the dynamic stress-coping process.

Other suggestions for future research
Because research has shown that long-term survivors of cancer during childhood only minimally differ from healthy peers [39-41] future research should consider the examination of adaptation mechanisms over a longer period of time. Interesting research questions are whether benefit finding continues in the
survival phase and whether dispositional pessimism returns to a level comparable with those of healthy children).

Moreover, there is much more work to be done in the context of parental adaptation. For example it would be interesting to know whether the mechanisms investigated in this thesis (e.g., shifts in dispositional optimism/pessimism, goal adjustment) also occur in parents of children with cancer. To our knowledge, this has not yet been investigated. Another area that deserves further exploration is the parent-child dyad. Possible venues for future research include dyadic benefit finding, the association between child benefit finding and parental adjustment and the relationship between parenting and goal adjustment (child).

Furthermore, examining the concepts of this thesis in other pediatric health conditions (and comparing the results between the conditions) warrants serious consideration.

Last, researchers may consider international collaboration. Not only because this entails the possibility to generate larger amounts of data, but also because it allows for generality checks (i.e., checking the universality or generality of one’s hypotheses and questions) and cross-cultural interpretation.

**Clinical implications**
The results of this thesis may be used to inform healthcare professionals about factors and mechanisms related to resilience and wellness, which may help them better understand patients’ and parents’ responses. It may also affect their professional role, as it can help them emphasize the importance of paying attention, supporting and bolstering patients'/parents' natural homeostatic stabilizing responses and strengths.

The fact that most families (child and parents) adapt relatively well to the stressors of childhood cancer does not mean that they do not need support during the cancer trajectory, in particular because some of the concepts covered in the thesis (caregiving stress, goal setting) are amenable to intervention. I agree with suggestions made by the Pediatric Psychosocial Preventative Health Model (PPPHM) [53] that care with a preventive focus (universal care) should be available for all families.

**Universal care**
Universal care focuses on tools and interventions that help children and their parents to use their natural coping mechanisms and strengths. These may include education, practical tools (e.g., a fact sheet with practical tips on managing side-effects or a copy of the treatment schedule), skills training, help with connections to needed resources, 24/7 availability of a nurse on call, systematic monitoring and risk assessment.
Based on the idea that individuals are embedded in multiple interconnected systems [54], healthcare professionals should take the child’s broader context (e.g., friendships, school, romantic relationships) into account. Systematic monitoring of health-related quality of life (HRQOL) (e.g., by means of the KLIK method developed by the Emma Children’s Hospital AMC in Amsterdam; www.hetklikt.nu) and patient’s progress toward personal meaningful goals may help detect and discuss problems in important life domains in an early stage. This in turn may guide the delivery of tailored interventions, for instance help with realistic goal setting, help with finding goal relevant means, and help with aligning personal goals with treatment goals.

A systematic evaluation of parents’ experiences with providing care can help in finding solutions for parental problems before they become severe. Appropriate interventions (e.g., help with financial difficulties, tools to discuss the illness with the child, facilitation of communication with the doctors) may not only contribute to a reduction of caregiving stress, but these interventions will also better prepare parents for their task of providing care to their child with cancer.

Risk assessment is a vital aspect of universal care. Despite the fact that the majority of children (and parents) seem to adjust relatively well (over time), a subset of children and parents may need extra help. Risk screening, for instance by means of the Pediatric Assessment Tool (PAT) [55, 56], could help identify families at risk of elevated/prolonged distress and deteriorated functioning. Targeted care, such as treatment by a psychologist which starts early on in the disease trajectory, may help prevent serious problems (i.e., PTSS) later on.

The organization of ‘universal care’
Effective support of families confronted with serious pediatric illnesses requires good multidisciplinary collaboration and networking. Case management [57] may be an appropriate model for the organization of care to children with cancer (and to children with other pediatric conditions). The central figure in case management is the case manager (often an experienced nurse or nurse practitioner) whose main task is to link patients to appropriate providers of care. As a member of the multidisciplinary pediatric oncology team the case-manager coordinates the patient’s care throughout the diagnosis, treatment and post-treatment phases. The advantage of case management is that one person is directing or overseeing the patient’s care. When appropriately trained, the case manager would be (best) suited to screen the child and his/her parents for problems. Depending on the type of problem and the opinion of the multidisciplinary team, the case manager may refer the child/parent to a specialized professional (e.g., educational consultant, psychologist) or solve the problem directly by speeding up an appointment with the specialist or by helping with the integration of rehabilitation goals in the activities of daily life.
Concluding remarks

This thesis, which aimed to get more insight into mechanisms underlying adaptive functioning of children and parents confronted with childhood cancer, suggests that a cancer diagnosis sets into motion a range of dynamic responses. In the literature, these responses (e.g., change in appraisal, shift in dispositions, meaning focused coping) are associated with better adjustment, and this may explain the adaptive competence of children and parents in pediatric oncology. It is evident that further research is required on the temporal dynamics of these responses and the pathways through which these responses affect adjustment. A better understanding of the mechanisms of resilience provides healthcare workers with tools to help the subgroup of children/parents who adjust less well to cancer.


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