FACULTY OF PSYCHOLOGY
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Families and childhood cancer: A study on crisis, resources and adaptation

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DANKWOORD

De voorbije jaren zag ik vele collega's indienen en verdedigen. De vraag "Wanneer is het jouw beurt?" werd vaak gesteld. Maar steeds leek het alsof ik nog een zee van tijd had. Maar kijk, na meer dan 7 jaar is het nu ook mijn beurt. Dat vraagt om een terugblik en een welgemeende *Dank je wel* aan allen die deze tijd mee onvergetelijk maakten.

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"All of the people around us they say
Can they be that close
Just let me state for the record
We're giving love in a family dose"
(Sister Sledge)

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GENERAL INTRODUCTION

This doctoral dissertation aims to gain insight into the impacts of childhood cancer on the ill child, his/her family members, and family (sub)system(s). In this general introduction, we will first discuss childhood cancer and its psychosocial consequences for the family as a whole, its members (patient, parents, siblings), and the parents' intimate relationship. Current gaps in our knowledge and limitations of the current literature will be identified. The specific aims of this doctoral dissertation, focused upon remediation of those issues, will then be presented as well as the theoretical framework guiding this dissertation, the double ABCX-model. Finally, we will provide an overview of the chapters to follow.

An Introduction to Childhood Cancer

What is Childhood Cancer?

Childhood cancer refers to a collection of related diseases that arise from abnormal, uncontrolled cell division. In normal circumstances, cell division and apoptosis (i.e., programmed cell death) are well regulated and balanced. However, in the case of cancer, cell growth proliferates and more new cells are formed than old cells demolished. This uncontrolled growth of cells can create a benign or a malignant (cancerous) tumor (National Cancer Institute, 2015a).

There are four differences between benign and malignant tumors. First, malignant tumors are characterized by infiltrative growth (i.e., cells spreading into surrounding tissues) while benign tumors are not. Second, the boundaries of malignant tumors are irregular and blurred, unlike those of benign tumors that have sharp, distinct anatomical boundaries. Third, the

degree of differentiation is lower in malignant than benign tumors. Fourth, differences exist in nuclear atypia, mitotic activity, and necrosis: in the case of a malignant tumor, cells are more distorted and no longer look like the original cells (nuclear atypia), grow faster (mitotic activity), and show a higher degree of cell death (necrosis) compared to benign tumors. There are two additional issues to note: (a) not all cancers involve tumors (e.g., pediatric blood cancer) and (b) sometimes it is impossible to distinguish between benign and malignant tumors. In that case, the term "borderline laesies" is used (Bosmans & Van Krieken, 2005).

Cancers can also "metastasize" and spread to new areas of the body. This occurs when cancer cells break away from the original (primary) tumor, travel through the lymph system or the blood, and form a new tumor in other tissues or organs of the body (American Society of Clinical Oncology, 2019). In the case of metastasized cancer, prognosis is worse and treatment is more intense.

Incidence, Mortality and Childhood Cancer Types

Worldwide, in 2018, an estimated 18.1 million people received a new cancer diagnosis and 9.6 million died of the disease (Naghavi et al., 2017; National Cancer Institute, nd.). Childhood cancer accounts for 1% of all cancers (American Cancer Society, 2019a), with 300,000 children across the globe receiving a cancer diagnosis every year (American Childhood Cancer Organization, nd.) and 341 children in Belgium (Belgian Cancer Registry, 2020). Although these numbers might seem to imply that this illness is rare and irrelevant for children, it is the leading medical cause of death for children and adolescents worldwide (Belgian Cancer Registry, 2020).

Fortunately, the 5-year survival rates for children with cancer increased in the last decades from 58% (mid-1970s) to 84/86% (USA 2019 and Belgium 2017, respectively), due to better knowledge of the disease, advances in medical treatment protocols, and delivery of treatment through

specialized multidisciplinary care teams (American Cancer Society, 2019a; Belgian Cancer Registry, 2020). Still, the survival rates vary depending on the type of cancer and other factors, such as treatment adherence.

The most common cancer diagnoses in children are leukemias (28% of all cancer diagnoses in children) and brain and spinal cord tumors (26%), with other diagnoses each affecting less than 10% of children with cancer (e.g., neuroblastoma – 6%, non-Hodgkin lymphoma – 5%; Wilms tumor – 5%, Hodgkin lymphoma – 3%; and rhabdomyosarcoma – 3%; American Cancer Society, 2019b). In this dissertation, to increase homogeneity of the sample, we focused on leukemia and Non-Hodgkin lymphoma. These types of cancer have similar, curative treatment protocols and together they represent one third of all cancer diagnoses in children.

Leukemia. Leukemia is characterized by disturbed production of white blood cells (WBc) in bone marrow. Large numbers of abnormal WBc crowd the bone marrow and obstruct the production of normal blood cells. Then, abnormal WBc spread through blood circulation and invade the lymphatic system and sometimes vital organs. There are four types of leukemia, based on two characteristics. First, the type of WBc impacted: lymphocytes (lymphatic leukemia) or granulocytes (myeloid leukemia). Second, the speed of proliferation of the abnormal blood cells: fast (acute leukemia) or slow (chronic leukemia). Combining these two characterizations, the four types of leukemia are acute lymphatic leukemia (ALL), acute myeloid leukemia (AML), chronic lymphatic leukemia (CLL), and chronic myeloid leukemia (CML) (Lardon, 2011).

With regard to the age of onset, ALL mainly occurs in children under the age of 14 and young adults, AML in adults, and the chronic types of leukemia (CLL and CML) mainly occur in middle-aged people. The 5-year survival rates for children with leukemia are 60-80% for CML, 65-70% for AML), and 90 % for ALL (Kom op tegen Kanker, 2018).

Non-Hodgkin lymphoma. Hodgkin lymphoma and non-Hodgkin lymphoma are forms of cancer that start in the lymphatic system. The

lymphatic system consists of lymph tissue and lymph nodes and plays an important role in the body's defense against pathogens. Specifically, lymphocytes – located in the lymph nodes – are white blood cells that defend the body against foreign substances, such as bacteria and viruses. When these lymphocytes develop abnormally and proliferate uncontrollably, the term lymph node cancer or "lymphoma" is used. The primary difference between Hodgkin and non-Hodgkin lymphoma is the type of lymphocyte that is affected. Hodgkin lymphoma is marked by the presence of Reed-Sternberg cells, while in non-Hodgkin lymphoma, these cells are not present.

Non-Hodgkin lymphoma has four stages. In the first stage, the cancer is found in a single region or organ, usually one lymph node and its surrounding area. When the cancer is found in two or more lymph node regions on the same side of the diaphragm, either above or below, the patient is in stage two. In stage three, the cancer is located in gland areas above *and* below the diaphragm. If the disease is in its final stage, the cancer has spread to other organs outside the lymph system, such as the lungs, liver, bone marrow or skin (Cancer Treatment Centers of America, 2020; Murphy et al., 1989).

The risk of non-Hodgkin lymphoma in children increases with age. It can occur at any age, but it is uncommon in children younger than three years old (American Cancer Society, 2019). The 5-year survival rate for children with non-Hodgkin lymphoma is 80-90% (American Cancer Society, 2017).

Treatment of Childhood Leukemia and non-Hodgkin Lymphoma

Childhood cancer treatment is aimed at completely removing the cancer (the tumor and/or circulating cancer cells) and any metastases, without damaging normal tissue (= curative treatment). In addition to curative treatments, there are palliative treatments. Palliative treatments are focused on improving quality of life, decreasing pain and other cancer-related symptoms, and prolonging the life of the child when cure is no longer possible.

Curative treatments for childhood leukemia and non-Hodgkin lymphoma generally involve one or more of the following treatments: radiotherapy, chemotherapy, and stem cell transplantation. The choice of treatment depends on several factors, such as the type of cancer, the extent of the disease, the age and the general condition of the patient, and the existence of other medical conditions.

Radiotherapy. In radiotherapy, ionizing rays are used to defeat the cancer. The radiation damages the genetic material of the cancer cell, which leads to cell death. Radiotherapy can take place in both curative treatment and palliative treatment. During or after radiotherapy, a number of side effects may occur depending on the place of radiation, the amount of radiation, and the patient's sensitivity. Possible side effects are nausea, diarrhea, fatigue, skin rash, and hair loss (Bracke et al., 2011).

Chemotherapy. Chemotherapy medicines have a cytostatic or cytotoxic effect on the cancer cells. This means that these medicines interrupt cancer cell division (cytostatic effect) or directly kill them (cytotoxic effect). Often a combination of chemotherapies are used, which are administered every three to four weeks using an injection or intravenous drip. Chemotherapy often has side effects, such as nausea, vomiting, fatigue, hair loss, and increased susceptibility to infections (Bracke et al., 2011).

Stem Cell Transplantation. A stem cell transplant is a procedure that involves destroying the stem cells within the bone marrow that are producing cancer cells and “transplanting” them with healthy stem cells. Patients first have their stem cells destroyed by very high doses of radiotherapy or chemotherapy. The healthy blood-forming stem cells are then administered through a needle in the vein. There are two types of transplantation: autologous transplantation (i.e., the stem cells come from the patient, are altered, and transplanted) or allogeneic transplantation (i.e., the stem cells are provided by a donor, who may be a blood relative or an anonymous donor). Allogeneic transplantation has a greater chance of severe side effects such as Graft-versus-host (GVH) disease. GVH occurs when the WBc generated from

the donor stem cells (the graft) recognize the cells in the patient's body (the host) as foreign and attack them. This can cause damage to the skin, liver, intestines, other organs, or even death (National Cancer Institute, 2015b).

State of the Art: What is the Current Evidence on the Psychosocial Consequences of Childhood Cancer?

The pediatric cancer literature illustrates how the turmoil and disruption created by childhood cancer reaches beyond the ill child and impacts other family members (parents, siblings), the family as a whole, and the parents' intimate relationship as well.

More specifically, the *child with cancer* often experiences a range of difficulties, such as pain, fatigue, reduced immunity, anxiety, and uncertainty (Voûte et al., 1997). *Parents* of children diagnosed with cancer often report significantly higher levels of distress, posttraumatic stress symptoms, parental conflict, emotional problems (anxiety, feelings of depression), and physical complaints (insomnia, fatigue), compared to parents with healthy children (Pai et al., 2007). *Siblings* often report poor quality of life in several domains (emotional, family, and social; Alderfer et al., 2010), and negative emotional reactions (shock, fear, worry, sadness, anger, and guilt). School-aged siblings often display more absenteeism and problems at school as compared to peers (Alderfer et al., 2010; Long et al., 2018).

Few studies have documented the impacts of childhood cancer on the *family as a whole* (see Pai et al., 2007 for an overview). Overall, quantitative studies revealed that most families function within normative ranges (e.g., adaptability, Pai et al., 2007; family support, Brown et al., 2003) or even report improved functioning in some realms (e.g., cohesion, Cornman, 1993). Qualitative studies, however, indicated a loss of normal family life (Bjork et al., 2009; Clarke-Steffen, 1997) and troubles balancing multiple family needs including those of siblings (Bjork et al., 2009).

Similarly few studies have examined the impacts of childhood cancer on family subsystems, like the *couple subsystem*. The few that are available (e.g., Hoekstra-Weebers et al., 1998; Patistea et al., 2000) reveal that although most couples adjust well to the crisis of childhood cancer in domains such as emotional closeness, couple support, and marital satisfaction, most couples do experience difficulties in the domains of sexual intimacy and conflict, both during and after treatment.

It should be noted that there is considerable *variability and inconsistency in findings* reported in the research literature regarding the individual adaptation of children diagnosed with cancer and their family members, the adaptation of the family, and the adaptation of the caregiver couple relationship. While most show resilience over time in many domains, some report adjustment problems after diagnosis. As a consequence, a growing number of studies has tried to explain why some family members, families, and couples adjust better than others, investigating the role of potential *resources*. These resources may arise at different levels within the social ecology of childhood: the individual level (e.g., personality; Erickson & Steiner, 2001), the intrafamilial level (e.g., family cohesion; Alderfer et al., 2009), and the contextual level (e.g., network support; Corey et al., 2008).

Limitations of the Current Literature and Research Aims of the Dissertation

To date, the issues of how childhood cancer affects the *family as a whole* and the *parents' intimate relationship* have received inadequate research attention. Also, there is little research on how childhood cancer affects family members other than the ill child and his or her parents, namely the *siblings* of the child with cancer. Furthermore, much of the existing research regarding the effects of childhood cancer has relied on cross-sectional rather than longitudinal designs, providing limited information about *patterns*

of adjustment across time. Finally, in the majority of existing pediatric cancer studies, no *theoretical frameworks* are specified as guiding the research questions or the selection of variables. Failure to use theoretical frameworks jeopardizes progression of the field as advances cannot be made if theories go untested and unrevised.

This dissertation arose from a series of questions: How do family members, families and couples respond to childhood cancer, and how do these responses change over time? Why do the strains of childhood cancer bring some families closer together and break other families apart? What resources help some families to cope better with the illness compared to others? And which theoretical framework can help us to best understand adaptation post-diagnosis and the existing variability in outcomes?

As answers on these questions are lacking, the *main aims* of the present dissertation were to examine (a) the short-term and long-term effects of childhood cancer on families, its members (patient, parents, and siblings) and the parents' intimate relationship and (b) the resources – situated at the individual level, intrafamilial level, and contextual level – that may help families and their members to recover from crisis and to adapt to the stressful circumstances resulting from childhood cancer. In addition, given our focus on all family members and the lack of research on and knowledge about siblings, the present dissertation will give special attention to (c) how siblings experience the illness and its treatment, and their everyday family life post-diagnosis.

The conceptual framework underlying the current research project is the *double ABCX model* (Fig.1; McCubbin & Patterson, 1983), a widely used family stress model (Becvar, 2013). This model is particularly useful for our research aims for three reasons. First, it models both *short-term and long-term* adjustment of families dealing with stress, thereby acknowledging the fact that individual, family, and couple responses develop over time (McCubbin & Patterson, 1983). Second, the model identifies variables that allow an understanding of why some family members/families/couples manage to

adapt better than others. By emphasizing the resources of families to adapt to stress, the double ABCX-model provides a *salutogenic* instead of a pathogenic view (Ho & Keiley, 2003). Third, it focuses on *individual-level* as well as *family-level variables* thereby coinciding with the growing consensus in the family stress literature that dealing with family crises – e.g., chronic disease – involves both family and individual processes (Boss, 1987; Burr, 1989), and that indicators at both levels are important for understanding and predicting the outcome of adaptation (Boss, 1987; Watson et al., 1988).

Theoretical Framework: the Double ABCX Model

The double ABCX model describes how a stressor (i.e., childhood cancer) impacts the adaptation of each family member (individual level), the family subsystems (family subsystem level), and the family as a whole (family level) over time and identifies variables that allow an understanding of why some family members, families and couples manage to adapt better than others (Weber, 2010). In addition, the model acknowledges that an individual's and a family (subsystem)'s response to a major stressor, like childhood cancer, develops over time and that family(subsystem)/individual adaptation is influenced by the family members' *resources* and the *appraisal* family members make of the stressful event (i.e., childhood cancer diagnosis). Specifically, "a" stands for the initial stressor, i.e., the childhood cancer diagnosis; "b" stands for the family's existing resources, situated at three levels (individual, intrafamilial, and contextual level); "c" stands for the subjective definition the family makes of the initial stressor: uncontrollable vs. manageable; and "x" stands for the indicators of short-term adaptation. According to this model, "a", "b", and "c" interact and lead to "x" in the short-term within each of the individual family members, the family subsystems and within the family as a whole. With respect to the long-term effects, "aA" stands for the stress pile-up; "bB" stands for the family's new and existing resources; "cC" stands for family's perception of the short-term adaptation,

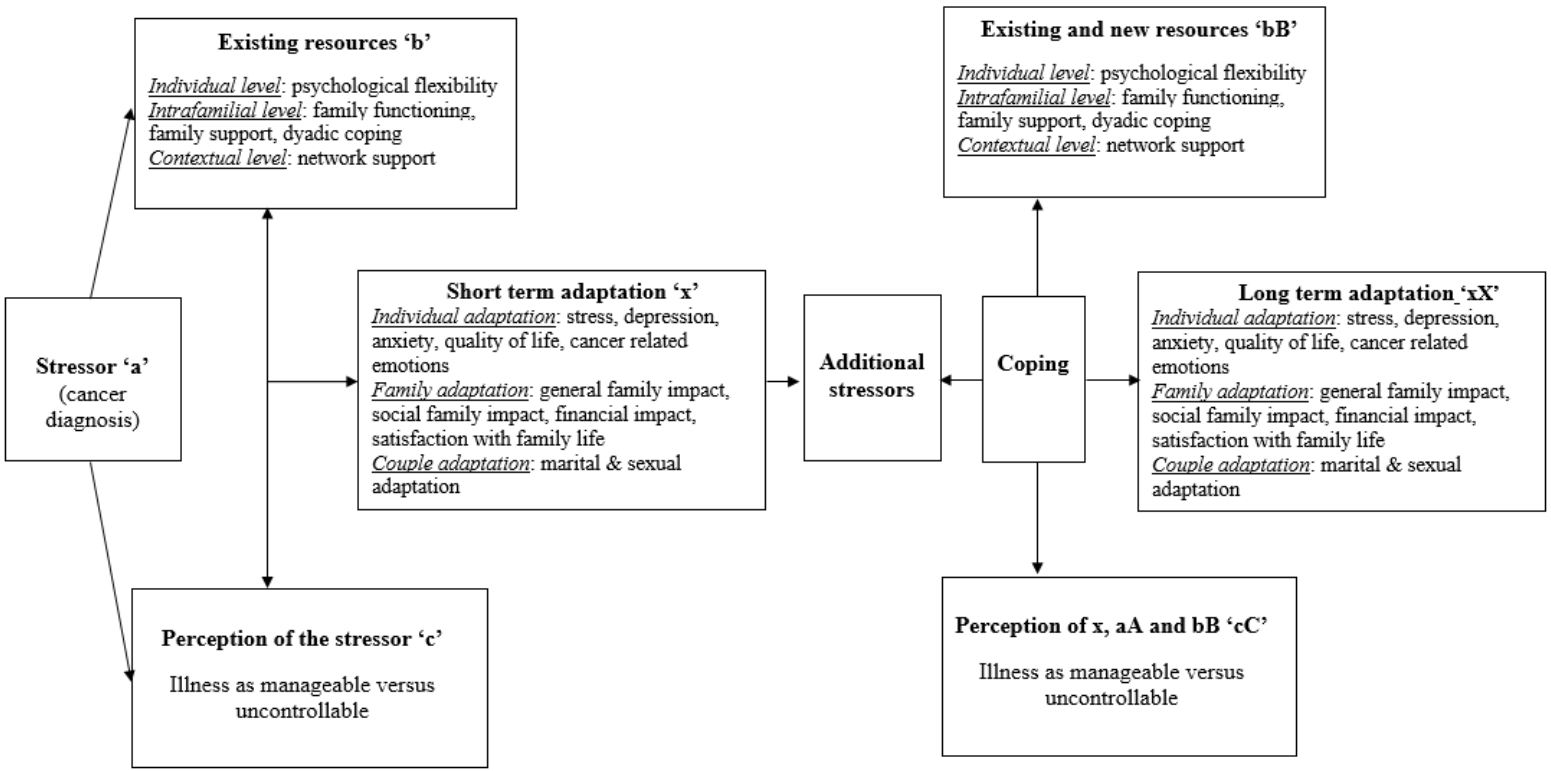

Figure 1. Double ABCX model (McCubbin & Patterson, 1983)

stress pile-up and existing/new resources; and "xX" stands for the indicators of long-term adaptation within each of the individual family members, the family subsystems, and within the family as a whole.

Drawing from the double ABCX model, predictors of adaptation are twofold: the appraisal/perception family members make of the stressor, and the family's resources. The *family's perception* of the disease refers to the tendency to define a stressor as uncontrollable (versus manageable) and makes families more (or less) vulnerable. So, lower levels of individual/family (subsystem) maladaptation can be expected when the disease is perceived as more manageable and less uncontrollable. The *family's resources* are factors that, by their presence, keep the individual/family (subsystem) from maladaptation and can be situated at three levels: the individual level, the intrafamilial level, and the contextual level. Based on the childhood cancer literature, this dissertation examines the following core resources: (a) each family member's psychological flexibility (i.e., the skill to flexibly adapt to fluctuating situational demands, being open and accepting of emotional experiences, and being willing to engage in difficult activities to persist in the direction of important values; Kashdan & Rottenberg, 2010) as an *individual resource*, (b) family functioning (e.g., support, cohesion, communication, conflict in the family and its subsystems) and dyadic coping (i.e., the extent to which parents deal with the stressor of pediatric cancer as a dyad; Bodenmann, 1995) as *intrafamilial* resources, and (c) the family's social network (e.g., friends, relatives) and the support received from them as *contextual* resources. So, we predict better adaptation at the individual and family (subsystem) level for families with higher levels of individual, intrafamilial, and contextual resources. In addition, the model predicts that families who are able to develop more resources over time (e.g., increased support from their social network) may evidence better adaptation on all family levels.

Chapter Overview

In order to address the research aims addressed above, the present dissertation is divided in three parts and includes 10 chapters (for an overview, see Table 1). The first part, "The impact on the family as a whole," focuses on the consequences of childhood cancer for the family system (i.e., *family adaptation*). The second part, "The impact on individual family members and the family as a resource", focuses on the individual adaptation of patients, parents, and siblings and on resources that can potentially explain why some individuals adapt better than others. The thread running through the chapters in the second part is the inclusion of a key intrafamilial resource: "family functioning" defined as the way in which the family as a whole deals with and responds to childhood cancer. So, in part 1 family functioning is conceptualized as an outcome variable, in part 2 family functioning is conceptualized as a resource that may contribute to individual adaptation of a patient, his or her parents and siblings when facing childhood cancer. The third part, "The impact on the parents' intimate relationship", focuses on the consequences of childhood cancer for the parents' intimate relationship. Below, every chapter is elaborated upon in more detail.

Part 1: The Impact on the Family as a Whole

First, this dissertation aims to gain insight into the consequences of childhood cancer for the family as a whole (i.e., family adaptation). In **Chapter 1**, we conducted a systematic review of quantitative and qualitative studies focusing on distinct aspects of family functioning (e.g., cohesion, conflict, adaptability, communication, family support) among families of children with cancer.

Table 1

Chapter Overview

	Methodology	Data/Participants	Method	Main research question
1	Review	43 qualitative and 35 quantitative studies	Systematic Review	Is there evidence of family resilience after a diagnosis of childhood cancer?
2	Quantitative	70 mothers and 53 fathers	Multilevel analysis	What is the role of parental psychological flexibility, dyadic coping, and network support in explaining family adaptation?
3	Qualitative	10 couples	Multi family member interview analysis	Which changes occur in family functioning after a pediatric cancer diagnosis?
4	Qualitative	10 siblings	Interpretative phenomenological analysis	How do siblings of children with cancer experience their everyday family life post-diagnosis?
5	Review	30 quantitative studies	Meta-Analysis	What is the association between family functioning and child adaptation after a childhood cancer diagnosis?
6	Quantitative	60 patients, 172 parents and 78 siblings	Multilevel analysis	What is the association between family functioning, cancer appraisal, and the individual adaptation of patients, parents, and siblings?
7	Quantitative	81 siblings	Multilevel analysis	What is the association between family functioning, family support, network support, and the individual adaptation of siblings?
8	Qualitative	4 families	Multi family member interview analysis	How do family members support each other when facing childhood cancer?

9	Review	17 quantitative and 13 qualitative studies	Systematic Review	What is the impact of a childhood cancer diagnosis on the couple functioning?
10	Quantitative	59 couples	Actor-Partner Interdependence Model	What is the association between dyadic coping and individual/relationship outcomes of parents in the context of childhood cancer?

The main aims in this review were to investigate family resilience after a diagnosis of childhood cancer and to examine theoretical, methodological, and statistical issues in the existing childhood cancer literature. We defined resilient families as those that return to, sustain, or achieve competent levels of family functioning in one or more family functioning domain (i.e., cohesion, adaptation, communication) after being challenged by childhood cancer, in line with the resilience definition of Hilliard et al. (2012).

The findings of this review allowed us to conceptualize recommendations for further research that we then followed to set up a large longitudinal survey study at four university hospital sites: Ghent, Leuven, Antwerp, and Brussels, i.e., "The UGent Families and Childhood Cancer Study". For this large-scale study, families of children diagnosed with leukemia or non-Hodgkin lymphoma (aged 0-18 years) were invited to take part. The ill child (only when s/he was aged 5-18 years; younger patients did not complete questionnaires), their biological parents, and any siblings (aged 5 years and more; younger siblings did not complete questionnaires) were asked to complete a set of questionnaires at five different time points (from diagnosis to 2.5 years post-diagnosis). All empirical, quantitative chapters described in this dissertation are based upon the findings of this large-scale study.

Chapter 2 describes a longitudinal questionnaire study with 70 mothers and 53 fathers of children with leukemia or non-Hodgkin lymphoma (80 families). Participants completed questionnaires at two measurement points (M_{T1} = 5.26 months; M_{T2} = 18.86 months). The aim of this study was to explore the role of protective factors at the individual (parental psychological flexibility), intrafamilial (dyadic coping), and contextual level (network support) in explaining *family adaptation* as perceived by parents of children with leukemia or non-Hodgkin lymphoma. We expected that higher levels of psychological flexibility (individual level), more adequate dyadic coping in the couple relationship (more stress communication, more

supportive dyadic coping, more common dyadic coping, less negative dyadic coping; intrafamilial level), and more (amount and satisfaction with) support from their social network (contextual level) would be associated with better *family adaptation*, both cross-sectionally and prospectively. A multilevel modeling approach was used to answer this research question.

In addition to the quantitative findings of chapter 2, chapter 3 and 4 provide more insight in the lived experiences of family members facing childhood cancer and its consequences for family life. In **Chapter 3**, a qualitative study is described reporting on the experiences of ten couples parenting a child diagnosed with leukemia or non-Hodgkin lymphoma. The aim of this study was to explore parents' perceptions of changes in family functioning after a childhood cancer diagnosis. Semi-structured interviews were conducted and a multi-family member interview analysis (MFMIA: Van Parys et al., 2017) was used to analyze the data. MFMIA allows a detailed and systematic analysis of shared family experiences (Smith, 1999; Van Parys et al., 2017) and has proved insightful in studies that analyze experiences shared by family members, particularly when assessing sensitive topics such as adaptation to illnesses (Eisikovits & Koren, 2010).

In **Chapter 4**, a qualitative study with ten siblings (10 – 16 years) of a child with leukemia or non-Hodgkin lymphoma is described. Semi-structured interviews were used to gain insight in how siblings experience their everyday family life post-diagnosis, allowing them to focus on particular aspects of the family that mattered most to them. Data was analyzed using interpretative phenomenological analysis (IPA; Smith et al., 2009). This approach comprises an in-depth exploration of the participants' lived experiences and how participants make sense of these experiences, while emphasizing the active role of the researcher in the process of the interpretative activity.

Part 2: The Impact on Individual Family Members and The Family as a Resource

Second, this dissertation aims to gain insight in why some family members adapt better than others. To answer this research question, both quantitative and qualitative studies were conducted to better understand the *individual adaptation* of different family members (patient, parents, siblings) and to explore the role of *family functioning* as a potential resource. In other words, does the functioning of the family as a whole in the context of childhood cancer influence the adaptation of individual family members?

In **Chapter 5**, a meta-analysis on the associations between family functioning and *child adaptation* (patient and siblings) after a childhood cancer diagnosis is described. In the analysis, 30 articles were included. Pearson's r correlations were the effect of interest. Omnibus and family functioning domain-specific random-effects meta-analyses were conducted.

In **Chapter 6**, a cross-sectional, quantitative study on the *individual adaptation of all family members* (60 patients, 172 parents and 78 siblings) is described. In this study, the aim was to explore the association between family functioning, cancer appraisal, and the individual adaptation of patients, parents, and siblings. We expected that better family functioning, perceiving the illness as more manageable and less uncontrollable, and their interaction, would be associated with better individual outcomes (i.e., less negative cancer-related emotions, more positive cancer-related emotions, and better quality of life) in patients, parents, and siblings. By making use of multilevel analyses, we were able to model interdependence in family relationships and to answer the research question.

In **Chapter 7**, a cross-sectional, quantitative study on the *individual adaptation of siblings* (N = 81) is described. In this study, we aimed to explore the association between family functioning, family support, network support, and the individual adjustment of siblings facing cancer in their brother/sister.

We expected that better family functioning, more family support, and more network support would be associated with better individual outcomes (i.e., less negative cancer-related emotions, more positive cancer-related emotions, and better quality of life) in siblings. A multilevel modeling approach was used to answer this research question.

Finally, as the previous chapters demonstrated the value of family functioning as a resource for the individual adaptation of patients, parents, and siblings, we wanted to explore *how exactly* family members support each other when facing childhood cancer. Therefore, in **Chapter 8**, a qualitative study with 4 families was conducted in order to explore the family practice of giving support. Individual interviews with 4 mothers, 3 fathers, and 5 siblings were performed; the data were analyzed using MFMIA (Van Parys et al., 2017).

Part 3: The Impact on the Parents' Intimate Relationship

Third, this dissertation aims to gain insight in the consequences for the *couple subsystem* when facing a diagnosis of cancer in their child. Indeed, a childhood cancer diagnosis not only impacts the family as a whole (part 1) and its members (part 2), but also the parents' intimate relationship.

In **Chapter 9**, we describe our systematic review of quantitative and qualitative studies focusing on distinct aspects of the *couple functioning* (e.g., emotional closeness, marital conflict, marital support, communication, sexual intimacy, marital satisfaction) among parents of children with cancer. The main aims of this review were to investigate couple functioning after a pediatric cancer diagnosis, and to examine theoretical and methodological issues in this literature.

In **Chapter 10**, a cross-sectional, quantitative study with 59 couples of children diagnosed with leukemia or non-Hodgkin lymphoma is described. The aim of this study was to explore the association between dyadic coping and the *individual* and *couple adaptation* of parents in the context of childhood cancer. We expected that adequate dyadic coping (i.e., more supportive dyadic

coping, more common dyadic coping, and less negative dyadic coping) would be associated with better individual outcomes (i.e., less negative emotions: less stress, anxiety and depression, and lower levels of childhood illness-related parenting stress) and relationship outcome (i.e., higher marital and sexual adaptation) within parents of children with cancer. The Actor-Partner Interdependence Model (APIM; Cook & Kenny, 2005) was used to analyze the data.

Taken together, throughout the ten chapters following this general introduction, the three aims of the current dissertation are addressed. More specifically we have gained insight in (a) the short- and long-term consequences of childhood cancer on the family, its members, and the parents' intimate relationship, (b) important resources impacting adjustment to childhood cancer, and (c) the experiences of siblings. We conclude this dissertation with a general discussion, in which we elaborate on our findings, discuss several theoretical and clinical implications, and provide directions for future research. To conclude, it should be noted that the present dissertation consists of ten research papers (10 chapters), of which nine have been published and one is accepted for publication. As each paper/chapter should be able to stand on its own, their contents may partially overlap.

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PART 1
THE IMPACT ON THE FAMILY AS A WHOLE

CHAPTER 1

FAMILY RESILIENCE AFTER PEDIATRIC CANCER DIAGNOSIS: A SYSTEMATIC REVIEW[1]

A systematic review was conducted to investigate family resilience in the context of pediatric cancer, and to examine theoretical, methodological, and statistical issues in this literature. Following guidelines for systematic reviews, searches were performed using Web of Science, Pubmed, Cochrane, PsycInfo, and Embase. After screening 5563 articles, 85 fulfilled inclusion criteria and were extracted for review. Findings indicated that most families are resilient, adapting well to the crisis of cancer diagnosis. However, a subset still experiences difficulties. Methodological issues in the current literature hamper strong nuanced conclusions. We suggest future research with a greater focus on family resilience and factors predicting it, based upon available theory, and conducted with attention toward unit of measurement and use of appropriate statistical analyses. Improvements in research are needed to best inform family-based clinical efforts.

[1]Van Schoors, M., Caes, L., Goubert, L., Verhofstadt, L. L., & Alderfer, M. (2015). Family resilience after pediatric cancer diagnosis: A systematic review. *Journal of Pediatric Psychology, 40*, 856-868. doi: 10.1093/jpepsy/jsv055

Introduction

Pediatric cancer is an unpredictable and uncontrollable stressor that puts the diagnosed child, his/her family members, and the family as a whole at risk for adjustment difficulties (Alderfer & Kazak, 2006). Research examining individuals' responses to the challenges posed by pediatric cancer, however, reveals resilience. For example, when compared with population norms, children diagnosed with cancer typically show no greater evidence of emotional maladjustment or psychological dysfunction (Phipps, 2007; Stam et al.,, 2001). In fact, in some studies, children with cancer demonstrate better emotional functioning than comparison groups and report benefit from their experience during treatment (Phipps et al., 2001) and positive changes within themselves, their relationships, and their plans for the future after treatment (Barakat et al., 2006). Similarly, most siblings of children with cancer score within normal limits on standardized measures of internalizing and externalizing disorders and may display increased empathy, maturity, and responsibility after cancer diagnosis (Alderfer et al., 2010; Houtzager et al., 1999). Most parents of children with cancer also exhibit resilience. After diagnosis, moderate levels of emotional distress, anxiety, and acute or posttraumatic stress symptoms are observed (Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008), but most improve within a matter of months to levels of distress comparable with normative samples (Dolgin et al., 2007; Ljungman et al., 2014). Parents also report posttraumatic growth, for example, being more patient and having a better understanding of what is important in life after their child's successful cancer treatment (Barakat et al., 2006).

Although there is evidence of resilience after childhood cancer diagnosis for individual family members, research into the *resilience of the family system* after diagnosis of pediatric cancer is less common. Given the presumptions that a family is more than the sum of its parts (Von Bertalanffy, 1973), and that a cancer diagnosis not only affects the individuals within the family, but also their relationships with one another and the way in which the

family functions (Alderfer & Kazak, 2006), it is important to understand the impact of cancer on the family as a whole. Furthermore, given the complex medical regimens of pediatric cancer, families must be able to alter roles and responsibilities, effectively communicate, manage emotions, and successfully work as a team to meet treatment demands. In short, family-level processes and outcomes are important in pediatric cancer.

While systematic reviews are available for the literature regarding family adjustment after a diagnosis of pediatric cancer (Long & Marsland, 2011; Pai et al., 2007), this work has not been conceptualized within the framework of family resilience theory. Consistent with Hilliard et al. (2012) , in which resilience was defined as achieving one or more positive outcomes despite exposure to significant risk, we defined resilient families as those that return to, sustain, or achieve competent levels of functioning in one or more domains of functioning (i.e., cohesion, adaptation, communication) after being challenged by childhood cancer. A systematic review was deemed necessary to synthesize the relevant empirical literature, which emerges across various disciplines (e.g., psychology, nursing, medicine) using divergent (i.e., qualitative and quantitative) methods. The primary aim of the review was to determine whether there is evidence of family resilience after a diagnosis of childhood cancer. The secondary aim was to examine theoretical, methodological and statistical issues in the existing literature and formulate recommendations for future family resilience research.

Method

As outlined by Eiser et al. (2000) and the Cochrane Collaboration (Deeks et al., 2011), we complied with a strict scientific methodology to ensure comprehensiveness, minimal bias and reliability of our systematic review. To this end, the following consecutive steps were taken: a) formulation of the scope of the review and research question, b) thorough

literature search, c) detailed data extraction, and d) integration of the major findings and implications. Meta-analysis was not conducted because of heterogeneity across studies in terms of sample characteristics (e.g., different stages of treatment) and outcomes measured, as well as our desire to integrate qualitative findings to ensure a comprehensive review.

Literature Search and Inclusion Criteria

Web of Science, Pubmed, PsycInfo, Cochrane, and Embase were searched using keywords selected in collaboration with a library information specialist and three researchers familiar with the field (details available upon request). Studies selected for inclusion examined aspects of functioning among families of children with cancer including: a) a subjective (qualitative) or objective appraisal of changes since diagnosis (longitudinal data); or, b) standardized scores, clinically meaningful categorization of families (e.g., based upon cut-scores), or a comparison with norms or control groups. Included studies investigated families of children diagnosed with any type of cancer before age 18. Studies published in languages other than English and nonempirical articles (i.e., reviews, case reports, books, book reviews, commentaries, practice guidelines, conference abstracts, and dissertations) were excluded. Articles concerning families of children who died from cancer were also excluded, as theirs is a distinct family experience. Reference lists of the selected papers were also reviewed to ensure inclusion of all relevant papers.

Study Selection

The database search was undertaken in July of 2014, identifying 5496 unique papers. The first and second author independently screened all titles for decisions regarding exclusion with 89% agreement. Disagreements were discussed and resolved. The 1592 remaining abstracts were then screened for

exclusion, with an agreement rate of 83%. Again, disagreements were discussed and resolved. Finally, the first author screened the full texts of the remaining 427 studies for final decisions regarding inclusion. The second author screened 25% with 93% agreement. Disagreements were discussed and, if necessary, a third reviewer was consulted; 72 studies were selected. Thirteen articles were added based on reference chaining, resulting in a final set of 85 papers (see Figure 1).

Data Extraction

The first and second author extracted data from the studies in a systematic and standardized way, summarizing study characteristics and general findings on abstraction sheets (available upon request). Year of publication, journal and database were recorded along with methodological characteristics such as type of design (cross-sectional or longitudinal), measures used, and sample achieved (e.g., sample size and demographics). In addition, the theoretical framework, unit of measurement used, and characteristics of the statistical analyses (interdependence of data reported by multiple respondents) were evaluated. Findings of the studies were extracted by summarizing the results in a few lines. The last author reviewed the information extracted against original publications to ensure accuracy. Authors were contacted for papers and information, as needed, but unpublished data were not requested.

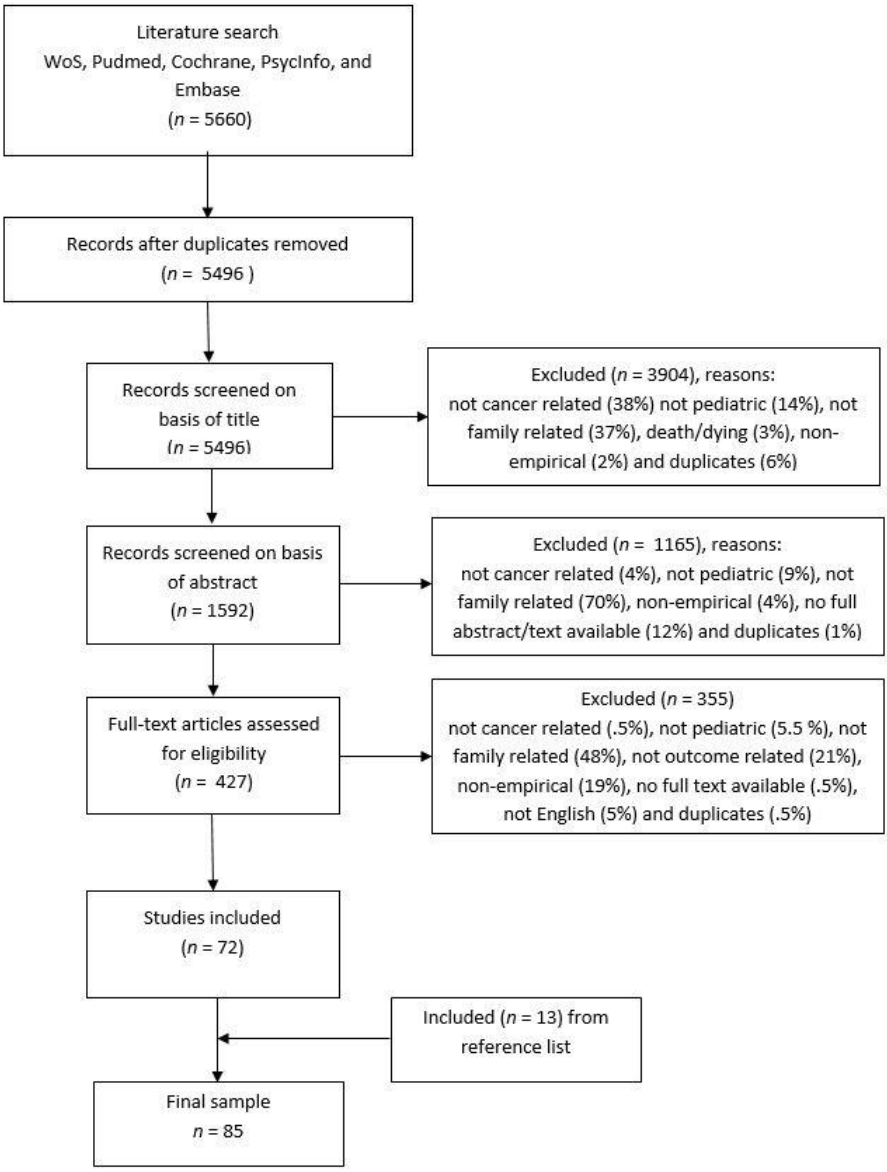

Figure 1. Flow chart

Results

Part I: Characteristics of the Studies in the Review

A Supplementary Table (i.e., Supplementary Table 1; at the end of this dissertation) summarizes the methods and findings of each reviewed study. The majority of the reviewed studies were quantitative (*n*=4351%); 41% used qualitative methods (*n*=35; marked in text with ^{QL}), and the rest used mixed methods designs (*n*=7; 8%; marked in text with ^{mix}). Most studies used cross-sectional designs (*n*=67; 79%). Sample size varied from three to 209 families (6 to 465 individuals). Among the studies with quantitative data, 20 (40%) included comparison groups and 14 (28%) used standardized norms; 16 studies (32%) did not make comparisons but provided longitudinal data or placed families into clinically meaningful groups (e.g., based on validated cut-scores). A wide variety of cancer diagnoses were included in the studies, with leukemia, lymphoma and brain tumors most frequently represented. Time since diagnosis ranged from one week to 18.6 years. No time frame was reported in 8 studies (8%) and was vague (e.g., "survivors") in 31 others (36%).

Part II: Narrative Summary of Reviewed Studies

The narrative review is organized by six relatively distinct aspects of family functioning that emerged from the literature: cohesion, conflict, adaptability, communication, family support, and overall family functioning. For each subsection, a brief explanation of the concept is given, followed by the number and types of relevant studies identified. Findings from the perspective of each family member (child with cancer, parents, and siblings) are then presented separately with qualitative results described before qualitative results.

Cohesion. Cohesion refers to the emotional bond between family members (Olson, 2000). Family resilience after pediatric cancer would be evident in a balance of connectedness and separateness, with possible increases in cohesion, whereas a lack of cohesion (disengagement) or too much cohesion (enmeshment) would be considered maladaptive. This construct was addressed in 17 qualitative, 17 quantitative, and 2 mixed method studies.

In qualitative work, *children with cancer* spoke about the illness drawing the family closer (Clarke-Steffen, 1997[QL]; Enskar et al., 1997[QL]; Woodgate & Degner, 2003[QL]). In quantitative work, many children reported strengthened bonds with parents (Kvist et al., 1991). In three studies, significantly greater levels of family cohesion were found as compared to control groups/standardized norms both during and after treatment (Beek et al., 2014; Cornman, 1993; Trask et al., 2003) and four additional studies of families who were off-treatment found no differences in child-reported cohesion in comparison to control groups/standardized norms (Kazak et al., 1994; Kazak & Meadows, 1989; Madan-Swain et al., 1994; Pelcovitz et al., 1998), all suggesting resilience. Only one study reported lower levels of survivor-reported cohesion among families of children with cancer than a normative sample; 40% of these teen survivors characterized their families as disengaged (Rait et al., 1992).

In qualitative studies, *parents* (Arabiat et al., 2013[QL]; Brody & Simmons, 2007[QL]; Clarke-Steffen, 1997[QL]; Koch, 1985[QL]; Neil-Urban & Jones, 2002 [QL]; Nicholas et al., 2009[QL]; Norberg & Steneby, 2009[QL]; Quin, 2004[mix]; Rocha-Garcia et al., 2003[QL]; Sloper, 1996[mix]; Woodgate & Degner, 2003[QL]) often indicated that family cohesion was strengthened by the illness, sometimes with a tendency toward enmeshment (Velasco et al., 1983[QL]), though a minority indicated the opposite in one study (Sloper, 1996[mix]). The bond between parents and the patient was specifically noted as becoming stronger (Kvist et al., 1991; Nicholas et al., 2009[QL]; Norberg & Steneby, 2009[QL]) but a minority of parents indicated that relationships with siblings

became weaker (Kvist et al., 1991; Quin, 2004[mix]). Thirteen quantitative studies compared parent-reported cohesion with norms/controls. Two studies, one with repeated assessments within 9 months post-diagnosis (Varni et al., 1996) and a second studying families toward the end of treatment and beyond (Cornman, 1993) found mean levels of parent-reported cohesion to be greater than norms. Nine additional studies with samples ranging from newly diagnosed families through long-term survivors (Beek et al., 2014; Carlson-Green et al., 1995; Cohen et al., 1994; Greenberg et al., 1989; Horwitz & Kazak, 1990; Kazak et al., 1994, Kazak & Meadows, 1989; Madan-Swain et al., 1994; Manne et al., 1995) found no differences between families of children with cancer and control groups/standardized norms. All of these studies suggest resilience. Finally, two studies suggested lower levels of cohesion among families of children with cancer, one investigating children on treatment (Morris et al., 1997) and a second of families after treatment (Rosenberg et al., 2014). Overall, findings generally point toward resilient outcomes (i.e., sustained or improved cohesion) from the parental perspective.

In qualitative studies, *siblings* also reported increased cohesion within the family (Chesler et al., 1991[QL]; Clarke-Steffen, 1997[QL]; Koch, 1985[QL]; Prchal & Landolt, 2012[QL]; Sargent et al., 1995[QL]; Sloper, 2000[QL]; Wiener et al., 2008[QL]; Woodgate & Degner, 2003[QL]; Woodgate, 2006a[QL]). One quantitative study found sibling-rated cohesion to be greater than norms (Cornman, 1993), also suggesting resilience. However, increased closeness was not always perceived as being inclusive of the siblings (Chesler et al., 1991[QL]).

In summary, most studies provide evidence for family resilience within the domain of cohesion after diagnosis of pediatric cancer, though siblings may experience being at the periphery of the family. We found no similarities among the few studies that suggested less cohesion among families of children with cancer nor any systematic differences between these studies and those suggesting resilience in terms of sample characteristics (e.g.,

diagnosis, time since diagnosis, treatment status, child age, country of origin) or methodology (e.g., respondent, measure, sample size, comparison group).

Conflict. Family conflict is openly expressed anger and discord among family members (Moos & Moos, 1994). Family resilience after pediatric cancer diagnosis would be evident if there were no increase in the amount of family conflict over time or in comparison to norms/controls. This construct was addressed in two qualitative and 12 quantitative studies.

Four quantitative studies compared family conflict reported by *children with cancer* with control groups/standardized norms, and findings were mixed. One of these studies indicated less child-reported conflict in families of children with cancer off-treatment compared with norms (Beek et al., 2014) and a second study indicated no difference between two such groups (Brown et al., 2003), both suggesting resilience. However, two additional studies indicated more child-reported conflict among families of children in treatment (Manne & Miller, 1998) and a sample including those on maintenance or off-therapy (Cornman, 1993) when compared to norms or controls.

In two qualitative studies, *parents* of children with cancer reported themes of family conflict across the illness trajectory (Patterson et al., 2004[QL]; Shortman et al., 2013[QL]) and two quantitative studies, one with families of children on treatment and a second with families in maintenance or off-therapy, indicated more parent-reported conflict compared with norms and controls (Cornman, 1993; Morris et al., 1997). However, seven studies, with samples ranging from one month through at least five years after diagnosis, indicated no differences between families of children with cancer and norms or controls on measures of conflict (Ach et al., 2013; Brown et al, 2003; Greenberg et al., 1989; Kronenberger et al., 1998; Noll et al., 1995; Varni et al., 1996) and two studies indicated less conflict for families of children with cancer, one studying families on treatment (Gerhardt et al., 2007) and the second studying families off-treatment (Beek et al., 2014), all suggesting

resilience. One study assessing sibling–reported family conflict in families off active therapy indicated greater levels of conflict when compared with norms (Cornman, 1993).

In summary, reports of increased conflict were not found in samples exclusively consisting of off-treatment families suggesting long-term resilience in this domain; however some conflicting results did arise when samples included families on treatment (including maintenance). Conflict was reported across both qualitative and quantitative studies and across family members, but not consistently. Sample characteristics (e.g., diagnosis, age of child, country of origin) and aspects of study design (e.g., measure, sample size) did not seem associated with outcome. It seems that being on treatment may be a risk factor for conflict.

Adaptability. Adaptability is the amount of malleability in the family's leadership, role relationships, and relationship rules (Olson, 2000). Well-functioning, resilient families would balance structure and flexibility after cancer diagnosis, possibly increasing in adaptability; poorly functioning families would be rigid (i.e., not enough adaptability) or chaotic (i.e., too much; Olson, 2000). This construct was addressed in 11 quantitative studies.

Six studies assessed adaptability from the perspective of the *child with cancer*. One, with a sample combining families on- and off-treatment revealed a greater family adaptability than norms (Trask et al., 2003) and five involving off-treatment families tended to find no differences in comparison to norms or controls (Kazak et al., 1994; Kazak & Meadows, 1989; Madan-Swain et al., 1994; Pelcovitz et al., 1998; Rait et al., 1992), suggesting resilience.

In regard to *parent-reported* adaptability, three studies of families of children on treatment (Cohen et al., 1994; Horwitz & Kazak, 1990; Manne et al., 1995) and two studies of families of children off treatment (Kazak et al., 1994; Kazak & Meadows, 1989) found no differences from norms or controls in level of adaptability. An additional study found a higher degree of

adaptability among families of survivors compared with norms (Rosenberg et al., 2014). All of these studies suggest resilience. However, one study of newly diagnosed families found that mothers tended to characterize their families as chaotic (Perricone et al., 2012), a second study of families on treatment found that a greater percentage of families of children with cancer than controls fell at the extremes for adaptability (i.e., either chaotic or rigid; Horwitz & Kazak, 1990), and a third study of families off-treatment noted that mothers were more likely than controls to characterize their families as rigid (Madan-Swain et al., 1994). No studies were found assessing family adaptability from the perspective of siblings.

Overall, it seems that most families of children with cancer are not different from norms/controls in terms of adaptability indicating resilience in this domain. While it is possible that a greater percentage adopt a chaotic way of functioning (near diagnosis) or a rigid style (during treatment and beyond), this may be a minority of families.

Communication. Communication, or the interchange of thoughts, feelings, experiences, and information within the family, is generally believed to be an important component of family functioning that can foster adaptation (Olson, 2000). Resilient families would maintain or increase communication within the family in response to cancer and their communication patterns would be open, clear, and effective. This construct was addressed in four qualitative studies, three mixed methods studies, and nine quantitative studies.

Four quantitative studies addressed expressiveness/communication within the family from the perspective of the *child with cancer*, all involving samples off active treatment. Greater expressiveness was reported in two of these (Beek et al., 2014; Cornman, 1993) and no differences were reported in a third (Madan-Swain et al., 1994), suggesting resilience. In the fourth study, more than 60% of a sample of adolescent survivors endorsed ‘unhealthy’ family communication patterns, characterized as vague and with masked

intent (Alderfer et al., 2009). It is unknown if this rate is different from families without cancer.

In a qualitative study, nearly 70% of mothers reported an open communication style with their children (Clarke et al., 2008[QL]). In two quantitative studies, *parents* of children with cancer reported more expressiveness within their families, as compared with norms, both during (Varni et al., 1996) and after active treatment (Cornman, 1993), and in six additional studies with both on- and off-treatment samples, no differences were found for expressiveness (Beek et al, 2014; Morris et al., 1997) or communication (Greenberg et al., 1989; Kazak et al., 1997; Madan-Swain et al., 1994; Streisand et al., 2003). All of these studies suggest resilience. However, in two studies with researchers characterizing communication patterns within families of children with cancer, 59% of newly diagnosed families were found to share minimal (40%) or ambiguous (19%) information (Clarke et al., 2005[QL]) and only about 30% of off-treatment families evidenced effective communication patterns (Adduci et al., 2012[mix]). An additional study found that about one third of parents of survivors rated their family communication patterns posttreatment as "unhealthy" (Alderfer et al., 2009). These latter studies did not include control groups, and so it is unclear whether these rates are unique to families of children with cancer.

Five studies assessed *siblings'* perceptions of family communication. Across three qualitative studies, most siblings reported being well-informed and satisfied with communication within their family (Havermans & Eiser, 1994[mix]; Prchal & Landolt, 2012[QL] ; Sloper, 2000[QL]). About two thirds of siblings in one sample, however, did want more information sooner (Sloper, 2000[QL]) and a minority across two other samples reported becoming tired of hearing about cancer when months into or after treatment (Havermans & Eiser, 1994[mix]; Prchal & Landolt, 2012[QL]). A fourth study, conducted in China, reported that 60% of the siblings in their sample claimed not to have a chance to talk about the illness with their parents or sick brother/sister during treatment (Wang & Martinson, 1996[mix]). This finding may be culturally-

specific. In one quantitative study from the United States, siblings' reports of expressiveness within their off-active-treatment families exceeded norms (Cornman, 1993).

In summary, when compared with norms/controls, children with cancer, their parents, and siblings reported equal or increased communication/expressiveness within their families, suggesting resilience. However, observations of families, classification based on cut-scores, and comments of siblings provide some evidence for poor communication patterns; it is unclear if the rates of these patterns are typical. Finally, cultural differences may be important in this domain of family functioning.

Family Support. Family support refers to assistance, encouragement, and caring from the family received or perceived by an individual (Walsh, 1998). Resilient families would be expected to maintain or increase support in response to cancer. Family support was addressed in 14 qualitative, two mixed method and 15 quantitative studies.

In qualitative studies, c*hildren with cancer* reported that family support was important in helping them get through cancer (Enskar et al., 1997a[QL]; Havermans & Eiser, 1994 [mix]; Kyngas et al., 2001[QL]; McGrath et al., 2005[QL]; Ritchie, 2001[QL]; Woodgate & Degner, 2003[QL]; Woodgate, 2006b[QL]). In fact, in one qualitative (Enskar et al., 1997[QL]) and three quantitative studies, they reported support or satisfaction with support from family/parents as being greater than that from any other source (i.e., friends, teachers; Kazak et al., 1994; Nichols, 1995; Trask et al., 2003). Three studies indicated that adolescents with cancer (Brown et al., 2003; Haluska et al., 2002) - and specifically those undergoing haematopoietic progenitor cell transplant (Barrera, Andrews, Burnes, & Atenafu, 2008) - reported more parental support than controls/norms, and three including those on- and off-treatment found no differences (Kazak & Meadows, 1989; Manne & Miller, 1998; Wesley et al., 2013), generally indicating resilience.

In qualitative studies, *parents* also reported that family support was important in the context of cancer (Beltrao et al., 2007[QL]; Brody & Simmons, 2007[QL]: Enskar et al., 1997a[QL]; Enskar et al., 1997b[QL]; Greenberg & Meadows, 1992[QL]; Jackson et al., 2008[mix]; McGrath et al., 2005[QL]; Nicholas et al., 2009[QL]; Shortman et al., 2013[QL]; Woodgate & Degner, 2003[QL]). Five studies comparing parents of cancer survivors with controls or norms with samples both on- and off-treatment indicated no differences in level of family support (Brown et al., 2003; Gerhardt et al., 2007; Kronenberger et al., 1998; Noll et al., 1995), suggesting resilience. Only one study, focused on parents of brain tumor survivors 1-5 years posttreatment, found levels of support lower than controls; this was for mothers' but not fathers' reports (Ach et al, 2013).

Finally, *siblings* also reported that family support was important in coping with cancer (Havermans & Eiser, 1994[mix]; Sloper, 2000[QL]; Woodgate & Degner, 2003[QL]). In one quantitative study, siblings' ratings of parental support were not different from norms (Barrera et al., 2008), suggesting resilience. Interestingly, friends and teachers were also frequently reported as support providers (Havermans & Eiser, 1994[mix]; Sloper, 2000[QL]). In one study, siblings rated support from parents as less important and available than support from friends and equal to that of teachers on both of these dimensions (Alderfer & Hodges, 2010).

Across studies, children with cancer, their parents, and siblings all reported that family support helped them cope with the cancer experience. Children with cancer consistently rated support within the family as being equal to or greater than norms/controls, suggesting resilience in this domain. Parents tended to report this too with one exception – mothers of brain tumor survivors. Late effects and the associated demands placed on mothers in this specific population may raise their support needs, so this finding may be important clinically. Finally, studies of siblings indicated that support from outside the family is also important and readily available to them.

General Family Functioning. Resilient families would maintain or improve on their general functioning patterns after cancer diagnosis. Perceptions of general family functioning across dimensions and domains was addressed in 20 qualitative, two mixed methods and 17 quantitative studies. Some studies assessing this construct combined data across family members. These findings are presented before data regarding individual family members' perspectives.

Qualitative studies combining *data across family members* revealed a shift in priorities and focus on the ill child that resulted in family disruption and loss of normal family life during treatment (Bjork et al., 2009[QL]; Clarke-Steffen, 1997[QL]; Koch, 1985[QL]; McGrath et al., 2005[QL]), as well as a struggle posttreatment to return to normality (Bjork et al., 2011[QL]). In one study, when asked about the impact of surviving cancer, about 10% of adolescent *survivors* reported general family functioning difficulties (Greenberg & Meadows, 1991[QL]). *Parents* specifically reported disruption of the family, stress between family members, and trouble balancing family needs including those of siblings (Arabiat et al., 2013[QL]; Bjork et al., 2009[QL]; Brody & Simmons, 2007[QL]; Enskar et al., 1997b[QL]; Ferrell et al., 1994[QL]; Patterson et al., , 2004[QL]; Quin, 2004[mix]; Rocha-Garcia et al., 2003[QL]; Sidhu, Passmore, & Baker, 2005[QL]; Sloper et al., 1996[QL]; Ward-Smith et al., 2005[QL]). *Siblings* reported disrupted family routines, being separated from the family due to treatment, and a general loss of family life (Chesler et al., 1991[QL]; Prchal & Landolt, 2012[QL]; Sargent et al., 1995[QL]; Sloper, 2000[QL]; Woodgate, 2006a[QL]).

Three quantitative studies combined *data across family members* to assess family functioning. The first found that families of children with cancer, at least two years post-diagnosis, were functioning similarly to control families across a range of areas (e.g., cohesion, communication, consideration, satisfaction; Sawyer et al., 1986). The second analyzed data from mothers, fathers, and survivors (not nested within family), and found that 41% of the sample characterized their family as well functioning (high cohesion, high expressive, low conflict), 46% placed their family in a moderate range, and

13% reported poor functioning (low cohesion, low expressiveness, high conflict); 26% of families had at least one member reporting poor functioning (Ozono et al., 2007). In a third study, using a family mean across parents and survivors, 35% of families were found to score in the 'unhealthy' range for general functioning (Alderfer et al., 2009). It is unknown whether these percentages are similar for families of children without cancer.

Turning to perceptions of *children with cancer*, across quantitative studies, including children on- and off- treatment, ratings of family functioning were no different from norms/ controls, suggesting resilience (Foley et al., 2000; Madan-Swain et al., 1993; Wesley et al., 2013; Yonemoto, et al., 2009),

Studies of *parents* also show no differences in general family functioning compared with norms/control groups, both during and after treatment (Foley et al., 2000; Kazak et al., 1997; Noll et al., 1995; et al., 2012; Sawyer et al., 1997; Sawyer, Antoniou, Rice, & Baghurst, 2000; Streisand et al., 2003) with relative stability across time from diagnosis through four years after diagnosis reported in longitudinal studies (Fife, Norton, & Groom, 1987; Sawyer et al., 1997; Sawyer et al., 2000). However, a subset does report problems. "Unhealthy" family functioning was reported by 26-38% of parents within 3 years of diagnosis (Long et al., 2013), 20% of parents on average 3.5 years after diagnosis (Martin et al., 2012), and 24%-38% of parents off-treatment (Alderfer et al., 2009; Peterson et al., 2012). Also, 11% of parents of long-term survivors reported problems with family harmony (Seaver et al., 1994[mix]).

One study compared *sibling* ratings of general family functioning during treatment to controls and found no differences (Madan-Swain et al., 1993); however 47% of siblings in a second study of families within three years of diagnosis reported "unhealthy" general family functioning (Long et al., 2013). It is unclear if this percentage is different from norms.

In summary, qualitative research clearly indicates that childhood cancer disrupts the functioning of the family in various ways; however, for

most families their general functioning, even in this time of stress, is within normal limits and similar to controls, suggesting resilience. Because control groups have not been consistently used, it is unclear whether the size of the subset of families experiencing "unhealthy" functioning is atypical.

Part III: Evaluation of the Literature

Theoretical Considerations. In the majority of the studies (*n* = 71, 84%), no theoretical framework was specified as guiding the research questions or selection of the variables. Failure to use theoretical frameworks risks limiting progression of the field, as advances cannot be made if theories go untested and unrevised.

Measurement Considerations. Even though the included studies focused upon family-level constructs, only 5 studies (6%) measured family functioning from the perspective of all immediate family members. In fact, more than half of the studies (*n* = 45; 53%) used a single family member as the reporter. Because the *unit of interest* should harmonize with the unit of measurement (Weber, 2011), one could argue that studies with a single informant did not adequately assess family functioning. Discrepancies in perceptions of family functioning across family members (e.g., Alderfer et al., 2009; Peterson et al., 2012) speak to the need to collect data from multiple family members, including siblings, to best capture this construct.

Statistical Considerations. In studies with data arising from multiple members within the same family, the interdependence of data within the family needs to be considered. Ignoring the dependency violates statistical assumptions of commonly used statistical approaches, generating inadequate test statistics (e.g., *t* or *F*), degrees of freedom and statistical significance values (i.e., the *p* value; Kenny, 1995). The majority of studies in our review (*n* = 70, 82%) avoided this issue through research design (e.g., qualitative analyses; single informant). Of the remaining studies, 11 (13%) avoided the issue by performing separate analyses for different family members without

combining their data. Only 4 (5%) took the interdependence into account by creating a summary score across respondents or by using appropriate statistical techniques to account for the dependency (e.g., multilevel modeling, actor-partner model).

Overall Quality. In addition to the issues mentioned above, certain characteristic of the research base make it particularly difficult to draw strong conclusions. For example, heterogeneity across and within studies in regard to sample characteristics and operationalizations of family functioning presents barriers to conducting meaningful meta-analysis. With rare exception, studies have small heterogeneous samples and gather data at a single time point, precluding identification of factors that may reliably predict which families experience the greatest difficulties meeting the challenges of pediatric cancer. While some studies used adequately sized demographically-matched control groups, these studies typically focused on comparing mean levels of functioning as opposed to comparing the percentages of families falling within dysfunctional ranges on the measures used, potentially masking important differences between groups on variables where both high and low scores may be problematic. Furthermore, nearly all studies relied on self-report of family functioning despite known drawbacks associated with this method (Schwarz, Groves, & Schuman, 1998). Observational assessment of family interactions could be indispensable in furthering our understanding of family-level adaptation in response to childhood cancer.

Discussion

This systematic review provides general evidence of family resilience after a pediatric cancer diagnosis; however, more work is needed to best understand this phenomenon. While we are starting to acknowledge and understand individual strengths, less is known about family-level strengths

after the experience of pediatric cancer (i.e., family resilience). To further this field of inquiry, future work should be theory-based, match the unit of measurement with the unit of interest (i.e., include all/many family members) and use appropriate statistical methods to nest data from family members within families.

The conclusions of this review are hampered by a few factors. We considered families as resilient if they returned to, sustained, or achieved competent levels of functioning after childhood cancer diagnosis. However, data regarding the functioning of the family before cancer, longitudinal data examining changes in the family over time after diagnosis, and criteria for judging whether the functioning of the family is "competent" were rarely available. We frequently relied upon comparisons between families of children with cancer and controls/norms to determine resilience; however, one could argue that competent functioning in the context of pediatric illness may be different from the functioning of families in which the children are healthy (see Alderfer & Stanley, 2012). For example, perhaps a more enmeshed or rigid pattern of functioning is adaptive in the face of cancer, at least for a certain period following diagnosis (Olson, 2000).

Our ability to draw conclusions about the resilience of families facing pediatric cancer was also hampered by the relative lack of studies using this framework as a basis for their research approach. In fact, other conceptualizations of family resilience could not be applied to the existing literature. For example, various family resilience theories (McCubbin & McCubbin, 1988; Patterson, 2002; Rolland & Walsh, 2006) do not see family functioning as the outcome of interest. Instead, family functioning is conceptualized as the process or means through which families achieve resilience. In these models, other outcomes are evidence of resilience, such as the family's ability to successfully meet future challenges (Rolland & Walsh, 2006), to maintain the family unit or to promote the development of individual members (Patterson, 2002). These outcomes are rarely, if ever, assessed in the context of pediatric cancer diagnoses. Other definitions of resilience in the

context of pediatric illness, such as successful management of illness and treatment demands (Mitchell et al., 2004), are rarely examined as an outcome of family-level processes (e.g., effective communication).

Suggestions for Future Research

To parallel the movement toward conceptualizing individual responses after pediatric cancer within a resilience framework, future research regarding family-level responses to pediatric cancer needs to adopt family resilience models. This change would require research to involve multiple members within families, assessed over time. More homogenous samples or samples large enough to examine heterogeneity (e.g., time since diagnosis, age of children) are recommended. Mixed qualitative and quantitative methods, along with observational methods, are needed to assess the full range of relevant constructs including objective and subjective characterization of the demands of pediatric cancer, capabilities, characteristics, and key processes of functioning within the family, and short and long-term family-level outcomes indicative of resilience. Research aimed at uncovering factors capable of identifying those families who might struggle to achieve resilience and isolating the mechanisms underlying family resilience would be most helpful for informing intervention.

Implications for Clinical Practice

Despite gaps in the current literature, adoption of a family resilience framework and the findings of our review have implications for clinical practice. First, attention should be focused upon the impact of cancer on the functioning of the family and family functioning should be routinely assessed in this population. Some families may need assistance in rallying their resources, developing a shared perspective of their experience and working together effectively to meet the demands of cancer. Such difficulties may

simultaneously jeopardize cancer treatment and important longer-term family outcomes. Relevant empirically based family-level intervention approaches are described in the literature (e.g., Rolland & Walsh, 2006; Saltzman et al., 2013). Second, clinical work with families should be sensitive to possible cultural differences, should consider the family within its larger socio-ecological context, and attend to subgroups that might be at elevated risk (e.g., families of children with brain tumors). Finally, based on our review, conflict within the family during treatment and communication with and support of siblings may be areas of common difficulty for families of children with cancer that should specifically be assessed and addressed as needed.

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CHAPTER 2

FAMILY ADJUSTMENT WHEN FACING PEDIATRIC CANCER: THE ROLE OF PARENTAL PSYCHOLOGICAL FLEXIBILITY, DYADIC COPING AND NETWORK SUPPORT[1]

Pediatric cancer is a life-threatening disease that poses significant challenges to the life of all family members and the family as a whole. To date, limited research has investigated family adjustment when facing pediatric cancer. The aim of the current study was to explore the role of protective factors at the individual (parental psychological flexibility), intrafamilial (dyadic coping), and contextual level (network support) in explaining family adjustment as perceived by parents of children with leukemia or non-Hodgkin lymphoma. In addition, we were interested to see whether these protective factors could be predictive for family adjustment at a later time point. Participants were 70 mothers and 53 fathers (80 families) of children with leukemia or non-Hodgkin lymphoma. Mean time since diagnosis was 5.26 (T1) and 18.86 (T2) months. Parents completed the Acceptance and Action Questionnaire II (to assess psychological flexibility), Dyadic Coping Inventory, a network support questionnaire, Impact on Family Scale and the Family Adjustment Scale. Both concurrent and prospective association models were tested. Psychological

[1]Van Schoors, M., De Paepe, A., Lemiere, J., Morez, A., Norga, K., Lambrecht, K., Goubert, L., & Verhofstadt, L. L. (2019). Family adjustment when facing pediatric cancer: The role of parental psychological flexibility, dyadic coping and network support. *Frontiers Psychology, 10*, 2740. doi: 10.3389/fpsyg.2019.02740

flexibility, dyadic coping, and network support proved to be cross-sectionally and positively related to parents' perception of family adjustment post-diagnosis; psychological flexibility and dyadic coping proved to predict better family adjustment over time. Our findings led to the conclusion that protective factors at all three levels (individual, intrafamilial, and contextual) are important for explaining family adjustment as perceived by parents facing a diagnosis of cancer in their child. Interventions targeting the individual, couple, as well as family level are warranted to enhance family adjustment.

Introduction

Advances in the medical treatment of pediatric cancer have resulted in an increased survival rate and a shift in research focus from death and grief into the adjustment of children diagnosed with cancer and their family. Although pediatric cancer is a major stressor, the current body of literature on the adjustment of the diagnosed child, his/her parents and possible siblings suggest that most family members adjust well, and only a subset experiences psychosocial problems during or after treatment (individual adjustment). For example, symptoms of anxiety, depression (Brinkman et al., 2016) and distress (Michel et al., 2010) have been observed in some patients. Post-traumatic stress symptoms, emotional distress, and anxiety are reported to a varying degree by parents (Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008), and worry, loneliness, sadness and post-traumatic stress symptoms are reported by a subset of the siblings (Alderfer et al., 2010; Long et al., 2018).

In addition to the impact on the family members' individual functioning, some studies have documented the impact of pediatric cancer on the family as a whole (family adjustment; see Pai et al., 2007; Van Schoors et al., 2015 for an overview). Overall, quantitative studies revealed that most families function within normative ranges (e.g., adaptability, Pai et al., 2007; family support, Brown et al., 2003) or even report improved functioning in

some realms (e.g., cohesion; Cornman, 1993). Being on-treatment and being a mother of a child with cancer, however, are risks factors for family conflict (Pai et al., 2007; Van Schoors et al., 2015). In contrast, qualitative studies into the family impact when facing pediatric cancer indicated a loss of normal family life (Bjork et al., 2009; Clarke-Steffen, 1997; Koch, 1985) and family rituals (Santos et al., 2018), troubles balancing multiple family needs including those of siblings (Bjork et al., 2009), and a shift in focus toward the diagnosed child at the cost of the family as a whole, the siblings, and the couple relationship (Van Schoors et al., 2018a, 2018b).

Given this variability in outcomes, both at the individual level and the family level (Kazak, 2006), a growing number of studies has tried to explain why some family members and families adjust better than others, investigating the role of potential protective factors. Based on existing research into pediatric oncology, protective factors that have been studied can be situated at three levels: the individual level (e.g., personality; Erickson & Steiner, 2001), the intrafamilial level (e.g., couple functioning; Santos et al., 2017; Van Schoors et al., 2019b), and the contextual level (e.g., network support; Corey et al., 2008).

To date, the current literature into individual and family adjustment when facing pediatric cancer is limited in four ways. First, most studies still tend to overlook outcomes at the family level (Van Schoors et al., 2015) and mainly focus on the consequences for individual family members facing pediatric cancer. Given the presumptions, however, that a family is more than the sum of its parts (Von Bertalanffy, 1973) and that a cancer diagnosis not only affects the individuals within the family, but also their relationships with one another and the way in which the family functions (Alderfer & Kazak, 2006), the family-level impact is undeniable. Second, most studies limit their scope to studying potential protective factors on one of the levels mentioned above (individual level, intrafamilial level, or contextual level). As a consequence, the existing studies mostly provide only a fragmented and partial explanation of the processes underlying post-diagnostic adjustment.

This fragmented approach, however, is conceptually not in line with most contemporary family stress models (see Weber, 2011), who consider all these three categories of resources crucial to understanding the varying effects of external stressors on families, ranging from family crisis to family adjustment. According to these models, protective effects may reside in characteristics of individual patients and family members, characteristics of some of the family subsystems, as well as the broader context in which the family is embedded. Gaining further insight into the question why some families more effectively meet the demands of facing pediatric cancer than other, requires research that conceptually and empirically takes into account the multi-level nature of families' resources. Third, most studies on adjustment after pediatric cancer relied on cross-sectional designs. As such, little is known on how family members/families adjust to the cancer diagnosis and its treatment over time. Fourth, within the pediatric cancer literature, most studies only include one single respondent (Van Schoors et al., 2015), rather than taking the perspectives of different family members into account, thereby neglecting the interdependence and bidirectional relationships between different family members.

The Present Study

In order to address these limitations, we conducted a study with two measurement points (T1 and T2) among parents (*mothers and fathers*) of children with leukemia or non-Hodgkin lymphoma to provide insight in the role of *individual*, *intrafamilial*, and *contextual* protective factors for the family adjustment at T1 (first aim, cross-sectional), as well as to provide insight in the role of these factors *over time* on the family adjustment (second aim, from T1 to T2, prospective). Family adjustment was operationalized as the economic consequences for the family (financial impact), the disruption in the family's normal social interactions (social impact), the disequilibrium experienced by the parents relating to the psychological burden of the illness

(e.g., difficulty of planning for the future; general family impact) and the parents' satisfaction with the family's way of life (satisfaction with internal family fit). With respect to *individual protective factors* (=possessed by individual family members), this study examined the role of the child's mother and father's psychological flexibility. Psychological flexibility generally refers to the willingness of an individual to experience unwanted or aversive stressors while pursuing one's values and goals, instead of avoiding unwanted or aversive stressors, thoughts, and feelings (Hayes et al., 1999). Psychological flexibility has received scientific and clinical attention during recent years and showed to predict better well-being in patients and their caregivers (Burke et al., 2014; Kashdan & Rottenberg, 2010). With respect to *intrafamilial protective factors* (= possessed by the collective members of the family) that contribute to better outcomes in families facing pediatric cancer, this study examined the role of the couple's dyadic coping. Dyadic coping refers to the extent to which partners deal with a stressor, like pediatric cancer, as a dyad (Bodenmann, 1995). Both theoretical (e.g., Systemic Transactional Model; Bodenmann et al., 2016) and empirical arguments (Badr et al., 2008) have illustrated the importance of couple variables, such as dyadic coping, within the context of health and illness-related issues. In addition, in a recent study (Van Schoors et al., 2019b), the importance of dyadic coping for the individual adjustment of parents facing leukemia or non-Hodgkin lymphoma in their child was illustrated. *Contextual protective factors* refer to the family's social network (e.g., friends and relatives) and the support (e.g., emotional, informational) received from them. In the current study, the amount of perceived network support as reported by mothers and fathers, as well as discrepancies/congruencies between desired and received parental support (i.e., parental satisfaction with the received support) were included. Indeed, previous cancer research has showed that network support helps the family to better cope with the illness (Woodgate & Degner, 2003; Woodgate, 2006) and even reduces individual adjustment problems post-diagnosis (Hoekstra-Weebers et al., 2001).

Taken together, we expected that higher levels of psychological flexibility in mothers and fathers of children diagnosed with cancer (individual level), more adequate dyadic coping in their couple relationship (more stress communication, more supportive dyadic coping, more common dyadic coping, less negative dyadic coping; intrafamilial level) and more (amount and satisfaction with) support they receive from their network (contextual level) would be associated with better family adjustment (i.e., lower financial impact, social impact and general family impact, and more satisfaction with internal family fit), both cross-sectionally (at the same moment in time) and prospectively (after some time had passed).

Method

Participants

The current sample consisted of 70 mothers and 53 fathers (80 families) where one child has been diagnosed with leukemia or non-Hodgkin lymphoma. All parents were Caucasian and living in the Flemish part of Belgium. The ill child's mean age at diagnosis was 6.96 years (SD = 5.05; Range = 0-18). In 58 families (72.5%), the diagnosed child had been diagnosed with acute lymphoblastic leukemia (ALL). In the remaining families, 7 children (8.8%) had been diagnosed with acute myeloid leukemia (AML), one child (1.3%) with chronic myeloid leukemia (CML), and 14 children (17.5%) with non-Hodgkin lymphoma. More details on the sample are listed in Table 1. Ethical approval from the University Hospitals of Ghent, Louvain, Brussels and Antwerp had been secured for the study and the appropriate written informed consent forms were obtained from all participants.

Table 1

Background Characteristics of the Study Sample

Demographic variable		
N (mothers; fathers)		123 (70; 53)
Age, mothers mean (*SD*)		37.61 (6.35)
Age, fathers mean *(SD)*		39.70 (6.41)
Age ill child at diagnosis, mean (*SD*)		6.96 (5.05)
Sex ill child, boys, *n (%)*		49 (61.3%)
Diagnosis[1], *n (%)*	ALL	58 (72.5%)
	AML	7 (8.8%)
	CML	1 (1.3%)
	Non-Hodgkin Lymphoma	14 (17.5%)
Time since diagnosis in months (*SD*; Range)	Time 1 (mothers)	4.74 (5.87; 0-28)
	Time 1 (fathers)	5.94 (6.95; 0-28)
	Time 2 (mothers)	19.46 (11.51; 3-45)
	Time 2 (fathers)	18.08 (12.10; 3-45)
Family status, *n (%)*	Married/Co-habiting	70 (87.5%)
	Divorced	7 (8.8%)
	Single parent	1 (1.3%)
	Stepfamily	2 (2.5%)
Number of children in the family, *n (%)*	One child	14 (17.5%)
	Two children	36 (45%)
	Three children	23 (28.7%)
	Four children	5 (6.3%)
	Five children	2 (2.5%)

Note. [1]ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia

Procedure

The present study is part of the 'UGhent Families and Childhood Cancer Study', a large study ongoing in Belgium, examining the impact of pediatric cancer on families (also see Van Schoors et al., 2019a, 2019b). For this large-scale study, families of children diagnosed with leukemia or

non-Hodgkin lymphoma (aged 0-18 years) were invited to take part in a survey study. The ill child (only when s/he was aged 5-18 years; younger patients did not complete questionnaires), their biological parents and any siblings (aged 5 years and more) were asked to complete a set of questionnaires at five different time points (from diagnosis to 2.5 years post-diagnosis). For the current study, parents with (at least) two measurements were included. If a parent had three or more measurements, his/her first and last measurement was taken into account. In the current study, mean time differences between measurement 1 (T1) and 2 (T2) were 15 and 12 months for mothers and fathers, respectively. Exclusion criteria for participation were: (1) not speaking Dutch, (2) relapse, and (3) the presence of a developmental disorder in the ill child. From the start of the study (September 2013), 137 families participated (65% of the eligible families); in 80 families at least two measurements per participant were available. The most important reasons for non-participation were lack of interest (41%), lack of time (27%) or being emotionally overwhelmed by the cancer (27%).

Measures

Psychological Flexibility. The Acceptance and Action Questionnaire II (Bond et al., 2011) was used to assess parents' ability to accept undesirable feelings and thoughts and to pursue their goals in the presence of potentially difficult experiences. The original questionnaire contains 10 items, rated on a 7-point Likert scale from 1 (*never true*) to 7 (*always true*) and is distributed across two factors. However, in accordance with Bond et al. (2011), the present study did not retain the three items on the second factor, as the predictive validity of the questionnaire was similar using a one-factor structure. All seven items were reversed, so higher scores indicate higher psychological flexibility. Total scores rage from 7 to 49. Example item are "Emotions cause problems in my life", "Painful memories prevent me from having a satisfying life", and "I'm afraid of my feelings". The AAQII has good

reliability and validity (Bond et al., 2011). In the present study, Cronbach's alpha coefficients were .89 and .93, for fathers and mothers, respectively.

Dyadic Coping. A short version of the Dyadic Coping Inventory (DCI; Bodenmann, 2008) was used to measure dyadic coping and stress communication. The questionnaire consists of 17 items, grouped into 6 subscales: Supportive Dyadic Coping (e.g., "S/he makes me feel that s/he understands me and is committed to me"; "S/he listens carefully and lets me speak, s/he responds appropriately to my stress or tries to lift me up"), Common Dyadic Coping (e.g., "We try to tackle the problem together and work together"; "We give each other emotional support"), Negative Dyadic Coping (e.g., "S/he does not take my stress seriously"; "S/he blames me for not being able to handle stress well"), Stress Communication (e.g., "When I feel overwrought, I show my partner that I feel bad and that I need his/her emotional support"), WE-Stress Appraisal (e.g., "If one of us is stressed, that is also "our" stress") and Individual Stress-Appraisal (e.g., "If my partner is stressed, that's his/her problem"). In the present study, (1) the two latter subscales were not included given our focus on dyadic coping strategies, and (2) the questionnaire was only completed by married or cohabiting parents. Response options for each item ranged from 1 to 5 (*very rarely* to *almost always*). Scores for the different subscales were obtained by summing the relevant items. The DCI has good reliability and validity (Ledermann et al., 2010). In the present study, Cronbach's alpha coefficients were .72/.78 (supportive dyadic coping), .91/.93 (common dyadic coping), .69/.81 (negative dyadic coping) and .85/.91 (stress communication) for fathers and mothers, respectively.

Network Support. Our measurement of network support was based on the Psychosocial Assessment Tool (PAT; Kazak et al., 2001, 2015), a screening instrument designed to investigate psychosocial risk in families of children diagnosed with cancer. The PAT consists of 20 items, assessing a constellation of risk and resource factors, including social support (Kazak et

al., 2001). For the present study, only the items relevant to network support were used. Participants had to indicate (*yes/no*) who they can count on to provide support, addressing six sources (Spouse/Partner, Patient's Grandparents, Extended family, Friend, Work Associates, Other, None) and five forms of support (Childcare/Parenting, Emotional Support, Financial Support, Information, Help with everyday tasks). In addition, in accordance with the existing literature on helpful support (Rafaeli & Gleason, 2009), we also assessed the extent to which the support they received from their network is in line with what they need/desire. Answer options were *more than I need, exactly what I need* and *less than I need*. In the present study, two network support indices were included: (1) the total amount of perceived network support (i.e., sum across all sources of support and forms of support) (2) the satisfaction with the received network support (i.e., categorical variable; 3 levels).

Family Adjustment. The Impact on Family Scale (Stein & Riessman, 1980) and the Family Adjustment Scale (Antonovsky & Sourani, 1988) were used to assess the adjustment of the family system, as perceived by parents facing a pediatric cancer diagnosis in their child. The Impact on Family Scale (Stein & Riessman, 1980) consists of 33 items, distributed across 4 subscales: (1) Financial Burden (3 items; e.g., "The illness is causing financial problems for the family"), (2) Disruption of Social Relations (9 items; e.g., "We see family and friends less because of the illness"), (3) General Family Impact (19 items; e.g., "I don't have much time left over for other family members after caring for my child") and (4) Mastery (4 items; e.g., "Because of what we have shared we are a closer family"). All items were rated on a 4-point Likert scale from 1 (*strongly agree*) to 4 (*strongly disagree*). Subscales were calculated as the sum of all relevant (reverse scored) items, and a higher score indicated higher family impact, thus worse family adjustment. The questionnaire contained good validity and reliability (Stein & Riessman, 1980). In the present study, the Cronbach's alpha coefficients were .66/.68

(Financial Impact), .74/.73 (Social Impact), .78/.71 (General Family Impact) and .38/.17 (Mastery), for fathers and mothers, respectively. Due to the low reliability of the latter subscale, this subscale was not included in the present study.

The Family Adjustment Scale (Antonovsky & Sourani, 1988) consists of 10 items and contains two subscales: satisfaction with internal family fit (8 items; e.g., "Are you satisfied with the family's way of life?") and family-community fit (2 items; e.g., "Are you satisfied with how your family fits into the neighborhood?"). Given our focus on family adjustment, the present study did not take into account the family-community fit subscale. All items are scored on a 7-point Likert scale from 1 (*totally unsatisfied*) to 7 (*totally satisfied*), with a higher score indicating higher satisfaction with internal family fit, thus better family adjustment. The FAS has good reliability (Antonovsky & Sourani, 1988). In the present study, Cronbach's alpha coefficients were .94 (fathers) and .91 (mothers).

Data Analytic Strategy

Reasons for selecting a multilevel modeling approach in the analysis of the data, rather than a single-level model, were twofold. First, the clustered sampling procedure (mothers and fathers from the same family) leads to non-independent observations: mothers and fathers from the same family tend to be more similar than mothers and fathers drawn at random from a population of parents. When using single-level methods (e.g., OLS multiple regression analysis) on non-independent data, standard errors tend to be underestimated. Such bias increases the rate of type I errors in statistical tests and may lead to incorrect statistical inference (Kenny & Judd, 1986). The multilevel approach, however, automatically adjusts for the effects of non-independent data and therefore more appropriate estimates of standard errors are obtained. Second, the multilevel approach enables us to address the relative contribution of

individual and familial influences. The relative sizes of variance components at individual (i.e., individual characteristics of the parent, differences within families) and family level (i.e., family s/he belongs to, differences between families) provide information about the level at which the main processes operate.

Four dependent variables were tested: *financial, social and general family impact* as measured by the Impact on Family Scale and *satisfaction with internal family fit* as measured by the Family Adjustment Scale. The dependent variables were predicted by covariates (*time since diagnosis, age ill child, age parent, sex parent, diagnosis, family situation (i.e., married/cohabiting, divorced, single parent or stepfamily))* and the variables of interest *psychological flexibility* (AAQ-II; individual protective factor), *dyadic coping* (supportive, common, negative dyadic coping and stress communication; DCI; intrafamilial protective factor) and *network support* (total amount of perceived network support and satisfaction with received network support; contextual protective factor). Both concurrent and prospective association models were tested for each of the four dependent variables. First, in the concurrent association models, we evaluated how baseline levels of the predictors were associated with baseline levels of the dependent variables. This relationship is assessed in the following equation:

$$Y_{ij}^{t1} = \beta_0 + b_i + \beta_1(Psychological\ flexibility)_{ij}^{t1} + \beta_2(Supportive\ DC)_{ij}^{t1} + \beta_3(Common\ DC)_{ij}^{t1} + \beta_4(Negative\ DC)_{ij}^{t1} + \beta_5(Stress\ communication)_{ij}^{t1} + \beta_6(Total\ social\ support)_{ij}^{t1} + \beta_7(Satisfaction\ with\ social\ support)_{ij}^{t1} + \beta_8(Time\ since\ diagnosis)_{ij}^{t1} + \beta_9(Diagnosis)_{ij}^{t1} + \beta_{10}(Age\ ill\ child\ at\ diagnosis)_{ij}^{t1} + \beta_{11}(Sex\ parent)_{ij}^{t1} + \beta_{12}(Age\ parent)_{ij}^{t1} + \beta_{13}(Family\ situation)_{ij}^{t1} + \varepsilon_{ij} \quad (1)$$

where there are j observations for i families. b_i is the random effect with $b_i \ \text{i.i.d.} \sim N(0, \sigma_b^2)$, allowing a different intercept for every family. In these

models, the superscript t1 indicates that only observations of time 1 are included in the analyses. ε_{ij} is the within-family error component with ε_{ij} i. i. d. $\sim N(0, \sigma_e^2)$.

Second, in the prospective association models, the dependent variables were predicted by the covariates and the variables of interest (as mentioned above), measured at the previous time-point. The time in between the two measurements varied between the participants from 1 to 32 months (M = 14, SD = 9) and was entered as an additional covariate. Time 2 measurements of the dependent variables were regressed on time 1 measurements of the predictors, following a blockwise hierarchical strategy. In the first block, covariates were entered along with time 1 status for each dependent variable to control for inherent stability. In the second block, the variables of interest were entered. We were interested in the amount of variance explained by the variables of interest that is not accounted for by previous status of the dependent variable. To formally test whether time 1 variables predicted the dependent variables at time 2 beyond initial status, we tested the statistical significance of the difference between block 1 (control for time 1 status) and block 2 (variables of interest) as indicated by the deviance statistic (-2*LogLikelihood). The model equation used in the second block was:

$$
\begin{aligned}
&Y_{ij}^{t2} \\
&= \beta_0 + b_i + \beta_1(Psychological\ flexibility)_{ij}^{t1} + \beta_2(Supportive\ DC)_{ij}^{t1} \\
&+ \beta_3(Common\ DC)_{ij}^{t1} + \beta_4(Negative\ DC)_{ij}^{t1} \\
&+ \beta_5(Stress\ communication)_{ij}^{t1} \\
&+ \beta_6(Total\ social\ support)_{ij}^{t1} \\
&+ \beta_7(Satisfaction\ with\ social\ support)_{ij}^{t1} + \beta_8(Time\ since\ diagnosis)_{ij}^{t1} \\
&+ \beta_9(Diagnosis)_{ij}^{t1} + \beta_{10}(Age\ ill\ child\ at\ diagnosis)_{ij}^{t1} \\
&+ \beta_{11}(Sex\ parent)_{ij}^{t1} + \beta_{12}(Age\ parent)_{ij}^{t1} + \beta_{13}(Family\ situation)_{ij}^{t1} \\
&+ \beta_{14}(Y_{ij}^{t1}) + \beta_{15}(T2 - T1)_{ij}^{t1} + \varepsilon_{ij}
\end{aligned}
\qquad (2)
$$

where there are j observations for i families. b_i is the random effect with $b_i \ \text{i.i.d.} \sim N(0, \sigma_b^2)$, allowing a different intercept for every family. In these models, the outcome is taken at time 2 (superscript t2), while the predictors are taken at time 1 (superscript t1). The outcome at the previous time-point was included as a predictor in the model (Y_{ij}^{t1}). ε_{ij} is the within-family error component with $\varepsilon_{ij} \ \text{i.i.d.} \sim N(0, \sigma_e^2)$.

All multilevel analyses were performed with the R-package *lmerTest* (Kuznetsova et al., 2017). Equations for the models are given in Supplementary Equations S1, S2. Continuous predictors were grand mean centered in order to aid interpretation (Schielzeth, 2010). Models were fitted with restricted maximum likelihood (REML) estimation. The ANOVA table was inspected to check for significant effects and specific hypotheses were tested. Satterthwaite's approximation was used to obtain the degrees of freedom (Sas Technical Report R-101, 1978). Model assumptions of linearity, independence, normality and homogeneity of variance were checked. The intra-class correlation coefficient (ICC) is reported as the amount of variance accounted for by differences between families rather than individual level components. For all statistical tests, significance levels were set at $p < .05$.

Results

Descriptive analyses

Table 2 shows the descriptive statistics and correlations of the variables in the present study.

Concurrent analyses

Regression coefficients and associated 95% confidence intervals (CI) for the models used are presented in Table 3.

Financial impact (IOF). Thirty percent of the variance in the model could be explained by differences between families, and 70% was caused by individual level components. None of the predictor variables were significantly associated with financial impact (all $F < 2.10$, all $p > .12$).

Social impact (IOF). Thirty six percent of the variance in the model could be explained by differences between families and 64% was caused by individual level components. Mothers reported more disruption of their social relations (higher social impact) than fathers ($F(1, 64.68) = 10.68$, $p = .002$). None of the other associations were significant (all $F < 2.93$, all $p > .09$).

General family impact (IOF). Forty percent of the variance in the model could be explained by differences between families, and 60% of the variance was caused by individual level components. More psychological flexibility was associated with less impact on the family, thus better family adjustment ($F(1, 92.72) = 5.18$, $p = .03$). In addition, higher levels of perceived network support was associated with less impact on the family, thus better family adjustment ($F(1, 92.80) = 4.35$, $p = .04$). Also, the satisfaction with the received support was of importance ($F(2, 92.23) = 4.77$, $p = .01$): parents receiving less support from their network than desired/needed (i.e. lower support satisfaction) showed a greater impact on the family, thus worse family adjustment, than those who reported to receive exactly the desired/needed amount of support ($p = .02$). Finally, the more time had passed since the diagnosis, the lower the impact on the family, thus the better the family adjustment ($F(1, 59.68) = 3.98$, $p = .05$), but this association was only marginally significant. None of the other associations reached significance (all $F < 2.94$, $p > .09$).

Table 2

Range, mean (M), standard deviation (SD) of the continuous variables of interest (psychological flexibility, dyadic coping and network support) and Pearson Correlation Coefficients between the two measurement points and between the variables of interest, aggregated over the two time-points

	Range	*M (SD)*	*N*	Cor (T1, T2)	1	2	3	4	5	6
1. Psychological flexibility	7-49	35.30 (7.56)	123	.71*	-	.16	.35*	-.32*	-.01	.15
Dyadic coping										
2. Supportive DC[1]	4-20	13.35 (2.50)	104	.64*	-	-	.73*	-.66*	.55*	.33*
3. Common DC	3-15	12.20 (2.09)	104	.55*	-	-	-	-.72*	.50*	.38*
4. Negative DC	3-15	5.97 (2.11)	104	.48*	-	-	-	-	-.31*	-.31*
5. Stress communication	2-10		105	.57*	-	-	-	-	-	.29*
		6.75 (1.76)								
6. Network support										
7. Total support	0-30	14.23 (5.09)	123	.75*	-	-	-	-	-	-

Note. [1]DC = Dyadic Coping; *p<.05

Table 3

Cross-sectional model with variables measured at baseline

Predictor	Financial impact *Coefficient B [CI]*	Social impact *Coefficient B [CI]*	General family impact *Coefficient B [CI]*	Satisfaction with internal family fit *Coefficient B [CI]*
Psychological flexibility	-.02 [-.07, .04]	-.07 [-.16, .03]	-.11 [-.21, -.02]*	.29 [.13, .44]***
Stress communication	.15 [-.08, .38]	.02 [-.39, .44]	.27 [-.13, .68]	-.06 [-.70, .59]
Supportive Dyadic Coping	-.05 [-.24,.14]	-.13 [-.47, .21]	-.15 [-.48, .18]	.44 [-.10, .97]
Common Dyadic Coping	.02 [-.24, .27]	.09 [-.37, .56]	.03 [-.42, .49]	.57 [-.15, 1.29]
Negative Dyadic Coping	-.04 [-.26, .18]	-.21 [-.61, .18]	-.19 [-.57, .19]	-.62 [-1.24, -.01]*
Total network support	-.05 [-.14, .03]	-.07 [-.22, .08]	-.16 [-.30, -.01]*	.04 [-.19, .27]
Satisfaction with network support (too few vs. enough)	1.12 [-.01, 2.26]	.83 [-1.25, 2.91]	2.57 [.48, 4.66]*	1.87 [-1.33, 5.08]
Satisfaction with network support (too much vs. enough)	-.35 [-1.54, .83]	-.67 [-2.84, 1.50]	-2.04 [-4.16, .08]	.87 [-2.48, 4.22]
Time since diagnosis	.002 [-.06, .07]	-.10 [-.22, .03]	-.13 [-.25, -.002]	-.09 [-.28, .10]
Age ill child	-.03 [-.16, .10]	-.21 [-.45, .03]	-.17 [-.41, .07]	-.06 [-.43, .30]
Diagnosis (AML[1] vs. ALL[2])	.73 [-.65, 2.12]	-.06 [-2.64, 2.51]	1.39 [-1.16, 3.94]	-.07 [-3.98, 3.83]
Diagnosis (CML[3] vs. ALL)	-1.19 [-5.39, 3.00]	-5.10 [-12.92, 2.71]	-2.98 [-10.73, 4.76]	15.52 [3.69, 27.35]*
Diagnosis[1] (Non Hodgkin vs. ALL)	-.36 [-1.60, .87]	.51 [-1.79, 2.81]	.62 [-1.68, 2.91]	0.60 [-2.88, 4.08]
Sex parent (women vs. men)	.51 [-.30,1.33]	2.44 [.98, 3.90]**	1.24 [-.18, 2.65]	0.44 [-1.87, 2.76]
Age parent	-.06 [-.15, .04]	-.05 [-.22, .12]	-.10 [-.27, .07]	.05 [-.21, .31]
Family status (Step family vs. Married)	1.00 [-1.52, 3.52]	-1.15 [-5.85, 3.54]	-2.67 [-7.31, 1.98]	-1.32 [-8.44, 5.79]
Family status (Divorced vs. Married)	-.10 [-3.11, 2.92]	-.80 [-6.34, 4.74]	-2.19 [-7.65, 3.26]	-15.29 [-23.81, -6.77]***

Note. * p < .05, ** p < .01, *** p < .001; [1] AML = Acute myeloid leukemia; [2]ALL = Acute lymphoblastic leukemia; [3]CML = Chronic myeloid leukemia; Only 1 family with CML was included in the analysis

Satisfaction with internal family fit (FAS). Twenty-nine percent of the variance in the model could be explained by differences between families and 71% was caused by individual level components. More psychological flexibility was associated with more satisfaction with internal family fit, thus better family adjustment ($F(1, 93.77) = 13.45$, $p < .001$), whereas more negative dyadic coping was associated with less satisfaction with the internal family fit, thus worse family adjustment ($F(1, 83.48) = 3.99$, $p = .049$). Finally, the family situation was also of importance ($F(2, 61.44) = 6.24$, $p = .003$): divorced parents reported less satisfaction with internal family fit, thus worse family adjustment, than married or co-habiting parents ($p < .001$). There was no significant difference between stepfamilies and nuclear families ($p = .14$). None of the other associations was significant (all $F < 2.22$, $p > .09$).

Prospective analyses

Regression coefficients and associated 95% confidence intervals (CI) for the models used for the prospective analyses are presented in Supplementary Tables (i.e., Supplementary Table 2-5; at the end of this dissertation).

Financial impact (IOF). There was a strong consistency for financial impact from time 1 to time 2 ($F(1, 92.12) = 43.29$, $p < .001$). Entry of the predictors of change improved the overall fit beyond that of time 1 status ($\chi^2(8) = 24.83$, $p = .002$). There was a significant predictive effect of psychological flexibility ($\beta = -.08$, 95% CI [-.13, -.03]; $F(1, 87.93) = 10.22$, $p = .002$): higher levels of psychological flexibility at time 1 were predictive of lower financial impact at time 2. There was also a significant predictive effect of stress communication ($\beta = -.25$, 95% CI [-.46, -.05]; $F(1, 89.61) = 5.89$, $p = .02$): more stress communication at time 1 was predictive for a lower financial impact at time 2. None of the other variables had a significant predictive effect (all $F < 3.05$, $p > .08$).

Social impact (IOF). There was a strong consistency for social impact from time 1 to time 2 ($F(1, 97.63) = 15.28$, $p < .001$). Entry of the predictors of change did not significantly improve the overall fit beyond that of time 1 status ($\chi^2(8) = 13.84$, $p = .09$), and none of the variables of interest had a significant predictive effect (all $F < 3.73$, $p > .05$).

General family impact (IOF). There was a strong consistency for general family impact from time 1 to time 2 ($F(1, 99.98) = 29.84$, $p < .001$). Entry of the predictors of change improved the overall fit beyond that of time 1 status ($\chi^2(8) = 15.61$, $p = .048$). There was a significant predictive effect of psychological flexibility at time 1 ($\beta = -.16$, 95% CI [-.26, -.06]; $F(1, 86.71) = 9.83$, $p = .002$), indicating that higher levels of psychological flexibility at time 1 were predictive for a lower impact on the family, thus better family adjustment, at time 2. There was also a significant effect of time since diagnosis ($\beta = -.07$, 95% CI [-.14, -.003]; $F(1, 74.50) = 4.17$, $p = .045$), indicating that the impact of the illness on the family was lower, thus better family adjustment, if more time had passed since diagnosis. None of the other variables had a significant predictive effect (all $F < 2.62$, $p > .10$).

Satisfaction with internal family fit (FAS). There was a strong consistency for satisfaction with internal family fit from time 1 to time 2 ($F(1, 97.97) = 21.18$, $p < .001$). Entry of the predictors of change did not significantly improve the overall fit beyond that of time 1 status ($\chi^2(8) = 9.75$, $p = .28$), and none of the variables of interest had a significant predictive effect (all $F < 2.61$, $p > .11$).

Discussion

The aim of the current study was to explore the role of potential protective factors at the individual (psychological flexibility), intrafamily (dyadic coping), and contextual level (network support) in explaining family

adjustment (i.e., financial impact, social impact, general family impact, satisfaction with internal family fit) as perceived by parents of children with leukemia or non-Hodgkin lymphoma. By taking into account protective factors at all three levels, we aimed to explain the existing variability in family outcomes when facing pediatric cancer. In addition, we investigated whether this variance was explained by individual and/or familial components; as well as the stability/changes in family adjustment across time.

Summary of Results

Psychological Flexibility and Family Adjustment. Our findings indicate that psychological flexibility, defined as an individual (here, the parents) willingness to experience unwanted or aversive stressors while pursuing one's values and goals (Hayes et al., 1999), is important for the family adjustment as perceived by parents facing leukemia/non-Hodgkin lymphoma in their child, both cross-sectionally and prospectively. This is in line with our prediction and with previous research on psychological flexibility in parents of children with cancer (Burke et al., 2014). However, different patterns of findings emerged for general and financial family consequences.

More specifically, we found that, both concurrently and prospectively, more psychological flexibility in parents was associated with a lower general impact on the family and, concurrently, higher satisfaction with internal family fit. In other words, the more a parent "accepts" his/her negative thoughts and emotions, the better the family adjustment, both concurrently and prospectively. This finding is in line with the idea that psychological flexibility is an important protective factor in predicting individual adjustment outcomes (Kashdan & Rottenberg, 2010). In addition, in the context of cancer, these negative thoughts and emotions may be centered around the illness and its treatment. Indeed, when facing pediatric cancer, psychological flexibility

may refer to a sense of acceptance of the diagnosis or the transition to be a cancer patient or to have a child with cancer, as well as the acceptance of the uncontrollable and possibly fatal nature of the illness. This "acceptance" has been shown to improve individual psychosocial outcomes in patients with cancer (Carver et al., 1993; Hulbert-Williams et al., 2015; Stanton et al., 2002) and parents of children with cancer (Burke et al., 2014).

Moreover, the present study extends previous research on psychological flexibility, as – to the best of our knowledge – it is the first investigating the association between psychological flexibility and *family* adjustment instead of individual adjustment. Based on existing literature, we know that pediatric cancer often causes parental distress post-diagnosis (i.e., anxiety, depression, post-traumatic stress symptoms, Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008), and that psychological flexibility can operate as a buffer for parental maladjustment (individual level; Burke et al., 2014). As parents play a cardinal role in their family, and parental functioning is linked to the way in which the family as a whole functions (theoretical argument: Social Ecology Model, Bronfenbrenner, 1977; empirical argument: Kashdan et al., 2004), we might assume that the underlying mechanism underneath the association between psychological flexibility and family adjustment may be the parents' individual functioning: the more parents accept their negative thoughts and emotions, the better their individual functioning (e.g., less anxiety; depression) and therefore the better the adjustment of the family as a whole. More research is needed, however, to confirm this hypothesis.

In addition, there was also a significant prospective association between psychological flexibility and the financial impact in families being confronted with pediatric cancer. So, the more parents "accept" their negative thoughts and emotions in the short term, the less they are worried about the financial consequences of the illness in the long term. It is possible that accepting negative thoughts/emotions in general, and cancer-related

thoughts/emotions in specific, helps parents to accept the financial impact as well, as these parents may potentially focus more on the well-documented "positive side-effects" of the cancer diagnosis (e.g., increased closeness within the family; Van Schoors et al., 2015; 2018a).

Dyadic Coping and Family Adjustment. Our findings indicate that dyadic coping can be linked to the adjustment of the family as perceived by parents being confronted with a cancer diagnosis in their child, both cross-sectionally and prospectively. Specifically, we found that more stress communication predicted a smaller financial impact in families being confronted with pediatric cancer (prospective finding). In other words, the more mothers and fathers shared their stress with their partner in the short term, the less they were worried about the financial consequences of the illness in the long term. Explanations are twofold. First, it is plausible to assume that couples sharing their illness-related stress, also share other worries, e.g., financial worries. As social sharing reduces stress (Rimé, 1995), we might assume that – although the objective financial impact stayed the same – the parental concerns about the financial consequences might decline with increased stress communication. Second, stress communication can be seen as a characteristic of "expressiveness" As a consequence, a parent sharing stress with his/her partner is likely to share stress with others (e.g., friends, grandparents of the diagnosed child) as well. When others know about possible financial problems in the family of the diagnosed child, they can help by, for example, giving/borrowing money or organizing benefits. This explanation is strengthened by the present data: we found a significant correlation of .27 between stress communication and the total amount of perceived network support.

Furthermore, there was an association between negative dyadic coping and satisfaction with internal family fit (cross-sectional finding). The more a parent experiences distancing, mocking or sarcasm from his/her partner when talking about the illness, the worse the perceived family

adjustment. This is in line with previous studies investigating the association between negative dyadic coping and negative (individual) outcomes in adult chronically ill populations (Meier et al., 2011) and parents of children with cancer (Van Schoors et al., 2019b). Surprisingly, however, there was no significant association between positive dyadic coping (supportive dyadic coping and common dyadic coping) and family adjustment. Explanations are twofold. First, the absence of a significant association between positive dyadic coping and family adjustment could be due to limited statistical power. Second, this finding may also suggest that positive dyadic coping is particularly important for the individual adjustment of parents being confronted with pediatric cancer (as previously found by Van Schoors et al., 2019b), and not the family adjustment. However, more research is needed to confirm this hypothesis.

Network Support and Family Adjustment. Our findings indicate that network support is important for the family adjustment as perceived by parents of children with cancer (cross-sectional finding). More specifically, we found that higher levels of network support as perceived by parents were related to a lower general impact on the family, thus better family adjustment. This finding emphasizes the importance of network support when facing pediatric cancer (Hoekstra-Weebers et al., 2001; Woodgate & Degner, 2003; Woodgate, 2006). In addition, when taking into account discrepancies and congruencies between desired and received parental support, we found that when parents received the exact amount of support they needed/desired, they reported a lower family impact, and thus better family adjustment, as compared to parents receiving less support than needed/desired. To note, no significant differences were found between parents receiving the exact amount of support and parents receiving more support than needed/desired. This is in contrast to some studies (e.g., Siewert et al., 2011) showing "the more, the better"; i.e. that the overprovision of support is related to higher well-being.

Other Findings. The results of the present study furthermore revealed the importance of *sex, time since diagnosis* and the *family situation* in the prediction of family adjustment. First, mothers reported a higher social impact of their child's illness than fathers (cross-sectional finding). Indeed, in most of the included families the mother (temporally) has quit her job to ensure that always one parent could accompany the diagnosed child to the hospital, whereas the father kept working to ensure financial security. As a consequence, whereas the mother's daily life changed completely, the father's daily activities stayed more or less the same as prediagnosis (Van Schoors et al., 2018b). Second, parents living in a family with a child who has been diagnosed more recently reported worse family adjustment than those who had been exposed to the illness for a more prolonged period of time, both cross-sectionally and prospectively. This can be linked to the treatment course of the cancer, and the intensity of the hospitalizations needed to cure the child. Whereas at diagnosis, intense treatment with long hospitalizations are needed, these hospitalizations decrease over time with only one-day care treatment after some months/a year. As especially being separated as a family (mother and ill child in the hospital vs. father and siblings at home) is hard to handle for the different family members (Van Schoors et al., 2018a, 2018b), decreased hospitalizations of the ill child can be linked to more time together as one family, and thus better family adjustment. Third, divorced parents reported a lower satisfaction with internal family fit, thus worse family adjustment, than married or cohabiting parents (cross-sectional finding). We might assume that working together as a team (mother and father) helps a parent to cope with the cancer diagnosis, and therefore helps the family as a whole to fulfill all family needs (e.g., individual needs of all family members including those of siblings, household needs, financial needs) (Van Schoors et al., 2018b), whereas divorced parents are mostly obliged to manage the cancer situation alone. This explanation is strengthened by the present study's finding that the family adjustment is comparable for nuclear families and

stepfamilies, emphasizing the need to divide family tasks in order to keep their head up in these difficult times.

Furthermore, for all outcomes of interest (financial impact, social impact, general family impact, satisfaction with internal family fit) both individual characteristics and the differences between families seem to be important. In other words, in predicting family adjustment, researchers should take into account *who* (individual characteristics of the parent) is reporting, as well as the *family* s/he belongs to. This is in line with a recent study on the individual adjustment of family members (patients, mothers, fathers, siblings) facing pediatric cancer (Van Schoors et al., 2019a).

Finally, the present study found that family adjustment at time 1 was an important predictor for family adjustment at time 2. This indicates that the relative adjustment of families compared to other families remains stable: families who score relatively high at time 1 will still score relatively high at time 2 and vice versa. In addition, within each family, the adjustment improves over time as evidenced by additional analyses that included *time* as predictor variable (general impact: β_{time} = -1.07, p = .004; financial impact: β_{time} = -.39, p = .03; social: β_{time} = -1.85, p < .001 ; satisfaction with internal family fit: β_{time} = -1.72, p = .03). This is in line with existing quantitative literature showing that although – over time – families return to "normal" again (van Buiren et al., 1998), discrepancies between families occur in the adaptation process post-diagnosis. Indeed, the Pediatric Psychosocial Preventative Health Model (PPPHM; Kazak, 2006) divides families of children in pediatric health care settings into three groups: (1) the so-called *Universal* group, which is the largest group and consisting of families showing at least moderately resiliency and possessing adequate to strong coping abilities, (2) the *Targeted* group includes those families at higher risk and in need of some services and (3) the *Clinical/Treatment* group refers to families at highest risk; showing more evident symptomatology. In order to facilitate

bon-adjustment post-diagnosis, family needs should be matched with clinical services.

Strengths and Limitations

A first strength of the present study is the longitudinal design. By taken into account two measurements, we were able to examine the temporal order of the associations under investigation. Second, although most studies in the pediatric cancer literature make use of a single-family member participant (Van Schoors et al., 2015), we included the perspectives of both parents. Third, protective factors at all three levels (individual level, intrafamilial level and contextual level) were included in the present study, whereas previous research mostly focused on only one of these levels. As a consequence, to the contrary of most existing studies who provided only a fragmented explanation of the processes underlying post-diagnostic family adjustment, we were able to present a more complete picture of factors fostering adjustment in families facing pediatric cancer.

Despite the strengths of the study, there were also some limitations. A first limitation is the small sample size. With only 70 mothers and 53 fathers, we can only draw limited conclusions regarding the association between psychological flexibility, dyadic coping, network support and family adjustment, as perceived by these parents. In addition, given the small sample size and the high number of included variables, non-significant results could also be due to limited power. Further research, with larger samples, are therefore needed to confirm our findings. Second, our sample consisted of Caucasian, heterosexual couples, thereby limiting the generalizability of our results. Future research should attempt to replicate these findings with more heterogeneous samples, e.g., homosexual couples. In addition, the Dutch language was an inclusion criterion for participation in the study. With respect to the current multicultural society, however, this language criterion might

have been a barrier for ethnic minorities. Third, we only focused on families with leukemia or non-Hodgkin lymphoma in one of the children. It is important to highlight that parents of children with other cancer diagnoses, e.g., brain tumors, may have different experiences. Fourth, mean time since diagnosis was 5.26 (T1) and 18.86 (T2) months post-diagnosis. In order to best capture the adaptation process post diagnosis, however, the first measurement should be as close as possible to the moment of diagnosis. In addition, researchers should include comparison groups (e.g., families with healthy children; families with a suspicion of pediatric cancer but no actual diagnosis, families where one child is diagnosed with a brain tumor), so more information about cancer specific processes vs. processes that are similar across specific health-related conditions can be explored. Fifth, the mean age of the patients in the present study was 6.96 years. Most of the patients were toddlers and primary school children. Further research with families of adolescents and young adults with cancer (AYA's) is needed, as the developmental stage of the children may indeed influence factors important for family functioning. Sixth, although we included predictive variables at all three levels (individual, intrafamily and contextual level), only one variable per level was selected. Further research should investigate other predictive variables, at all three levels. Finally, as one of the main reasons for non-participation was being emotionally overwhelmed by the diagnosis, we might assume that especially more resilient families participated in our study (selection bias). Other findings might occur for emotionally distressed families.

Clinical Implications

Our findings provide evidence that a pediatric cancer diagnosis not only impacts the individual functioning of the different family members, but also the family functioning. Three specific recommendations arise from the study findings. First, clinical interventions should be tailored to gender

differences and specific characteristics of mothers and fathers facing pediatric cancer. Indeed, our findings suggest that mothers might be in greater need of psychosocial support, as they perceived the social disruption post-diagnosis as more severe. Second, across our findings, especially the association between psychological flexibility (individual protective factor) and family adjustment seems to be important. As a consequence, families could in particular benefit from interventions targeting the promotion of acceptance of unwanted negative thoughts and emotions, e.g., using Cognitive Behavioral Therapy or Acceptance and Commitment Therapy (Hayes et al., 2012). Third, when facing pediatric cancer, a holistic approach - including individual, couple and family interventions - is needed to best help families to cope with this severe stressor. Indeed, the findings of the present study showed that protective factors at all three levels (individual, intrafamilial, and contextual level) are important for the adjustment of the family as a whole. Moreover, as family adjustment is both explained by individual characteristics ("who filled in the questionnaire?") and differences between families ("the family s/he belongs to"), both the individuality of each family members, as well as the mutual and bidirectional influences within families should be taken into account by clinicians.

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Supplementary Equation - S1

For the concurrent association models, this was the general equation:

$$Y_{ij}^{t1} = \beta_0 + b_i + \beta_1 X_{ij}^{t1} + \cdots + \varepsilon_{ij}$$

There are j family members for i families. b_i is the random effect with b_i i. i. d. $\sim \mathrm{N}(0, \sigma_{\mathrm{b}}^2)$, allowing a different intercept for every family. In these models, the superscript t1 indicates that only observations of time 1 are included in the analyses. ε_{ij} is the within-family error component with $\varepsilon_{ij}\ i.i.d. \sim N(0, \sigma_e^2)$.

Supplementary Equation - S2

For the prospective association models, this was the general equation:

$$Y_{ij}^{t2} = \beta_0 + b_i + \beta_1 X_{ij}^{t1} + \beta_2 Y_{ij}^{t1} + \cdots + \varepsilon_{ij}$$

There are j family members for i families. b_i is the random effect with b_i i. i. d. $\sim \mathrm{N}(0, \sigma_{\mathrm{b}}^2)$, allowing a different intercept for every family. In these models, the outcome is taken at time 2 (superscript t2), while the predictors are taken at time 1 (superscript t1). The outcome at the previous time-point was included as a predictor in the model ($\mathrm{Y}_{\mathrm{ij}}^{\mathrm{t1}}$) . ε_{ij} is the within-family error component with $\varepsilon_{ij}\ i.i.d. \sim N(0, \sigma_e^2)$.

CHAPTER 3

PARENTS' PERSPECTIVES OF CHANGES WITHIN THE FAMILY FUNCTIONING AFTER A PEDIATRIC CANCER DIAGNOSIS[1]

Pediatric cancer is a life-threatening disease that challenges the life of the diagnosed child, the parents, and possible siblings. Moreover, it also places considerable demands on family life. The aim of this study was to explore changes in the family functioning after a pediatric cancer diagnosis. Ten couples parenting a child with leukemia or non-Hodgkin lymphoma were interviewed individually about their experiences. Interviews were semistructured and the data were analyzed using Multi Family Member Interview Analysis. Three themes emerged from the data: (a) Family Cohesion: Strengthened Versus Fragmented; (b) Educational Norms and Values: Overindulgence Versus Being Stricter, and (c) Normality: Loss Versus Preservation. The conflicting dynamics present in these emerging themes exemplify the complexity of this process of family adaptation. This study illustrates the need to take into account the family level, as well as the conflicting feelings parents may experience after a pediatric cancer diagnosis.

[1]Van Schoors, M., De Mol, J., Morren, H., Verhofstadt, L. L., Goubert, L., & Van Parys, H. (2018). Parents' perspectives of changes within the family functioning after a pediatric cancer diagnosis: A multi family member interview analysis. *Qualitative Health Research, 28,* 1229-1241. doi: 10.1177/1049732317753587

Introduction

Pediatric cancer is the second most common cause of death in children in developed countries (Kaatsch, 2010). Although this disease used to be mostly fatal, an increasing number of children now survive – currently around 82% of all cancer-affected children survive for five or more years (Cancer Research UK, 2010). Like all chronic diseases, cancer may have a significant impact on the life of the diagnosed child (Kazak et al., 2001; Kestler & LoBiondo-Wood, 2012) and the family members (Alderfer et al., 2010; Grootenhuis & Last, 1997; Pai et al., 2007). Therefore, a growing number of studies have focused upon detrimental and protective factors for the adaptation of patients (e.g., Gliga et al., 2016), siblings (e.g., Barrera et al., 2004), and parents (e.g., Caes et al., 2014; Wijnberg-Williams et al., 2015), in order to promote long-term resilience in all family members and help families cope with the disease more effectively. In addition, according to recent systematic reviews, certain family characteristics (e.g., cohesion and adaptability) may determine the family members' ability to adapt to life after diagnosis (Long & Marsland, 2011; Van Schoors et al., 2017). Indeed, according to the family psychology literature (Carr, 2012) children are embedded in a family, and within families, individual family members influence each another. This idea is also embedded within various family systems models often applied to chronic illness populations (Van Schoors et al., 2016). For example, the Social Ecology Model (Bronfenbrenner, 1977) illustrates how the child is nested within and influenced by the family system in addition to other social systems; whereas the double ABCX model (McCubbin et al., 1980) posits that certain aspects of family functioning can either foster or undermine individual adjustment to illness or disability. In the case of a childhood cancer diagnosis, families must be flexible in their roles and responsibilities, communicate effectively, manage emotions, and successfully work as a team in order to meet treatment demands (Kazak et al., 2004; Marcus, 2012), demonstrating the impact on the family level and the role of family functioning as predictor

of individual family member adaptation (Van Schoors et al., 2017; Van Schoors et al., 2015).

The existing research in pediatric cancer is limited in three ways. First, most research on the subject to date has assessed the connection between detrimental and protective factors and the participants' adaptability to life after diagnosis, using questionnaires and heterogeneous samples, covering a broad range of diagnoses, child ages, and time periods since diagnosis or treatment (Van Schoors et al., 2015). These methods, however, cannot capture the unique experience of a family confronted with such a diagnosis, as well as the meaning that family members give to their unique situation (Smith et al., 2009). Second, despite a growing awareness regarding the role of family functioning in the context of pediatric cancer, most studies tend to overlook the family system level, and focus solely on the individual level (e.g., the diagnosed child or their parents). This approach has limitations when applied to a clinical context or screening strategies, as, for example, literature has already revealed associations between (mal)adaptive family functioning and child adjustment problems (Van Schoors et al., 2017). Third, the majority of studies that focus on the family functioning only included responses from a single family member. This approach, however, may not adequately reflect the family life in its entirety (Van Schoors et al., 2015).

To address these limitations, the current qualitative study was conducted among parents of children with leukemia or non-Hodgkin lymphoma (a) to provide insight into personal accounts of parents' experiences, and (b) to obtain in-depth descriptions of parents' perspectives on changes in family functioning after a pediatric cancer diagnosis. In addition, (c) one-to-one interviews were conducted with the mother and father separately. This allowed each parent to provide their own perspective on shifts in family life post-diagnosis (Eisikovits & Koren, 2010), without having to factor in their partner's feelings (Morris, 2001).

Method

Multi Family Member Interview Analysis (MFMIA; Van Parys et al., 2017) was used as a methodological framework to analyze the individual interviews, focusing on the couple as the unit of analysis. This approach takes into account ethical and methodological challenges inherent to interviewing couples (Taylor & de Vocht, 2011; Ummel & Achille, 2016) and has proved effective in studies that analyze experiences shared by a couple, particularly when assessing sensitive topics such as adjustment to an illness (Eisikovits & Koren, 2010).

Participants

Ten married couples with children diagnosed with leukemia or non-Hodgkin lymphoma participated in the study. They were all Caucasian, living in the Flemish part of Belgium and aged between 37 and 56 years of age, representing a reasonably homogenous sample that conforms to the requirements of Interpretative Phenomenological Analysis (IPA; Smith et al., 2009). The children (seven males and three females) were either diagnosed with acute lymphoblastic leukemia (N = 6), acute myeloid leukemia (N = 1) or non-Hodgkin lymphoma (N = 3). The diagnosed child's age ranged from four to 16 years. Time since diagnosis varied from six to 33 months (M = 21.6). In two families, the diagnosed child was their only child. The remaining families had either two (three families), three (three families), or four (two families) children. Ethical approval from the University Hospitals of Ghent, Brussels, Antwerp, and Louvain had been secured for the study and the appropriate informed consent forms were obtained.

Data Collection

This study is part of a larger ongoing study in Flanders (Belgium) examining the impact of a pediatric cancer diagnosis on families, that is, the "UGhent Families and Childhood Cancer study". For this large-scale study, families of children diagnosed with leukemia or non-Hodgkin lymphoma between the age of one and 18 years were invited to take part in a longitudinal survey. Exclusion criteria were as follows: (a) not speaking Dutch, (b) expression of a developmental disorder in the diagnosed child, and (c) relapse. All participating parents (N = 173 individuals, including 55 couples) were subsequently invited to complete an interview about the impact of the cancer diagnosis on the functioning of their family. In 33 of the participating couples (60%), both partners agreed to attend an interview. Ten of these couples were randomly selected and contacted by H.M.. All interviews were conducted by the same interviewer (H.M.), were audio recorded and lasted 60 to 120 minutes. Verbatim transcripts of these interviews served as the raw data for this study. All interviews were based on an interview schedule and consisted of open-ended questions about (a) the experience of the diagnostic and treatment process, (b) the impact of the diagnosis on the parent, (c) the family relationships, and (d) the family functioning (interview details available upon request). The participants' experiential accounts were facilitated by prompts, in order to encourage the participants to give personal accounts (Smith et al., 2009).

Analysis

Data consisted of one-to-one interviews with each mother and father separately about the impact of the cancer diagnosis on their family functioning. In addition to the transcripts, further data were supplied by a task that required the participants to demonstrate the emotional bond between their family members through arranging puppets (i.e., figural technique based on

the Family System Test; Gehring & Wyler, 1986): The closer they positioned the puppets, the stronger the family cohesion. The results of the task were referred to throughout the interview and informed data analysis.

Inspired by IPA (Smith et al., 2009) and dyadic interview analysis (Eisikovits & Koren, 2010), MFMIA (Van Parys et al., 2017) allows detailed and systematic analysis of shared family experiences (Smith, 1999; Van Parys et al., 2017). In a first phase, all interviews were analyzed separately, using the principles of IPA. Each transcript was read a number of times by M.V.S. in order to familiarize herself with the participant's account. The transcript was then annotated with initial observations. Next, these initial notes (e.g., "it seems important for this father to continue the siblings' hobbies") were translated into more general themes (e.g., "life should go on"). Then, parallels were explored between these emerging themes. This analytical and theoretical step results in a clustering of themes for each of the cases. This process was repeated for each case. At the second stage, when each individual transcript had been analyzed, themes that were relevant to each couple, so *within couples*, were identified by combining the themes of both partners. In a third phase, we searched for parallel themes between couples from different families. The final list of subordinate and superordinate themes reflects patterns of convergence between different couples, so *across couples*, based on analysis of unique aspects of each parent's and each couple's experiences. As a consequence, we were not interested in gender differences, but only in the complex feelings experienced by a couple following a pediatric cancer diagnosis. Finally, all themes were translated into a written account, elaborating on the analysis and illustrating it with direct quotes from the participants. Pseudonyms were given to protect the anonymity of the participants.

As interpretations may be influenced by personal experiences and one's own theoretical background, a team of auditors (H.V.P. and J.D.M.) was invited to challenge the way M.V.S. constructed themes and subthemes at several points in the analysis (Hill et al., 1997), and to assess to what extent

the analysis has been conducted systematically, transparently, and credibly (see Smith et al., 2009 for more details on IPA). M.V.S., who analyzed the transcripts, is a clinical psychologist and PhD student. She is also trained in Psycho-Oncology, and through her PhD is in regular contact with staff and families in pediatric cancer departments in Flanders. H.V.P. is a clinical psychologist and postdoctoral researcher with experience in qualitative research in the field of family psychology and family therapy. She was the first auditor for this study. J.D.M. is a clinical psychologist and associate professor who specializes in qualitative research. In the study, he functioned as the second auditor and notably contributed to the analysis of emergent themes.

Results

The changes in the family functioning perceived by parents were clustered into three main themes: (a) Family Cohesion: Strengthened Versus Fragmented, (b) Educational Norms and Values: Overindulgence Versus Being Stricter, and (c) Normality: Loss Versus Preservation. Each of these themes comprised several subordinate themes (see Figure 1). In addition, the complexity of the family adaptation process after a pediatric cancer diagnosis was marked by conflicting dynamics within these emerging themes. Specifically, in the first theme, the family is perceived as a stronger unit. However, at the same time, fragmentations in the family unit are also experienced, including a shift in focus toward the diagnosed child, at the cost of attention on the family as a whole, the siblings, and the couple themselves. In the second theme, parents identify the need for a new parenting approach, one that compensates for the suffering of the diagnosed child by overindulgence. At the same time, however, parents believe the child will heal and feel responsible for the child becoming a responsible adult. Therefore, parents adopt a stricter parenting approach than pre-diagnosis, in order to

compensate for their overindulgence. The third theme articulates the overwhelming impact of the cancer diagnosis on the family, which is often described by the parents as "nothing is normal anymore". However, at the same time, families tend to strive for normality and try to safeguard the normal life of family members.

Theme 1: Family Cohesion: Strengthened vs. Fragmented

Subtheme 1a: Being closer as a family. For most parents, the illness drew their family members closer together. This increased closeness was, for some parents, most notable at the difficult moments throughout the illness, as at those times family members stuck together and supported each other.

> *I do think that, in the end, we were a closer family, we were a closed circle and not much could come between us. (Mother of a boy, 14 years)*

For this mother, support was provided by the family itself. Visualized as a closed circle, family members stood close together, with limited space for others to join "the circle" or to come between them. As a consequence, it may be difficult for others (e.g., friends) to understand how these families feel and how they could help. In addition, some parents not only described their family as growing closer post-diagnosis, but also as playing a more important role. They recalled an increased desire to spend more time together as a family, instead of (for example) focusing on their careers.

> *The world stopped turning. I enjoy life more. Let's say, I used to live for my job and my career, but now I want to enjoy things more. Enjoying it for the full 100% and going on a holiday with the children. (Father of a boy, 4 years)*

These parents started to change their attitude to life: their family came to play a major role in their sense of self, and extra-familial things became less important.

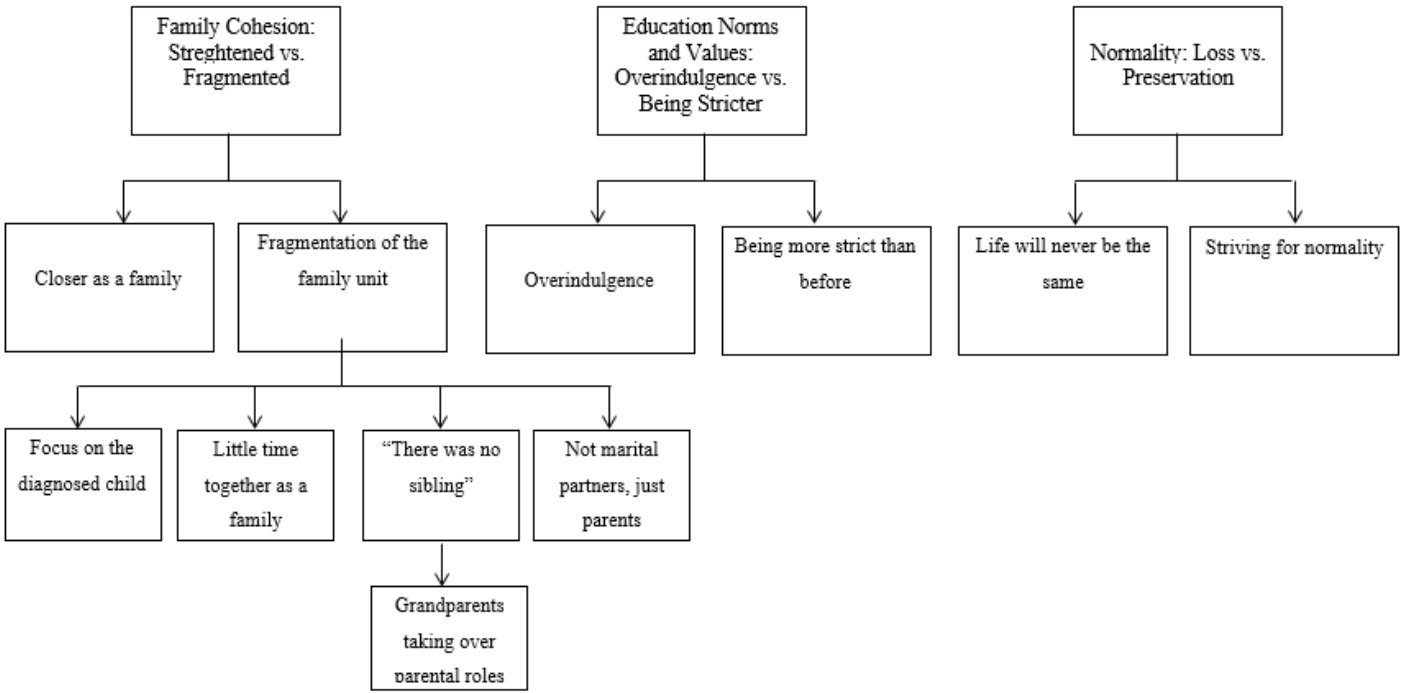

Figure 1. Superordinate and subordinate themes

Subtheme 1b: Fragmentation of the Family Unit. From the moment of the cancer diagnosis, the diagnosed child became the center of focus in the family. As a consequence, all parental time and attention were focused on this child, strengthening the bond between parent(s) and patient. At the same time, however, this shift in focus often puts a strain on relationships with healthy siblings, and as marital partners, creating fragmentations in the idea of "the family as one unit".

Focus on the diagnosed child. Due to the fatal character of a cancer diagnosis, the parental desire to divide time and attention equally over the children changed in a merely unique focus on the diagnosed child. This was, for example, described by both parents of a 14-year-old boy:

> *You just focus on that child. Like being there for him when he feels down, to cheer him up again. (Mother).*
>
> *Rick actually always comes first. (Father).*

In all families, the pediatric cancer diagnosis resulted in a stronger emotional bond between the parent(s) and diagnosed child, while the relationship between parent and siblings remained unchanged.

> *Victor made me a father, and I'm very grateful to him for that. He used to be a real pain in the ass, believe me. I loved him and he didn't love me. And what happened with the illness, we became a lot closer. (Father of a boy, 6 years)*

Parents seemed to struggle with this shift in attention to one child at the expense of the others. Given that such parental behavior differs from the general normative expectations that each child will be loved in the same way, some parents tried to rationalize their behavior. For example, parents explained the increased closeness between themselves and the diagnosed child as a result of the child's increased vulnerability. After all, due to the side effects of the treatment, most children undergoing chemotherapy could no longer take care of themselves.

> *I suppose that now I have a much stronger bond with my son than most parents would have with their eldest child. Because, right before puberty, so drastic, wiping his bum again … (Father of a boy, 14 years)*

In some families, a different impact on the bond between diagnosed child and each of the parents was identified. Parents attributed this to the fact that on a couple level, one parent became the main caregiver of the diagnosed child and quit his or her job in order to accompany the child to the hospital, whereas the other parent continued his or her work in order to guarantee financial security. The bond between the diagnosed child and the main caregiver was strengthened, whereas the impact on the bond with the other parent was less pronounced.

> *He is much more attached to my wife these days because she has been at home for the whole period. She has always been with him so. But I think it's normal, that the one they see the most … (Father of a boy, 4 years)*

Finally, the continual presence of the carer tended to result in enmeshment. Children may become used to the constant presence and help of this parent, making the transition to autonomy a greater challenge.

> *Due to the fact that you're together 24/7 for half a year, and also supporting her in difficult moments, because those injections are not much fun. So Mummy's there for everything and in the long run Mummy needs to be there for the stupidest things, things she could do perfectly well herself. (Mother of a girl, 5 years)*

Little time together as a family. In many families, the parents worked hard to ensure that one of them was always at hospital, accompanying the diagnosed child, while the other stayed at home with the siblings, or went to work. These roles were often switched regularly, so both parents could support the diagnosed child and take care of the siblings.

> *My husband and I alternated: I stayed with Talia in the hospital for a couple of days and then I went home for a moment so the other children could see me as well and then my husband was in the hospital. (Mother of a girl, 5 years)*

Aside from being preoccupied with the care of the diagnosed child and the desire to spend as much time as possible with this child, parents were also concerned with their parental duty to any siblings. They seemed to struggle with their desire to always accompany the diagnosed child, therefore not providing adequate care for the siblings. In addition, parents reported feeling guilty and obligated to divide their time and attention between all children. This pursuit of a balance was a common theme among the couples.

> *At the beginning, it's really hard, you need to find a balance between the hospital admissions and time at home with the other children, somehow trying to be one family. (Mother of a boy, 4 years)*

The words "trying to be *one* family" are notable and recur in other interviews. However, the impossibility of caring for the diagnosed child and maintaining their parental role towards the siblings could cause fragmentation of the family unit. The emotional struggle aside, it also was practically impossible for parents to be simultaneously at the hospital and at home. Consequently, it seemed unavoidable for most families that the family relationships would become strained

> *In the beginning, your family life falls apart; boom, you fall down an abyss so to speak. (Father of a boy, 14 years)*

"There was no sibling". The disease not only resulted in less family time but specifically in less parental time and attention for any siblings. During treatment, siblings were "in the background" of the family.

> *A huge amount of your time and attention goes to the one child undergoing treatment, and the other children get, yeah, they're a little bit in the background. (Father of a girl, 5 years)*

Although parental attention was mainly focused on the diagnosed child, families differed in their approaches to the siblings. In two families, the sibling was a newborn baby, and because of breastfeeding, the baby was always with the mother, whereas the father barely saw the baby. Both parents of a 6-year-old boy reported:

> *I was at the hospital with two children, because at that time I still breastfed her, so it all was a bit crazy. (Mother)*
>
> *There was no relationship with [name sibling] that really was something, there was no daughter right. (Father)*

In addition, parents indicated that the siblings had to cope with this extreme stressor with only limited parental support. And although parents were aware of this situation and felt guilty about it, they saw no other solution at that time.

> *The treatment is so intensive and relatively little attention was dedicated to [name sibling]. That's what I feel guilty about. He had to cope without us. I really struggle with that. I just hope that he will not blame us for it later, that we weren't there enough for him. And if it gets that far, and he takes it badly, then I will be very humble and not try to find excuses. Then I will say "you're right. But I don't know how we could have done it differently". (Father of a boy, 9 years)*

Apart from feelings of guilt, parents also expressed a worry that they would later be blamed by their other children. This not only seemed unavoidable, but also understandable to the parents. In addition, we noted that in the context of pediatric cancer, parents are confronted with overwhelming feelings of helplessness and situations in which they need to depend on others. For example, in the case of treatment of the diagnosed child, parents depend on the medical team; and to fully meet the needs of the siblings, parents depend on others to take care of them (see "*Grandparents taking over parental roles*"). Consequently, parents did what they thought was best, and could only hope the sibling would understand, both now and later in life.

Grandparents taking over parental roles. In most families, other family members took care of the siblings, helping them to cope with this life event.

> *I think the biggest change was for the two eldest, because in that period, they were mostly looked after and brought up by their grandparents. (Father of a girl, 5 years)*

It is unclear from the data whether involved grandparents enabled parents to spend as much as possible in the hospital or whether they merely filled the parental vacuum. Nevertheless, parents always remained committed to the siblings' well-being, as even in their absence they tried to make the best possible arrangements for them.

> *I thought it was important that the siblings could stay at home, I didn't want them to go from one set of grandparents to the other, I preferred that they stayed at home and the grandparents came to them. (Mother of a girl, 5 years)*

Although the grandparent's care was usually practical and exerted little influence on the relationship between the parents and the siblings, one family experienced a degree of estrangement between parents and child.

> *[Name sibling] has been with my Mum a lot at that time. So, one time when Victor was doing very badly, I tried to go to her. She was afraid of me and she crawled to my Mum… (Mother of a boy, 6 years)*

It seemed that this mother was rather upset by the observation that her child temporarily formed a closer bond with the grandparent than with her. After all, every parent wants their children to love them, even in the context of pediatric cancer where parents feel obligated to focus their time and attention on one child. Furthermore, after treatment is completed, parents may have to deal with the aftermath of this disruption to family life. Siblings may have become accustomed to living with the grandparents and difficulties arise when the sibling has to move home again.

> *He got used to being with his grandparents all the time. And it was very difficult to get him to come back home. (Mother of a girl, 9 years)*

The help and support the parents get from other family members seemed to be necessary to fulfill not only their own needs (i.e., spending as much time as possible with the diagnosed child) but also the needs of the siblings. However, grandparents taking care of the siblings may also disrupt family functioning.

Not marital partners, just parents. The focus on the diagnosed child also has consequences for the parents' intimate relationship. As both parents tried to accompany the diagnosed child as much as possible to the hospital and divided their remaining time between the siblings, their jobs, and the household, little time was left to spend as marital partners.

> *It's been either my husband who came here (to the hospital) or myself, we always split it up, we were seldom here together. (Mother of a girl, 9 years)*

Parents rarely spent time together and they felt like their lives as partners, beyond their lives as parents, had disappeared.

> *We used to have many shared activities, like going to theatre or making city trips together without the children. We really tried to look for moments where we could "do our thing" together. This became harder to do. Going out together sometimes is a problem; we always ask ourselves "is she ok?" Is anything wrong? She also fainted a couple of times and actually that is enough reason to never leave her alone. (Father of a girl, 16 years)*

Rather than a lack of love, parents reported that worries about their child's health prevented them from spending time together. In addition, most parents downplayed the impact of the cancer diagnosis on the couple subsystem and emphasized that this event was just one of many affecting their relationship.

> *Whether many things changed? I don't know, I don't think so. Let's say we'd known each other for 15 years and now we've known each other for 17 years. I mean, I don't think so actually. (Father of a boy, 4 years)*

In contrast, for some parents, the disease did mark the relationship and made the couple subsystem less clearly defined. One parent described that their focus was redirected toward the children, resulting in a greater emotional distance between the parents.

> *As a couple we are a bit distanced from each other these days. While we used to feel like "we have our three children, and then there's us and then there's the family". Lynn, well not Lynn but the illness, has meant that my wife and I have grown a bit apart from each other, and that our focus is more on our three children. (Father of a girl, 16 years)*

So, during cancer treatment, it became even harder to combine a parental role with a partner role. Their love and time for the diagnosed child was unconditional, even at the cost of their own intimacy. However, despite these obstacles, almost all of the couples indicated that the cancer diagnosis did not threaten their marital relationship.

Theme 2: Educational Norms and Values: Overindulgence Versus Being Stricter

Subtheme 2a: Overindulgence. Parents indicated that the illness necessitated a different approach to child- rearing.

> *You need to adapt your parenting style completely, not just a little bit but completely. I don't know, is it 180 degrees, yes – otherwise we're back, so 180 degrees. Completely changing it. (Father of a boy, 6 years)*

Parents started to indulge the diagnosed child more, especially shortly after diagnosis. To justify this overindulgence, several reasons were given (e.g., to compensate for the suffering, to persuade the child to eat). Furthermore, it seemed like this overindulgence was an attempt not only to compensate for the illness but also to make life easier (both during hospital stays and at home)

and to avoid family conflict. Given the demanding nature of a cancer diagnosis, parents may after all lack the energy to maintain their pedagogical principles. On a couple level, couples mainly gave the same reasons for this overindulgence.

> *Victor used to be raised quite strictly. We intended to do everything like it should be done. No Coca-Cola, DVDs, IPad, ….. In retrospect this was a stupid idea, but ok. The advantage was that once he had to go to the hospital, he was allowed for once to watch a movie and … Because there is no other way, you need to keep him busy. (Father of a boy, 6 years)*
>
> *With regard to rearing, I think it was harder to determine what was allowed and what not. Victor was allowed to do things that before I could never have imagined for a three or four year old. But you need to keep him busy. That's a form of compensation. (Mother of a boy, 6 years)*

Parents emphasized that this behavior occurred unconsciously: although they did not want to let go of all their pedagogical principles and they did not want to favor one child, the cancer situation forced them to do so.

> *Of course, the one who's ill keeps on requiring your attention. And that one will be allowed a little bit more than the two others, unconsciously. You will protect him more. But will you privilege him? Consciously? No. Unconsciously? Yes, because he has gone through so many things, our little boy… (Father of a boy, 4 years)*

Parents seemed to make a distinction between rearing the diagnosed child and rearing their siblings. They were not only more concerned about the diagnosed child but also indulged this child more. In rearing the diagnosed child, the parents had to consider the possibility of losing the child, as well as their responsibility as a parent to set limits. In contrast, when rearing any siblings, parents could focus on their long-term responsibilities – their strict behavior could be justified in the long run and accidental conflicts could be resolved. In addition, this favoritism was not only a parental concern; it also had an

actual impact on the siblings' behavior. Some parents described their other children as showing feelings of jealousy towards the diagnosed child, as well as resentment that their parents' attention was exclusively focused on the diagnosed child.

> *The big ones resent me for that sometimes, especially [name sibling], she tells Talia once in a while "Just because you have cancer doesn't mean that you can do everything" or "that you can claim Mummy". (Mother of a girl, 5 years)*

In addition, an undermining of parental authority was reported.

> *Even my authority is affected a little bit, I guess. Although when I really tell him off, he takes it seriously. My wife's authority is affected dramatically. (Father of a boy, 12 years)*

The fact that the authority of the main caregiver was particularly affected may be linked to the fact that they spent most of the time together, and this caregiver was a daily witness to the child's suffering.

Subtheme 2b: Being more strict than pre-diagnosis. Although in the short-term overindulgence may have positive effects on the child (e.g., comforting the child) and the parents (e.g., avoiding conflict), parents were also worried about the potential negative consequences of overindulgence on their child's development, as this may produce undesirable and immature behavior.

> *You feel compassion for your child, so you give in more. But also, you realize "we're aiming for recovery here, so after this, we need to make sure that we can still manage him". (Father of a boy, 12 years)*

One way to deal with this concern is trying to "find a balance" between overindulgence and setting rules.

> *It really is an adaptation and it's difficult to find a balance again. Because, he was so sick, you would, let's say, allow a lot of things. Punishing a child is something you don't do in that kind of moment. (Mother of a boy, 4 years)*

Two things are notable. First, finding balance is hard. Parents feel torn between an awareness of the dangers of overindulgence and a desire to comfort their child. Although the overindulgence may have a positive short-term effect – it makes the child happy – and a negative long-term effect – behavioral problems down the line – it can be reversed with the adoption of a stricter approach to parenting after treatment. Indeed, setting limits produces desirable behavior in the long term, but may be difficult to impose in the short term, as it may create conflict between parent and child. Furthermore, it seemed that this balance is only achieved *after* the intensive treatment period. Rather than alternating between an indulgent and a strict approach to parenting during the cancer treatment, parents tended to indulge their child during treatment and discipline them after the cancer treatment.

> *I realize that I'm more strict now, 'cause I think he was spoiled last year and we need to make that right. (Mother of a boy, 14 years)*

Parents try to compensate for all the things they allowed shortly after diagnosis, by adopting a stricter approach to parenting than before diagnosis. Thus, both overindulgent and strict approaches are magnified in this context.

Theme 3: Normality: Loss vs. Preservation

Subtheme 3a: Life will never be the same. As a result of the cancer diagnosis, family life changed.

> *I have moved a stone in the river and the river will never flow in the same way again. That's a song. Actually the illness is the same. We will always be that family, but this has changed the flow and so it's going to flow differently. When Lynn is better, we won't return to the same place. (Father of a girl, 16 years)*

And although parents emphasized that life would be different, most did not mention whether this change was good or bad. For some families, the diagnosis even improved their family functioning.

> *I'm gonna say something, but I know that at this point, it's a weird or misplaced comment: "I hope that in one year, I will be able to say that in fact it's been a very bad period, but it has had a positive influence." I can't say I'll be 'glad', because everybody is suffering, especially Lynn. But if it has to be like this, then we've done a good job and we can look back at the course of treatment with satisfaction. (Father of a girl, 16 years)*

When we looked into detail which aspects of life are in particular changed after diagnosis, all families experienced increased anxiety about the health of the diagnosed child. Although previously child illness was just a part of life, every sign of illness became a reason to panic. Notably, this catastrophizing was only about the health of the diagnosed child, and not the health of the siblings.

> *In the old days, when the other two children had 40-degree fevers, I didn't panic. Now, with him, I panic: I will call the pediatrician and I will insist that his blood is tested. (Mother of a boy, 4 years)*

Subtheme 3b: Striving for "normality". Although parents realized that their family life would never be the same as before, they recalled a constant striving for normality. Parents tried to live a normal life, although the diagnosis had changed everything.

> *There were times when I thought everything was going fine, that everything would be alright. I almost pretended as if we had a normal life. (Mother of a boy, 6 years)*

For these parents, "normal" seems to be the same as their life pre-diagnosis. Striving for normality might therefore be a form of comfort, creating a feeling of stability and hope. Moreover, "normal" behavior and "normal" situations were seen as a blessing. Parents reported appreciating the smaller things more; they valued their time together as a family more.

> *She is on a strict diet. So one cannot go to a restaurant, she cannot sit in the sun, nothing's normal anymore. So when something is normal,*

> *then it's a gift from God. We're not at all religious, but it simply is a gift. (Mother of a girl, 16 years)*

Parents made a distinction between the impact of the diagnosis on themselves and the diagnosed child, on the one hand, and on the siblings, on the other.

> *The illness has had a very big impact and then again not, because life did go on. For the other children, everything needs to continue as normal as possible, their lives cannot be turned upside down because our lives have been turned upside down or because Talia's life has been turned upside down. (Mother of a girl, 5 years)*

Parents strived to preserve a normal lifestyle for the siblings, even though the impact of the cancer was undoubtedly present. However, this "normal lifestyle" was based upon going to school and hobbies, outside of (changes within) family life.

Discussion

Pediatric cancer is a life-threatening disease, one that is extremely difficult for the diagnosed child, his or her family members, and the family as a whole to adjust to (Alderfer & Kazak, 2006). The aim of this study was to explore how parents perceive changes in functioning of the family after a pediatric cancer diagnosis, using MFMIA (Van Parys et al., 2017). The analysis has provided insight into the conflicting dynamics parents experience in association with these changes. In the first theme, *Family Cohesion: Strengthened Versus Fragmented*, we saw, on the one hand, that family cohesion was strengthened by the illness, and that parents reported valuing their family more. This is in line with previous qualitative studies (Clarke-Steffen, 1997; Woodgate & Degner, 2003), quantitative studies (Beek, et al., 2015; Trask et al., 2003), and systematic reviews (Van Schoors et al., 2015). However, at the same time, the strength of the family unit was threatened by an overwhelming parental focus on the diagnosed child. Parents felt the need

to shift all attention toward the diagnosed child (cf. previous qualitative studies; for example, Prchal & Landolt, 2012), even at the cost of time and attention allocated to any siblings, the family as a whole or their couple subsystem. Consequently, these parents may struggle to meet prevailing cultural values and standards of "good parenting". Indeed, although West-European parents are expected to divide their time and attention equally among all children, and love each child equally (Ganong & Coleman, 2017), these principles are challenged in the context of pediatric cancer and may result in parental feelings of guilt, shame, frustration, and distress (Long & Marsland, 2011). Moreover, the parents in our study seemed to question whether, in this context, a "good parent" is one that accompanies the diagnosed child no matter what or one managing to care equally for all children. In addition, previous research into multiple roles (i.e., the role-strain approach; Goode, 1960) has revealed that the greater the number of parental roles, the greater the demands and role incompatibility and the greater the strain and psychological distress (Voydanoff & Donnelly, 1999). We could posit, however, that in the context of pediatric cancer – in which the parental role dominates all others – parents experience the same emotional strain. Indeed, these parents indicated that their paid worker role, their partner role, their friend role, and so on had been subsumed by their parental role and their parental duty to the diagnosed child in particular. Although this predominance of the parental role may seem self-evident, it may also give rise to negative feelings or thoughts, for example, the idea that they are letting their other children down (Grootenhuis & Last, 1997). In conclusion, the findings of the first theme are consistent with those of other studies. However, this study contributes to the current body of evidence by showing that both subordinate themes emerge *at the same time*, and that it is specifically this dialectical experience that parents grapple with. In the second theme, *Educational Norms and Values: Overindulgence Versus Being Stricter*, parents described the impact of the cancer diagnosis on the rearing of the diagnosed child. As with the first theme, parents were confronted with two conflicting dynamics.

Specifically, shortly after diagnosis, parents started to spoil their child, a finding that has been reported in other qualitative studies as well (e.g., Enskar et al., 1997; Norberg & Steneby, 2009; Quin, 2004). Parents wanted to comfort their child and alleviate their suffering. In addition, parents might want to compensate for their own feelings of powerlessness. After all, a stricter upbringing may seem irrelevant and undesirable when their child is suffering from a life-threatening illness. However, at the same time, parents claimed to believe that their child could recover and to be aware that this spoiling may be beneficial in the short term but also may produce undesirable behavior in the long term. Once they had realized this possibility, they tried to compensate for their overindulgence by being even stricter with the child than they had been pre-diagnosis. Consistent with previous research, this study found that this indulgent behavior is only applied to the diagnosed child and not to the siblings (e.g., Van Dongen-Melman et al., 1998). In conclusion, this study builds on previous search with the finding that both behaviors (i.e., overindulgence and being strict) do not appear simultaneously, but rather occur in succession, as well as that both behaviors are magnified compared to pre-diagnosis standards. In a third theme, *Normality: Loss Versus Preservation*, parents described the idea that the family is irreversibly changed due to the cancer diagnosis. This change in family functioning has already been extensively documented in existing research (see several systematic reviews: Long & Marsland, 2011; Pai et al., 2007; Van Schoors et al., 2015). At the same time, however, parents described striving for normality. The concept of normality or the life they led pre-diagnosis may comfort the parents, as well may give them hope and courage. In addition, parents strive above all to maintain a sense of normality for the siblings. They seemed to believe that by maintaining normal routines, the impact on these other children could be minimalized. However, research has shown that the experiences of siblings cannot be separated from that of the family (Carpenter & Levant, 1994), and that they too can struggle to adjust to life post-diagnosis (Alderfer et al., 2010). Therefore, we can posit that siblings may not experience

"normal" life but share the overwhelming impact of the cancer diagnosis on the family. Future research should try to document the experiences of siblings post-diagnosis through in-depth interviews. In conclusion, this study not only confirms the major impact of cancer diagnoses on family functioning but also highlights parents' desires to preserve normality within their families and outlines the dialectical experiences of parents post-diagnosis.

Methodological Considerations

Some limitations of this study need to be addressed. First, as we report on a small-scale qualitative study of parents, we do not intend or claim to be representative. Rather, we tried to understand processes using a specific sample in a specific context, which could help uncover some of the processes underlying the impact of a pediatric cancer diagnosis on the family functioning. Second, conform to the requirements of IPA and MFMIA, our sample consisted of a homogeneous group: Only parents of children with leukemia or non-Hodgkin lymphoma were included. Although this homogeneous sample can be considered an advantage of our study, it is important to highlight that parents of children with other cancer diagnoses may have different experiences. Third, time since diagnosis varied between the couples, ranging from six to 33 months. As all parents were questioned about the first six months after diagnosis, the potential biases inherent in such retrospective methods could have influenced their responses (e.g., forgetting, defensiveness). Fourth, we focused exclusively on a sample of Belgian, Caucasian parents. As Belgium is only a small country, it is likely that the experiences of parents in other countries or with other nationalities differ (Chapple & Ziebland, 2017). In addition, every country has its own system of medical insurance or treatment procedures, which will also influence families' experiences. Fifth, in this MFMIA study we focused on the couple's experiences after a pediatric cancer diagnosis. Although this approach has many benefits (Van Parys et al., 2017), it does not take into account gender

differences within a couple. Given that research has already revealed that mothers and fathers may respond differently to a cancer diagnosis (Hoekstra-Weebers et al., 1998; Yeh, 2002), it is probable they would report different experiences of the impact on the family functioning too. Sixth, by focusing on the couples' experiences, we did not include the perspectives of ill children and healthy siblings. Discrepancies in perceptions across family members (Alderfer et al., 2009; Peterson et al., 2012), however, speak to the need to collect data from all individuals. Finally, this study does not take into account other family structures than nuclear two-parent families. As families with same-sex parents, multi-generational caregivers, and single-parent households become more represented within the society (Galvin, 2006), more research is needed to explore their unique experiences.

Clinical Implications

This study confirms the impact of a pediatric cancer diagnosis on the family functioning, as well as the necessity of routine assessment of family functioning (Long & Marsland, 2011; Van Schoors et al., 2015). Three specific recommendations arose from the study. First, awareness of the conflicting dynamics parents are confronted with may help clinicians to better understand these parents, while helping them to normalize their own behavior and feelings. For example, parents may feel guilty about devoting disproportionate attention and time to the diagnosed child and not the siblings, and/or about their difficulties in finding a balance between indulgent and strict parenting. Helping the parent to understand the extremity of the cancer context may therefore not only reduce negative parental feelings but also assist the child's adjustment (Robinson et al., 2007). Second, across the three themes, parents made a distinction between the impact of the cancer diagnosis on the diagnosed child and themselves, on the one hand, and their other children, on the other hand. In the first theme, an increase in perceived connectedness was only described between parent(s) and patient, not with the siblings. In the

second theme, parents only discussed the impact of the diagnosis on the rearing of the diagnosed child, and in the third theme, parents indicated that, in contrast to their own lives and the life of their diagnosed child, the lives of the siblings were rather unaffected by diagnosis. As a consequence, clinicians should be aware of possible enmeshment between the parents and the diagnosed child. Furthermore, together with the parents, they can explore the meaning and impact of the illness for the siblings and broaden the idea that a cancer diagnosis particularly affects the parent-patient dyad. Third, clinical work with families affected by pediatric cancer should be aware that certain individuals and relationships might be vulnerable, for example, the siblings or the couple subsystem. Throughout the study, siblings were described as being on the periphery of the family. As some siblings may also experience difficulties as a result of the cancer diagnosis (Alderfer et al., 2010; Houtzager, Grootenhuis, & Last, 1999), this subgroup should also be addressed. In addition, as marital satisfaction may seem secondary to the support of the diagnosed child, marital issues may be overlooked by psychosocial providers in oncology or even downplayed by the couple themselves. However, as these problems might negatively impact the adjustment of the child and his/her treatment, it is also important to screen for and remedy such problems (Van Schoors et al., 2017).

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CHAPTER 4

SIBLINGS' EXPERIENCES OF EVERYDAY LIFE IN A FAMILY WHERE ONE CHILD IS DIAGNOSED WITH BLOOD CANCER[1]

Childhood cancer not only presents challenges to the life of the child with cancer but also to the siblings' daily family life. The aim of the current study was to gain a better understanding of siblings' experiences of living in a family where one child has been diagnosed with blood cancer. Ten siblings of children with leukemia or non-Hodgkin lymphoma completed a semi-structured interview about their everyday family life experiences post-diagnosis. The verbatim transcripts of the interviews served as the raw data for an interpretative phenomenological analysis. The results showed that overall the siblings experienced a continuity in many aspects of their family life: they still experienced their family as an important source of support and information/communication, as warm and loving, and as a safe harbor where family members aim to protect each other. However, at the same time, the participating siblings also expressed that some things felt unmistakably different post-diagnosis: They felt that their family as a whole had been ripped apart, with a greater focus on the diagnosed child and changing responsibilities for each family member. This study informs parents and clinicians about the daily family life experiences from the siblings' perspective, a perspective that

[1]Van Schoors, M., De Mol, J., Laeremans, N., Verhofstadt, L. L., Goubert, L., & Van Parys, H. (2019). Siblings' experiences of everyday life in a family where one child is diagnosed with blood cancer: A qualitative study. *Journal of Pediatric Oncology Nursing, 36*, 131-142. doi: 10.1177/1043454218818067

is often overlooked. A focus on challenges as well as continuities within family life, the wish for connection expressed by the siblings, and the uniqueness of every sibling's experiences is what can be taken away from this study by psychosocial workers in the field.

Introduction

Pediatric cancer is a major life event that presents many challenges to the life of the child receiving the diagnosis, the parents, and any siblings (Alderfer & Kazak, 2006). To date, plenty of research has focused on the impact of a pediatric cancer diagnosis on the physical and psychosocial well-being of the different family members. The current body of literature shows that, across family members, most adjust well over time, with a minority of patients, parents, and siblings showing social or emotional problems during or after treatment. For example, symptoms of anxiety, depression, antisocial behavior (Brinkman et al., 2016), and distress (Michel et al., 2010) can be found in patients, post-traumatic stress symptoms, emotional distress, and anxiety (Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008) in parents, and worry, sadness, and posttraumatic stress symptoms (Alderfer et al., 2010; Long et al., 2018) are sometimes reported by siblings. To optimize interventions for those experiencing difficulty, it is important to better understand the impact of a pediatric cancer diagnosis on all family members.

Within the past decade, research on sibling adjustment has steadily grown (Long et al., 2018). However, up till now, most of the research on pediatric cancer still focuses upon the diagnosed children and their parents

plan et al., 2013). As siblings are embedded in the family and therefore ınfluenced by the illness as well as by the way in which the other family members respond to pediatric cancer (*Social Ecology Model*; Bronfenbrenner, 1977), more research on siblings is needed in order to best capture their unique experiences.

Previous Literature on Siblings

The adjustment and experiences of siblings of children with cancer are – to date – summarized in different systematic and integrated reviews (Alderfer et al., 2010; Long et al., 2018; Van Schoors et al., 2017b; Van Schoors et al., 2015; Wilkins & Woodgate, 2005; Yang et al., 2016; Zegaczewski et al., 2015), illustrating a predominance of quantitative compared with qualitative studies (Long et al., 2018). Quantitative studies on siblings showed that, overall, mean levels of anxiety, depression, and general adjustment are similar across siblings and comparisons (Long et al., 2018). However, for a significant subset of these children, negative emotional reactions (e.g., fear, worry, sadness, and helplessness) and poor quality of life in emotional, family, and social domains were found (Alderfer et al., 2010; Zegaczewski et al., 2015). Moreover, two thirds of the siblings endorsed moderate to severe levels of post-traumatic stress symptoms, illustrating their risk for psychosocial adjustment problems when facing pediatric cancer (Long et al., 2018). In addition, school-aged siblings show poorer academic functioning and more absenteeism compared with peers (Alderfer et al., 2010; Long et al., 2018). In qualitative studies that have examined siblings' views of the effects of pediatric cancer on their lives, there are two predominant themes. First, most qualitative studies have focused on the impact of the cancer diagnosis on the self (individual level), indicating worry about the diagnosed child and fear of death (Nolbris et al., 2007; Prchal & Landolt, 2012) as well as the presence of negative emotions like sadness, anger and jealousy (Woodgate, 2006). Indeed, the cancer experience is emotionally potent for siblings and intense negative emotions are often elicited (see Wilkins & Woodgate, 2005, for an overview). Second, some studies have examined the impact of the cancer diagnosis on family life as perceived by the siblings (interpersonal level). For instance, there is a preliminary evidence on siblings' perspectives on the impact of a cancer diagnosis on specific aspects of family life (e.g., family communication; Sloper, 2000 or family support;

Woodgate & Degner, 2003) and on the changes within family functioning post-diagnosis (Björk et al., 2005; Long et al., 2015; Van Schoors et al., 2015; Yang et al., 2016). Qualitative research from the siblings' perspective clearly indicates that childhood cancer disrupts the functioning of the family in various ways, for example, decreased parental attention, family separations, and disintegration of familiar family dynamics (see Van Schoors et al., 2015 and Wilkins & Woodgate, 2005 for an overview). Surprisingly, however, (a) less research has looked at siblings' experiences of *daily family life* after facing a pediatric cancer diagnosis. In addition, (b) in the majority of the existing qualitative studies, the specific aspects of family functioning that were included in the study were selected by the researchers. As a consequence, the siblings themselves got little freedom to talk about what really mattered to them, and therefore those unique aspects of family life that felt unmistakably different for siblings post-diagnosis could possibly have been missed.

The Present Study

The aim of this study was to gain an increased understanding of (a) how siblings experience their *everyday family life* post-diagnosis and (b) allowing them to put their *own* emphasis on particular family aspects that *matter to them*. To this end, a qualitative study with interpretative phenomenological analysis (IPA; Smith, Flowers, Larkin, 2009) based on in-depth semistructured interviews was selected. IPA is a qualitative research method which draws on the theoretical principles of phenomenology, hermeneutics, and idiography. This approach comprises of an in-depth exploration of the participant's lived experiences and how participant makes sense of these experiences (phenomenology), while emphasizing the active role for the researcher in the process of interpretative activity (hermeneutics). An idiographic focus means that only a limited number of cases are included and that each case is scrutinized in its own right before moving on to an analysis on a group level.

IPA has been applied successfully in the context of health psychology in general (Smith, 2011) and living with cancer more specifically (Reynolds & Lim, 2007), as well as on the lived experiences of children (Kvale & Brinkmann, 2009). Interviewing children allows them to voice their own experience and helps us understanding their lifeworld. In addition, IPA studies are often about experiences that have a strong impact on people's lives. In this context, we focus on the lived experience of a sibling becoming ill, the moment of diagnosis, the period of intense treatment, and the consequences for family life. The central research question was "How do siblings of children with cancer describe their everyday family life when one child had been diagnosed with cancer?"

Method

Procedure

The present study is part of a larger ongoing study in Flanders (Belgium) examining the impact of a pediatric cancer diagnosis on families, that is, the 'UGhent Families and Childhood Cancer study'. For this large-scale study, children diagnosed with leukemia or non-Hodgkin lymphoma between the age of one and 18 years, their parents and any siblings were invited to take part in a longitudinal survey study. Exclusion criteria were: a) not speaking Dutch, b) a developmental disorder in the diagnosed child and c) relapse. All participating siblings, aged between 10 and 16 years (N = 27), were subsequently invited to complete an interview about their experiences regarding the influence of the cancer diagnosis on their everyday family life. Fifteen of the participating siblings (56%) agreed to participate in this interview study, ten of whom were randomly selected for participation.

Participants

For this study, ten siblings (six girls and four boys) of children with leukemia or non-Hodgkin lymphoma were interviewed. They were all Caucasian, living in the Flemish part of Belgium and aged between ten and 16 years, representing a reasonably homogeneous sample appropriate to the requirements of IPA (Smith et al., 2009). Their ill brother or sister was either diagnosed with acute lymphoblastic leukemia (n = 6), acute myeloid leukemia (n = 1), chronic myeloid leukemia (n = 1), or non-Hodgkin lymphoma (n = 2); and aged between three and 16 years. Time since diagnosis varied from two to 26 months (M = 8). In two families, the parents were divorced; the parents of the other siblings were married. More details on the sample are listed in Table 1. Ethical approval from the University Hospitals of Ghent, Brussels, Antwerp, and Louvain had been secured for the study. Written informed consent of both parents and assent of the child was obtained before each interview took place.

Data Collection

Semistructured interviews were conducted at the siblings' home by the third (n = 8) and the last (n = 2) author. Both interviewers are clinical psychologists and are trained in psycho-oncology and family therapy, respectively. The interviews were audio-recorded, lasted between 40 and 107 minutes and consisted of three parts (full interview guideline available on request from the corresponding author).

The first part included open-ended questions about their understanding of the diagnosis and its treatment (e.g., "What do you know about your brother/sister's illness?"). In the second part, open-ended questions were provided about the influence of the cancer on the life of the sibling (individual level; e.g., "How is it for you to have an ill brother/sister?"). The last part included open-ended questions about the sibling's perspective on

Table 1

Background Characteristics of Siblings

Name_S	Age_S	Age_DC	Diagnosis_DC	Gender_DC	TSD	Marital status	#
Thomas	10	8	ALL	boy	24	Married	2
Daniella	16	16	CML	girl	5	Divorced	3
Melissa	10	8	ALL	Boy	4	Divorced	3
Nicole	13	12	ALL	Boy	5	Married	2
Barbara	11	3	ALL	Boy	26	Married	3
Ulric	13	16	AML	Boy	2	Married	2
Una	15	16	non-Hod	girl	5	Married	3
Ulfred	14	16	non-Hod	girl	5	Married	3
Fanny	14	9	ALL	Boy	3	Married	3
Bert	12	9	ALL	Boy	3	Married	3

Note. S = Pseudonym for the sibling; DC = Diagnosed Child ; # = number of children in the family; ALL = Acute Lymphoblastic Leukemia; CML = Chronic Myeloid Leukemia; AML = Acute Myeloid Leukemia; non-Hod = non-Hodgkin Lymphoma; TSD = time since diagnosis (months).

living in a family where one child has been diagnosed with cancer (family level; e.g., "How did your family life change post-diagnosis?"). The participants' experiential accounts were facilitated by means of prompts (Smith et al., 2009). Pseudonyms have been given in order to protect the anonymity of the participants. After interviewing, all interviews were transcribed verbatim. These transcriptions served as the raw data of the study's analysis.

Analysis

The siblings' interviews were analyzed one by one by the first author using the step-by-step approach for IPA, as described by Smith and Osborn (2015). First, for each interview separately, the transcript was read a number of times to obtain familiarity with the cases. Second, first interpretations and reflections ("notes") were written down in the margin of the text. This annotating in IPA ensures that both descriptive, linguistic and conceptual comments are registered. In contrast to descriptive comments, linguistic and

conceptual comments allow for interpretation of the data by the researcher, albeit staying close to the participants' phrases. Third, combining the three types of notes (e.g., "this siblings emphasize that it is important to talk about emotions") with the data then guided the phases of initial coding (e.g., code: talking is important) and the construction of emergent themes at a higher level of abstraction (e.g., theme: talking about emotions helps) which resulted in a more interpretative stance. Then, connections between these emerging themes were explored. This analytical and theoretical step resulted in a clustering of themes for each of the cases. This process was repeated for every case. When each individual transcript had been analyzed, the coded transcripts were reviewed for potential themes *across* siblings. To this end, convergences and divergences between the individual emergent themes were sought. In a final step, all themes were translated into a narrative account, explaining in more detail the data and illustrating them with verbatim extracts from the participants.

To enhance the trustworthiness of the study (Hill et al., 1997) and to assess to what extent the analysis has been conducted systematically, transparently, and credibly (see Smith et al. (2009), for more details on IPA), a team of auditors was invited to challenge the way the first author had constructed main themes and subthemes at several points in the analysis. The last author was the first auditor for this study. She is a clinical psychologist and postdoctoral researcher with expertise in qualitative research in the field of family psychology and family therapy. The second author was the second auditor and is a clinical psychologist and associate professor in clinical child and adolescent psychology who specializes in qualitative research. In addition to these strategies, the Yardley criteria (Yardley, 2000) were also taken into account to ensure the quality of the study: (a) sensitivity to context (e.g., first author's expertise in pediatric oncology literature; Van Schoors et al., 2015, 2017a, 2017b); (b) commitment and rigor (e.g., the precision/completeness of the analysis undertaken and the appropriateness of the sample); (c) transparency and coherence (e.g., the description of the subsequent steps in

the analysis); and (d) impact and importance (e.g., clinical implications of the study).

Results

Based on the IPA, the siblings experiences of their everyday family life when one child had been diagnosed with cancer can be clustered into two main themes: *Continuity within Family Life* (Theme 1) and *Beyond the Familiar: Facing Illness-Related Challenges* (Theme 2). Overall, in comparison to pre-diagnosis, the siblings in our study experienced continuity in many aspects of their family life (Theme 1). More specifically, they still experienced their family as an important source of support and information/communication, as warm and loving, and as a safe harbor where family members aimed to protect each other. In addition, due to the cancer diagnosis, the siblings indicated that these key features became even *more* pronounced within the family: They became more aware of their families' resources and vulnerabilities. However, at the same time, the siblings also referred to the challenges they were confronted with due to the cancer diagnosis, expressing that some things felt unmistakably different (Theme 2): Many felt that the family as a whole had been ripped apart post-diagnosis, with a greater focus on the diagnosed child and changing responsibilities for each family member (see Figure 1).

Theme 1: Continuity within Family Life

Subtheme 1a: The family as a source of support. For most siblings, the family was an important source of support both during times of treatment and pre-diagnosis.

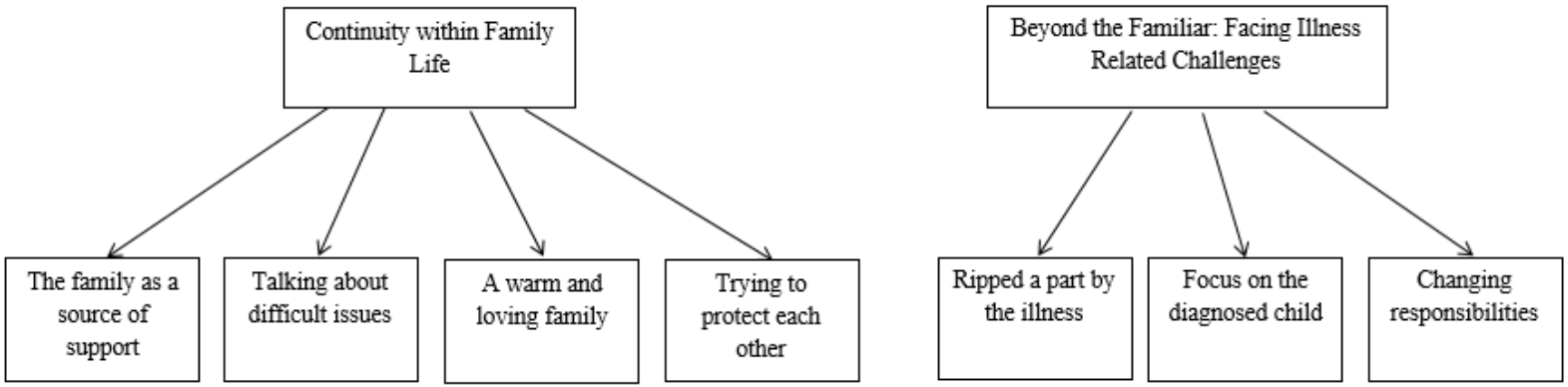

Figure 1. Themes and subthemes

> *We are always there for each other. Because we really need each other sometimes. (Ulfred, 14 years)*

For this sibling, family members being available to help each other was deemed self-evident as well as necessary. In addition, most siblings seemed to experience a lot of intimacy in their family. They had the feeling that, within their family, they could count on each other, and that the other family members were available to share worries, emotions, and experiences.

Verbal as well as nonverbal methods of support were reported across the siblings' accounts. Moreover, it seemed like families with younger children were more likely to make use of nonverbal support, whereas families with older children tended to combine verbal and nonverbal support. Overall, whatever the age of the sibling, all siblings felt supported by their parents and every family was characterized by their own way of providing support.

> *When for example, somebody felt bad, we would give a hug. We said things like "it'll be alright." (Una, 15 years)*

Two differences between support provided by mothers and fathers were reported. First, for most siblings in this study, the mother was the most important source of support within the family.

> *I'm really close to my mum because she always takes care of me and yeah … she's the person I would go to first. (Nicole, 13 years)*

Second, mothers and fathers seemed to differ in the type of support they tended to provide to the siblings. While the mother was mainly consulted on emotional issues and addressed as a person they could talk to, the father took care of providing distraction or joy in times of worry or sadness.

> *My mum and I can talk together very well. With my dad it's not the same. But with him I often do gymnastic tricks. (Melissa, 10 years)*

So, according to the siblings, mothers and fathers engaged in different supportive behaviors and focused on various aspects of the siblings' well-being. Moreover, siblings seemed to know who they could rely on, depending on what they needed (e.g., talking and distraction).

Finally, when confronted with pediatric cancer, the importance of family support was further reinforced.

> *We supported each other when something was going on. It was also like that before, but now I think it was even more. (Ulfred, 14 years)*

This sibling described that family support had always been apparent but became even more essential post-diagnosis. Moreover, it seemed that siblings not only needed more support from their parents, they were now also confronted with their parents needing support (emotional or practical).

> *I gave my mum and dad a lot of support, and my mum actually said that that wasn't necessary, but I just did it (laughs) and they supported me in return. (Melissa, 10 years)*

This sibling talked about both receiving *and* giving support within the family, describing this mutual support as something obvious to her. And although she expressed this reciprocal process of caregiving as neither needed nor expected by her parents, she believed that she could do her part too. Indeed, siblings indicated that they not only provided practical support, such as helping in the household, but also provided help on an emotional level as they assisted their parents with coping with the cancer experience. For example, some siblings described that they helped their parents by giving them a hug or by talking; according to others, simply being present was the most helpful.

Subtheme 1b:Talking about difficult issues. Talking about the cancer diagnosis and its consequences was often perceived as hard by the siblings in our study. They highlighted three things of importance in talking about the illness. First, the siblings found it important to get answers to their questions concerning the illness, either from their parents or from the medical staff.

> *When we asked questions, they [parents] always tried to answer them directly. And when they didn't know the answer, we went with them to the hospital and then they [hospital staff] explained it to us there. (Ulfred, 14 years)*

Second, the siblings found it also important that their parents made time for them to talk about the illness and its consequences.

> *I sometimes wanted to talk myself, because I wanted to know how he was. Sometimes my mum herself said things like "Shall we talk about it?" And then I always answered "yes" because that would be a load off my mind. (Thomas, 10 years)*

This quote illustrates the beneficial effects of talking about the illness for this sibling. Additionally, the fact that his mother *invited him* to talk was received positively. Indeed, being invited to talk about the illness possibly lowered the threshold to introduce the cancer subject and therefore facilitated finding a way of coping with the distress it caused in the sibling. However, across the siblings' accounts, it seemed that siblings did not always feel like talking about the illness themselves. Sometimes they preferred not to talk or think about it.

> *Interviewer: And how do you try to cope with that? Bert: By not thinking about it. Even though you know, you just don't even say something about it or think about it (12 years).*

Finally, the siblings in our study pointed at a process of regulating the level of information being exchanged within the family. The siblings indicated that, in talking about difficult issues (e.g., problems at school), they had always taken into account the capabilities and the workload of their parents and their brothers/sisters. However, from the moment of the cancer diagnosis onward, they seemed to take into consideration which and how much information they were willing to share even more than pre-diagnosis. For example, for most siblings, knowing all the details about the illness, the treatment and the prognosis of their brother or sister was not necessary. What they needed was a brief and accurate update.

> *Well, the doctors tell it to my parents, to my mum in particular. I'm not present there very often. And then my mum and dad sometimes tell me something, but yeah, not everything. And I really don't need to know everything. (Nicole, 13 years)*

Subtheme 1c: A warm and loving family. Due to the many hospital stays, the family as a whole spent less time together. As a consequence, most siblings really looked forward to being together with the diagnosed child, and used every free moment to visit their ill brother or sister in the hospital.

> *Especially during the last hours in class, I would be keeping an eye on the clock, for example, on Wednesdays because I knew that in the afternoon, we would go and visit my brother. And then I would be counting down the seconds until the bell rang and, yeah, eating really quickly and then leaving for the hospital. (Nicole, 13 years)*

In addition, side effects of the cancer treatments (e.g., fatigue or nausea) complicated time together at home. The diagnosed child was often limited in his or her possibilities to play together with the healthy sibling, which sometimes elicited feelings of sadness or loneliness in the sibling.

> *He can't do anything, so why would I, yeah … I can't have fun then. Because before, I'd done everything in my life together with my brother. He came pretty soon after me, so we know each other very well and we are best buddies. (Thomas, 10 years)*

However, despite the obstacles for being together presented by the illness or the treatment, all siblings described the love within their family. The siblings felt loved by their parents and the diagnosed child, and loved them in return. Whether it was in words or simply by hugging each other, each family seemed to have its own ways to express this love.

Finally, most siblings indicated that family cohesion was even strengthened by the illness. The siblings spoke about the illness drawing the family closer together and appreciated time spent together more.

> *The illness not only has negative influences, you know. Well, there are a lot of negatives, but it can also bring your family closer together. In my case, it was already good, and now it's just really good, let's say even better. (Ulric, 13 years)*

Subtheme 1d: Trying to protect each other. Most siblings described that within their family, all family members looked after each other, for example, by trying to prevent the others from feeling sadness or distress. Parents tried to prevent the sibling from being confronted with the impairment of the diagnosed child: when the ill child was too sick (e.g., nauseous), parents tended to not want the sibling to visit their brother/sister in the hospital.

> *I myself have never seen him ill, I've never seen him throwing up. I only saw him very tired and weak. Most of the times that I wanted to go, my mum and dad would keep me at a distance so that I would not see this. (Ulric, 13 years)*

The siblings themselves also tried to protect their parents and the diagnosed child. Some siblings believed that by sharing their own worries or sadness, they would make the others more upset. As a consequence, they sometimes did not share their emotions with their parents and the diagnosed child, and tried to cope with it alone or with others outside the family.

> *I don't talk about the illness with my brother. That makes you, yeah, I don't know. Maybe it could make him feel sad. So I don't see any reason to talk about it. (Ulric, 13 years)*

Moreover, one sibling not only avoided sharing negative emotions in order to protect her family but also avoided sharing the positive things she experienced, for instance, the contacts she had with others while her sister was in hospital.

> *And when they [friends] said: "Say hello to her from me" I actually didn't do that. Because … I don't know. I felt sorry for her. Then it would have been like: "I had to say hello from this person and I talked to that person". And Nadine didn't get to see anybody. Maybe it would do her good, but I thought it was hard to tell her when she herself didn't see anybody. I saw a friend who she hadn't seen in a long while and she said "say hello to her". Then I answered: "Maybe you could send her a text." (Una, 15 years)*

This sibling seemed to believe that if she shared her social encounters, the diagnosed child would become more aware of her own impairment and that this confrontation would be painful for her. The discrepancy between the diagnosed child's daily life and that of the siblings made it hard for siblings to share these experiences, as siblings might feel ashamed that their lives continued, while the life of the diagnosed child temporally seemed to be on hold. Furthermore, this quote also illustrates how this sibling tried to maintain the diagnosed child's social network. By encouraging friends to keep in touch with her ill sister, she tried to ensure that external support for the diagnosed child was sustained.

Finally, in these critical times of cancer diagnosis and treatment, the wish to protect each other became even more pronounced. At the same time, however, siblings felt the impossibility to protect their loved ones all the more. Indeed, from the moment of the cancer diagnosis onward, siblings seemed to be confronted with the idea that in some cases it is not possible to protect their family. They became more aware of the vulnerability of life and reported having catastrophic thoughts about losing their parents or brother/sister.

> *I am more worried. My father does 10,000 steps a day, and these days I don't like it when he goes for a walk at night, because I imagine that something might happen to him. And I felt the same when August [other sibling] left for his camp: "Imagine something would happen to him" (...) And also when mum and Ken drove to the hospital I thought "I hope nothing happens to them". Before, the fact that something could happen didn't come to my mind. And now that something has happened, it does. (Fanny, 14 years)*

Theme 2: Beyond the Familiar: Facing Illness-Related Challenges

Subtheme 2a: Ripped apart by the illness. Whereas, in the first theme, siblings described continuity in many aspects of their family life, they were at the same time also confronted with many challenges and expressed

that some things felt different post-diagnosis (Theme 2). For example, due to the many hospital stays, the family members were hardly ever all together. Most of the time, the diagnosed child was accompanied by the parent(s) to the hospital, and the siblings were taken care of by others or took care of themselves.

> *When Nadine had to go to the hospital, our family was separated. When Nadine was back, we were reunited. So yeah, then there really was a feeling of "we're back together again." That was good. (Ulfred, 14 years)*

This sibling described that he felt most happy when everyone was at home. Indeed, for most siblings, being separated from the rest of their family was hard: They missed (the help of) their parents.

> *Sometimes we also needed them [our parents], but they weren't there. But we looked after ourselves. (Ulfred, 14 years)*

In addition, when the siblings talked about their family being ripped apart by the diagnosis, they often referred to all the small things that had changed. In the siblings' daily routine, for instance, the absence of the diagnosed child was tangible.

> *The couch used to be full in the evenings, and then there was a lot of space available. That was really weird. (Ulfred, 14 years)*

Experiencing the absence of the family members was alienating for most siblings. They were used to being together and to doing things together, so not being together was not "normal" for them.

Subtheme 2b: Focus on the diagnosed child. From the moment of the cancer diagnosis onward, all eyes were on the child diagnosed with cancer. The survival of that child became a priority and, due to the life-threatening character of the diagnosis, the parents' desire to divide time and attention equally between the children gave way to a greater focus on the diagnosed child.

> *There used to be more equal attention for all of us children. We know that that is not possible now. (Una, 15 years)*

Although all siblings showed understanding for this shift in attention, for some siblings this was difficult to cope with.

> *Sometimes I hated that everything was about Nadine. (Una, 15 years)*

The diagnosed child was the center of the family, and that sometimes provoked jealousy in the siblings. In addition, some siblings felt pity for themselves. From one moment to the next, they were expected to do things on their own, things they had never done by themselves before.

> *For example, when it came to studying, I really had to become more independent. I had never learned to study and my mum and dad always helped with that before. Then, I had to study by myself, which was and still is quite difficult. (Ulfred, 14 years)*

This increased independence and responsibility was also reported by other siblings.

> *I think we, Tom [another sibling] and I, have become more independent in the things we have to do, you know, we became a little bit more grown up. (Nicole, 13 years)*

While some siblings saw this increased maturity as one of the few advantages of the cancer diagnosis and continued these new behavior over the course of the cancer treatment, others suffered from this enforced independence and were above all happy when parental help returned after a while.

Subtheme 2c: Changing Responsibilities. The illness and its treatment resulted in changing responsibilities within the family. Some siblings indicated that they were expected to help more, or to do more little tasks at home. After all, as the parent(s) were in the hospital frequently, and often combined the hospital stays with their jobs, little time was left to manage the household.

> *Sometimes I have to do more at home, small things, such as cleaning up or doing things in the kitchen. (Nicole, 13 years)*

The siblings not only indicated changing responsibilities for themselves but also within the parental relationship. While pre-diagnoses, the parents seemed to have clear roles and expectations of each other, siblings noticed that this changed after diagnosis. They described that the hospital stays, the emotional impact, and the unpredictability of the illness forced their parents to rethink their contributions to family life, and to make new arrangements.

> *My dad (laughs) doesn't do very much in the household, so my mum has to do everything. And before that wasn't a problem, but now she has to go the hospital most of the time, he has to do the grocery shopping and then he forgets half of the things and he – well maybe half is a bit exaggerated – but then he forgets a little something which is still quite important and then mum gets angry and so you have fights between the two of them. (Nicole, 13 years)*

This sibling witnessed marital distress in her parents' relationship that she linked to the changing responsibilities within the parental relationship. Irritation caused by changes in the family routines were also noticed by another sibling whose father lived somewhere else due to a divorce.

> *We were not really used to him being there every day, but then all of a sudden he [returned into our lives] … And sometimes, my sister and I didn't like it so much because he had his say in everything. (Daniella, 16 years)*

This sibling was already used to the absence of the father, and her mother taking on a larger parental role. In her opinion, it was her father's increased involvement that caused difficulties, rather than the splitting up of the family or the redistribution of household duties. More generally, it seemed that siblings in particular experienced difficulties when old routines, habits, and roles changed, and regretted the fact that the cancer diagnosis changed so many aspects of familiar family life.

Discussion

In this study, we used a qualitative research method in order to gain a better understanding of siblings' experiences of everyday family life post-diagnosis. Two themes emerged from the current data. In the first theme, *Continuity Within Family Life*, siblings indicated that many aspects of their family life stayed the same post-diagnosis and were sometimes even reinforced by the diagnosis. Four subordinate themes could be distinguished here. In subtheme 1a, the family served as an important source of support, a finding that has also been reported by previous qualitative studies (Havermans & Eiser, 1994; Sloper, 2000; Woodgate & Degner, 2003). Moreover, not only did siblings receive support from their parents, they also described ways in which they supported their parents. This is in line with research on bidirectionality and reciprocity in parent-child relationships (Crouter & Booth, 2003; De Mol & Buysse, 2008; Kuczynski, 2003), in which the co-occurrence of both directions of influence is emphasized (Kuczynski, 2003). Indeed, parents do not only have impact on their children, children too influence many aspects of parent(ing) and family functioning (De Mol & Buysse, 2008; Grusec & Goodnow, 1994). In addition, although the reciprocal support between sibling and parent(s) was commonly described in the siblings' accounts, less was said about the support provided by and given to the diagnosed child. Although the importance of support between brothers/sisters after a pediatric cancer diagnosis has been commonly stressed in previous qualitative research (Havermans & Eiser, 1994), the siblings in our study might not have talked about it because this was not explicitly asked for. One other explanation could be that the physical distance between the diagnosed child and the sibling(s) could have impeded the children from supporting each other: Patient and sibling(s) were often hardly together (see Theme 2) living parallel lives instead where the diagnosed child stayed at the hospital, and the siblings were at home, at school, or taken care of by others.

In Subtheme 1b, family communication about the illness was described as helpful in coping with the cancer experience. This is in line with previous qualitative research (Sloper, 2000; Wilkins & Woodgate, 2005) and a recent meta-analyses (Van Schoors et al., 2017b) which has shown that greater family expressiveness is associated with better child adjustment (e.g., less posttraumatic stress, anxiety, and behavioral problems). In addition, this study builds on previous research documenting that siblings prefer so-called selective communication. For instance, although siblings generally appreciated being informed about the diagnosed child's state of health (Woodgate, 2006), our study indicated that they wanted their parents to limit the medical information they disclosed as there was no need to know all medical details. In addition, the siblings sometimes did not feel like talking about the illness, thus, family communication could also be selective in terms of moments in which the illness is discussed.

In Subtheme 1c, siblings described the love within their family. In addition, in line with previous qualitative research, siblings spoke about the illness bringing the family members closer to one another (Clarke-Steffen, 1997; Van Schoors et al., 2015). This increased level of cohesion is sometimes seen as one of the few positive things associated with a pediatric cancer diagnosis (Prchal & Landolt, 2012; Sloper, 2000). However, based on existing literature, we know that this increased degree of closeness is not always perceived as inclusive of the siblings (Van Schoors et al., 2015). In other words, siblings can feel that they are at the periphery of the family as family life after the cancer diagnosis is determined by the ill child's treatment and this can result in regular absences of parents and diagnosed child and a reduction in time spent together as a family (Prchal & Landolt, 2012).

In Subtheme 1d, the siblings described that within their families, all family members looked after each other. One way to do this, was by trying to protect the others from feeling sadness or distress. For example, the parents protected the sibling from confrontation with impairment of the diagnosed child and did not want the sibling to visit the ill child when she or he was

extremely sick. We could question, however, whether siblings feel protected by this parental behavior, or whether it rather caused feelings of exclusion from the family (Van Schoors et al., 2015). In addition, the siblings also tried to protect their parents and the diagnosed child, believing that by sharing their own worries and emotions, they could make the others (more) upset. Two remarks can be added. First, it is possible that siblings are less likely to share their emotions with their parents because they find it difficult to handle the emotional impact of the illness on their parents, as well as the emotions evoked by such conversations (Prchal & Landolt, 2012). Second, as the previous literature has illustrated that after a pediatric cancer diagnosis siblings often suffer from intrusive worries about the ill child's health and prognosis (Nolbris et al., 2007; Woodgate, 2006), other sources of support for siblings may be of great value, such as other relatives, friends, or teachers (Havermans & Eiser, 1994; Sloper, 2000; Van Schoors et al., 2018).

In the second theme, *Beyond the Familiar: Facing Illness-Related Challenges,* the siblings described the challenges they were confronted with due to the cancer diagnosis. Some aspects of their family life felt different from the moment of the diagnosis onward. Three subordinate themes could be distinguished here. In Subtheme 2a**,** the siblings experienced their family as being ripped apart by the illness. They were separated from their ill brother or sister, as well as from their parents, as typically one parent stayed at the hospital long-term, and the other parent spent considerable time there on visits. This is in line with the findings of previous qualitative research (Prchal & Landolt, 2012) and systematic reviews (Alderfer et al., 2010; Wilkins & Woodgate, 2005). In addition, the apparent conflicting dynamic of feeling closer together (Subtheme 1c) as well as feeling ripped apart as one family (Subtheme 2a) is in line with a recent qualitative study in parents of children with leukemia and non-Hodgkin lymphoma (Van Schoors et al., 2018), illustrating the complexity of the process of family adaptation after a pediatric cancer diagnosis.

In Subtheme 2b, the central focus on the diagnosed child was experienced as a major change to the daily family life post-diagnosis. While pre-diagnosis, parents divided time and attention equally between the children (Ganong & Coleman, 2017), this inevitably changed to a merely unique focus on the diagnosed child; a finding that has also been reported by other qualitative studies (Alderfer & Hodges, 2010; Prchal & Landolt, 2012). In addition, the physical impossibility of parents being both at the hospital and at home sometimes forced siblings to manage alone at home. This experience of increased responsibility can be seen as an example of posttraumatic growth (D'Urso et al., 2017; Kamibeppu et al., 2010).

In Subtheme 2c, the siblings indicated a shift in responsibilities within the family due to the cancer diagnosis. From the cancer diagnosis onward, daily routines were challenged, as family life became determined by the health of the diagnosed child. More specifically, siblings reported that they took over household duties, such as cooking and cleaning (cf. previous qualitative research: Prchal & Landolt, 2012). In addition, siblings also witnessed tension between their parents, related to changes in the division of tasks. This finding is in line with a recent systematic review illustrating that a pediatric cancer diagnosis impacts on a couple's relationship as well (Van Schoors et al., 2017a).

Limitations of the Study and Suggestions for Further Research

Some limitations of the current study need to be addressed. First, as we report on a small-scale qualitative study of siblings, we do not intend or claim to be representative. Rather, appropriate to the requirements of IPA (Smith et al., 2009), a small number of participants was included to understand specific processes in a specific context. Furthermore, also in line with IPA (Smith et al., 2009), we selected a homogenous sample (e.g., diagnosis). Although this homogenous sample can be seen as an advantage of our study, it is important to highlight that siblings of children with other cancer

diagnoses may have different experiences. In addition, we also limited the age range of the included siblings from ten to 16. We can assume that younger or older siblings may have different experiences. Second, our focus was limited to only the sibling's experiences of daily family life post-diagnosis. As there may be discrepancies in perceptions across family members (Alderfer et al., 2009; Peterson et al., 2012), interview studies with parents and/or the diagnosed child are needed to get insight into their experiences as well. In addition, to better understand experiences on a *family* level, we need to include and integrate the experiences of different family members, for example, by making use of Multi Family Member Interview Analyses (Van Parys et al., 2017). This might further our understanding of the complexity of families (Van Parys et al., 2017) and broader family dynamics (Reczek, 2010). Third, the design of our study was retrospective and cross-sectional. As time since diagnosis varied from two to 26 months, siblings' reports may be limited to their memories as well as by the extent to which they are willing to share their experiences about the cancer experience. Fourth, the reporting of our interpretations of the siblings' accounts was challenged by language differences: While the interviews were conducted in Dutch, the results were written in English.

Clinical Implications

This study affirms that the life of all family members is affected by a pediatric cancer diagnosis and that the psychosocial needs of siblings too should be recognized and addressed by professionals (Alderfer et al., 2010). Three specific recommendations arise from this study. First, awareness of both the continuity and the challenges within family life that siblings are confronted with may help clinicians to better understand how siblings adapt. Moreover, as siblings emphasized that overall their daily family life stayed the same as pre-diagnosis, and that this continuity helped them to cope, clinical workers as well as parents should strive to retain this continuity within the siblings'

lives. In addition, clinical workers and parents should be aware that some key features (support, communication, love, and protection) become more pronounced within the family post-diagnosis (e.g., support is more needed or love is more palpable), and this awareness can help in normalizing siblings' feelings and behavior. Second, across the themes, the (importance of the) connection between the siblings and the other family members was stressed. Moreover, in the second theme, every challenge can be reframed as a call for togetherness. For example, in Subtheme 2a, a call for pulling together as one family can be recognized, as siblings described the feeling that their family was being split in two. In the Subtheme 2b, a call for togetherness between sibling and parents can be recognized, as the siblings felt a shift in focus and questioned their own position within the family. In Subtheme 2c, a call for togetherness between the parents can be noticed, as siblings linked the revision of parental tasks to witnessed relationship stress. As a consequence, given the centrality of this concept, clinicians are encouraged to screen and focus on (difficulties in) family cohesion, taking into account evidence-based standards for family therapy and psychosocial care in pediatric oncology (Wiener et al., 2015). Third, clinicians working with families affected by pediatric cancer find evidence in this study to (further) take into account the fact that every family and every sibling is unique. For example, while some siblings prefer to talk about the cancer, others prefer not to talk or think about it. Taking this individuality into account would therefore foster the family adaptation.

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PART 2
THE IMPACT ON INDIVIDUAL FAMILY MEMBERS AND THE FAMILY AS A RESOURCE

CHAPTER 5

ASSOCIATIONS BETWEEN FAMILY FUNCTIONING AND CHILD ADJUSTMENT AFTER PEDIATRIC CANCER DIAGNOSIS: A META-ANALYSIS[1]

A systematic review and meta-analysis was conducted to investigate associations between family functioning and child adjustment (patient/siblings) after pediatric cancer diagnosis. Database searches were performed using Web of Science, Pubmed, Cochrane, PsycInfo and Embase. After screening 5563 articles, 35 were identified regarding this topic; 30 contributed data for meta-analyses. Pearson's *r* correlations were the effect of interest. Omnibus and family functioning domain-specific random-effects meta-analyses were conducted. Data are depicted in forest plots. A statistically significant association was found between family functioning and child adjustment (patient/siblings) after cancer diagnosis. Greater family cohesion, expressiveness, and support and less family conflict were each associated with better child adjustment outcomes. Limitations in the existing literature preclude strong conclusions about the size of these effects and potential moderators.

[1]Van Schoors, M., Caes, L., Knoble, N., Goubert, L., Verhofstadt, L. L. & Alderfer, M. (2017). Associations between Family Functioning and Child Adjustment after Pediatric Cancer Diagnosis: A Meta-Analysis. *Journal of Pediatric Psychology*, *42*, 6-18. doi: 10.1093/jpepsy/jsw07

Introduction

Pediatric cancer is a highly stressful experience that can challenge the whole family system, as well as the adjustment of the child receiving the diagnosis and other children within the family (Alderfer & Kazak, 2006). While there is evidence that most patients adjust well, some may experience social or emotional problems during (Kestler & LoBiondo-Wood, 2012) or after treatment (Kazak et al., 2001). Similarly, while most siblings adjust with time, some siblings show elevated levels of posttraumatic stress symptoms, negative emotional reactions, and poor quality of life when compared to norms or control groups (Alderfer et al., 2010). To optimize interventions for the children who experience difficulty, it is important to better understand factors that influence their adjustment.

The way in which the family as a whole responds to pediatric cancer is generally assumed to impact the adjustment of children within the family. Indeed, when faced with childhood cancer, families need to deal with intense emotions, communicate effectively, and renegotiate roles and responsibilities to accommodate the demands of treatment (Kazak et al., 2004; Marcus, 2012). While most families are resilient to these challenges (Van Schoors et al., 2015), children in poorly functioning families who struggle with these demands may be at greater risk for adjustment problems (e.g., Long, Marsland, & Alderfer, 2013; Myers et al., 2014).

This key principle is embedded within various family-systems models often applied to chronic illness populations. For example, the *Social Ecology Model* (Bronfenbrenner, 1977) illustrates how the child is nested within and influenced by the family system in addition to other social systems. The *Double ABCX-Model* (McCubbin et al., 1980), the *Disability-Stress-Coping Model* (Wallander & Varni, 1998), and the *Family Adjustment and Adaptation Response Model* (FAAR; Patterson, 2002) each propose that aspects of family functioning can be risk or protective factors for individual adjustment to illness or disability. Additionally, the *Circumplex Model* (Olson & Gorall,

2003), the *Adolescence Resilience Model* (Haase, 2004), the *Process Model of Stress and Coping* (Armstrong et al., 2005), and the *Family Resilience Process model* (Walsh, 2002; 2003) each propose specific aspects of general family functioning that impact child adjustment such as family cohesion, conflict, adaptability, belief system, communication, organizational patterns, problem-solving ability, and social support.

While various reviews have summarized the impact of pediatric cancer on family functioning and/or child adjustment (Alderfer et al., 2010; Long & Marsland, 2011; Pai et al., 2007; Van Schoors et al., 2015), to date, there are no known systematic reviews or meta-analyses that summarize the empirical evidence investigating *associations* between family functioning and child adjustment to pediatric cancer. The primary aim of this paper is to fill that gap by providing an analysis, summary, and commentary on the current evidence regarding associations between the functioning of the family as a whole and child adjustment to pediatric cancer.

Method

This review is part of a series of systematic reviews of family functioning after childhood cancer (Van Schoors et al., 2015), sharing a single search strategy and following strict scientific methodology (Eiser et al., 2000; Higgins & Green, 2011).

Literature Search and Inclusion Criteria

Literature searches in Web of Science, Pubmed, PsycInfo, Cochrane, and Embase were undertaken using the following search terms: (cancer OR tumor OR malignancy OR oncolog*) AND (child* OR pediatric) AND (family OR parental), AND (psycholog* OR adaptation OR adjustment). Studies selected for analysis examined associations between constructs

capturing the functioning of the family as a whole (e.g., cohesion, flexibility, conflict, communication) and child (patient, sibling) adjustment (e.g., behavioral problems, anxiety, depression, psychosocial quality of life, posttraumatic stress) after cancer diagnosis. To maintain a focus on the family as a whole, studies examining parent-child relationship variables were not included. Eligible studies were quantitative, written in English, empirical (i.e., no reviews, case reports, commentaries, books, practice guidelines, conference abstracts, and dissertations), and involved families of children diagnosed with any type of cancer before age 18. Studies focused upon distress related to a medical procedure or appointment and those involving bereaved families were excluded, as these experiences are different from general adjustment to cancer diagnosis and treatment.

Study Selection

The original database search was undertaken in July 2014; a total of 5496 unique papers were identified. The first and second author independently screened 5496 titles (89% agreement) and identified 1592 potentially relevant abstracts for further review. Review of abstracts resulted in eliminating all but 427 manuscripts. Those full texts were then screened for final decisions regarding inclusion by the first author. The second author screened 25% with 87% agreement. Disagreements were discussed and, if necessary, a third reviewer was consulted. Reference lists of the selected papers were reviewed, and one additional relevant paper was identified. To ensure up-to-date search results, a second database search was undertaken in November 2015, identifying 157 new papers. After the process above was repeated, one study was added, resulting in a final set of 35 papers (see Figure 1).

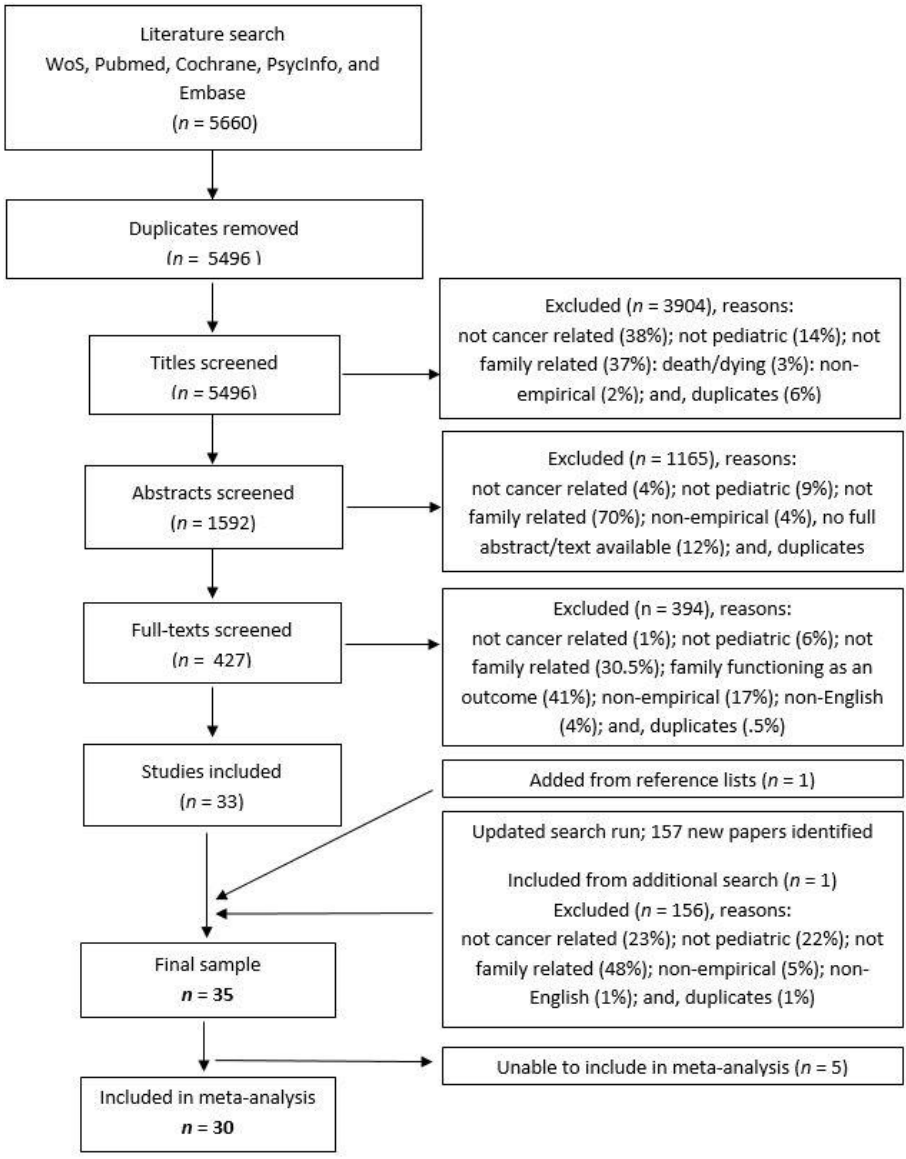

Figure 1. Steps in the research process

Data Extraction

Data extraction was conducted in a systematic and standardized way, summarizing basic study information (e.g., year of publication, target population: patients/siblings, family functioning and adjustment variables assessed), theoretical framework, aspects of methodology, and general findings on abstraction sheets (available upon request). In addition, quantitative data were collected for the purpose of the meta-analysis, specifically sample size and bivariate correlations between family functioning and child adjustment variables or group sample size, means, standard deviations, and statistical data comparing independent groups (e.g., adjustment of children in "enmeshed" vs. "balanced" families). Contact was attempted with the authors of 17 papers to gather missing statistical data. Three author groups provided information, eight indicated that they no longer had access to the relevant data, three author groups did not respond and valid contact information was unavailable for the authors of the remaining three studies. The last author checked all information extracted against original publications to ensure accuracy.

Quality Assessment

The first author rated the scientific merit and potential bias of each included study based upon the criteria published in Alderfer and colleagues (2010). This system evaluates nine aspects of quantitative studies (i.e., explicit scientific purpose, design and analysis appropriate to question posed, measurement reliability, statistical power and approach, internal and external validity, appropriate discussion, contribution to knowledge) on 3-point scales (1 = *no or little evidence in fulfilling the criterion or low quality* to 3 = *good evidence or high quality*). Individual aspect scores were averaged to obtain a total scientific merit score for each paper. The third author rated 33% of the included papers to assess reliability of the scientific merit evaluation. The

single measure and average measures intraclass correlation coefficients across the two raters were .83 and .91, respectively, demonstrating good interrater reliability.

Data Analyses

The statistical information extracted from each study or provided by the authors was entered into Comprehensive Meta-Analysis (CMA) 3.0 statistical software (Borenstein et al., 2015) for analysis. Group comparison results were converted to Pearson's *r* (Lipsey & Wilson, 2001). The Pearson's *r* values were transformed into Fisher's Z correlations with calculation of the corresponding standard error for meta-analysis, then (back) transformed to Pearson's *r* for interpretation and creation of forest plots. When authors indicated nonsignificant findings but did not provide statistical data, an effect size of 0 was used (Rosenthal, 1995). When authors indicated significant relationships without providing Pearson's *r* or a specific *p* value, CMA was used to calculate the smallest possible statistically significant value for the sample size using a nondirectional test (Faul et al., 2007).

The first level of analysis took an omnibus approach summarizing data across all domains of family functioning and all child outcomes. The sign of the correlation was standardized so that a positive value indicated that better family functioning (e.g., more communication, less conflict) was associated with better child adjustment (e.g., more social competence, fewer internalizing symptoms). A random-effects model meta-regression (method of moments) was used to account for nonnormally distributed effect sizes, and methodological heterogeneity across studies that could introduce significant random error (Borenstein et al., 2015; Cooper, 2017). Multiple effect sizes within studies/ samples were averaged for this analysis. The *Q* statistic was used to assess heterogeneity in the effect (Borenstein et al., 2015). To assess and adjust for the possibility of publication bias, funnel plots were created and the "trim and fill" algorithm (Duval & Tweedie, 2000) was used. To determine

if combining patient and sibling data in this analysis was justified, child role (patient/sibling), a between-studies variable, was examined as a potential source of signficant heterogeneity in the analysis. The association between scientific merit rating and effect was also examined.

Because the omnibus analysis simply averaged effects within studies when multiple domains of family functioning were measured, subgroup meta-analyses were conducted to better estimate the size of associations between specific family functioning domains and child adjustment. These analyses were only conducted when at least five studies were available assessing a specific family functioning domain and when the number of estimated associations represented less than one-third of the available data in an effort to ensure reliability of the estimated effect (Valentine et al., 2010). Data are summarized in forest plots created with Microsoft Excel (Neyeloff et al., 2012).

Results

PART 1: General Characteristics of the Studies in the Review

The methods and findings of the 35 studies retained for this review are summarized in a Supplementary Table (i.e., Supplementary Table 6; at the end of this dissertation). Most were cross-sectional (n = 27; 77%); only 23% (n = 8) were longitudinal. Sample size varied from 30 to 778 individuals, involving 30 to 389 families. The cancer-related time frame of these studies ranged from newly diagnosed families to those 30 years post-treatment. Leukemia, lymphoma, and brain tumors were the most frequently represented cancer diagnoses across studies. Patients were the focus of 28 of these studies.

The scientific merit ratings of the studies ranged from 1.39 to 2.67 on the 3-point scale used (Alderfer et al., 2010). Overall, the average quality rating across studies fell in the "good" range (M = 2.24) with four studies

scoring below 2.0 (see Supplementary Table 6). The most common weaknesses across studies were the psychometric properties of the measures used (e.g., low internal consistency), internal validity (e.g., measuring predictor and outcome at same point in time), and external validity (e.g., poor enrollment rates, potentially biased samples limiting generalizability). Common strengths across studies included well-justified objectives and use of methods appropriate to address the stated study purpose.

Many aspects of family functioning have been examined in the literature as predictors of child adjustment after diagnosis of pediatric cancer, including cohesion/affective involvement/affective responsiveness, expressiveness/communication, conflict, adaptability, support, roles, problem-solving, control, organization, and overall family functioning. The most frequently investigated child adjustment outcomes included internalizing, externalizing, and total behavioral problems, posttraumatic stress, quality of life, and social competence.

Thirty of the 35 identified studies provided data for meta-analysis (Supplementary Table 6). One publication reported on two separate samples (Maurice-Stam et al., 2007), and these were treated as independent in analysis; data from two manuscripts reporting on the same sample (Ozono et al., 2007; 2010) were combined. Most studies reported multiple effects of interest (range: 1-45, M = 6.6, SD = 9.3) due to measurement of multiple family functioning domains, multiple forms of child adjustment, multiple reporters (parent, child) for single constructs, and multiple time points of assessment producing both cross-sectional and lagged associations. Five studies did not provide the statistical information needed to characterize bivariate associations between family functioning and child adjustment; three of these reported associations after adjusting for covariates (Barakat et al., 1997; Houtzager et al., 2004; Rait et al., 1992).

PART 2: General Association Between Family Functioning and Child Adjustment

Family Functioning and Child Outcomes – Omnibus Meta-analysis. Across the 30 studies identified, 199 associations of interest were reported. A total of 22 associations were estimated within seven of these 30 studies. Significant heterogeneity beyond sampling error was apparent across studies within the omnibus meta-analysis, (Q [29] =48.79, p =.012) validating the use of the random effects model. The summary estimate of the correlation between family functioning and child adjustment was 0.19 with a 95% confidence interval (95% CI) of 0.13 to 0.24. This value was significantly different from 0 (Z =6.4, p < . 0001); as family functioning improved, so did the adjustment of the child. Using a random-effects model, the trim and fill approach suggested that publication bias resulted in three missing effects; after imputation of these missing data, the coefficient was 0.16 (95% CI: 0.10-0.22).

Child Role. Child role (patient/sibling) was not a significant contributor to the heterogeneity observed across the summarized studies (Q [1] =2.20, p =.14). The summary estimate of the correlation within studies (k =24) investigating patient adjustment (0.16, 95% CI: 0.10-0.23) was not significantly different from the summary estimate of the correlation within studies (k = 6) investigating sibling adjustment (0.26, 95% CI: 0.15-0.38). Significant heterogeneity was present among the studies investigating patients (Q [23] =37.88, p < .03), but not siblings (Q [5] =7.68, p =.18). Using a random-effects model, the trim and fill approach suggested no publication bias among the patient studies, but that two sibling studies were probably missing. After imputation the sibling coefficient was 0.20 (95% CI: 0.08-0.32).

Scientific Merit. Meta-regression indicated that the size of the association was significantly related to the scientific merit of the study (Q [1] =4.91, p <.03). As scientific merit improved, the size of the association

between family functioning and child adjustment got smaller (-0.25, 95% CI: -0.48 to -0.03). Scientific merit accounted for 8% of the variance in effects (ΔI^2 =.082) and significant heterogeneity remained (Q [27] =40.86, p < .05).

PART 3: Specific Family Functioning Domains and Child Adjustment

Five family functioning domains were investigated in five or more studies: cohesion, expressiveness/communication, conflict, adaptability, and support. The adaptability meta-analysis was not conducted because four of 11 data points required estimation. Results for each of the remaining domains are presented below.

Cohesion. Within the family functioning literature, cohesion is defined as the emotional climate within the family or the emotional bond between family members (Olson, 2000). This construct (operationalized as cohesion, affective involvement, or affective responsiveness) was investigated in 17 independent samples across the 30 studies included in the omnibus analysis, producing 51 associations of interest. Seven associations across five studies were estimated. Across studies, indices of child adjustment included internalizing symptoms, externalizing symptoms, posttraumatic stress, total behavioral problems, social competence, anxiety, and resilience. Figure 2 displays results from the individual studies contributing to the meta-analyses along with sample size and a brief description distinguishing multiple effects within studies.

Across adjustment outcomes, greater cohesion was significantly associated with better child adjustment (0.20, 95% CI: 0.11-.29; Z =4.32, p < .0001). There was, however, significant heterogeneity in the effect across studies (Q[16] =37.93, p =.002). The trim and fill analysis indicated that three studies needed to be imputed to account for probable publication bias. The adjusted association was estimated as 0.14 (95% CI: .04-24).

Expressiveness/Communication. Communication or expressiveness can be defined as the interchange of thoughts, feelings, experiences, and

information within the family (Olson, 2000). This construct was addressed in 10 of the 30 studies in the omnibus meta-analysis, producing 42 associations of interest; five of these originating from two studies were estimated. Child adjustment outcomes assessed across these studies included internalizing and externalizing symptoms, posttraumatic stress, total behavioral problems, social competence, and anxiety.

Data from these studies are presented in Figure 3. The meta-analysis indicated that greater expressiveness within the family was associated better child adjustment (0.15, 95% CI: 0.06 to 0.23, $Z = 3.32$, $p < .001$). There was no significant heterogeneity across these studies (Q [9] = 12.10, $p = .21$). The trim and fill method suggested that two studies needed to be imputed to offset probable publication bias. The adjusted association was .12 (95% CI: 0.03 to 0.21).

Conflict. Conflict within the family can be defined as openly expressed anger and discord among family members (Moos & Moos, 1994). Associations between family conflict and child adjustment after cancer were examined in seven of the 30 studies included in the omnibus analysis and 33 associations of interest; none were estimated. Adjustment outcomes assessed in these studies included internalizing and externalizing symptoms, posttraumatic stress, total behavioral problems, social competence, and anxiety.

Results of the individual studies are presented in Figure 4. The meta-analysis provides evidence that conflict within the family is significantly associated with poorer child adjustment (-0.25, 95% CI: -0.37 to -0.13, Z =3.92, $p < .0001$). There was no significant heterogeneity in the effects across these studies (Q[6] =9.22, p =0.16). The trim and fill method suggested that two studies needed to be imputed to account for probably publication bias. The adjusted coefficient was -0.19 (95% CI: -0.32 to -0.04).

Family support. Family support refers to practical assistance and encouragement and caring from the family received or perceived by an individual (Walsh, 2002). This construct was assessed in six of the 30 studies

in the omnibus analysis, providing 18 associations of interest; two of which arising from a single study were estimated. Across these studies, child adjustment outcomes included internalizing and externalizing symptoms, total behavioral problems, anxiety, depression, posttraumatic stress, negative affect, and social competence.

Results from the individual studies are presented in Figure 5. The meta-analysis provided evidence that greater support is associated with better child adjustment (0.23, 95% CI: 0.04-0.40, p =.019); however, significant heterogeneity was present among the studies (Q[5] = 18.87, p =.002). The trim and fill analysis indicated that two studies needed to be imputed to address probably publication bias; the adjusted coefficient was 0.30 (95% CI: 0.13-0.46).

Discussion

To our knowledge, this is the first meta-analysis investigating associations between family functioning and child adjustment after pediatric cancer diagnosis. The results of our meta-analysis generally indicate that better family functioning and specifically greater family cohesion, support, and expressiveness and less family conflict are associated with better child adjustment. These general finding are consistent across patients and siblings. The evidence, however, is not overwhelming, and the sizes of the summary correlations are not large. As such, our conclusions are tentative and not without qualifications. Below, we address the quality and limitations of the current literature and our analysis, provide recommendations for further research, and discuss clinical considerations.

Quality and Limitations of the Current Literature and Our Analyses

The scientific merit of the included studies ranged from poor to exceptional, with the average rating across all studies falling slightly above the mid-point on the scale (Alderfer et al., 2010). In these individual studies, the most commonly noted weakness included small, heterogeneous samples characterized by a broad range of diagnoses, child ages, and time since cancer diagnosis or treatment. Furthermore, the psychometric properties of the measures used to assess family functioning were sometimes a concern (e.g., low internal consistencies) and threats to both internal and external validity were apparent.

Meta-analysis was applied to the results of these studies to attempt to pool the data and circumvent problems with small sample sizes and heterogeneous samples. The benefits of using meta-analysis are discussed in more length by Valentine, Pigott, and Rothstein (2010) and Cooper (2017), including the ability to go beyond tallying significant and nonsignificant findings by estimating confidence intervals for effect sizes across studies. However, the limitations of this method also need to be appreciated. The studies summarized here were heterogeneous in regard to design (i.e., cross-sectional, prospective) and specific family functioning and child outcomes assessed – distinctions that are theoretically and empirically important, but were lost in the omnibus analysis. Dependencies in the data precluded analyses to determine whether specific family functioning domain and specific child adjustment outcome accounted for significant heterogeneity in the associations found and to determine the relative strength of associations between various family functioning domains and various child adjustment outcomes.

It should be noted that our method of estimating associations when statistical data were not provided was conservative and likely led to underestimation of the association, but this is preferable to introducing bias by ignoring null or incompletely reported results (Rosenthal, 1995). Further,

our analysis demonstrated that associations between family functioning and child adjustment were larger in studies with poorer scientific merit. This likely reflects bias in our publication practices. While well-designed studies are likely to be published regardless of results, more poorly designed studies may only be published when large, significant effects are reported. Statistical adjustments were made as needed within analyses to offset likely publication bias.

Additionally, it should be noted that the associations between family functioning and child adjustment uncovered in our analyses are specific to families of children with cancer. These findings may or may not generalize to other illness populations, and it is unclear whether these associations are similar to those for typically developing children in the general population. While our analysis only focused upon the functioning of the family as a whole, associations between parent-child relationships variables and child adjustment should be investigated.

Recommendations for Future Research

Theory should underlie the design of research and our research should aim to refine theory. One example of the lack of attention to this issue emerges in studies that acknowledge that, in theory, both high and low levels of certain family functioning variables are problematic, but then use research designs and statistical techniques based upon linear instead of curvilinear models. Second, attention needs to be paid to the conceptualization and measurement of family functioning constructs. Some measures of family functioning have low internal consistency in pediatric populations. Further, a small group of measures (e.g., Family Environment Scale (FES), Family Adaptability and Cohesion Scale (FACES)) are typically used. Aspects of family functioning that could uniquely promote resilience for children facing cancer (e.g., managing strong emotions, experiencing and expressing gratitude, repairing relationship rifts) may therefore go unexamined. Third, more research into the

associations between family functioning and child adjustment is needed. Many of the studies uncovered were from the past century. These data may not reflect current patterns, leaving important questions unanswered. Future research should include more homogenous or larger sample sizes, potentially through multi-site studies, to draw stronger conclusions regarding the associations between family functioning and child adjustment in specific contexts or to systematically investigate the role of moderators or mediators. For example, certain aspects of family functioning may be more important to child adjustment at certain time points (e.g., near diagnosis, coming off treatment), for those with specific biological risk profiles (e.g., central nervous system disease), during different developmental stages, or for families embedded in different cultures. Comparing associations between family functioning and child adjustment across populations (e.g., illness and nonillness groups) would also be informative. Lastly, basic statistical information needs to be published in individual studies to support future meta-analyses including the values of all significant and nonsignificant statistical analyses and associations between constructs.

Clinical Considerations

Most children adapt well after pediatric cancer, although an important subset experiences problems (Alderfer et al., 2010; Kestler & LoBiondo-Wood, 2012). The results of this meta-analysis indicate that better family functioning supports child adjustment. Therefore, we recommend assessing the unmet needs and providing support to *all* family members *and the family as a whole* when a child is diagnosed with cancer. Difficulties in the way in which the family is functioning after pediatric cancer may, indeed, have implications for the adjustment of all individuals within the family, and interventions at the level of the family may serve to help ameliorate or prevent adjustment problems for *all* children. A universal preventative model integrating screening and identifying risk and protective factors (Kazak et al.,

2001) across the family may be most efficient and support long-term adaptation. Indeed, focusing on building the family's strengths such as their emotional bonds with one another, ability to communicate openly, and resolve conflict may promote child adjustment while fostering family resiliency and growth.

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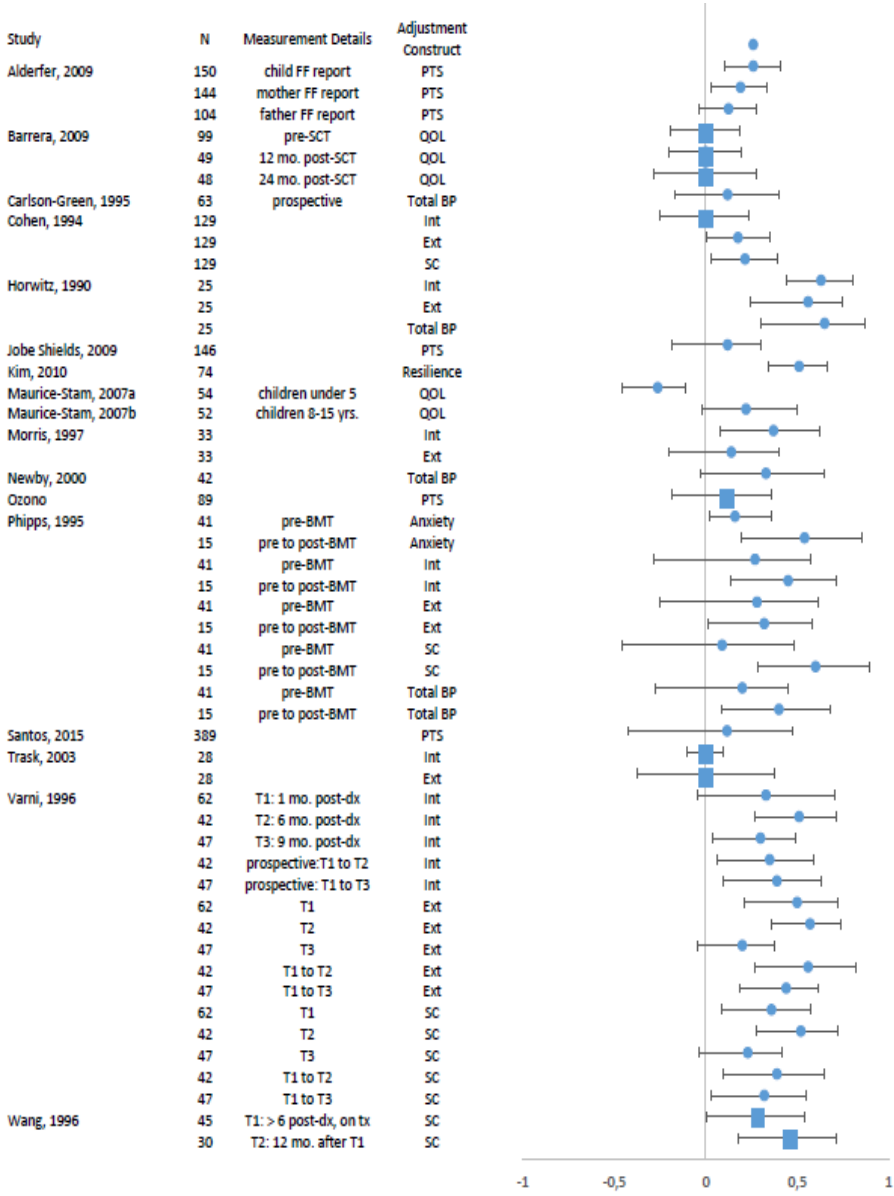
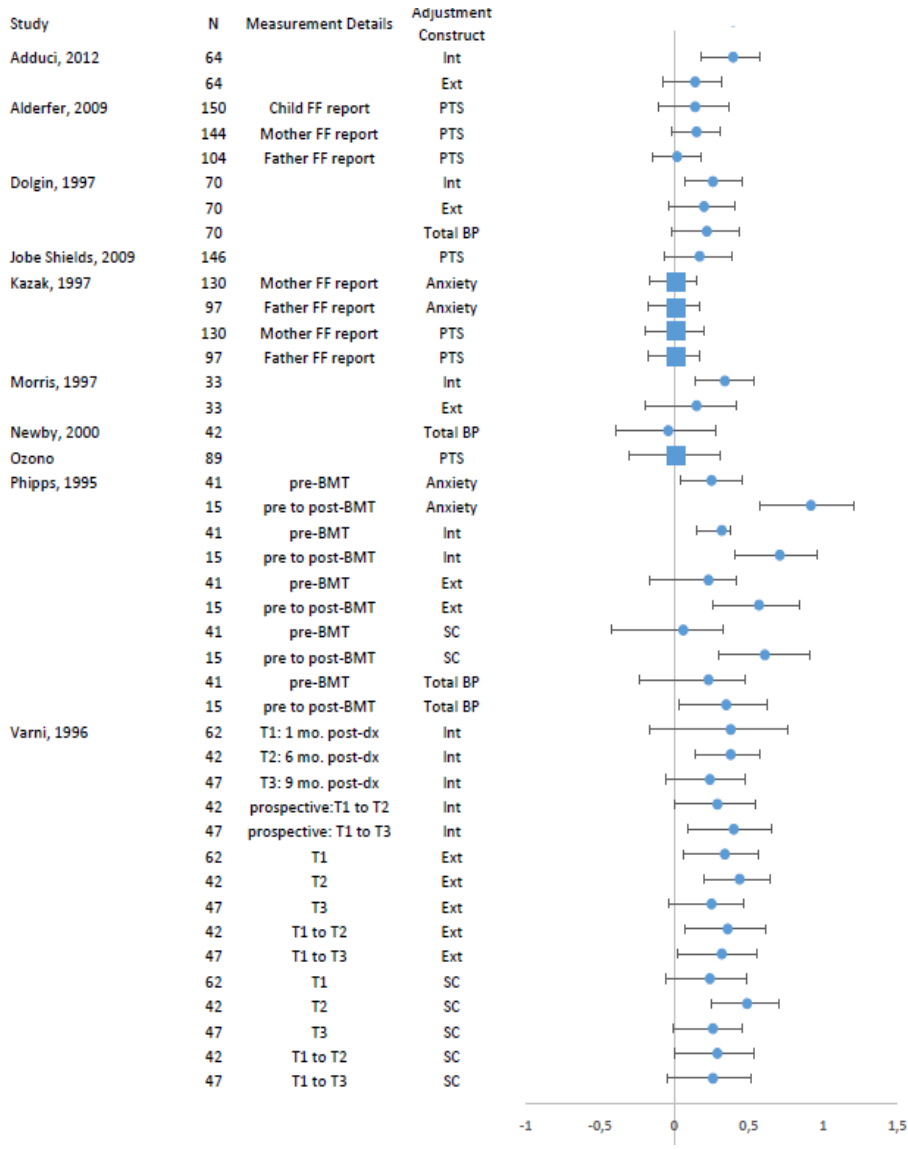
Figure 2. Forest Plot: Cohesion and child adjustment



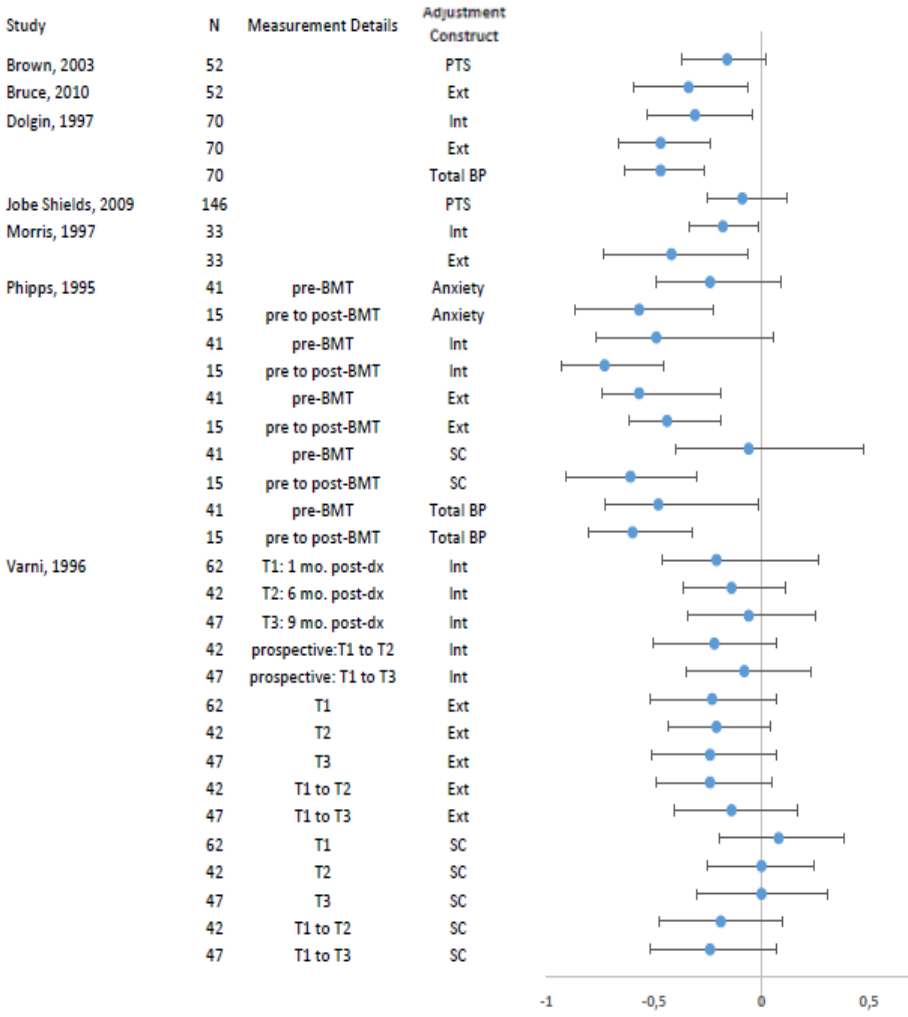
Note. Int = Internalizing Symptoms; Ext = Externalizing Symtpoms; PTS = Posttraumatic Stress; Total BP = Total Behavioral Problems; SC = Social Competence; FF = Family Functioning; BMT = Bone Marrow Transplant; mo. = months; dx = cancer diagnosis; Positive correlations indicate that more cohesion is associated with better child adjustment.

Figure 3. Forest Plot: Expressivity and child adjustment



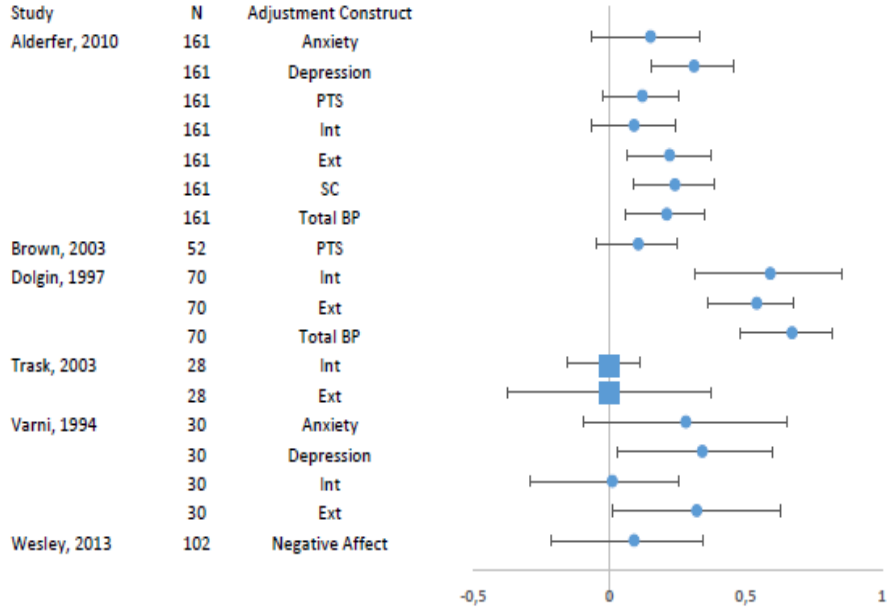
Note: Int = Internalizing Symptoms; Ext = Externalizing Symtpoms; PTS = Posttraumatic Stress; Total BP = Total Behavioral Problems; SC = Social Competence; FF = Family Functioning; BMT = Bone Marrow Transplant; mo. = months; dx = cancer diagnosis; Positive correlations indicate that more expressivity/better communication is associated with better child adjustment.

Figure 4. Forest Plot: Conflict and child adjustment



Note: Int = Internalizing Symptoms; Ext = Externalizing Symtpoms; PTS = Posttraumatic Stress; Total BP = Total Behavioral Problems; SC = Social Competence; FF = Family Functioning; BMT = Bone Marrow Transplant; mo. = months; dx = cancer diagnosis; Negative correlations indicate that greater conflict is associated with poorer child adjustment.

Figure 5. Forest Plot: Support and child adjustment



Note: Int = Internalizing Symptoms; Ext = Externalizing Symtpoms; PTS = Posttraumatic Stress; Total BP = Total Behavioral Problems; SC = Social Competence; FF = Family Functioning; BMT = Bone Marrow Transplant; mo. = months; dx = cancer diagnosis; Positive correlations indicate that more support is associated with better child adjustment.

CHAPTER 6

FAMILY MEMBERS DEALING WITH CHILDHOOD CANCER: A STUDY ON THE ROLE OF FAMILY FUNCTIONING AND CANCER APPRAISAL[1]

Childhood cancer is a life-threatening disease that poses significant challenges to the life of the diagnosed child and his/her family members. Based on the ABCX-model, the aim of the current study was to explore the association between family functioning, cancer appraisal, and the individual adjustment of patients, parents, and siblings. Participants were 60 children with leukemia or non-Hodgkin lymphoma, 172 parents, and 78 siblings (115 families). Time since diagnosis varied from zero to 33 months. Family functioning and the appraisal of the cancer diagnosis proved to be related to patients', parents', and siblings' cancer-related emotions and quality of life post-diagnosis. In addition, family members differed in their perception of some family functioning domains, the appraisal of the cancer diagnosis, positive feelings and quality of life. The differences across members within one family and differences between families speak to the need of screening all family members and intervening at the level of individual as well as the family unit.

[1]Van Schoors, M., De Paepe, A., Norga, K., Cosijns, V., Morren, H., Vercruysse ,T., Goubert, L., & Verhofstadt, L. L. (2019). Family members dealing with childhood cancer: A study on the role of family functioning and cancer appraisal. *Frontiers in Psychology, 10*, 1405. doi: 10.3389/fpsyg.2019.01405

Introduction

Every year, approximately 300,000 children are diagnosed with cancer worldwide (Steliarova-Foucher et al., 2017). Although there has been a huge improvement in survival rates in the last decades – with currently a 5-year survival rate of 83.9% (National Cancer Institute, 2014) – the psychosocial impact of childhood cancer cannot be underestimated. Children diagnosed with cancer are often confronted with social and/or emotional problems during or after treatment (Brinkman et al., 2016; Kazak et al., 2001; Michel et al., 2010). Previous studies also revealed that the turmoil and disruption created by childhood cancer reach beyond the diagnosed child and impact the parents and possible siblings as well (Kazak et al., 2001; Kestler & LoBiondo-Wood, 2012; Van Schoors et al., 2017). More specifically, parents often report feelings of posttraumatic stress, uncertainty, anxiety, and depression, especially shortly after diagnosis (Patino-Fernandez et al., 2008; Vrijmoet-Wiersma et al., 2008). In addition, some siblings show increased symptoms of posttraumatic stress, negative emotional reactions, and poor quality of life when compared to norms or control groups (Alderfer et al., 2010; Long et al., 2018).

It should be noted, however, that the research literature on the individual adjustment of children diagnosed with cancer and their family members documents a considerable variability in outcomes: while most show resiliency, some report adjustment problems after diagnosis. This idea of variability in adjustment to stressors is a key principle of the so-called ABCX-model (Hill, 1958; Figure 1), one of the major family-stress models (Weber, 2011). This model assumes that a stressor ("a") interacts with the family members' crisis-meeting resources ("b"), and the appraisal ("c") family members make of the stressful event, and that this interaction produces the amount of crisis or maladjustment ("x") in each family member (Weber, 2011). In other words, how an individual (the ill child and his/her family members) responds to or deals with childhood cancer is the result of an

interaction between his/her available resources, and his/her perception of the illness: the more resources and the more one perceives the illness as

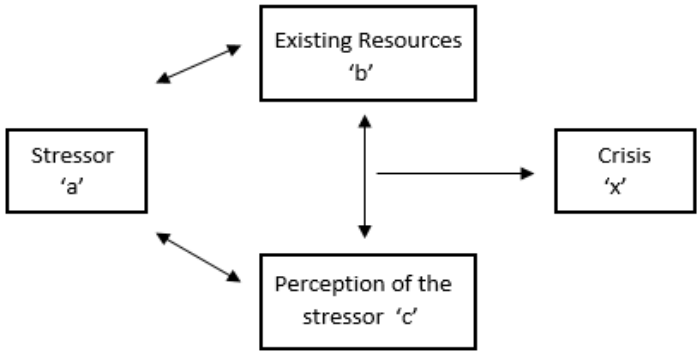


Figure 1. ABCX model (Hill, 1958)

manageable instead of uncontrollable, the better the individual adjustment. Resources can be interpreted as factors that, by their presence, keep the individual from crisis or, by their absence, urges a family member into crisis. Resources can be situated at three levels: the individual level (e.g., personality; Erickson & Steiner, 2001), the family level (e.g., family functioning; Van Schoors et al., 2017), and the contextual level (e.g., network support; Corey et al., 2008).

Existing research on the individual adjustment of children diagnosed with cancer and their family members is limited in three ways. First, most research is a-theoretical (i.e., not based on a theoretical framework; Van Schoors et al., 2015), so the selection of the variables within studies (type and their role) and the interpretation of the results is rather arbitrary. Second, up till now, most of the research that tried to explain why some family members adjust better than others after a diagnosis of childhood cancer focused on individual and contextual resources, and less research attention has been paid to family resources. However, the way in which the family as a whole deals with and responds to childhood cancer ("family functioning") is generally assumed to impact the adjustment of all members within the family (e.g., Van

Schoors et al., 2017). Indeed, when facing childhood cancer, family members need to cope with intense emotions, communicate effectively, and renegotiate roles and responsibilities to accommodate the demands of treatment (Kazak et al., 2004; Marcus, 2012; Van Schoors et al., 2015), and poorly functioning families who struggle with these demands may be at greater risk for adjustment problems (e.g., Long et al., 2013; Myers et al., 2014; Van Schoors et al., 2017). Third, within the childhood cancer literature, most studies only include one single respondent (e.g., Van Schoors et al., 2015) rather than considering the perspectives of *all* family members. As a consequence, the interdependence between family members and the bidirectional relationships within families are, to date, mostly neglected.

Addressing these three limitations, the aim of the present study was twofold. First, relying on the ABCX model as theoretical framework, we aimed to investigate how the interplay of family functioning (a key family resource; "b") and the appraisal of the cancer diagnosis (perception/definition; "c") predicts cancer-related emotional well-being and perceived quality of life (individual adjustment, "x") in patients, parents, and siblings when facing childhood cancer. More specifically, we expected that better family functioning and perceiving the illness as more manageable and less uncontrollable, as well as the interplay between both, will be associated with better individual outcomes (i.e., less negative cancer-related emotions, more positive cancer-related emotions, and better quality of life) in patients, parents, and siblings. The secondary aim was more explorative in nature and concerns the investigation of similarities and differences in the appraisal of the cancer diagnosis, the perception of family functioning, cancer-related emotions and perceived quality of life across members within one family.

Method

Participants

The sample consisted of 115 families where one child has been diagnosed with leukemia or non-Hodgkin lymphoma. All families were Caucasian and living in the Flemish part of Belgium. Across the families, time since diagnosis varied from 0 to 33 months (*M* = 6,90, *SD* = 8,05). The ill child's mean age was 6,60 (*SD* = 4,84; Range = 0–19). In 24 families (21%), the diagnosed child was the only child. The remaining families had either two (52 families; 45%), three (28 families; 24%), four (9 families; 8%) or five (2 families; 2%) children.

Due to the questionnaires' age limits (e.g., the Family Environment Scale (FES) is only applicable for children aged 11 and above) and the willingness of the different family members to participate, data from 60 ill children, 172 parents and 78 siblings were included in the present study. More details on the sample are listed in Table 1. Ethical approval from the University Hospitals of Ghent, Brussels, Antwerp, and Louvain had been secured for the study. Written informed consent forms were obtained from all the participating parents in this study, as well as all the participating children above the age of 12. Parental consent was obtained for all participating children under the age of 16.

Procedure

The current study is part of a larger ongoing study examining the impact of childhood cancer on families, that is, the "UGhent Families and Childhood Cancer study". For this large-scale study, families of children diagnosed with leukemia or non-Hodgkin lymphoma between the age of zero and 18 years at the moment of diagnosis were invited to take part in a

longitudinal survey study. Specifically, all children (patients and siblings) aged 5 years and more and both parents were asked to complete a set of questionnaires at five different time points (diagnosis to 2.5 years post-diagnosis). For this study, only the first measurement of all family members was included. Exclusion criteria for participation were: (a) not speaking Dutch (N = 20), (b) expression of a developmental disorder in the diagnosed child (N = 9), and (c) relapse (N = 6). Over a period of 4 years, 115 families participated (56% of the eligible families). The most important reasons for non-participation were being overwhelmed by the diagnosis and lack of time.

Measures

Patients, parents, and siblings separately filled out a similar set of questionnaires, as described below. However, due to a minimum age limit of the questionnaires, some younger children did not complete all questionnaires. For each questionnaire, the minimum age and the number of participants excluded for the questionnaire based on this minimum age ("N_{age}") are reported.

Family Functioning. The Dutch version of the Family Environment Scale (FES; Moos & Moos, 1994) was used to evaluate family functioning. The questionnaire contains 77 "yes–no" items, distributed across seven subscales: (1) cohesion (e.g., "we support each other anyway"), (2) expressiveness (e.g., "we have many spontaneous conversations in our family"), (3) conflict (e.g., "we quarrel a lot at home"), (4) organization (e.g., "we take care that our rooms are cleaned up"), (5) control (e.g., "we pay close attention to being at home on time"), (6) norms and values (e.g., "working first than playing is a rule in our family"), and (7) social orientation (e.g., "everyone has hobbies in our family"). Two composite scores can be calculated as well: the family relation index (FRI, cohesion + expressiveness - conflict) and the family structure index (FSI, organization + control),

Table 1

Background Characteristics of the Study Sample

		Demographic variable		
Families		Age ill child, mean (*SD*)		6,60 (4,84)
N = 115		Sex ill child, boys, *n (%)*		69 (60%)
		Diagnosis, *n (%)*	Acute lymphoblastic leukemia	85 (73,9%)
			Acute myeloid leukemia	8 (7%)
			Chronic myeloid leukemia	2 (1,7%)
			Non-Hodgkin Lymphoma	20 (17,4%)
		Time since diagnosis in months (*SD*; Range)		6,90 (8,05; 0 – 33)
		Family status, *n (%)*	Married/Co-habiting	100 (87%)
			Divorced	8 (7%)
			Single parent	3 (3%)
			Stepfamily	4 (3%)
Participating	Ill child	N		60
Family		Sex, boys, *n (%)*		34 (56,7%)
members[1]		Age, mean (*SD*)		9,90 (3,76)
	Parents	N		172
		Sex, men, *n (%)*		73 (42%)
		Age, mothers mean (*SD*)		37,58 (6,31)
		Age, fathers mean *(SD)*		40,18 (6,46)
	Siblings	N		78
		Sex, boys, *n (%)*		37 (47,4%)
		Age, mean (*SD; range*)		10,82 (4,92; 5-25)

Note. [1]Only the characteristics of the participating family members are summarized

reflecting the affective nature of the family relationships and the extent to which the family is structured and open to change, respectively. Higher FES composite scores reflect higher emotional closeness within the family (FRI; more cohesion and expressiveness and less conflict) and a more firm family structure (FSI; more control and organization). The FES is applicable for children aged 11 and above (N_{age} = 82; 37 patients, 45 siblings), and has good reliability and validity (Jansma & De Coole 1995). In the present study, the overall Cronbach's alpha reliabilities ranged from .71 (fathers) to .76 (siblings) for the relation index and from .57 (mothers) to .67 (siblings) for the structure index. The low Cronbach's alphas for the FSI subscale could not be improved by dropping one or more items.

Appraisal of the Cancer Diagnosis. The Perceived Stress Scale (PSS; Cohen et al., 1983) measures the extent to which a person perceives the last month as unpredictable, uncontrollable, and overloading. For this study, the instruction of the questionnaire was adapted and the participant was asked to rate the extent to which s/he perceives her/his *life since the cancer diagnosis* as unpredictable, uncontrollable, and overloading. The questionnaire consists of 10 items, rated on a 5-point Likert scale from 0 (never) to 4 (very often). Total scores range from 0 to 40 and higher scores indicate perceiving the illness as more uncontrollable and less manageable. An example item is "since the cancer diagnosis, how often did you feel that things were going as you wanted?" The PSS is applicable for children aged 10 and above (N_{age} = 71; 32 patients, 39 siblings), and has good reliability (e.g., Golden-Kreutz et al., 2005). In addition, 3 participants older than 10 years (1 patient and 2 siblings) did not complete the questionnaire. In the present study, Cronbach's alpha coefficients were .74, .54, .51, .63, for patients, mothers, fathers and siblings respectively. The low Cronbach's alphas for the mothers and the fathers could not be improved by dropping one or more items.

Cancer-Related Emotions. The Situation-Specific Emotional Reactions Questionnaire (SSERQ; Grootenhuis & Last, 1997; Houtzager et al., 2004) is developed to assess emotional reactions in families where one

child has been diagnosed with cancer. Different versions are available for patients (30 items), parents (30 items), and siblings (26 items), but all are divided in four subscales: (1) loneliness (e.g., "I feel lonely"), (2) uncertainty (e.g., "I am afraid to lose my child"), (3) positive feelings (e.g., "I am proud that I persevere"), and (4) helplessness/emotional involvement (e.g., "I regret that my parents have to undergo this"). This latter subscale is called "helplessness" in the patients' and parents' version, and "emotional involvement' in the siblings" version. However, given the consensus on a content level, and in agreement with the authors of the subscales, this subscale will further be referred to as "helplessness". All items are rated on a 4-point Likert scale from 0 (almost never) to 3 (almost always). The higher the scores, the more emotional reactions, both negative (loneliness, uncertainty, and helplessness) and positive (positive feelings). The questionnaire is applicable from the age of 7 (N_{age} =24; 13 patients, 11 siblings) and has satisfactory to good validity and reliability (Grootenhuis & Last, 2008; Houtzager et al., 2004). In addition, 5 participants older than 7 years (1 patient and 4 siblings) did not complete the questionnaire. In the present study, Cronbach's alpha coefficients ranged from .77 (patients) to .88 (siblings) for uncertainty, from .68 (patients) to .88 (siblings) for helplessness, from .67 (patients) to .92 (mothers) for loneliness and from .58 (siblings) to .81 (fathers) for positive emotions.

Quality of Life_Child (QoL). The pediatric quality of life inventory (PedsQL) and the general life satisfaction subscale of the Maudsley Marital Questionnaire (MMQ) were used to assess quality of life in children and parents, respectively. The PedsQL (Varni et al., 1999) measures children's health-related quality of life. Different versions of the questionnaire are available, for example, the PedsQL™ 3.0 Cancer Module (children diagnosed with cancer) and PedsQL™ Generic Core Scales (healthy children). In this study, the PedsQL™ 3.0 Cancer Module measured the diagnosed child's quality of life and is composed of 27 items comprising eight dimensions: (1) Pain and Hurt (e.g., "I have a lot of pain"), (2) Nausea (e.g., "I feel too

nauseous to eat"), (3) Procedural Anxiety (e.g., "I get scared when blood has to be taken"), (4) Treatment Anxiety (e.g., "I get scared when I have to go to the doctor"), (5) Worry (e.g., "I worry about the side effects of the medical treatments"), (6) Cognitive Problems (e.g., "I have trouble remembering what I read"), (7) Perceived Physical Appearance (e.g., "I am ashamed when others see my body"), and (8) Communication (e.g., "it's difficult to ask nurses and doctors questions"). The PedsQL™ Generic Core Scales measured the siblings' quality of life and is composed of 23 items comprising four dimensions: (1) Physical functioning (e.g., "it's hard for me to run"), (2) Emotional functioning (e.g., "I feel angry"), (3) Social functioning (e.g., "other kids tease me") and (4) School functioning (e.g., "I forget things"). Within both questionnaires, all items are scored on a five-point Likert-scale (0 = never to 4 = almost always). Each of the item scores is reversed and rescaled to a 0-100 scale: a score of 100 represents the best quality of life possible, a score of 0 the worst quality of life possible. Scale scores, as well as the sum score, are computed by adding together the different item scores and dividing this obtained score by the number of items answered. The questionnaire is applicable from the age of 5 (N_{age} = 0) and has sufficient to good validity and reliability (Varni et al., 2001). Five participants older than 5 years (1 patient and 4 siblings) did not complete the questionnaire. In the present study, Cronbach's alpha coefficients were .89 and .89, for patients and siblings, respectively.

Quality of Life_Parent (QoL). The Maudsley Marital Questionnaire (MMQ; Arrindell et al., 1983) evaluates life in general (e.g., "Are you competent and successful at your job and your housework?") and the marital/sexual relationship (e.g., "How much are you committed to this marriage?"). The MMQ contains 20 items, each of which is rated on a 0–8 scale, with 0 representing the optimum response. Higher scores indicate more maladjustment. The MMQ has good reliability and validity and the psychometric qualities of the Dutch version were also found to be satisfactory (Arrindell et al., 1983; Orathinkel et al., 2007). In the present study, the MMQ

was not completed by single or divorced parents (N = 15; 9 mothers and 6 fathers) and only the subscale measuring general life satisfaction (i.e., satisfaction with life, household and social network) was taken into account, with a Cronbach's alpha reliability of .70 (mothers) and .72 (fathers).

Parents' scores on the MMQ were reversed, so for all participants (patients, siblings, mothers, fathers) higher scores (on the PedsQL and MMQ respectively) indicate better quality of life.

Data Analytic Strategy

A multilevel (or hierarchically nested) approach was used to structure the data. This means that observations at one level of analysis (individual family members) were nested within another level of analysis (family). Multilevel modeling was preferred over ordinary-least-squares (OLS) methods, such as ANOVA, because it provides better parameter estimates with nested data (Kenny et al., 1998). The R-package *lme4* (Bates et al., 2015) was used to analyze multilevel data. The amount of variance attributable to each of the grouping structures were calculated using the function *icc* of the R package *sjstats* (Lüdecke, 2019). Continuous predictor variables were centered around their mean value to improve interpretability of the regression coefficients (Schielzeth, 2010).

To investigate whether family functioning and the appraisal of the cancer diagnosis affect *cancer-related emotions* and perceived *QoL*, separate models were fitted with SSERQ scores and the QoL score, respectively, as outcome variables. For *cancer-related emotions* four separate models were fitted for the subscales of the questionnaire (loneliness, uncertainty, positive emotions and helplessness). For *QoL* two separate models were fitted, one for the mothers and fathers (with scores on the MMQ as outcome variable) and one for the patients and siblings (with scores on the PedsQL as outcome variable).

Predictor variables of interest were FES scores as a measure of *family functioning* and the PSS score as a measure of the *cancer appraisal*. In a first step, family functioning composite scores were entered (i.e., *FRI* and *FSI;* Fowler, 1981). In a second step, the model was refitted with the seven family functioning subscales (cohesion, expressiveness, conflict, organization, control, norms and values, and social orientation) to get more insight into the specific aspects of the family relationships and structure. *Diagnosis* (ALL, AML, CML, and Non-Hodgkin Lymphoma), *time since diagnosis* (in months), *number of children*, *sex* (of the respondent), *family member* (patient, mother, father, and sibling), *age* (of the ill child at diagnosis) and *family situation* (married, divorced, single parent, step family) were included in all models as covariates. In order to investigate whether the associations differed between family members, interaction effects between the two predictors of interest, and the covariate family member were included in the model. In addition, in accordance with the ABCX model (Hill, 1958), we also investigated whether the interaction of *family functioning* and the *appraisal of the cancer diagnosis* predicted cancer-related emotions and quality of life. If the interaction effects were not significant, they were left out of the final model.

In order to investigate similarities and differences in the perception of *cancer-related emotions* and *quality of life* across members within one family, the covariate *family member* (patient, mother, father, and sibling) was included in the multilevel analysis (as described above). Next, in order to investigate similarities and differences in the perception of *family functioning* and the *appraisal of the cancer diagnosis*, two separate models were fitted with the FES scores and the PSS score as outcome variable and *family member* as predictor variable. As for the previous research question, *diagnosis* (ALL, AML, CML, Non-Hodgkin Lymphoma), *time since diagnosis* (in months), *number of children*, *sex* (of the respondent), *age* (of the ill child at diagnosis) and *family situation* (married, divorced, single parent, and step family) were included as covariates. If *family member* was significant within the model,

post hoc comparisons were conducted using Tukey's all-pair comparisons as implemented in the package "multcomp" in R (Torsten, Bretz & Westfall, 2008) to assess which family members differed significantly from each other.

Models were fitted with restricted maximum likelihood (REML) estimation. Since most of the missing data was caused to age restrictions of the questionnaires, we assumed that the data are missing completely at random (MCAR). Therefore, listwise deletion was used. The ANOVA table was inspected to check for significant main and interaction effects and specific hypotheses were tested. Satterthwaite's approximation was used to obtain the degrees of freedom (Sas Technical Report R-101, 1978). Model assumptions of linearity, independence, normality and homogeneity of variance were checked. Significance was evaluated at the 5% significance level. To get insight into the magnitude of the effects, 95% confidence intervals (CI) are reported.

Results

Table 2 shows the means, standard deviations, and observed range for the variables in our study.

Family Functioning, Cancer Appraisal and Cancer-Related Emotions

The final models for the associations between family functioning, cancer appraisal and cancer-related emotions are shown in Table 3.

Loneliness. The interaction effects between *family functioning (FRI and FSI)* and *family member* (*FRI:* $\chi^2(3) = 5.54$, $p = .14$; *FSI:* $\chi^2(3) = 2.79$, $p = .43$), between *cancer appraisal* and *family member* ($\chi^2(3) = 5.34$, $p = .15$) and between *family functioning* and *cancer appraisal* (*FRI:* $\chi^2(1) = 1.13$, $p = .29$; *FSI:* $\chi^2(1) = 2.30$, $p = .13$) were not significant and were

Table 2

Descriptive Statistics of the Study Variables

		Patient			Mother			Father			Sibling		
		M	*SD*	Range	*M*	*SD*	Range	*M*	*SD*	Range	*M*	*SD*	Range
Cancer Appraisal		18.81	5.31	8-28	21.03	6.55	9-39	17.97	6.28	5-32	20.82	6.19	10-36
Family Functioning	FRI	56.22	7.91	37-68	53.76	7.99	28-68	52.66	7.78	26-68	54.82	8.04	37-68
	FSI	54.09	7.73	39-68	49.68	7.55	20-64	49.34	8.41	18-64	51.06	8.34	35-65
Cancer-related Emotions	Loneliness	5.91	3.63	1-14	7.82	6.81	0-30	5.34	5.13	0-22	5.49	4.70	0-18
	Uncertainty	5.65	3.78	0-15	8.88	4.26	0-18	7.40	3.82	0-15	7.29	5.56	0-24
	Helplessness	12.87	4.70	1-23	13.36	4.67	3-21	11.23	4.51	1-21	13.37	5.14	1-21
	Positive Emotions	8.85	3.50	3-16	9.11	3.30	2-18	7.56	3.36	0-15	4.56	2.26	0-9
Quality of Life		69.94	13.76	35 - 95	12.62	6.56	2 -34	10.88	6.04	0 -30	73.44	14.99	35-95

Table 3

Final models for the associations between family functioning, cancer appraisal and cancer-related emotions

	Loneliness (N = 220; 20 patients, 28 siblings, 99 mothers, 73 fathers)[1]			**Uncertainty (N = 220; 20 patients, 28 siblings, 99 mothers, 73 fathers)[1]**			**Helplessness (N = 220; 20 patients, 28 siblings, 99 mothers, 73 fathers)[1]**			**Positive feelings (N = 220; 20 patients, 28 siblings, 99 mothers, 73 fathers)[1]**		
	B	**95% CI**	**p value**	**B**	**95%**	**p value**	**B**	**95% CI**	**p value**	**B**	**95% CI**	**p value**
FES – FRI	-.15	[-.25, -.05]	.003*	-.03	[-.10, .03]	.34	.001	[-.08, .08]	.98	-.17	[-.36, .02]	.07
Cohesion[2]	-.05	[-.58, .48]	.85	-.02	[-.40, .37]	.93	.03	[-.41, .47]	.90	-1.46	[-3.19, .28]	.10
Expressiveness[2]	-.49	[-.84, -.13]	.008*	-.19	[-.45, .08]	.17	-.07	[-.37, .24]	.67	-.83	[-1.69, .03]	.06
Conflict[2]	.02	[-.29, .33]	.88	-.06	[-.27, .16]	.61	-.10	[-.35, .14]	.40	.006	[-1.30, 1.31]	.99
FES – FSI	-.006	[-.10, .09]	.90	.03	[-.03, .09]	.40	.07	[-.003, .15]	.06	-.004	[-.06, .06]	.91
Organization[2]	-.16	[-.54, .21]	.40	-.13	[-.40, .14]	.36	.02	[-.30, .33]	.92	.88	[-1.03, 2.79]	.37
Control[2]	.006	[-.39, .40]	.98	.19	[-.10, .48]	.20	.20	[-.13, .53]	.24	-.20	[-1.52, 1.11]	.76
FES – Norms[2]	-.05	[-.42, .32]	.79	.10	[-.18, .37]	.49	.28	[-.03, .59]	.08	.40	[-.66, 1.46]	.46
FES – Social orientation[2]	-.31	[-.62, .01]	.06	.06	[-.16, .29]	.58	.07	[-.19, .32]	.62	-.52	[-1.34, .30]	.22

PSS – Cancer Appraisal	.48	[.37, .58]	<.001**	.40	[.33, .47]	<.001**	.38	[.29, .46]	<.001**	-.03	[-.10, .04]	.43
Control variables												
Family member (Mother vs. Patient)	-1.85	[-4.34, .64]	.15	2.47	[.50, 4.45]	.02*	-.33	[-2.61, 1.94]	.77	-.79	[-2.78, 1.20]	.44
Family member (Father vs. Patient)	-.78	[-3.40, 1.84]	.56	2.04	[-.02, 4.10]	.05	-.04	[-2.42, 2.34]	.97	-1.98	[-4.01, .04]	.06
Family member (Sibling vs. Patient)	-2.72	[-5.29, -.15]	.04*	.60	[-1.40, 2.60]	.56	1.56	[-.70, 3.82]	.18	-5.37	[-7.48, -3.26]	<.001**
Diagnosis (AML vs. ALL)	.31	[-2.93, 3.56]	.85	.05	[-1.87, 1.98]	.96	-.38	[-2.59, 1.83]	.74	1.37	[-.58, 3.32]	.17
Diagnosis (CML vs. ALL)	1.37	[-4.57, 7.31]	.65	2.81	[-.31, 5.93]	.09	-.43	[-3.94, 3.08]	.81	.14	[-3.15, 3.43]	.93
Diagnosis (Non Hodgkin vs. ALL)	1.39	[-1.04, 3.82]	.27	-.05	[-1.44, 1.33]	.94	-.60	[-2.18, .98]	.46	.85	[-.56, 2.26]	.24
TSD	-.04	[-.13, .05]	.39	-.08	[-.14, -.03]	.005*	-.13	[-.19, -.06]	<.001**	.04	[-.02, .10]	.22
# Children	-.18	[-1.08, .72]	.70	.16	[-.37, .70]	.56	-.28	[-.89, .33]	.37	-.06	[-.60, .49]	.84
Family situation (single parent vs. stepfamily)	3.11	[-4.08, 10.30]	.40	-1.11	[-5.16, 2.95]	.59	-.61	[-5.26, 4.03]	.80	1.10	[-3.02, 5.23]	.60
Family situation (divorced vs. stepfamily)	2.52	[-2.97, 8.02]	.37	.42	[-2.74, 3.57]	.80	-.67	[-4.28, 2.95]	.72	.50	[-2.71, 3.70]	.76

Family situation (married vs. stepfamily)	2.50	[-1.84, 6.84]	.26	.11	[-2.34, 2.56]	.93	-.46	[-3.26, 2.35]	.75	.57	[-1.92, 3.06]	.66
Age (of ill child at diagnosis)	-.22	[-.41, -.02]	.03*	.01	[-.10, .13]	.82	.07	[-3.26, 2.35]	.33	-.14	[-.26, -.02]	.03*
Sex (female vs. male)	2.38	[-.07, 4.82]	.06	-.24	[-2.19, 1.70]	.81	1.04	[-1.20, 3.28]	.36	.46	[-1.27, 2.19]	.60

Note. [1]Only 48 children could be included in the analyses, due to the age restrictions of some of the questionnaires (FES and PSS)
Note. [2]Obtained by fitting a second model, including the subscales of the FES, instead of the FRI and FSI.

subsequently left out of the final model. In the final model, 32% of the variance in *loneliness* was attributable to differences between family members (regardless of which family one belonged to) and 36% was attributable to differences between families. Within the same family, there was a correlation of .53 between the different family members in their reports of loneliness.

A significant effect of *family relation index (FRI)* upon loneliness was found ($\chi^2(1) = 9.03$, $p = .003$): higher emotional closeness within the family (more cohesion and expressiveness, less conflict) was related to lower levels of loneliness in all family members. In addition, when refitting the model with the FES subscales instead of the two composite scores, there was a significant effect of *expressiveness* ($\chi^2(1) = 7.26$, $p = .007$). In other words, when a participant perceived his/her family as more expressive, s/he reported to feel less lonely. None of the other FES subscales were significantly related to loneliness (all $\chi^2 < 3.7$, all $p > .05$). Furthermore, there was a significant effect of *cancer appraisal* ($\chi^2(1) = 81.83$, $p < .001$): the more one perceived the illness as uncontrollable and the less as manageable, the more s/he reported to feel lonely. This was the case for all family members. Finally, there was also a significant effect of the *age of the ill child at diagnosis* ($\chi^2(1) = 4.58$, $p = .03$): the older the ill child was at diagnosis, the less all family members reported to feel lonely. None of the other variables were significantly related to loneliness (all $\chi^2 < 3.7$, all $p > .05$).

Uncertainty. The interaction effects between *family functioning (FRI and FSI)* and *family member* (*FRI:* $\chi^2(3) = .92$, $p = .82$; *FSI:* $\chi^2(3) = 2.55$, $p = .47$), between *cancer appraisal* and *family member* ($\chi^2(3) = 2.82$, $p = .42$) and between *family functioning (FRI and FSI)* and *cancer appraisal* (*FRI:* $\chi^2(1) = 1.08$, $p = .30$; *FSI:* $\chi^2(1) = 1.60$, $p = .21$) were not significant and were subsequently left out of the final model. In the final model, 18% of the variance in *uncertainty* was attributable to differences between family members (regardless of which family one belonged to) and 0% was attributable to differences between families.

There was a significant effect of *cancer appraisal* upon uncertainty in all family members ($\chi^2(1) = 118.66$, $p < .001$): the more one perceived the illness as uncontrollable and the less as manageable, the more s/he reported to feel insecure. There was also a significant effect of *time since diagnosis* ($\chi^2(1) = 8.20$, $p = .004$]), indicating that participants reported less uncertainty if more time had passed since diagnosis. Finally, there was also a significant effect of *family member* ($\chi^2(3) = 9.99$, $p = .02$). This will be explained below (see section "Similarities and Differences Across Members Within One Family"). None of the other variables were significantly related to uncertainty (all $\chi^2 < 1.0$, all $p > .30$).

Helplessness. The interaction effects between *family functioning (FRI and FSI)* and *family member* (*FRI:* $\chi^2(3) = 3.42$, $p = .33$; *FSI:* $\chi^2(3) = 3.47$, $p = .32$), between *cancer appraisal* and *family member* ($\chi^2(3) = 2.30$, $p = .51$) and between *family functioning (FRI and FSI)* and *cancer appraisal* (*FRI:* $\chi^2(1) = 1.02$, $p = .31$; *FSI:* $\chi^2(1) = .73$, $p = .39$) were not significant and were subsequently left out of the final model. In the final model, 0% of the variance in *helplessness* was attributable to differences between family members (regardless of which family one belonged to) and 0% was attributable to differences between families, indicating that clustering based on family members and families cannot explain the variance in helplessness.

A significant effect of *cancer appraisal* upon helplessness was found ($\chi^2(1) = 78.13$, $p < .001$). In other words, the more one perceived the illness as uncontrollable and the less as manageable, the more s/he reported to feel helpless. There was also a significant effect of *time since diagnosis* ($\chi^2(1) = 14.96$, $p < .001$), indicating that participants reported less helplessness with increasing time since diagnosis. None of the other variables were significantly related to helplessness (all $\chi^2 < 3.6$, all $p > .06$).

Positive feelings. The interaction between the *family relation index (FRI, family functioning)* and *family member* was significant ($\chi^2(3) = 8.79$, $p = .03$). The other two interactions with *family member* were not significant (interaction with *FSI*: $\chi^2(3) = 3.49$, $p = .32$; interaction with *cancer appraisal*:

$\chi^2(3) = 4.54$, $p = .21$), nor were the interactions between *family functioning* and *cancer appraisal* (*FRI:* $\chi^2(1) = .31$, $p = .58$; *FSI:* $\chi^2(1) = .0001$, $p = .99$). Only the significant interaction was kept in the final model. In this model, 70% of the variance in *positive feelings* was attributable to differences between family members (regardless of which family one belonged to) and 3% was attributable to differences between families. Within the same family, there was a correlation of .04 between the different family members in their reports of positive feelings.

There was a significant main effect of *family member* ($\chi^2(3) = 33.99$, $p < .001$), as will be explained below (see 3.3). There was also a significant effect of the *ill child's age at diagnosis* ($\chi^2(1) = 5.07$, $p = .02$): the older the ill child was at diagnosis, the less all family members reported to experience positive emotions. None of the other variables were significantly related to positive emotions (all $\chi^2 < 3.30$, all $p > 0.07$). Of note, when excluding the non-significant interactions (interaction with FSI, interaction with cancer appraisal, interaction between family functioning and cancer appraisal), the interaction effect between *FRI* and *family member* did no longer reach significance ($\chi^2(3) = 6.60$, $p = 0.09$).

Family Functioning, Cancer Appraisal and Quality of Life

The final models for the associations between family functioning, cancer appraisal and quality of life for mothers and fathers on the one hand and patients and siblings on the other hand are shown in Table 4.

Table 4

Final models for the associations between family functioning, cancer appraisal and reported quality of life

	QoL mothers and fathers (N = 157; 90 mothers, 67 fathers)			**QoL patients and siblings (N = 48; 20 patients, 28 siblings)[1]**		
	B	**95% CI**	**p value**	**B**	**95% CI**	**p value**
Variables of interest						
FES - FRI	.26	[.12, .39]	<.001**	.04	[-.46, .55]	.86
Cohesion[2]	.15	[-.66, .95]	.72	-.48	[-2.94, 1.96]	.70
Expressiveness[2]	.73	[.16, 1.30]	.01*	.14	[-1.32, 1.62]	.85
Conflict[2]	-.42	[-.85, .006]	.06	.17	[-1.35, 1.71]	.82
FES - FSI	-.03	[-.17, .10]	.62	-.26	[-.74, .24]	.32
Organization[2]	-.24	[-.77, .29]	.37	-.33	[-2.31, 1.64]	.74
Control[2]	.12	[-.49, .73]	.69	-.87	[-2.60, .87]	.34
FES - Norms[2]	.31	[-.27, .88]	.30	1.26	[-.38, 2.90]	.14
FES - Social orientation[2]	.30	[-.16, .77]	.20	2.30	[.79, 3.81]	.006*
PSS - Cancer Appraisal	-.27	[-.42, -.12]	<.001*	-1.46	[-1.97, -.94]	<.001**
Control variables						
Family member (Father vs. Mother) or (sibling vs. patient)	1.26	[-.41, 2.94]	.14	12.18	[6.44, 17.93]	<.001**
Diagnosis (AML vs. ALL)	.28	[-3.54, 4.11]	.89	-19.30	[-39.00, .39]	.08
Diagnosis (CML vs. ALL)	5.47	[-5.71, 16.65]	.34	-11.93	[-31.84, 7.99]	.26
Diagnosis (Non Hodgkin vs. ALL)	.64	[-2.35, 3.64]	.67	-19.73	[-13.59, -7.87]	.004*
TSD	.08	[-.04, .21]	.19	.56	[.09, 1.03]	.03*

# Children	-1.21	[-2.36, -.06]	.04*	-1.40	[-5.50, 2.71]	.51
Family situation (single parent vs. stepfamily)	6.68	[-6.21, 19.57]	.31	10.16	[-17.43, 37.74]	.48
Family situation (divorced vs. stepfamily)	4.81	[-8.17, 17.80]	.47	-16.72	[-40.24, 6.79]	.18
Family situation (married vs. stepfamily)	1.24	[-4.90, 7.38]	.69	-3.23	[-22.05, 15.58]	.74
Age (of ill child at diagnosis)	.08	[-.16, .32]	.51	1.76	[.47, 3.04]	.01*
Sex (female vs. male)[3]				5.04	[-1.09, 11.16]	.12

Note. [1]Only 48 children could be included in the analyses, due to the age restrictions of some of the questionnaires (FES and PSS)
Note. [2]Obtained by fitting a second model, including the subscales of the FES, instead of the FRI and FSI.
Note. [3]S*ex* was redundant and was left out of the model assessing quality of life for mothers and fathers, since the variable *Family member* (father vs. mother) was identical in this case

Mothers and fathers. The interaction effects between *family functioning (FRI and FSI)* and *family member* (*FRI:* $\chi^2(1) = .58$, $p = .45$; *FSI:* $\chi^2(1) = .64$, $p = .43$), between *cancer appraisal* and *family member* ($\chi^2(1) = 2.67$, $p = .10$) and between *family functioning* (*FRI and FSI)* and *cancer appraisal* (*FRI:* $\chi^2(1) = 1.10$, $p = .29$; *FSI:* $\chi^2(1) = 1.53$, $p = .22$) were not significant and were subsequently left out of the final model. In the final model, 27% of the variance in *quality of life* was attributable to differences between families[2].

There was a significant effect of the *family relation index* (*FRI*) upon quality of life ($\chi^2(1) = 13.49$, $p < .001$) , indicating that higher emotional closeness within the family (more cohesion and expressiveness, less conflict) was associated with better *quality of life* in mothers and fathers. In addition, the model was refitted with the FES subscales instead of the composite scores. This analysis revealed that the subscale *expressiveness* ($\chi^2(1) = 6.26$, $p = .01$) was significantly associated with *quality of life:* when a parent perceived his/her family as more expressive, s/he reported better quality of life. None of the other FES subscales were significantly related to quality of life. Furthermore, there was a significant main effect of the *appraisal of the cancer diagnosis* ($\chi^2(1) = 12.78$, $p < .001$) in both parents: the more one perceives the illness as uncontrollable and the less as manageable, the worse his/her quality of life. The effect of the *number of children in the family* was also significant ($\chi^2(1) = 4.27$, $p = .04$). This means that families with more children reported worse parental *quality of life*. None of the other variables were significantly related to quality of life (all $\chi^2 < 4.00$, all $p > .10$).

Patients and siblings. The interaction effects between *family functioning (FRI and FSI)* and *family member* (*FRI:* $\chi^2(1) = 3.57$, $p = .06$; *FSI:* $\chi^2(1) = .69$, $p = .41$), between *cancer appraisal* and *family member* ($\chi^2(1) = .58$, $p = .44$) and between *family functioning* (*FRI and FSI)* and *cancer*

[2] In this model only a random intercept for *family* was included, since the variance in the random intercept for *family member* was completely confounded with the residual variance.

appraisal (*FRI:* $\chi^2(1) = .02$, $p = .88$; *FSI:* $\chi^2(1) = .66$, $p = .42$) were not significant and were subsequently left out of the final model. In the final model, 0% of the variance in *quality of life* was attributable to differences between family members and 48% was attributable to differences between families.

For the FES subscales, there was a significant effect of *social orientation (*$\chi^2(1) = 8.93$, $p = .003$*)*: when a child perceived his/her family as more socially oriented, s/he reported better quality of life. There was also a significant main effect of the *appraisal of the cancer diagnosis* ($\chi^2(1) = 30.43$, $p < .001$): the more one perceives the illness as uncontrollable and the less as manageable, the worse his/her quality of life. The effect of the *family member* was also significant ($\chi^2(1) = 17.27$, $p = .<.001$). This will be explained below (see section "Similarities and Differences Across Members Within One Family"). There was a significant effect of the *age of the ill child at diagnosis* ($\chi^2(1) = 7.15$, $p = .008$): a higher age was associated with higher quality of life in patients and siblings. There was also a significant effect of *time since diagnosis* ($\chi^2(1) = 5.47$, $p = .02$): the more time had passed since the diagnosis, the higher the quality of life. Finally, there was a significant effect of *diagnosis* ($\chi^2(1) = 11.80$, $p = .008$), indicating that quality of life was lower with a diagnosis of Non-Hodgkin lymphoma, compared to a diagnosis of ALL. None of the other variables were significantly related to quality of life (all $\chi^2 < 3.00$, all $p > .10$).

Similarities and Differences Across Members Within One Family.

Mean scores for family functioning (scores on the FES subscales), appraisal of the cancer diagnosis (PSS scores), cancer related emotions (scores on the SSERQ subscales) and quality of life (PedsQL scores and MMQ scores) per family member are presented in Table 5. Mean scores for mother, father, sibling and patients were compared.

Across the *family functioning* subscales, the perception of the mothers tended to differ from the perception of the patients and/or the siblings. Specifically for the *cohesion* subscale, mothers experienced less emotional togetherness within the family compared to the patients (β = -5.00, p = .02) and the siblings (β = -5.05, p = .008). None of the other comparisons were significantly different (all p > .25). For the subscale *organization*, mothers scored significantly lower than the patients (β = -5.46, p = .03). In other words, the child with cancer experienced significantly more family rules, tasks and duties compared to his/her mother. None of the other comparisons were significantly different (all p > .25). For the subscale *norms*, mothers scored significantly lower than siblings (β = -4.28, p = .02) : according to the siblings, more norms and standards were being pursued within the family than according to the mother. None of the other comparisons were significantly different (all p > .08). For the subscale *control*, there was a significant main effect of *family member* (χ^2 (3) = 10.34, p = .02). However, none of the paired comparisons between family members reached significance (all p > .08). For the subscales *expressivity*, *conflict* and *social orientation*, there were no significant differences across members within one family (all χ^2 < 4.60, all p > .20). For the *appraisal of the cancer diagnosis,* fathers scored significantly lower than siblings (β = -4.62, p = .006), indicating that fathers experienced the illness as significantly more manageable compared to the healthy siblings. None of the other comparisons were significantly different (all p > .09).

With regard to the *cancer related emotions*, siblings reported less positive emotions than patients (β = -5.37, p < .001), mothers (β = -4.58, p < .001) and fathers (β = -3.39, p = .004). None of the other comparisons were significantly different (all p > .21). For *uncertainty*, there was a significant main effect of *family member* (χ^2 (3) = 9.99, p = .02).

Table 5

Mean scores for cancer appraisal (PSS scores), family functioning (FES subscale scores), cancer related emotions (SSERQ subscale scores) and quality of life (standardized PedsQL and MMQ scores) for the different family members.

		Patient *M (SD)*	**Mother** *M (SD)*	**Father** *M (SD)*	**Sibling** *M (SD)*
Cancer appraisal		18.81 (5.31)	21.03 (6.55)	17.97 (6.28)	20.82 (6.19)
Family Functioning	**Cohesion**	56.17 (5.32)	51.55 (7.66)	53.03 (7.21)	53.79 (6.65)
	Expressiveness	52.52 (7.78)	53.06 (9.15)	51.37 (10.05)	52.73 (7.97)
	Conflict	44.52 (11.92)	45.26 (9.47)	47.25 (10.11)	45.33 (10.25)
	Organization	54.61 (6.97)	49.56 (8.35)	50.10 (10.24)	49.76 (8.87)
	Control	51.78 (7.93)	49.44 (7.60)	48.18 (7.97)	51.76 (8.66)
	Norms	53.09 (5.54)	48.88 (7.46)	50.48 (6.48)	52.91 (5.22)
	Social orientation	48.35 (11.62)	48.64 (11.45)	48.38 (9.76)	51.18 (10.03)
Cancer-related emotion	**Loneliness**	5.91 (3.63)	7.81 (6.81)	5.34 (5.13)	5.49 (4.70)
	Uncertainty	5.65 (3.78)	8.88 (4.26)	7.40 (3.82)	7.29 (5.56)
	Helplessness	12.87 (4.70)	13.36 (4.67)	11.23 (4.51)	13.37 (5.14)
	Positive Emotions	8.85 (3.50)	9.11 (3.30)	7.56 (3.36)	4.56 (2.26)
Quality of Life (standardized)		-0.13 (0.95)	-0.11 (1.03)	0.16 (.95)	0.11 (1.03)

However, none of the paired comparisons between family members reached significance (all $p > .06$). For loneliness and helplessness, no differences across members within one family were found (all $\chi^2 < 4.70$, all $p > .15$). For *quality of life*, siblings ($\beta = 12.18$, $p < .001$) reported higher quality of life than patients. For parents, there was no significant difference between mothers and fathers ($\beta = 1.26$, $p = .14$).

Discussion

Based on the ABCX model (Hill, 1958) and using a multi-level approach (R-package *lme4*; Bates et al., 2015), the present study sought to examine whether family functioning and the appraisal of the cancer diagnosis, as well as the interplay between both, was related to individual outcomes (i.e., cancer-related emotions and perceived quality of life) in patients, parents, and siblings facing cancer in one of the children. In addition, similarities and differences between family members within one family were explored.

Summary of results

Family functioning, cancer appraisal and cancer-related emotions. Our findings indicate that both family functioning and the appraisal of the cancer diagnosis matter for the emotional well-being of family members being confronted with childhood cancer. This is in line with our prediction and with previous quantitative studies on family functioning (Van Schoors et al., 2017) and stress (Hamama et al., 2000) in the context of childhood cancer. However, different patterns of findings emerged for both predictors.

More specifically, we found that more **emotional closeness** within the family (more cohesion and expressivity, less conflict) was associated with lower levels of loneliness in all family members. In other words, when a family member perceived his/her family as warm and loving (cohesion), open to talk about experiences and emotions (expressivity) and there were little

conflicts, s/he reported to feel less lonely. This is in line with the idea that family functioning is important for the adjustment of children (see Van Schoors et al., 2017 for an overview) and parents (Fuemmeler et al., 2003) when facing childhood cancer. In addition, when taking into account the family functioning subscales, there was a significant association between expressiveness and loneliness: the more family members can share their experiences within the family, the less loneliness in all family members. This finding illustrates the importance of family communication (Van Schoors et al., 2018a).

Furthermore, we found – for all family members – that when a family member perceived the illness as more uncontrollable and less manageable (i.e., *cancer appraisal*), s/he reported more negative emotional reactions (i.e., feelings of loneliness, uncertainty, and helplessness). This is in line with the idea that the meaning a person gives to a certain stressor has an impact on the stressor's consequences (e.g., the role of catastrophizing; Caes et al., 2011). Remarkably, there was no significant association between the appraisal of the cancer diagnosis and positive emotions. This interesting finding should be explored in further research.

Family functioning, cancer appraisal and quality of life. Our findings indicate that both family functioning and cancer appraisal matter for patients', parents' and siblings' quality of life when facing childhood cancer. More specifically, more *emotional closeness* within the family (more cohesion and expressivity, less conflict) was associated with better *parental* quality of life, a finding that has also been reported by several quantitative studies in parents (Ozono et al., 2010; Santos et al., 2015). When considering the family functioning subscales, a significant association between expressiveness and parental quality of life; and between social orientation and children's quality of life was found: the more a parent perceived his/her family as expressive and the closer a child is to his/her social environment (e.g., friends), the better his/her quality of life. These findings emphasize the importance of sharing

experiences within the family, especially for parents (Van Schoors et al., 2018a) and with the social network, especially for children (Beltrao et al., 2007; McGrath et al., 2005). Furthermore, we found that – for all family members – *cancer appraisal* was related to quality of life: perceiving the illness as more uncontrollable and less manageable was related to worse quality of life, in parents and in children (patients and siblings). This is in line with existing quantitative studies. For example, according to Witt et al. (2010), the experience of a child with cancer is not in itself related to poor quality of life, but it is related to an increased level of perceived stress, which may in turn adversely impact parental (quality of) life.

Similarities and Differences Across Family Members Within Families. Family member differences as well as important family member similarities in the perception of cancer appraisal, family functioning, cancer-related emotions and perceived quality of life emerged from our data. For the appraisal of the cancer diagnosis, we found that fathers are more likely than siblings to experience the illness as more manageable and less uncontrollable. Possible explanations are twofold. First, in most of the included families and in line with the Western idea that especially mothers are responsible for the childcare, the father kept working to ensure financial security, whereas the mother (temporally) quit her job to ensure that always one parent could accompany the diagnosed child to the hospital (Van Schoors et al., 2018b). As a consequence, the father's daily activities stayed more or less the same as pre-diagnosis and potentially protecting him from catastrophizing about the illness as being unsurmountable. For siblings, however, the impact on their daily life is huge: from one day to another, they are confronted with less parental attention, the need to become more responsible and independent, and others (e.g., grandparents) taking over parental roles (Van Schoors et al., 2018a). These sudden and major disruptions of siblings' lives may them feel more overwhelmed by the illness. Second, the cancer appraisal (more manageable and less uncontrollable) can also operate as a protecting

mechanism for fathers: as fathers are obligated to continue to go to work in order to assure finical certainty, they cannot afford to head down. By believing the illness is manageable and the child will cure, they can concentrate more on their job, and thus, on the family's financial certainty.

With regard to family functioning, mothers rated their family functioning after diagnosis significantly worse – less close, less organized, less strict in following norms – than the children (patients, siblings). Possible explanations are twofold. First, this is in line with the idea that parents – and especially mothers – may struggle to meet prevailing cultural values and standards of "good parenting": while West-European parents are expected to divide their time and attention equally among all children, and love each child equally (Ganong & Coleman, 2017), these principles are challenged in the context of pediatric cancer and may result in parental feelings of guilt, shame, frustration and distress (Long & Marsland, 2011) and rating the family functioning as less adaptive. Second, the finding that mothers reported lower levels of organization and norms within their family, as compared to the children, makes sense, given the demanding character of the cancer treatment, e.g., isolation, invasive procedures and all obligations/responsibilities for the patient within his/her healing process, as well as the possible changes in the daily life of the siblings (Van Schoors et al., 2018a). However, our finding on family cohesion (i.e., siblings experienced more cohesion compared to mothers) is not in line with existing qualitative studies, showing that most parents and patients - but not siblings - experience an increase in family cohesion post-diagnosis (Prchal & Landolt, 2012; Van Schoors et al., 2015, 2018).

Regarding cancer-related emotional responses, we found that siblings experienced less positive emotions compared to patients, mothers and fathers. This is in line with several systematic reviews, emphasizing the possible negative impact of a childhood cancer diagnosis on siblings (Alderfer et al., 2010; Yang et al., 2016; Zegaczewski et al., 2015). Moreover, this finding can be linked to a recent systematic review on family resiliency (Van Schoors et

al., 2015) and two recent qualitative studies (e.g., Van Schoors et al., 2018a; 2018b) showing that siblings often feel at the periphery of the family, as family life post-diagnosis is determined by the ill child's treatment and this often results in regular absences of the parents and the diagnosed child and a reduction in time spent together as a family (Prchal & Landolt, 2012). For quality of life, the siblings' quality of life was found to be higher than the quality of life of the patient, affirming the severe impact of the illness on the patient (e.g., physical effects, Eiser, 1998).

Furthermore, not only the differences and the similarities in the family members' mean scores on our study variables (as described above) were considered, we also investigated whether the *associations* of interest (i.e., cancer appraisal/family functioning and cancer-related emotions/quality of life) were similar/different for patients, parents, and siblings. Across our findings, no indication for an interaction effect with the type of family member was found. This illustrates that, for all family members, comparable associations between predictors and outcomes were found. This is in line with the idea that a childhood cancer diagnosis impacts all family members, and that the same predictors are important for all family members.

Finally, for uncertainty and positive emotions, especially the differences between family members seem to be relevant, instead of the differences across families. In other words, in predicting uncertainty and positive emotions, it seems to be more important *which family member* (patient, parents, sibling) it is, than the *family* s/he belongs to. Only for loneliness, significant correlations between family members within the same family were found, making loneliness a rather shared family experience. In addition, differences between families were important in the prediction of quality of life. So, how satisfied someone is with his/her life after diagnosis depends mainly on the characteristics of the family s/he belongs to.

Other Findings. The results of the present study furthermore revealed the importance of time since diagnosis and age of the ill child at diagnosis in

the prediction of cancer-related emotions. First, family members living in a family with a child who has been diagnosed more recently showed greater uncertainty and helplessness (all family members) and reported worse quality of life (children) than those who had been exposed to the illness for a more prolonged period of time. This is in line with the concept of habituation: responses - such as negative emotions - to a certain stressor might decrease after repeated or prolonged presentations (Bouton, 2007). Indeed, when time goes on, the diagnosed child and his/her family may get gradually used to the hospital staff, long hospitalizations and medical procedures, with a decrease in negative emotions as a result. Second, there was a significant association between the age of the ill child at diagnosis on the one hand and loneliness, positive feelings and quality of life in children on the other hand: the older the ill child at diagnosis, the less loneliness and the less positive feelings in all family members; and the better the patients' and the siblings' quality of life. This finding adds to the current, inconsistent body of literature regarding the influence of the diagnosed child's age on the individual adjustment of patients, parents and siblings after facing childhood cancer (e.g., Yalug et al., 2011 vs. Phipps et al., 2005) and is – to the best of our knowledge – the first presenting the influence of age at diagnosis on the adjustment of *all* family members together (patient, parents, siblings).

Furthermore, the number of children in a family and the ill child's diagnosis was related to perceived quality of life. More specifically, the more children in a family, the worse the parental quality of life. Possible explanations are twofold. First, this finding confirms the general idea that having children negatively impacts parental quality of life, especially the first years of parenthood (Myrskylä & Margolis, 2014). Second, from the moment of the cancer diagnosis onward, the diagnosed child becomes the center of focus in the family. When the ill child is the only child, the whole family organization can more easily be adapted to the needs of that child. However, when siblings are present, the siblings' needs have to be recognized as well (Prchal & Landolt, 2012), and parents may struggle with the desire to focus

merely on the ill child (Van Schoors et al., 2018b). There was also a significant impact of the type of diagnosis on quality of life: patients diagnosed with ALL as well as their siblings reported better quality of life than patients with non-Hodgkin lymphoma and their siblings. This is in surprising, as children with leukemia are - in general - more hospitalized than children with non-Hodgkin lymphoma.

Finally, across our findings, no interaction effect between cancer appraisal and family functioning was found to be significant. In other words, contrary to the prediction of the ABCX model (Hill, 1958), only the main effects of the resources (i.e., family functioning; a key family resource "b") and the perception (the appraisal of the cancer diagnosis, "c") were found to be important when facing childhood cancer, and not the interplay between both. This somewhat unexpected but nevertheless consistent finding would be worthwhile to explore in future research.

Strengths and Limitations

A first strength of the present study is that it makes use of the ABCX-model as underlying theoretical framework guiding the selection of variables and the interpretation of the results. Second, although most studies in the childhood cancer literature make use of one single family member participant (Van Schoors et al., 2015), we included the perspectives of *all* family members, i.e., patient, parents, and siblings. As a consequence, we were able to investigate similarities and differences across family members within the same family for both individual level variables (cancer appraisal, cancer-related emotions, and quality of life) and family level variables (family functioning). Third, by making use of multi-level analyses, we were able to model the interdependence in the family relationships.

The present findings must be considered within the scope of some important limitations. First, only Dutch speaking families were invited for participation. With respect to the current multicultural society, however, this

language criterion might have been a barrier for ethnic minorities. Second, we only focused on children diagnosed with leukemia and non-Hodgkin lymphoma. As a consequence, it is important to highlight that families of children with other cancer diagnoses may have different experiences. In addition, as ALL was diagnosed in 73.9% of our families and this diagnosis is most common in early childhood, peaking between 2 and 5 years of age, most ill children were too young to be invited to our study (see method section: "all children aged 5 years and more and both parents were asked to complete a set of questionnaires at five different time points"; mean age at diagnosis = 6.6 years). As a consequence, our sample only consisted of 60 children with cancer. Third, as being overwhelmed by the cancer diagnosis was one of the most important reasons for non-participation, we can question whether more stressed families in general were more likely to refuse participation (i.e., selection bias). Fourth, as the associations described in this study are correlational in nature, the temporal order of the variables under investigation could not be tested with the present data. As a consequence, inverse associations (e.g., higher QoL predicting more adequate family relationships) are also possible. Fifth, for this study, we adapted the timeframe of the PSS from "in the last month" to "since the cancer diagnosis". This might have consequences for the questionnaire's psychometrics. A final limitation is the low reliability coefficients for the FSI subscale (FES) and the PSS scale (mothers; fathers), which could not be improved by dropping one or more items. For the FES, this is in line with previous literature (Hildenbrand & Alderfer, 2019). So, caution is warranted when interpreting these (sub)scales and further research is needed to confirm our findings.

Clinical implications

Our findings provide evidence for the fact that the life of all family members is impacted by a childhood cancer diagnosis and that, therefore, the psychosocial needs of *all family members* should be recognized and addressed

by the multidisciplinary intervention team. Multiple specific recommendations arise from the present study. First, our findings provide further empirical support for existing social ecological prevention and intervention models in child health. For example, our findings on the association between family functioning on the one hand and emotional well-being and quality of life in cancer-affected families on the other hand, fully support the recommendations of the pediatric psychosocial preventative health Model (PPPHM; Kazak, 2006) that all families of children diagnosed with cancer should be screened for factors potentially predisposing them for maladjustment or distress, including family risk factors (e.g., family conflict, family structure). Accordingly, clinical interventions for cancer-affected families can then be tailored to these family risk factors, the families' specific care needs, and the care expectancies of these families (ranging from standard psychosocial care to more intensive individual or family therapy; see Kazak, 2006 for greater detail). Second, clinical interventions should also be sensitive to some important individual characteristics of patients, parents and siblings facing childhood cancer. For example, the age of the diagnosed child, as less positive feelings, less loneliness and better quality of life is reported when the diagnosed child is older. Third, as cancer-related emotions proved to be mostly explained by the differences between family members (and not the differences between families), and as for example, siblings experience less positive emotions than patients, mothers and fathers, interventions should also take into account the potential differences and specific intervention needs of each family member. This may imply that individual family members may particularly benefit from social contact with fellow sufferers to share their experiences (e.g., via group therapy). Finally, discrepancies in perceptions across family members as well as our findings on the role of family functioning speak to the need to involve all family members in intervention, both with respect to individual level variables (emotions and quality of life) and family level variables (family functioning). More specifically, to facilitate and enhance family communication as well as to help families to get insight

in every family member's perspective, appraisal of the cancer diagnosis, and subjective meaning making, interventions at the family level –in addition to individual or group therapy- would be particularly suited for families facing pediatric cancer.

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CHAPTER 7

SIBLINGS DEALING WITH PEDIATRIC CANCER: A FAMILY- AND CONTEXT-ORIENTED APPROACH[1]

Pediatric cancer is a severe life-threatening disease that poses significant challenges to the life of the siblings. Based on the Social Ecology Model, the aim of the current study was to explore the association between intrafamilial (family functioning, family support) and contextual (network support) resources, and the individual adjustment of siblings facing cancer in their brother/sister. Participants were 81 siblings of children with leukemia or non-Hodgkin lymphoma. The mean siblings' age was 10.32 years. Siblings completed the Family Environment Scale, the Social Support Questionnaire for Children, the Situation-Specific Emotional Reactions Questionnaire and the Pediatric Quality of Life Inventory. Data was analyzed using a multi-level approach. Family functioning, family support and network support proved to be related to siblings' cancer-related emotional reactions post-diagnosis, but not the siblings' quality of life. In addition, the present study suggests to take into account the gender of the ill child and the age of the siblings. Our findings led to the conclusion that resources at both the intrafamilial level and the contextual level are important for explaining sibling adjustment post-diagnosis. Interventions targeting the sibling, the family and the external network are warranted to enhance sibling adjustment.

[1]Van Schoors, M., Sels, L. Goubert, L., & Verhofstadt, L. L. (2020). Siblings dealing with pediatric cancer: A family- and context-oriented approach. *Manuscript accepted for publication in Journal of Pediatric Oncology Nursing.*

Introduction

Pediatric cancer is a severe life-threatening disease with every year 300,000 new diagnoses worldwide (Steliarova-Foucher et al., 2017). Given the highly interdependent nature of family relationships, all family members, including siblings, are impacted by the illness (Van Schoors et al., 2019b). Previous studies showed that siblings of children with cancer often suffer from the absence of their parents and their ill brother/sister at home, changes in their day-to-day routines (e.g., grandparents taking over parental roles), and increasing household chores and responsibilities (Alderfer et al., 2010; Van Schoors et al., 2019a). Two systematic reviews, incorporating 168 empirical studies, documented the impact of childhood cancer on siblings' individual functioning (Alderfer et al., 2010; Long et al., 2018). Specifically, these studies indicated that while there is no evidence for elevated prevalence of psychiatric disorders (e.g., anxiety and depression) in siblings, they often suffer from severe levels of post-traumatic stress symptoms, especially in the first months after diagnosis (Alderfer et al., 2010; Long et al., 2018). In addition, they often report poor quality of life in several domains (i.e., emotional, family, and social; Alderfer et al., 2010) and negative emotional reactions (i.e., shock, fear, worry, sadness, anger, and guilt) during cancer treatment. Finally, school-aged siblings often display more absenteeism and problems at school than peers (Alderfer et al., 2010; Long et al., 2018).

Because adjustment problems appear to occur only in a subset of the siblings studied, researchers started to focus on possible resources and tried to explain why some siblings adapt better than others. Resources that have been studied in the context of childhood cancer in general can be situated at three levels: the individual level, the intrafamilial level, and the contextual level. For example, existing research on the individual adjustment of siblings when facing a cancer diagnosis in their brother/sister indicated that maintaining positive expectations regarding the illness (coping; individual resource; Houtzager et al., 2005), lower levels of family conflict (family functioning;

intrafamilial resource; Van Schoors et al., 2017), and more network support (contextual resource; Barrera et al., 2004) is associated with better sibling adjustment.

The present study

In the current study, we focused on resources situated at the intrafamilial and contextual level. In other words, a family- and context-oriented approach was applied to investigate siblings' adjustment to their brother'/sister's cancer diagnosis. This approach was deemed necessary, as every child (ill child, sibling) is embedded in a broader social context with mutual influences between the stressor (i.e., cancer diagnosis), the child, and his/her social context (e.g., family, external network; Social Ecology Model; Bronfenbrenner, 1997). More specifically, we focused on *family functioning* as a first *intrafamilial resource*. There is abundant empirical evidence that the way in which the family as a whole deals with and responds to childhood cancer ('family functioning') impacts the adjustment of the siblings (Long et al., 2013; Van Schoors et al., 2017; 2019b). For example, families need to redefine their relationships, communicate effectively (i.e., "emotional closeness within the family") and flexibly renegotiate roles and responsibilities (i.e., "family structure") to accommodate the demands of cancer, and poorly functioning families who struggle with these demands may be at risk for adjustment problems in all family members, including siblings (Long et al., 2013; Van Schoors et al., 2017; 2019b).

As a second *intrafamilial resource,* we focused on *family support*. Family support refers to practical assistance, encouragement, and caring within the family, as perceived by the sibling (Walsh, 1998). The family has been shown to be an important source of support for siblings facing childhood cancer, with the mother being identified as the most important source of support (Van Schoors et al., 2019a). In addition, based on a recent meta-analysis (see Van Schoors et al., 2017 for an overview), more family support

is associated with better adjustment (e.g., less anxiety, depression, post-traumatic stress symptoms) in siblings.

As a third and final resource, we focused on *network support* (contextual resource). Network support refers to emotional, practical, and informative support and help from an individual outside the family (i.e., external network), as perceived by the sibling (Gordan, 2011). Due to the fatal character of the cancer diagnosis, parents primarily focus on the ill child, which is at the cost of time and attention allocated to the siblings (Prchal & Landolt, 2012). While parents accompany the diagnosed child to the hospital, others (like grandparents or neighbors) take care of the siblings and help them cope with the illness and its consequences (Van Schoors et al., 2018; 2019a), making these "others" important sources of sibling-support. In addition, in line with the developmental age of the siblings (Greenberg et al., 1983), friends and peers are important sources of sibling-support as well (Barrera et al., 2004).

To the best of our knowledge, there are no studies investigating family functioning, family support, and network support together as predictors when facing childhood cancer. From a social-ecological perspective (Bronfenbrenner, 1977), however, adjustment cannot be understand by solely focus on the individual; adjustment is the product of a constellation of forces from the family and the external network. In other words, including intrafamilial (family functioning, family support) and contextual (network support) resources are needed to best understand sibling adaptation when facing childhood cancer and to get insight in (the importance of) the social context siblings are living in. Therefore, the aim of the current paper is to investigate the association between *family functioning*, *family support*, *network support*, and the *individual adjustment* (quality of life; cancer related emotions) of siblings facing cancer in their brother/sister. More specifically, we expected that better family functioning (more emotional closeness within the family and a more firm family structure), more family support, and more network support would be associated with better individual outcomes (i.e.,

less loneliness, less uncertainty, more emotional involvement in the illness process, more positive cancer-related emotions, and better quality of life) in siblings.

Method

Participants

Details on the sample are listed in Table 1. Our sample consisted of 81 siblings of children with leukemia (N = 67) or non-Hodgkin lymphoma (N = 14). The mean ill child's age at diagnosis was 7.60 years (SD = 4.91; Range = 1-18). The mean siblings' age was 10.32 years (SD = 4.57; Range = 5-25). All siblings were Caucasian and living in Belgium. Ethical approval from the University Hospitals of Ghent, Louvain, Antwerp, and Brussels had been secured for the study. Written informed consent forms were obtained from all participating siblings above the age of 12. Parental (written) consent was obtained for all participating siblings under the age of 16.

Procedure

The present study is part of a larger study examining the impact of pediatric cancer on families, that is the 'UGhent Families and Childhood Cancer Study'. For this large-scale project, families of children with leukemia or non-Hodgkin lymphoma aged zero to 18 years were invited to take part in a longitudinal questionnaire study. All family members aged five years and above (patient, siblings, mother, father) were asked to complete a set of questionnaires at 5 different time points (diagnosis – 2.5 years post-diagnosis). Exclusion criteria were not speaking Dutch, expression of a developmental disorder in the diagnosed child, and cancer relapse. For the present study, only

Table 1
Background Characteristics of the Study Sample

Demographic variable		
N siblings (*n* boys, *%*)		81 (39, 48%)
Age, siblings, mean *(SD)*		10.32 (4.57)
Age ill child at diagnosis, mean (*SD*)		7.60 (4.91)
Sex ill child, *n* boys *(%)*		45 (56%)
Diagnosis[1], n (%)	ALL	60 (74%)
	AML	5 (6%)
	CML	2 (3%)
	Non-Hodgkin Lymphoma	14 (17%)
Family status, *n (%)*	Married/Co-habiting	68 (84%)
	Single parent	8 (10%)
	Stepfamily	5 (6%)
Number of children in the family, *n (%)*	Two children	24 (30%)
	Three children	43 (53%)
	Four children	13 (16%)
	Five children	1 (1%)

Note. [1]ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia

the repeated measurements of the sibling data were used (see Data Analytic Strategy for more details). Collection of the data was conducted between June 2014 and January 2020, with a response rate of 65%. The most frequent reasons for non-participation were being too overwhelmed by the diagnosis and lack of time.

Measures

Due to a minimum age limit for the questionnaires, some younger siblings did not complete all questionnaires. For each questionnaire, the minimum age and the number of siblings *excluded* for the questionnaire based on the minimum age ("N_{age}") are reported.

Family Functioning. The Dutch version of the Family Environment Scale (FES; Moos & Moos, 1994) was used to measure family functioning. The questionnaire contains 77 '*yes–no'* items, across seven subscales: (1) cohesion (e.g., "At home we do everything together the entire weekend"), (2) expressiveness (e.g., "We have many spontaneous conversations in our family"), (3) conflict (e.g., *"*We argue a lot at home"), (4) organization (e.g., "When we do something we always prepare well*"*), (5) control (e.g., "We make sure that everyone in the family keeps to the agreements"), (6) norms and values (e.g., "We believe in competition and believe that the best must win"), and (7) social orientation (e.g., "We think it is important to be aware of politics*"*). From these subscales, two composite scores were calculated by summing the relevant item scores: the family relation index (FRI; cohesion + expressiveness – conflict) and the family structure index (FSI = organization + control), reflecting the affective nature of the family relationships ("emotional closeness within the family") and the extent to which the family is structured ("family structure") , respectively. Higher FES composite scores reflect more emotional closeness within the family (FRI; more cohesion and expressiveness and less conflict) and a more firm family structure (FSI; more control and organization). These composite scores were included in the analyses as indicators for family functioning. The FES is applicable for children aged 11 and above (N_{age} = 48) and has good reliability and validity (Jansma & De Coole, 1995). In the present study, the Cronbach's alpha reliabilities were .76 for the FRI and .77 for the FSI.

Family and network support. The Social Support Questionnaire for Children (Gordon, 2011) assesses the amount of social support as perceived by the sibling. The questionnaire has five factors representing distinct sources of support: parents (e.g., "A parent makes sure I have what I need"), relatives (e.g., uncle, grandparent; "I have a relative who gives me good advice"), non-relative adults (e.g., coach, teacher; "An adult cares about my feelings"), siblings (e.g., "I have a sibling I can trust to keep a secret"), and peers (e.g., classmate, close friend; "A peer comforts me when I am upset*"*) and consists

of 50 items. All items were rated on a 4-point Likert scale from *never true* to *always true*. Two composite scores were calculated by summing the different item scores, reflecting the total amount of perceived family support (i.e., support from parents and siblings*)* and network support (i.e., support from relatives, non-relative adults and peers*)*. These composite scores were included in the analyses as indicators for family and network support, with higher scores reflecting higher levels of perceived social support from the family and the external network, respectively. The questionnaire is applicable from the age of 7 (N_{age} = 15) and has satisfactory to good validity and reliability (Gordon, 2011). In the present study, Cronbach's alpha coefficients were .94 (family support) and .96 (network support).

Cancer-Related Emotions. The Situation-Specific Emotional Reactions Questionnaire - Siblings (SSERQ-S; Houtzager et al., 2004) is developed to assess emotional reactions in siblings after facing a pediatric cancer diagnosis in their brother/sister. The questionnaire consists of 26 items, divided in four subscales: (1) loneliness (e.g., "I feel alone"), (2) uncertainty (e.g., "I worry about the future"), (3) emotional involvement (e.g., "I regret that my parents have to go through all this"), and (4) positive cancer-related feelings (e.g., "I am proud that I can keep up with it"). All items are rated on a 4-point Likert scale from *almost never* to *almost always*. Higher sum scores represent more emotional reactions. The questionnaire is applicable from the age of 7 (N_{age} = 9) and has satisfactory to good validity and reliability (Houtzager et al., 2004). In the present study, Cronbach's alpha coefficients were .79 (loneliness), .87 (uncertainty), .88 (emotional involvement), and .68 (positive feelings).

Quality of Life (QoL). The Pediatric Quality of Life Inventory (Varni, Seid & Rode, 1999) measures children's health-related quality of life. Different versions of the questionnaire are available, for example the PedsQL™ 3.0 Cancer Module (children with cancer) and PedsQL™ Generic Core Scales (healthy children). In this study, the PedsQL™ Generic Core Scales measured the siblings' quality of life. The questionnaire is composed

of 23 items comprising 4 dimensions: (1) physical functioning (e.g., "It's hard for me to lift big things"), (2) emotional functioning (e.g., "I feel sad"), (3) social functioning (e.g., "Other kids tease me"), and (4) school functioning (e.g., "It is hard to pay attention at school"). All items are scored on a five-point Likert-scale from *never* to *almost always*, reversed and rescaled to a 0-100 scale: a score of 100 represents the best quality of life possible, a score of 0 represents the worst quality of life possible. Scale scores, as well as the sum score, are computed by adding together the different item scores and dividing this obtained score by the number of items answered. Only the sum score (i.e., general quality of life) was included in the analysis. The questionnaire is applicable from the age of 5 (N_{age} = 0) and has sufficient to good validity and reliability (Varni et al., 2001). In the present study, the Cronbach's alpha coefficient was .87 for the sum-score.

Data Analytic Strategy

We investigated the associations between family functioning (emotional closeness within the family and family structure), family support, network support, and siblings' individual adjustment. We modeled the effects of family functioning, family support, and network support on five adjustment indicators: quality of life and the cancer-related emotions loneliness, uncertainty, emotional involvement, and positive feelings. Because our data were clustered, with measurement occasions (level 1, which ranged from 1 to 5 for each participant) that are nested within siblings (level 2), and siblings nested within families (level 3), we first investigated dependencies between observations by empty three-level models (in accordance with Hoffman & Stawski, 2009). Specifically, between-person and between-family variances were estimated for quality of life and cancer-related emotions. Because the between-family variances were negligible in most models (e.g., the total proportion of variance in quality of life between families was .002 %) and the inclusion of this level led to estimation problems, we decided to omit this third

level from the main analyses. In the reported two-level models, we allowed a random intercept, which varied for each sibling within each family, and indicated that observations were repeated across time. To account for missing data, efficient estimates were obtained through maximum likelihood estimation procedures. Analyses were carried out with Statistical Package for the Social Sciences version 11.5 (SPSS Inc., 2003).

Given high correlations between our key predictors of interest (see Table 2), we modeled the effects of emotional closeness (i.e. family functioning), family structure (i.e. family functioning), family support, and network support on adjustment separately, controlling for covariates. The covariates time since diagnosis, number of children in the family, age of the ill child, gender of the sibling, gender of the ill child, and age of the sibling were considered in preliminary models, but only the latter two were correlated with our outcome variables, and thus integrated in the final models. All continuous predictors were grand-mean centered.

Results

Table 2 shows the descriptive statistics and correlations of the variables in our study.

The final models for the associations between family functioning, family support, network support, quality of life and cancer-related emotions are shown in Table 3.

Table 2

Descriptive statistics and correlations of the variables

	Range	*M (SD)*	*N*	1	2	3	4	5	6	7	8	9
1. Emotional closeness	11-30	22 (4.78)	65	-	.59**	.62**	.23	.31*	-.30*	.08	.38**	.41**
2. Family structure	3-20	13 (4.25)	65	-	-	.49**	.37**	.32**	-.18	-.04	.22	.42**
3. Family support	8-60	40.05 (11.90)	127	-	-	-	.56**	.11	-.09	.23**	.52**	.25**
4. Network support	8-60	36.56 (12.04)	127	-	-	-	-	.13	-.28*	-.13	.21*	.29**
5. Quality of life	34.69-97.50	73.63 (13.50)	172	-	-	-	-	-	- .50**	- .48**	- .23**	.002
6. Loneliness	3-21	15.77 (4.13)	148	-	-	-	-	-	-	.59**	.29**	-.003
7. Uncertainty	0-24	17.03 (5.56)	148	-	-	-	-	-	-	-	.57**	.05
8. Emotional involvement	0-20	8.12 (5.17)	148	-	-	-	-	-	-	-	-	.26**
9. Positive feelings	0-9	4.53 (2.41)	148	-	-	-	-	-	-	-	-	-

Note. *Correlation is significant at the .05 level; ** Correlation is significant at the .01 level.

Table 3

Final models for associations between family functioning, family support, network support, quality of life and cancer-related emotions

	Quality of Life		Cancer-related emotions: loneliness		Cancer-related emotions: uncertainty		Cancer-related emotions: emotional involvement		Cancer-related emotions: positive feelings	
	B [CI]	p value	B [CI]	p value	B [CI]	p value	B [CI]	p value	B [CI]	p value
Predictors										
Family structure	.53 [-.25,1.32]	.18	-.12 [-.40, .16]	.40	-.06 [-.40, .28]	.72	.30 [.02, .58]	.03*	.15 [.01, .28]	.03*
Gender ill child	2.86 [-5.38, 11.11]	.49	-2.39 [-5.04, .26]	.08	2.16 [-1.12, 5.44]	.19	3.73 [1.33, 6.13)	.004*	-.19 [- 1.46, 1.08]	.76
Age sibling	-.05 [-1.01, .90]	.91	-.02 [-.34, .30]	.91	.08 [-.31, .48]	.67	.19 [-.11, .50]	.21	-.05 [-.20, .10]	.51
Emotional closeness	.57 [-.09,1.22]	.09	-.28 [-.51, -.05]	.02*	-.14 [-.42, .14]	.32	.30 [.07, .52]	.01**	.14 [.03, .25]	.01**
Gender ill child	2.41 [-5.58, 10.40]	.54	-2.02 [-4.54, .50]	.11	2.29 [-1.01, 5.59]	.17	3.48 [1.04, 5.91]	.007**	-.26 [-.1.49, .96]	.67
Age sibling	-.22 [-1.13, .68]	.62	.03 [-.27, .32]	.86	.10 [-.28, .47]	.61	.09 [-.20, .38]	.541	-.10 [-.24, .05]	.18
Family support	.09 [-.11, .29]	.38	-.07 [-.13, -001]	.05*	.004 [-.07, .08]	.91	.19 [.12, .26]	.000***	.05 [.02, .09]	.004**
Gender ill child	-1.93 [-8.11, 4.26]	.54	-.32 [-2.18, 1.54]	.73	1.22 [-1.01, 3.44]	.28	1.75 [-.22, 3.73]	.08	.006 [-.89, .90]	.99
Age sibling	.36 [-.34, 1.06]	.31	-.07 [-.29, .15]	.53	.04 [-.22, .29]	.77	.11 [-.12, .34]	.34	-.18 [-.29, -.08]	.001***
Network support	-.01 [-.19, .21]	.92	-.08 [-.14, -.01]	.02*	-.004 [-.08, .07]	.93	.19 [.12, .26]	.000***	.05 [.01, .08]	.009**
Gender ill child	-1.68 [-7.89, 4.54]	.59	-.46 [-2.30, 1.37]	.62	1.23 [-.99, 3.45]	.27	2.21 [.19, 4.24]	.03*	.13 [-.77, 1.03]	.77
Age sibling	.41 [-.29, 1.10]	.25	-.08 [-.29, .13]	.46	.04 [-.21, .29]	.75	.14 [-.09, .38]	.23	-.16 [-.27, -.06]	.003**

Note. *p<.05 **p<.01 ***p<.001

Quality of Life. None of the predictor variables (emotional closeness within the family, family structure, family support, network support, and gender ill child, age sibling) were significantly associated with quality of life (all $p > .05$).

Cancer-Related Emotions.

Loneliness. A significant negative association between emotional closeness within the family and the siblings' feelings of loneliness was found ($p = .02$): more emotional closeness within the family (more cohesion and expressiveness, less conflict) was related to less feelings of loneliness in the siblings. In addition, more perceived social support from the family ($p = .047$) and the external network ($p = .02$) was associated with less feelings of loneliness in the siblings. None of the other predictor variables (family structure, gender ill child, age sibling) were significantly associated with loneliness (all $p > .05$).

Uncertainty. None of the predictor variables (emotional closeness within the family, family structure, family support, network support and gender ill child, age sibling) were significantly associated with uncertainty (all $p > .05$).

Emotional involvement. A significant positive association between family functioning, both the emotional closeness within the family ($p = .01$) and the family structure ($p = .03$), and the siblings' reported emotional involvement in the illness process was found: More emotional closeness within the family (more cohesion and expressiveness, less conflict) and a more firm family structure (more clear family organization and more parental control) was related to more siblings' emotional involvement in the illness process.

In addition, there was a significant positive association between the amount of perceived social support from the family ($p < .001$) and the external network ($p < .001$), and the siblings' emotional involvement in the illness process: the more perceived support (both from the family and the external

network), the more emotional involvement the siblings reported in the illness process. Furthermore, across the models[2], there was a positive association between the gender of the ill child and emotional involvement (all $p < .05$): brothers and sisters of a girl with leukemia or non-Hodgkin lymphoma reported more emotional involvement in the illness process as compared to brothers and sisters of an ill boy. The predictor variable age sibling was not significantly associated with emotional involvement (all $p < .05$)

Positive cancer-related feelings. A significant positive association between the family functioning, both the emotional closeness within the family ($p = .01$) and the family structure ($p = .03$), and the siblings' reported positive feelings was found: more emotional closeness within the family (more cohesion and expressiveness, less conflict) and a more firm family structure (more clear family organization and more parental control) was related to more positive feelings in the siblings. In addition, there was a significant positive association between the amount of perceived social support from the family ($p = .004$) and the external network ($p = .009$), and the siblings' positive feelings: the more support (both from the family and the network), the more positive feelings the siblings reported. Furthermore, in the network support model, the older the sibling, the less positive feelings s/he reported ($p < .003$). The predictor variable gender ill child was not significantly associated with positive feelings (all $p < .05$).

Bonferroni correction. Because five different outcome variables were modelled, a Bonferroni corrected threshold for significance of $p = .01$ can be applied. When taking into account this Bonferroni corrected threshold, only the associations between emotional closeness, social support (from the family and external network), the ill child's gender and emotional involvement in the illness process; and between emotional closeness, the sibling's age and positive feelings were preserved (see Table 3). Caution is

[2] This significant positive association between gender ill child and emotional involvement was *not* found in the model of network support and emotional involvement (p = .08); see Table 3.

warranted when interpreting the other associations ($.05 < p < .01$) and further research is needed to confirm these findings.

Discussion

Based on the Social Ecology Model (Bronfenbrenner, 1977) and using a multi-level approach (Hoffman & Stawski, 2009), the present study sought to examine whether intrafamilial resources (family functioning, family support) and contextual resources (network support) were related to the individual adjustment (quality of life and cancer-related emotions) of siblings facing cancer in their brother/sister.

Summary of results

Our findings indicate that both family functioning (emotional closeness & family structure), family support, and network support matter for the adaptation of siblings being confronted with childhood cancer. This is in line with our prediction and with previous studies on family functioning (Van Schoors et al., 2017; 2019b) and support (Brown et al., 2003; Dolgin et al., 1997) in the context of childhood cancer.

More specifically, we found that more *emotional closeness* within the family (more cohesion and expressivity, less conflict) was associated with lower levels of loneliness and higher levels of positive cancer-related feelings. In other words, when a sibling perceived his/her family as warm and loving (cohesion), open to talk about experiences and emotions (expressivity), and there were little conflicts, s/he reported to feel less lonely and more positive regarding the illness and its consequences. These findings are in line with the idea that family functioning is important for the adjustment of children when facing childhood cancer (see Van Schoors et al., 2017 for an overview). In addition, more emotional closeness within the family was associated with

higher levels of emotional involvement in the illness process: The better the family bounds, the more the sibling was worried about and committed to his/her ill brother/sister and parents. This association makes sense: a stable characteristic of the family (emotional closeness) is reflected in the involvement with the illness and the ill child/parents at one specific moment measured. In other words, it is the translation of a family characteristic into family members' interactional behavior and involvement in times of stress.

Furthermore, *family structure* was positively associated with the level of emotional involvement in the illness process and positive cancer-related feelings in the siblings: The more clear family rules, the more predictability in the household (organization), and the more parental control, the higher the emotional involvement in the illness process and the more positive cancer-related emotions the siblings reported. Possible explanations are twofold. When facing childhood cancer, the family's world is turned upside-down. The family's focus is allocated to the health of the ill child, at the cost of time and attention for the family as a whole and the siblings (Van Schoors et al., 2018). The sibling is often left to his/her lot and feels lost (Prchal & Landolt, 2012). As a consequence, the more siblings have the idea that their parents are still in control, and the more rules and predictability they have in the organization of their "new" life, the more siblings might feel comfortable and positive regarding the illness. Second, the age of the siblings should be taken into account. Ninety-one percent of the included siblings were younger than 16, and all included siblings were living together with their ill brother/sister and parents. For most of these siblings, a more firm family structure, with parents taking the mean family decisions, is conform the principles of the family life cycle (Minuchin et al., 1998): The younger the child, the more the parent takes the lead; the older the child, the more there is a balanced hierarchy between parent and child.

Furthermore, we found that siblings receiving more *support* from their family and the external network reported lower levels of loneliness, and higher levels of emotional involvement in the illness process and positive cancer-

related feelings. This is in line with previous studies illustrating that social support can buffer maladjustment after a childhood cancer diagnosis (Van Schoors et al., 2017). Moreover, this study shows that both social support from the family *and* the external network are needed to best help siblings: when parents are emotionally unavailable due to their own intense emotions or focus on the diagnosed child, the external network can provide sibling support; and vice versa.

Finally, the results of the present study suggest to take into account the *gender of the ill child* and the *age of the siblings*. First, siblings of an ill girl were more emotionally involved in the illness process than siblings of an ill brother. This is in line with the cultural idea that men/boys are perceived as "stronger" than women/girls, and thus that ill men need less help/care. In addition, the study of Bendelow (1997) showed that the pain expression of girls is higher than those of boys. In other words, girls show more pain than boys, and thus ill girls may attract more help/concerns in the other family members, as compared to ill boys. Second, older siblings reported less positive cancer-related feelings than younger siblings. This is in line with the idea that most cancer-related medical details are not shared with younger siblings, nor by the parents, nor by the medical team. As a consequence, older siblings may be more aware of the life-threatening character of the illness, and the possibility their brother/sister could die from it. Moreover, a fully understanding of the concept "death" is only reached at age 10 (Cox et al., 2005).

Surprisingly, none of the predictors of interest (family functioning, family support, network support) were significantly associated with quality of life. Possible explanations are twofold. First, the current study is characterized by a small sample (N = 85) and thus small power. It is possible that associations with smaller effect sizes were not detected. Second, whereas the SSERQ is a cancer specific questionnaire assessing cancer-related emotional reactions, the PedsQL is a population based questionnaire assessing general quality of life. In line with Alderfer et al. (2008) and Hildenbrand et al. (under

revision), we might question whether population based instruments are applicable in the context of chronic pediatric illnesses. Indeed, making use of population based measurements might ignore the understanding that what is dysfunctional in general population might be functional when facing a chronic child illness.

Strengths and Limitations

A first strength of the present study is the focus on siblings of children with leukemia or non-Hodgkin lymphoma. Up till now, most studies focused on the ill child or his/her parents (Alderfer et al., 2010). Second, in line with the Social Ecology Model (Bronfenbrenner, 1997), a family- and context-oriented approach was applied in the present study, including resources at the intrafamilial (family functioning, family support) and the contextual (network support) level. As previous research mostly focused on resources situated at only one level (individual, intrafamilial, contextual level) rather than combining these resources, they only provided a fragmented explanation of the processes underlying post-diagnostic sibling adjustment. In contrast, we were able to present a broader picture of the social context that might foster sibling adjustment when facing pediatric cancer. Third, by making use of multi-level analyses, we were able to take into account the nested structure of the data.

The current findings must be considered within the scope of some limitations. First, with only 81 included siblings, we can only draw limited conclusions regarding the association between family functioning, family support, network support and the siblings' adjustment. Further research, with larger samples, is therefore needed to confirm our findings. Second, as the associations described in this study are correlational in nature, the temporal order of the variables under investigation could not be investigated. Longitudinal analyses were considered, but were ultimately not carried out because only 13 siblings provided longitudinal data, and such a small sample

would have led to severe power issues. Third, only Dutch speaking siblings were included. Given the current multicultural society, research including different languages and ethnic populations is needed to increase the generalizability of the findings on sibling adjustment. In addition, we only focused on siblings of children diagnosed with leukemia and non-Hodgkin lymphoma. It is important to highlight that siblings of children with other cancer diagnoses may have different experiences. Fourth, as this study is part of a larger project including measurements of all family members, research assistants invited the ill child's parents to participate to the study. It is possible that families (including siblings) with severe adjustment problems declined for participation (i.e., most important reason for non-participation was being too overwhelmed by the diagnosis), or that more sibling-data could have been collected if the siblings themselves were asked to participate. Finally, given the criticism that a Bonferroni correction might be too conservative and may lead to reject results which actually are meaningful, this correction was addressed in the result section, but not in the discussion section. Overall, caution is warranted when interpreting associations with a p-value between .05 and .01, and further research is needed to confirm these findings.

Clinical Implications

Four clinical recommendations arise from the current study. First, the current results provide further empirical evidence for existing social ecological prevention and intervention models in child health, conform the clinical practice guidelines for families facing childhood cancer (Wiener et al., 2015). In line with these guidelines, specific clinical attention for siblings is needed, as some siblings might adapt worse than others. Second, clinical interventions should be sensitive to some individual characteristics of siblings facing childhood cancer. For example, the age of the sibling should be taken into account, as less positive cancer-related feelings are reported when the sibling is older. Third, given the importance of a clearly structured family life

(i.e., a clear family organization and more parental control) and family support post-diagnosis, psycho-education can be given to parents, patients, and siblings. During this psycho-education, clinicians should invite the family as a whole, give the family members easy-to-follow advices and emphasize the importance of the *family* for the adaptation of the siblings, taking into consideration the current shift in parental focus to the ill child and the parental guilt that can accompany this shift (Van Schoors et al., 2018). Fourth, as network support is an important contextual resource, clinicians should map the existing social network of the sibling and help siblings to ask for (emotional or practical) help where needed.

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CHAPTER 8

THE FAMILY PRACTICE OF SUPPORT-GIVING AFTER A PEDIATRIC CANCER DIAGNOSIS[1]

Pediatric cancer presents many challenges to the life of the child diagnosed with cancer and his/her family. Among the studies investigating risk and protective factors, social support has emerged as an important construct. However, little is known on *how* family members support each other in this particular context. In order to further explore this process, interviews were performed separately with mothers, fathers and siblings. Multi Family Member Interview Analysis was used as the methodological framework to analyze the individual interviews, allowing a detailed and systematic analysis of shared family experiences. The analysis of the data revealed three themes: "Being together matters": the families identified the need of being physically together; "Finding support in (not) talking": the complexity of sharing emotions was explained, and "Working together as a team": the families described working together as a team in order to get everything organized. This study broadens our understanding of the interpersonal process of family support-giving when facing pediatric cancer.

[1]Van Schoors, M., De Mol, J., Verhofstadt, L. L., Goubert, L. & Van Parys, H. (2019). The family practice of support giving after a pediatric cancer diagnosis: A multi-family member interview analysis. *European Journal of Oncology Nursing, 44*, 101712. doi: 10.1016/j.ejon.2019.101712

Introduction

Pediatric cancer is an unpredictable and uncontrollable stressor for the diagnosed child and his/her family members. There are a number of pediatric cancers, with blood cancer, including leukemia (30%) and lymphoma (8%), as the most common type (American Cancer Society, 2016). Current pediatric cancer treatments are very intensive, including hospitalization, painful and invasive procedures, surgery, chemotherapy, and/or radiation therapy (Alderfer et al., 2009). The impact of the illness and its treatment on the physical and psychological wellbeing of all family members is therefore undeniable.

There is a growing body of literature on the psychological adjustment of families dealing with pediatric cancer. In the current literature, it is concluded that a significant subset of patients, parents, and siblings are at risk of adjustment difficulties. For example, some children with cancer experience social or emotional problems during (Kestler & LoBiondo-Wood, 2012) or after treatment (Kazak et al., 2001). In addition, feelings of uncertainty, anxiety, depression, and posttraumatic stress symptoms can be observed in parents of children with cancer shortly after diagnosis (Vrijmoet-Wiersma et al., 2008). Similarly, some siblings show elevated levels of post-traumatic stress symptoms, negative emotions or report poor quality of life when compared to siblings of healthy children (Alderfer et al., 2010; Long et al., 2018). It should be noted, however, that the research described above also revealed considerable variability, across and within studies, in individual outcomes for children being confronted with pediatric cancer and their family members. More specifically, some children and family members seem to adjust better than others.

Given this great variability of outcomes, a growing number of researchers have tried to explore why this is the case. Among the studies investigating detrimental and protective factors influencing the adjustment of families being confronted with pediatric cancer, social support has emerged

as an important construct (Alderfer & Hodges, 2010). Moreover, as children are embedded in a family (Carr, 2012; Fiese, 1997) and the family is an important social support system for children and adolescents (e.g., Newman et al., 2007), the importance of *family support* in the context of pediatric cancer has frequently been emphasized. Indeed, according to several empirical studies (e.g., Alderfer & Hodges, 2010; Varni et al., 1994; Zegaczewski et al., 2015) and a recent meta-analysis (Van Schoors et al., 2017) greater perceived family support is associated with better child adjustment, for both the diagnosed child and their siblings.

Notwithstanding the growing body of evidence emphasizing the importance of family support in the context of pediatric oncology, little is known on *how* exactly family members support each other after a pediatric cancer diagnosis. A qualitative interview study was therefore set up to allow an in-depth exploration of the *specific ways* in which family members support each other when facing pediatric cancer. Furthermore, the concept of family support was assessed from the perspectives of multiple family members. Existing qualitative studies on (the importance of) family support typically make use of a single family member as the informant (Van Schoors et al., 2015). Because the unit of interest (i.e., support at the family level) should harmonize with the unit of measurement (Weber, 2011), we argue that studies with only a single informant do not adequately capture support within the family.

To fill this gap, we completed a study using one-to-one interviews with *multiple* family members (i.e., mothers, fathers and siblings), focusing on family support-giving. Multi Family Member Interview Analysis (MFMIA; Van Parys et al., 2017) was used as a methodological framework to analyze the individual interviews, focusing on families as the unit of analysis. Inspired by Interpretative Phenomenological Analysis (Smith et al., 2009) and Dyadic Interview Analysis (Eisikovits & Koren, 2010), MFMIA allows a detailed and systematic analysis of shared family experiences (Van Parys et al., 2017). This approach has proved insightful in studies that analyze

experiences shared by family members, particularly when assessing sensitive topics such as adjustment to an illness (Eisikovits & Koren, 2010). The research question that guided our interviews and data-analysis was '*How exactly do family members support each other when facing a pediatric cancer diagnosis?'*.

Method

Participants

The present study is part of a larger ongoing study in Flanders (Belgium), examining the impact of a pediatric cancer diagnosis on families (see also Van Schoors et al., 2018, 2019). Using a purposive sampling strategy, children diagnosed with leukemia or non-Hodgkin lymphoma between the age of one and eighteen, their parents and any siblings were invited to take part in a longitudinal survey. Exclusion criteria were: (1) not speaking Dutch, (2) expression of a developmental disorder in the diagnosed child, and (3) relapse. All participating parents (N = 173 individuals, including 55 couples) and siblings, aged between 10 and 16 (N = 27), were subsequently invited to complete an interview about their experiences regarding the influence of the cancer diagnosis on their family life. Thirty-three couples (60%) and 15 siblings (56%) agreed to participate in this interview study. Ten couples and ten siblings were then randomly selected and contacted by the first author. Ten mothers, ten fathers and ten siblings were interviewed separately. For the purpose of this study, only data from the four families in which both parents and at least one sibling participated were analyzed as this allowed for an analysis at the family level. Table 1 shows the participating families' characteristics. Data-collection was done by two onco-psychologists under the supervision of the last author (H.V.P.) who is specialized in family interview techniques. Approval of the Ethics Committee of the University

Table 1

Families' characteristics

		Daisy	Lien	Bob	Ruben
Diagnosed Child	Age	16	16	4	8
	Gender	Female	Female	Male	Male
	Diagnosis[1]	CML	Non-Hod	ALL	ALL
	Time since diagnosis (months)	5	5	26	24
Parents	Age mother	48	45	38	42
	Age father	49[2]	44	37	44
	Marital status	Divorced	Married	Married	Married
Siblings	Number of siblings	2	2	2	1
	Age first participating sibling	16	15	11	10
	Gender first participating sibling	Female	Female	Female	Male
	Age second participating sibling	-	14	-	-
	Gender second participating sibling	-	Male	-	-

Note. [1]CML = Chronic Myeloid Leukemia; Non-Hod = non-Hodgkin lymphoma ; ALL = Acute Lymphoblastic Leukemia; [2] = non-participating father

Hospitals of Ghent, Brussels, Antwerp and Louvain was obtained, both for the longitudinal survey and the interview study. All participants received written and verbal information about the study. Confidentiality was assured during all phases of the study.

Data collection

Interviews took place at the participants' homes. Parents gave their written informed consent at the time of the interview. For the sibling interviews, written informed consent of both parents and assent of the child were obtained.

Participants' (parents and siblings) interviews consisted of questions about their experiences of the diagnostic and treatment process and their perspectives on their family relationships and family functioning post-diagnosis (Table 2; detailed interview guide available upon request). All family members were interviewed separately as this allowed them to share their own perspective on the research topic without having to take into account the feelings of other people in the room (Morris, 2001). Parent interviews lasted between 59 and 143 min; sibling interviews lasted between 40 and 107 min. Each interview was audio-taped and transcribed verbatim using pseudonyms.

Table 2
Family Interview questions

Parent interview	Sibling interview
To what extent did the disease affect your family?	How has the disease affected your family?
How did you as a family experience/endure this period?	What changed in your family when your brother/si became ill?
What has changed in your family due to this diagnosis/illness?	To what extent could you talk about the disease with your parents? With your brother/sister?
To what extent was there an influence on your relationship with your children?	How did this period affect your relationship with your parents? Or with your brother/sister?
How would you describe social support within the family?	How did you experience the support within your family?

Qualitative Analysis

Inspired by Interpretative Phenomenological Analysis (IPA; Smith et al., 2009) and Dyadic Interview Analysis (Eisikovits & Koren, 2010), Multi Family Member Interview Analysis (MFMIA; Van Parys et al., 2017) facilitates the understanding of broader family dynamics by obtaining and combining the perspectives of multiple family members. MFMIA consists of three phases (Fig. 1; for a more in depth description of this method, see Van Parys et al., 2017, p. 395).

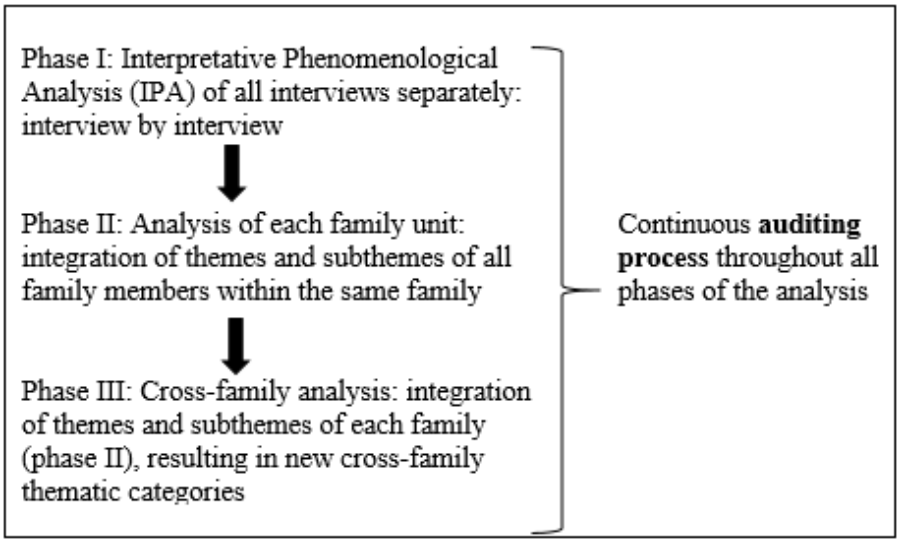

Figure 1. Overarching data analysis strategy

In the first phase, the interviews were analyzed separately, which included (a) adding initial notes based on interview observations and theoretical concepts to each transcript, (b) line-by-line coding based on the research question (‘*how do family members support each other exactly when facing a pediatric cancer diagnosis?’)*, (c) clustering of the codes (e.g., “it seems important for this father to accompany the ill child to the hospital) into higher level themes (e.g., being physically together is important), and (d) writing up a narrative/summary for each of the interviews. The second phase consisted of a within-family analysis in which themes of the interviews with

the mother, father and sibling(s) of one family were combined in a theme structure for the family. In a third phase, we searched for parallel themes between the different families. Thus, the final list of main themes and subthemes reflects patterns of convergence between the different families, whilst doing justice to divergences and unique outcomes as well. The merit of this approach lies in the focus on the family level: by first doing the MFMI-analysis within the families, and then across the families, we are able to say something about what typifies *familie*s, rather than what typifies mothers, fathers or siblings.

To enhance the trustworthiness of the study, a team of auditors (listed here as co-authors) was invited to challenge the way the first author constructed themes and subthemes at several points in the analysis (Hill et al., 1997). Based on extensive research reports, these auditors verified whether the analysis had been conducted systematically and transparently, and whether the research report was credible (Smith et al., 2009). More specifically, the first auditor (last author; H.V.P.) is specialized in family interview techniques and qualitative data-analytic methods, and was the principal investigator's academic mentor. The second author (J.D.M.) functioned as the second auditor and notably contributed to the construction of emergent themes at a theoretical and conceptual level.

Results

Analysis of the data on the family practice of support-giving after a pediatric cancer diagnosis could be clustered into three main themes. Specifically, in the first theme, the families identified the need of being physically together, both as a family and with the diagnosed child. The second theme articulated the eagerness of the families to talk about the illness and its impact, as this was experienced as a relief. However, talking about emotions was sometimes also experienced as hard and some family members preferred

not sharing these experiences. In the third theme, the families described working together as a team in order to get everything organized (see Fig. 2).

Theme 1: Being Together Matters

The families in our studies indicated that they felt supported by each other's presence. They identified a need of being physically together as one family unit, and they ensured that always (at least) one of them accompanied the diagnosed child to the hospital, regardless of the child's age.

Subtheme 1a: Feeling strengthened by being physically together. From the moment of the cancer diagnosis onwards, the families in our study felt separated: The diagnosed child and one or both parent(s) stayed at the hospital, whereas the siblings stayed at home or were taken care of by others.

> *When Lien had to go to the hospital, our family was torn apart. When Lien came back, we were reunited. So yeah, it really felt like 'we're back together again'. (Sibling, 16 years)*

As a consequence, for most families, the moments they were all together were scarce and thus were appreciated more. They enjoyed spending time as *one* family.

> *I liked it when we all watched a movie together, or when we went out for a picnic, or when my brother, my dad and I were playing football together. It never lasted long, but it was really cool and then my mother would watch us with a smile. (Sibling, 10 years)*

In addition, the importance of physical contact in order to cope with the cancer experience was emphasized. Parents and siblings gave hugs to express their love and to comfort each other.

> *Every day there were hugs. The children cuddled each other, just as we cuddled the children, and I hugged my wife. (Father)*

Similarly, a mother in another family described that embracing the ill child

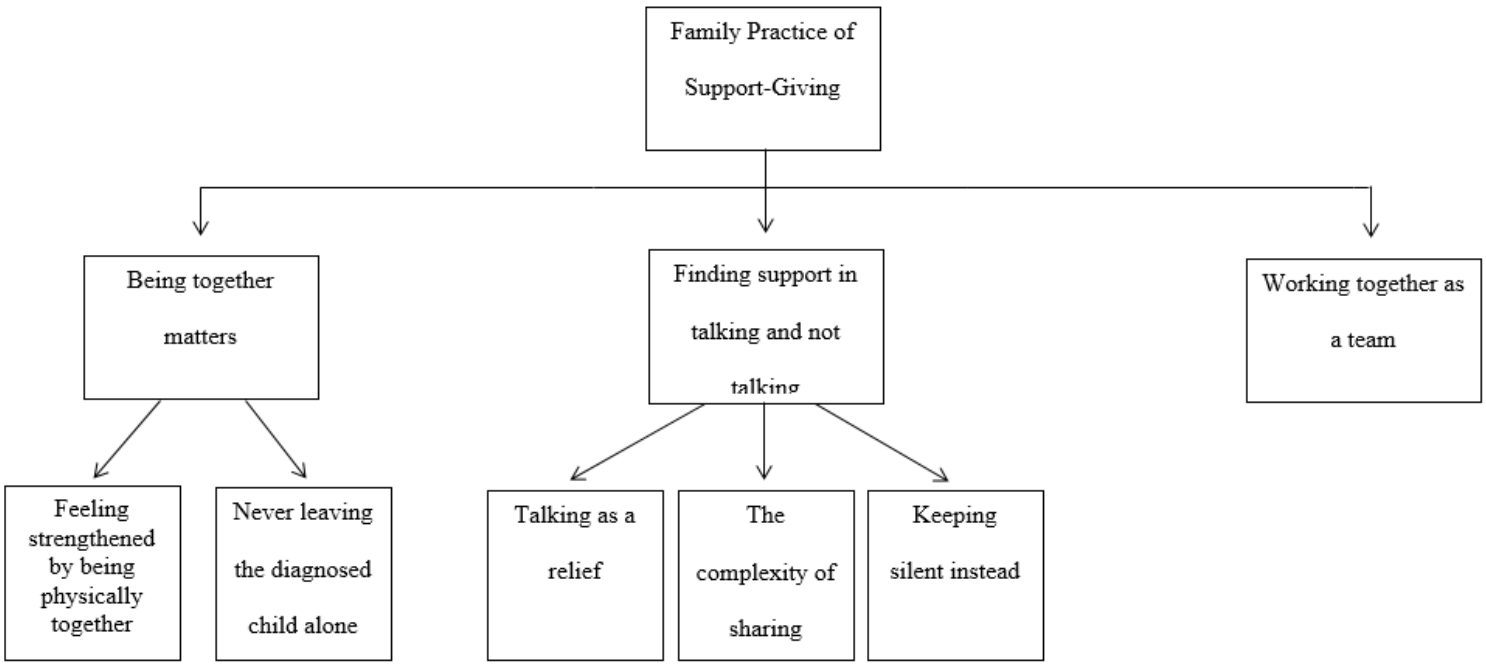

Figure 2. Main themes and subthemes

was all she could do, as words and other attempts to help were often insufficient.

> *If your sixteen year old daughter cries constantly … The only thing you can do is take her into your arms and lay down in the bed together with her. (Mother)*

It seemed that parents and children tried to support each other by being physically together: by their presence, they showed companionship, availability, and emotional support.

Subtheme 1b: Never leaving the diagnosed child alone. The parents in our study ensured that always one of them accompanied the diagnosed child to the hospital and that the ill child was never left alone.

> *Bob has never been by himself. [He was] either with me or my wife. (Father)*

Regardless of the age of the diagnosed child, accompanying the ill child during the hospital stays was deemed important by the parents of all four families. The parents indicated that the cancer treatment was too severe for the child to endure alone.

> *When you have a child with cancer, even when this child is sixteen years old, you don't leave her alone. She wouldn't have been able to. It was too heavy. (Mother)*

In addition, the siblings also tried to visit their ill brother or sister as much as possible.

> *Yeah, I visited her quite often. There is no other way right, cause she's my sister. (Sibling, 14 years)*

For this sibling, visiting his ill sister showed that he was concerned about her health and that she wanted to help her with his presence. Parents accompanied the diagnosed child day and night and never let that child alone, but this was different for the siblings. They reported having a need to continue their daily life as well, and searched for a balance between supporting their ill brother/sister, attending school, and maintaining their own social life.

> *I didn't feel like, I mean, maybe that sounds a little selfish: 'I will visit Lien often but I won't be there every day'. (Sibling, 15 years)*

Finally, in one family, the mother considered the implications of going out as a family while the diagnosed child was at the hospital. Taking into account fatalistic scenarios, this family decided to never leave the diagnosed child alone and therefore never went on a trip with just the siblings.

> *My father wanted the four of us (parents and two siblings) to go to an amusement park one day. But I really didn't feel like it, just the idea, maybe it sounds strange, but imagine that something happens to the four of us, that we have an accident, then he [diagnosed child] would stay behind alone. I just couldn't think of that. Let's say, I didn't want that. (Mother)*

Theme 2: Finding Support in Talking and not Talking

Most families in our study found it important to talk about the cancer experience, and perceived this talking as a relief. However, for some family members, talking about this emotional experience was not easy, and therefore not talking was preferred. This theme shows the complexity of communication about the pediatric cancer diagnosis and treatment within families.

Subtheme 2a: Talking as a relief. The families in our study found it important to talk about the illness and its consequences.

> *We kept on talking to each other. And that's really important, that's really really important. (Father)*

Moreover, talking about the cancer diagnosis was perceived as helpful in coping with the experience. For one sibling, this was even the most helpful thing in coping with his brother's illness.

> *Interviewer: What do you think helped you the most? Sibling (10 years): Uhm, talking about it with my family.*

It seems that family members in particular understood each other's worries and fears, and that, therefore, family members appreciated each other's help more than help from outsiders.

> *It's still odd when I talk about it or when I hear about it (cries). I also told my wife. I couldn't think of anyone else I would have been able to overcome this period with. So I must kiss both my hands that I have a fantastic wife and two fantastic children. (Father)*

In addition, talking about the cancer and its consequences was also perceived as a relief. In one family, this was described by both the mother and the sibling.

> *I think that's always been a strength of my husband and me, that both of us are people that talk. Neither of us bottle up. That's how we always did things, and it really helped us this time. (Mother)*
>
> *Sometimes we also had talks about the illness in the hospital room or something, how it was going with my brother and that was a relief, when we had a talk like that. (Sibling, 10 year)*

For this family, being able to talk about the cancer experience was described as one of their family's strengths. And although this strength was already present pre-diagnosis, it became even more pronounced post-diagnosis.

Subtheme 2b: The complexity of sharing emotions. Although all of the families in our study talked about the cancer, in every family, communication seemed to have its own characteristics and challenges. For example, in one family, parents and siblings found it hard to talk about emotions, instead talking about the medical details of the cancer and its treatment.

> *We did talk about the illness, but let's say, not about feelings or anything. (Mother)*
>
> *Interviewer: Were you able to talk about the illness with your parents?*
> *Sibling (11 year): Yes, when I had questions, I was. But apart from that, well, there used to be questions like 'how did it go' and things like that, but apart from that, we didn't talk about it a lot.*

The complexity of sharing emotions was also experienced by another family. The father described the concern that sharing his worries could elicit worries in the other family members.

> *What I'm also afraid of is, when I touch upon that fear or emotion, that I will elicit it in somebody else as well. When I tell you [the interviewer] that this fear is huge, then it doesn't induce fear in you. As an outsider, you can listen to it. If I would say it to my wife or the children or to Lien, then I kind of create, I guess, the same fear and emotion in them. (Father)*

This quote is illustrative of the ambivalence often reported by families facing pediatric cancer. More specifically, it may be considered necessary to talk about the cancer in order to cope with the experience and even helpful or preferable by parents and children. However, at the same time, when a family talks about the cancer, it is not only about receiving support from one another, but also about taking into account the emotions elicited by such conversations. Family members do not want to upset each other and, therefore, sometimes rather prefer not to share these experiences within the family, but talk with friends or clinicians. After all, as the diagnosed child is a shared loved one, all family members share the same fear of losing that child. This fear of death, the commonality of this fear and how difficult it was to share was expressed by the following father:

> *It's really hard to express this in the family. One kind of hides it a little bit. It's a really difficult period. Sometimes you protect yourself and the others by not naming or discussing the very anxious things, even though they are there. Everybody struggles with it. (Father)*

It seemed that talking about death and loss only occurred when the actual possibility of death became real. The families only talked about these intense emotions when, as a whole, they could no longer hide behind the hope and conviction the ill child would heal.

> *At the beginning Lien caught an infection of which we didn't know the cause. She had an almost forty degree fever and then it came really*

> *close. Then you do have to talk about it. It's only in moments like that, that you cannot get around it anymore. Those are really heavy emotions. (Father)*

Subtheme 2c: Keeping silent instead. Every family had its own way of coping with the cancer experience. For some, sharing might have been a relief (cf. subtheme 2a). However, others preferred not to talk about the cancer or the emotions evoked by such an intrusive situation.

> *Interviewer: Could you talk about Lien's illness to your mum and dad?*
>
> *Sibling (15 years): Uhm, actually I didn't do that a lot.*

Furthermore, in some families, differences in preferred coping style occurred between family members and these differences (i.e., talking vs. not talking) were sometimes hard to handle.

> *My husband and I were very different. Emotions were very hard for him. Very hard. For me that was less of a struggle. That [the difference] was not easy. (Mother)*

In addition, even in families who were used to sharing their daily stresses, the cancer situation sometimes forced them not to talk, as they were too tired or too emotional at that time.

> *One of our principles used to be: 'when there's something between the two of us, or something that's on our mind, whether it's legitimate or not, we won't go to sleep before we talk things through'. And yeah, that rule, we had to drop it now and then because we were just too tired, both physically and mentally. Yeah, then you feel like: 'we cannot do this right now, let's get back to it tomorrow'. (Father)*

The complexity of giving words to emotions was also described by other families. In this respect, it seemed that the more families struggled to find words, the more they found solace in the physical proximity of the other family members (see also subtheme 1a "Being together matters").

Theme 3: Working Together as a Team

The families in our study described working together as a team, to get everything organized. More specifically, due to the many hospital stays and the unpredictability of the illness, family members were forced to re-think their contribution to the family life and to make new arrangements. This was described by the father and the sibling of one family.

> *Before, I almost never went grocery shopping, then [post-diagnosis] I went a lot more. I did bit more of this, then I vacuum cleaned… But that's obvious, right, when she [his wife] was in the hospital. (Father)*
> *I didn't have much time off; we had to help more with things at home, cause otherwise you didn't manage alone. (Sibling, 11 year)*

This family described the self-evidence of helping more in the household, as the mother was mostly in the hospital accompanying the diagnosed child. Indeed, whereas pre-diagnosis, the family members seemed to have clear and distinct roles and expectations of each other, they started to work more together as a team from the diagnosis onwards.

> *Well, my wife was there, and I was here. Things had to move on. Then you vacuum cleaned sometimes, and you did this, and you went to the shop so there was food. Teamwork is what we call it. She put things in the dishwasher and the washing machine and I hung the clothes to dry and emptied the dishwasher. (Father)*

In addition, as parents indicated they were both responsible for the care of the diagnosed child and the financial situation of the family, parents divided all the work and made arrangements to accomplish both tasks.

> *Because women are often mums, they're better at caregiving. And because they're better at it, and we notice that they do it well, that's a reassurance. Not that I ever worried about it. That's a reassurance. That's why you leave it to your partner. The advantage is that I could empty a part of my hard disk. The fact that I could outsource this care*

completely to my wife, enabled me to function 'normally' in my job. (Father)

According to this father it was by working together that the family as a whole was able to get everything organized. Moreover, only by dividing the family tasks the individual family members were able to manage, and to keep their head up in these difficult times.

Discussion

When a child is treated for cancer, the lives of all family members change. In order to best cope with this stressful situation, family support has been put forward as an important resource (e.g., Zegaczewski et al., 2015). In the present study, we used a phenomenological-hermeneutic research method to interpret the narratives from the parents' and siblings' interviews in order to gain a better understanding of the specific ways in which family members support each other when facing pediatric cancer. This qualitative method was chosen with the aim of capturing the lived experiences of families regarding the processes of family support-giving in the context of pediatric oncology. Multi-family member interview analysis was used to integrate the perspectives of the different family members within one family, before moving on to the convergences across families, allowing agreement between the unit of interest (family level of support) and the unit of analysis.

Three themes emerged from the analysis. In the first main theme, *Being together matters*, parents and siblings indicated that they felt supported by each other's presence. This is in line with previous qualitative studies (e.g., Brody & Simmons, 2007; Sloper, 2000), a recent systematic review (Van Schoors et al., 2015) and meta-analysis (Van Schoors et al., 2017), illustrating that family support helps families to cope with cancer. In addition, this study contributes to the current body of evidence by emphasizing the importance of being *physically* together: by their presence, the participants in our study

showed availability and emotional support, as being together was sometimes all they could do to help the other.

The second main theme, *Finding support in talking and not talking*, illustrates the need to share cancer related experiences within the family, emphasizing the idea of 'social sharing of emotions': people who experience an intense emotion describe an imperious need to share this experience and to talk about it (Rimé, 2009). Moreover, parents and siblings in this study perceived talking about the illness as helpful and as a relief. This is in line with previous qualitative studies (e.g., Prchal & Landolt, 2012) and a meta-analysis showing that greater expressivity within the family is associated with better child adjustment (Van Schoors et al., 2017). However, talking about emotions was not always perceived as easy and sometimes not talking was preferred. This illustrates the complexity of family communication post-diagnosis. Indeed, although a family member may have felt the desire or need to talk about the cancer experience, they only shared their experiences when the other was perceived as emotionally strong enough and fully available. This study adds to the current body of research that, regarding the possibility of losing the diagnosed child, families sometimes perceived it easier to talk with strangers (i.e., friends, clinicians) than with family members, because sharing these intense emotions of threatening loss could elicit the same negative emotions in the others. In other words, due the fear to elicit painful thoughts and feelings in the others, family members sometimes decided to keep silent instead of sharing.

In the third main theme, *Working together as a team*, family members were forced to rethink their contributions to family life. They felt they needed to work together even better than pre-diagnosis; a finding that has also been found in previous qualitative studies (e.g., Prchal & Landolt, 2012; Van Schoors et al., 2018).

Taken together, we aimed to address two specific gaps in existing research. First, we focused on *how* family members supported each other after a pediatric cancer diagnosis. Second, using MFMIA, we were able to produce

family level interpretations from the individual interviews, integrating the perspectives of multiple family members and creating a multi-faceted understanding of family support in the context of pediatric cancer.

Methodological Considerations

Some limitations in the current study need to be addressed. First, the number of included families (N = 4) was small, which could elicit questions of transferability. However, like all qualitative studies, we did not aim to achieve generalizable findings, instead aiming to get a better understanding of specific family processes using a specific sample in a specific context. Second, the present study is part of a large-scale project. As a consequence, the number of included families was fixed and the principle of data saturation (i.e., reaching a point in the analysis that sampling more data will not lead to more information related to the research question) could not be applied. Third, our sample consisted of Belgian, Caucasian families. It is likely, however, that the experiences of families in other countries or with other nationalities differ. Fourth, we did not include the perspective of the ill child. This was due to the fact that the diagnosed child already participated to the longitudinal survey study, and we did not want to fatigue the ill child. Discrepancies in perceptions across family members (Alderfer et al., 2009; Stegenga et al., 2018), however, speak to the need to collect data from all involved individuals. For example, by including the diagnosed child, the reciprocity of family support could have been investigated in more detail and/or other ways of support giving could have been identified. As a consequence, to best capture family level constructs such as family support, the perspectives of all family members should be taken into account. Fifth, interpretations may be challenged by language differences: while the interviews were conducted in Dutch, the results were written in English. Sixth, in order to assure network confidentiality, the dissemination of our results had to be at a general level, rather than at a family level (Ummel & Achille, 2016). In other words, the strength of this analysis (being able to

offer an in-depth understanding of shared family experiences) at the same time encompasses the method's main shortcoming: we simply cannot exemplify all new insights at a family level when seriously considering our responsibility as researchers to protect (family) confidentiality.

To address these limitations, future research should further explore the idea of family support using the principles of data saturation and incorporate the perspectives of all family members (diagnosed child, mother, father, siblings), in heterogeneous families and heterogeneous cultures/countries.

Implications for Nursing Practice

The findings of this study may help clinicians to better understand how families adapt after a pediatric cancer diagnosis. Three specific recommendations can be put forward. First, when dealing with pediatric cancer, clinicians should offer to meet the family as a whole. Only by taking into account the perspectives of all family members, experiences at a family level could be understood. Second, as families indicated that they felt supported by each other's presence, efforts should be made to sustain this physical togetherness. For example, the parental desire to accompany the diagnosed child in the hospital around the clock (Van Schoors et al., 2018) illustrates the importance of hospitals providing (the possibility of) rooming in and flexible visiting hours for siblings. Third, clinicians working with families affected by pediatric cancer should be aware of the complexity of the topic when talking about the cancer experience: while some prefer to talk about the cancer, others find it hard to talk about the emotional impact. As families share their emotions by verbal as well as non-verbal strategies (e.g., physical proximity), the creativity of clinicians is challenged, and taking this individuality into account would therefore foster the family adaptation process. Fourth, not talking is important too as its exploration can give access to unvoiced concerns and worries of the family members (Rober, 2002).

Moreover, while both the families and the clinicians should respect the silence and even appreciate it as a way to care for each other, clinicians can provide a safe environment where families can try new ways of supporting each other.

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PART 3
THE IMPACT ON THE PARENTS' INTIMATE RELATIONSHIP

CHAPTER 9

COUPLE FUNCTIONING AFTER PEDIATRIC CANCER DIAGNOSIS: A SYSTEMATIC REVIEW[1]

A systematic review was conducted to (a) investigate couple functioning after a pediatric cancer diagnosis and (b) examine theoretical and methodological tendencies and issues in this literature. Searches of Web of Science, Pubmed, Cochrane, PsycInfo, and Embase resulted in inclusion of 32 qualitative, quantitative or mixed method papers. Findings of these papers were extracted for summary. Most couples adapt well to the crisis of a pediatric cancer diagnosis in domains such as emotional closeness, support, marital satisfaction, and general marital adjustment. However, most experience difficulties in the domain of sexual intimacy, and reports on conflict are mixed across qualitative and quantitative studies. This review illustrates the need for future research with a greater focus on the impact of a pediatric cancer diagnosis on the couple's functioning, conducted with use of appropriate theoretical frameworks and based on both partners' reports. Improvements in research are needed to best inform couple-based interventions.

[1]Van Schoors, M., Caes, L., Alderfer, M., Debeuf, T., Goubert, L., & Verhofstadt, L. (2016). Couple functioning after pediatric cancer diagnosis: A systematic review. *Psycho-Oncology, 26*, 608-616. doi: 10.1002/pon.4204

Introduction

Becauce of advances in treatment, approximately 76% of children diagnosed with cancer survive (Cancer Research UK, 2013), many with long-term and late effects. Thus, pediatric cancer is now considered a chronic illness. Like all chronic illnesses, pediatric cancer impacts not only the diagnosed child (Kestler & LoBiondo-Wood, 2012) but also the other family members (Alderfer et al., 2010; Kazak et al., 2004). Family members and the family system as a whole need to adapt to the unpredictable and uncontrollable course of cancer and its treatment (Alderfer & Kazak, 2006).

Research has been accumulating focused upon individual adaptation of patients (Stam et al., 2001), parents (Grootenhuis & Last, 1997; Patino-Fernandez et al., 2008), and siblings (Alfderfer et al., 2010; Houtzager et al., 1999). There have also been recent efforts at summarizing the literature on the adaptation of the family system as a whole after pediatric cancer diagnosis (Van Schoors et al., 2015), providing evidence that most families return to, sustain, or achieve adaptive levels of family functioning after this challenge. However, research into the adaptation of family subsystems is less common. Subsystems within the family are relational units marked by invisible interpersonal boundaries based upon specific characteristics (e.g., age or generation) or function. Subsystems within the family often have different relationship rules and patterns of interaction (Minuchin, 1974). For example, children within the family form one subsystem, parents form a subsystem in their role of providing and caring for the children within the family, whereas the adult couple form yet another subsystem. In particular, little is known about how the *couple subsystem* – more specifically the intimate relationship of the diagnosed child's parents – is affected by pediatric cancer. This apparent gap in the research literature is somewhat surprising given that three related but distinct areas within the family psychology literature (i.e., social ecology, stress and coping, intimate relationship science) point toward the likelihood that the couple subsystem will be impacted by childhood cancer.

First, the *Social Ecology Model* (Bronfenbrenner, 1977) postulates that an individual is embedded in a broad social context and that a stressor (like pediatric cancer) will influence the development and adaptation of the individual (i.e., the child with cancer) as well as the context in which this individual lives and the subsystems with which she or he interacts, including the couple subsystem. Second, *family stress and coping* models (e.g., *Double ABCX-model*, McCubbin & Patterson, 1983; *FAAR-model*, Patterson, 1988), specify pathways through which external stressors impact family systems and their subsystems, including the couple subsystem (see Bradbury & Karney, 2014). Third, most *theories on how intimate relationships succeed or fail* focus upon the powerful role that circumstances outside the relationship can play in shaping experiences within the relationship (Bradbury & Karney, 2014). Taken together, within the broader *family psychology* literature, the relationship between married or cohabiting partners has become one of the most frequently studied and measured components of the family system (Spanier & Lewis, 1980) and has been considered to be the actual core element of the family system (Schaer & Bodenmann, 2010).

Engagement in a wide range of coping and coping assistance strategies is reported by parents of a child with cancer (Hildenbrand et al., 2011). Consequently, it is plausible to assume that the stressors accompanying pediatric cancer and its treatment require both material and emotional resources (e.g., time and emotional availability) from the child's parents, which then cannot be invested in the maintenance of their intimate relationship. Moreover, the stressors may also give rise to conflict within the couple. Across research studies, findings reveal both reduced and enhanced levels of relationship quality after pediatric cancer (Greenberg & Meadows, 1992). However, to date, no attempts have been made to provide a systematic and critical integration of the available evidence. As such, a systematic review addressing the couple subsystem in the context of pediatric oncology would add substantial value to our understanding of how couples adapt to pediatric cancer diagnosis. The primary aim of this review was to investigate the impact

of a pediatric cancer diagnosis on couple functioning. A secondary aim was to examine theoretical and methodological patterns and issues in the literature and to formulate recommendations for future research and clinical practice.

Method

The current review is the third in a series of systematic reviews summarizing qualitative and quantitative evidence of family and couple functioning after a pediatric cancer diagnosis (Van Schoors et al., 2015; Van Schoors et al., 2017). While the previously published reviews focused on family-related and individual child functioning in the context of pediatric cancer, the focus of the current review is on relationship functioning within the couple subsystem. All reviews followed a strict scientific method, as outlined by Eiser et al. (2000) and the Cochrane Collaboration (Higgins & Green, 2011), to conduct a rigorous systematic search and provide a reliable and unbiased overview of the findings (see Van Schoors et al., 2015 for more details). A literature search was conducted in July 2014 and was updated in October 2015 to include the most recent published articles on this topic.

Literature Search and Inclusion Criteria

Web of Science, PubMed, PsycINFO, Cochrane, and Embase were searched using the following search terms: (cancer OR tumor OR malignancy OR oncolog*), (child* OR pediatric), (family OR parental OR marital OR marriage OR sexuality OR couple), and (psycholog* OR adaptation OR adjustment). Studies were retained if the article (a) examined the impact of pediatric cancer diagnosis (0 -18 years; any type of cancer) and treatment on any aspect of couple functioning, (b) was written in English, (c) presented new, empirical qualitative or quantitative data (i.e., reviews, case reports, commentaries, books, practice guidelines, conference abstracts, and

dissertations were excluded), and (d) did not *exclusively* focus on palliative care or bereavement, as these experiences are distinct from general adjustment to pediatric cancer and may have a different impact upon couple functioning. Studies focusing on both curative and palliative care were retained. No restrictions were placed on studies related to publication date.

Study Selection

The original search, in July 2014, identified a total of 5660 papers, which were independently screened by the first and second authors in three separate steps: (a) title screening (100% screened by the first and second author; 1592 articles retained with 89% agreement); (b) abstract screening (100% screened by the first and second author; 427 articles retained with 83% agreement); and (c) full-text screening (100% screened by the first author and 25% by the second author; 87% agreement). This 3-step selection process resulted in a total of 29 articles retained for the purpose of this review. Across our 3-step selection process, for 191 articles (although judged as not relevant on the basis of title or abstract) no full text was available. Disagreements between screeners were discussed and resolved; where necessary, a third reviewer was consulted. In addition, the reference lists of the retained articles were scanned to identify additional relevant articles, resulting in the addition of two papers. Lastly, to ensure up-to-date results, the search was repeated in October 2015 and 449 new potentially relevant papers were identified. The same 3-step screening process was followed for this updated search: 68 were retained on the basis of their title (87% agreement between the first and second author); 17 were retained on the basis of their abstract (75% agreement between first and second author); and 1 was retained after full-text screening. In sum, the current review is based on a final set of *32 articles* (see Figure 1).

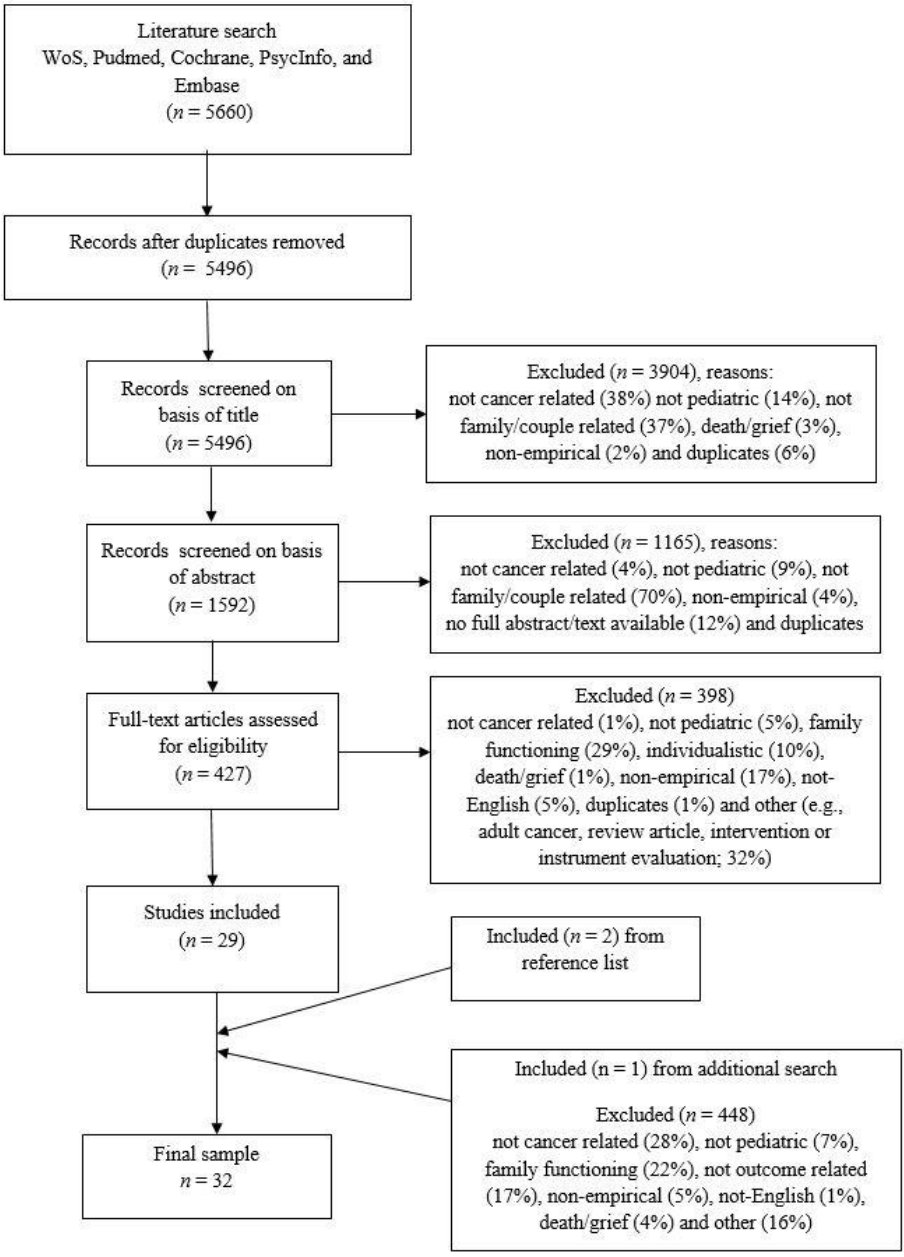

Figure 1. Flow chart

Data Extraction

The main purpose of this systematic review was to provide a narrative synthesis of the current state of knowledge on couple functioning after childhood cancer diagnosis. This was done by adopting a textual approach with evaluation of the scientific merit of the available evidence. A data abstraction sheet was developed to ensure systematic and standardized data extraction (available upon request). The data abstraction sheet identified the following study aspects: (a) study characteristics such as year of publication, journal, and database; b) which, if any, theoretical framework was used; (c) methodological and statistical aspects, such as design (e.g., cross-sectional versus longitudinal), sample size, unit of measurement, and assessment of interdependence (i.e., appropriately accounting for the interdependence of partner reports within couples); and (d) a summary of the general findings. The first author conducted the data extraction for all the included articles. To ensure accuracy, the second author conducted data extraction for 25% of the articles (i.e., full-text screening; 87% agreement).

A meta-analysis of the gathered data was deemed inappropriate as we preferred to include qualitative findings to ensure a comprehensive review. Furthermore, with regard to the quantitative studies, either too few studies were identified assessing a particular construct to warrant a meta-analysis (i.e., less than 2 studies identified for the same construct) or too much heterogeneity was observed in sample characteristics and outcomes.

In addition, each included study was rated by the second author with respect to its scientific merit using the criteria published in Alderfer and colleagues (2010). Quantitative studies were evaluated for explicit scientific purpose, appropriateness of design and analysis, measurement reliability, statistical power and approach, internal and external validity, appropriate discussion, and knowledge contribution. Qualitative studies were evaluated for explicit scientific purpose, appropriateness of design and analysis, grounding results in examples, integration of results into a framework,

specification of author's perspective, accurate and understandable topic coverage, application of credibility checks, and appropriateness and description of sample (Alderfer et al., 2010). Each aspect was rated on a 3-point scale ranging from 1 = "no or little evidence in fulfilling the criterion or low quality" to 3 = "good evidence or high quality". An overall score for scientific merit was obtained for each study by averaging the individual aspect scores. Reliability of the quality assessment was assured by double coding of 33% of the included studies by the first author. This revealed excellent interrater reliability as evidenced by a single measure and average measures intraclass correlation coefficients across the two raters of .92 and .96, respectively.

Results

Part I: Characteristics of Reviewed Studies

The methods and findings of the 32 reviewed studies are summarized in a Supplementary Table (i.e., Supplementary Table 7; at the end of this dissertation). About half of the reviewed studies were quantitative (n = 17, 53%). Thirteen studies (41%) used qualitative methods; two used mixed methods designs (6%). About two-thirds of the studies were cross-sectional (n = 21), and the rest were longitudinal (n = 11). Sample size ranged from 8 individual partners to 328 partners/164 couples. In 35% of the studies, only one partner participated (female partner: n = 4 studies; male partner: n = 2 studies; gender not specified: n = 8 studies). In the other studies, couples (n = 8 studies) or a combination of couples and individual partners (n = 10 studies) were included. Ten studies (31%) only included married couples, whereas 17 studies (53%) used a mix of married, cohabiting, or other couples (i.e., divorced parents, single parents, step-families, remarried parents, and widowed parents). In five studies (16%), marital status was not reported. A

wide variety of cancer diagnoses were included, with leukemia, lymphoma and brain tumors as the most frequently represented. Time since diagnose ranged from new diagnoses to 7 years post-diagnosis, but was not reported in two studies and vague in seven others ("long-term survivors" and "in treatment').

Part II: Narrative Summary of Reviewed Studies

Seven distinct dimensions of *couple functioning* emerged from the research literature: emotional closeness, marital conflict, marital support, communication, sexual intimacy, marital satisfaction and general marital adjustment. Within each of the following subsections, a brief explanation of the dimension is given, followed by the number and type of included studies identified and a narrative summary of the findings across studies. Qualitative results are presented before quantitative results.

Emotional Closeness. Emotional closeness refers to the feeling of positive connectedness between partners, varying from acquaintanceship to complete absorption of self and other into oneness (Davis, 2009). This dimension was investigated in five qualitative studies, one quantitative study, and one mixed method study.

Across four qualitative studies, all including samples of families both on and off treatment, participants often indicated that couple connectedness was strengthened by the illness (Beltrao et al., 2007; Brody & Simmons, 2007; Enskar et al., 1997; Khoury et al., 2013). In two studies involving on-treatment families, however, a range of responses was revealed. One of these studies reported that 60% of the participants (n = 23) indicated an increase in couple connectedness since diagnosis, 34% (n = 13) reported no change, and 5% (n = 2) reported a decrease in connectedness (Barbarin, et al., 1985[mix]). In the second study, 45% of the participants (n = 32) reported an increase, 17% (n = 12) reported no change, and 38% (n = 27) reported a decrease in emotional

closeness (Patistea et al., 2000). Variability in the experience of closeness has been proposed to reflect baseline differences across families (Patistea et al., 2000) or differences in the illness and treatment challenges faced by couples (Enskar et al., 1997). The one quantitative study of couple's closeness indicated that female partners reported a significant increase in emotional closeness with their partners from diagnosis to one year postdiagnosis (Tremolada et al., 2013).

In summary, most studies provided evidence for increased emotional closeness within the couple after diagnosis of pediatric cancer. However, this pattern clearly does not characterize all couples. More research into which couples draw closer and which do not is needed. Closeness before diagnosis and disease/treatment characteristics might play an important role.

Marital Conflict. Marital conflict refers to overt opposition or disagreement between partners that is identified as a source of difficulty in the relationship (Fincham, 2009). This dimension was addressed in five qualitative studies and three quantitative studies.

Across the qualitative studies, subsets of participants reported increased conflict during treatment (Patistea et al., 2000; Wills, 1999), after treatment (Greenberg & Meadows, 1992; Patterson et al., 2004), and across the illness trajectory (Khoury et al., 2013). However, an increase in conflict frequency was not reported by all couples in these studies: some reported no changes, whereas others indicated having fewer arguments, as they channeled all their energy into caring for the sick child and had no time to argue (Wills, 1999).

In included quantitative work, one study compared the yearly divorce rate of families with pediatric cancer with the rate in the general population and found no significant differences (Lansky et al., 1978). The second quantitative study indicated that only 8% of female participants (n = 2) and 5% of male participants (n = 1) experienced regular difficulties with their partner within three months after diagnosis. Nine months later, 21% of the

female participants (n = 4) and 7% of the male participants (n = 1) reported regular difficulties, a nonsignificant difference across time (Lahteenmaki et al., Salmi, 2004). In the third quantitative study, some couples reported an improvement in conflict resolution skills after their child's cancer diagnosis (Lavee & Mey-Dan, 2003).

In summary, reports of conflict were found in samples consisting of both on and off treatment families. However, the question remains whether this frequency of conflict transcends the frequency of conflict inherent to all couples. Sample characteristics (e.g., diagnosis, age of child, and country of origin) and aspects of study design (e.g., measure and sample size) did not seem to be associated with findings regarding conflict.

Marital Support. Marital support refers to assistance, encouragement, and caring provided by one partner and received or perceived by the other (Walsh, 1998). This dimension was addressed in nine qualitative studies, two quantitative studies, and one mixed method study.

Across the included qualitative studies, the partner was described as a highly important source of support (Barbarin et al., 1985 [mix]; Beltrao et al., 2007; Enskar et al., 1997; Greenberg & Meadows, 1992; McGrath, 2001; Patistea et al., 2000; Patterson et al., 2004; Wills, 1999, 2009) and sometimes even the most relied upon source of support (Mercer & Ritchie, 1997), both during and after treatment. Participants turned to their partners for all kinds of support, including emotional (e.g., listening to each other) and practical (e.g., maintaining the home) support (McGrath, 2001; Patistea et al., 2000). However, within these same studies, some individual participants reported that their partners were so impacted by the diagnosis or so focused on the ill child that they could not provide adequate partner support (McGrath, 2001; Patistea et al., 2000; Wills, 1999, 2009). For example, in one study, 14% of the participants (n = 9) described their partner as not at all supportive during treatment (Barbarin et al., 1985[mix]).

The first included quantitative study investigating partner support indicated that the spouse was one of the most frequently used sources of support and the most helpful source of support (Morrow et al., 1982). The second study indicated that perceived partner support was consistent across time from diagnosis until 1 year post-diagnosis (i.e., at diagnosis, and 6 months and 12 months later; Penn et al., 2009). These results are consistent with the qualitative studies.

In summary, across qualitative and quantitative studies, most partners reported that their partner's support was important, available to them, and helped them cope with the cancer experience. However, not every partner was equally able to provide such support, sometimes resulting in unmet support needs.

Communication. Communication is the interchange of thoughts, feelings, experiences, and information within the couple (Olson, 2000). This dimension was investigated in three quantitative studies.

The first quantitative study addressed couple agreement in reports of communication style at diagnosis and demonstrated that male and female reports did not differ in the amount of perceived avoidance (e.g., withdrawal, avoiding conflict, silence), incongruent (e.g., dishonesty or preference for talking to others), and destructive (e.g., insulting, irritated, or abusive) communication. However, female partners did experience less mutual understanding and sharing in their relationship than did male partners (Wijnberg-Williams et al., 2015). According to the second study, only presenting data of couples *with* interspousal agreement on communication patterns, 29% of the couples (n = 10) reported positive changes since the cancer diagnosis, 20% (n = 7) reported no changes, and none reported negative changes (Lavee & Mey-Dan, 2003). In a third study, mothers of children undergoing hematopoietic stem cell transplantation reported equal amounts of partner criticism and avoidance during the first six months after hematopoietic

stem cell transplantation, with no significant changes over time (Manne et al., 2003).

In summary, most available studies focused on comparison between partner reports on communication, revealing few differences. However, the evidence regarding changes in communication patterns between partners after a child's cancer diagnosis is too sparse to draw strong conclusions. There have been no comparisons between communication patterns among couples with children with cancer and couples with healthy children and no qualitative reports.

Sexual Intimacy. Sexual intimacy involves physical closeness between partners (Canary et al., 1977). This dimension was investigated in three qualitative studies and one quantitative study.

All three qualitative studies indicated that the pediatric cancer diagnosis had a negative impact on the sexual relationship of the participants. More specifically, participants indicated that intimacy had "gone out the window" (Enskar et al., 1997; Ferrell et al., 1994), as there was no privacy or time to spend together because of the constant attention and care needed by their child (Ferrell et al., 1994). Of note, one study found that worry – not lack of love – was the reason that couples reported being too drained for sex during their child's illness trajectory (Greenberg & Meadows, 1992). The one included quantitative study revealed that in nearly half of the couples both partners reported a deterioration in their sexual relationship. In this same study, only one couple reported improvements (Lavee & Mey-Dan, 2003).

In summary, across qualitative and quantitative studies, couples reported that the cancer diagnosis negatively affected their level of physical intimacy and sexuality. The magnitude of this effect and full understanding of the underlying reasons, however, remain elusive.

Marital satisfaction. Marital satisfaction refers to partners' global sentiment or happiness with their relationship (Lawrence et al., 2009). This

dimension was investigated in six quantitative studies and one mixed method study.

Across studies, a decrease in marital satisfaction was reported during the first year after diagnosis by both male (Hoekstra-Weebers et al., 1998a) and female partners (Hoekstra-Weebers et al., 1998a; Wijnberg-Williams et al., 2015), with the highest level of dissatisfaction during the first two months after diagnosis (Yeh, 2002) and significantly higher levels of satisfaction after treatment completion (i.e., 2 years after diagnosis; Brown et al., 1992). When compared with that of population-based control groups, no significant differences were found during (Lahteenmaki et al., 2004; Wijnberg-Williams et al., 2015) or after treatment (Hoekstra-Weebers, et al., 1998a; Wijnberg-Williams et al., 2015). At diagnosis, levels of marital satisfaction were lower than those of well-adjusted couples in the general population, but higher than couples referred for couples therapy (Fife, Norton, & Groom, 1987). At one year postdiagnosis, marital satisfaction was also higher for those couples with a child with cancer compared with couples referred for therapy (Hoekstra-Weebers et al., 1998a). Surprisingly, parents of children who had relapsed reported levels of marital satisfaction similar to those of parents of newly diagnosed children (Yeh, 2002), as well as those of parents of children who had survived (Wijnberg-Williams et al., 2015).

Some inconsistencies emerged across studies when examining gender differences in marital satisfaction. Two studies, both involving a mix of on- and off-treatment families, found no differences between male and female participants regarding their reported level of marital satisfaction (Hoekstra-Weebers et al., 1998a; Wijnberg-Williams et al., 2015). One study (Yeh, 2002) conducted in Taiwan, found that female partners were more dissatisfied than were male partners. A third study (Shapiro & Shumaker, 1987 [mix]) from the United States conducted repeated-measures analyses separately for males and females, then compared results, and found that male partners were more dissatisfied than female partners.

In summary, a U-shaped curve for marital satisfaction emerged over

time, with a decrease in marital satisfaction in the first year after diagnosis and a gradual increase across later stages of the illness trajectory. In addition, participants reported levels of marital satisfaction that were comparable with those of population-based control groups, lower than those of well-adjusted couples and higher than those of samples with recognized marital problems. Gender differences in marital satisfaction remained unclear, although culture may play a role.

General marital adjustment. General marital adjustment refers to a broad scope of outcomes, including a consideration of marital processes and marital outcomes (Lawrence et al., 2009). While general marital adjustment overlaps with all above-addressed outcomes, the studies summarized in this section assessed marital adjustment as a general construct without providing details on different specific dimensions of marital functioning. This general construct was addressed in three qualitative studies, seven quantitative studies, and one mixed method study.

Two qualitative studies investigated the impact of childhood cancer survival on general marital adjustment, revealing that only a minority reported the marital relationship was in jeopardy as a consequence of the diagnosis (Fletcher & Clarke, 2003). While equal reports of positive change (e.g., "This even made our marriage stronger", 23% of the participants (n = 21)) and marital difficulties (25% of participants (n = 24)) emerge, among those reporting difficulties, 46% (n = 11) remained married, 33% (n = 8) reported divorce as a direct result of the child's illness, and 21% (n = 5) reported divorced due to problems prior to the illness. The last qualitative study addressed similarities in perception between male and female partners and found that 79% of spouses (n = 23) agreed regarding the impact of cancer on the quality of their marital relationship (Patistea et al., 2000).

The quantitative studies indicated that only a minority of couples reported that pediatric cancer diagnosis had a major negative impact on their marital adjustment and hardly ever resulted in divorce (Lansky et al., 1978).

In one study, 72% of the participants (*n* = 46) reported that the marital relationship presented no problem during treatment, and only 5% (*n* = 2) indicated marital adjustment to be a major concern (Wills, 2009[mix]).

Similar reports of marital distress were observed across gender with 25% of female partners (*n* = 17) and 28% of male partners (*n* = 19) reporting clinically elevated levels of marital distress during the first weeks after diagnosis (Dahlquist et al., 1993). Twenty months later, these numbers were 19% (*n* = 8) and 24% (*n* = 10), respectively (Dahlquist et al., 1996). However, inconsistencies emerged regarding differences in reports of marital quality as a function of gender. While Barbarin and colleagues (1985), Cornman (1993) and Lavee (2005) did not find any differences between male and female partners' reports of (changes in) marital quality, Lavee and Mey-Dan (2003) found that men tended to perceive more positive changes than their female partners.

Inconsistencies also emerged regarding changes over time in marital quality. While one study reported an absence of significant change across time (Dahlquist et al., 1996), others reported either a curvilinear change (i.e., a deterioration during the first year followed by improvements across years two and three postdiagnosis; Lavee & Mey-Dan, 2003) or both positive or negative change across the illness trajectory (Lavee, 2005).

When marital adjustment during cancer treatment was compared with population-based norms or control groups, parents of children with cancer were found to be similar to parents of healthy children (Wittrock, Larson, & Sandgren, 1994), but less well-adjusted than the norms of married American couples (Cornman, 1993; Lansky et al., 1978). Parents of children with cancer, however, have been consistently found to be better adjusted than divorced couples (Cornman, 1993) or couples experiencing marital problems (Lansky et al., 1978).

In summary, research indicates that childhood cancer has the potential to negatively affect marital adjustment. However, for most couples, their marital adjustment, even in this time of stress, is within normal limits and

similar to that of controls. More research is needed to unravel the reasons for and impact of different reports across gender and time.

Part III: Evaluation of Reviewed Studies

On average, the scientific merit of the included studies was good (*M* = 2.31, range = 1.67-2.78) with five studies scoring below 2.0 on a 3-point scale (Alderfer et al., 2010). The most common weaknesses across studies were related to power (e.g., small sample size), failure to integrate the findings within a theoretical framework, internal validity (e.g., predominantly cross-sectional designs), and external validity (e.g., limited detail on saturation techniques and non-responders limiting generalizability). Areas of strength included well-justified objectives, selection of appropriate research methods, and providing example quotes.

Theoretical considerations. In the majority of the studies (*n* = 25, 78%), no theoretical framework was specified as guiding the research questions or selection of the variables. In the remaining studies *with* an underlying theoretical framework (*n* = 7, 22%), however, it should be noted that none of the models used (i.e., ABCX-model, family empowerment model, time-bound model, pediatric medical traumatic stress model, family stress theory, and family adjustment and adaptation response model) were specifically developed to understand how an external stressor like pediatric cancer diagnosis affects *couples* and their functioning. Instead, the models used in the reviewed studies were general stress and coping models, describing the general impact of a stressful situation on an individual and the entirety of the family context and subsystems with which she or he interacts, including the couple subsystem.

Measurement considerations. Even though the included studies focused upon marital constructs, only 18 studies (56%) assessed couple functioning from the perspective of both partners. Eight of those studies only included data from couples (i.e., reports of both partners), whereas 10 studies

included data from couples and individual partners. Studies including reports of both partners almost never analyzed the dyad (Cook & Kenny, 2005), but rather did separate analyses for male and female partners. Because the *unit of interest* should harmonize with the *unit of measurement* (Weber, 2011), one could argue whether the reviewed studies adequately assessed couple functioning. Indeed, discrepancies in perceptions across *family members* (e.g., Alderfer et al., 2009; Peterson et al., 2012) and *couple members* (Bradbury & Karney, 2010) speak to the need to collect data from both male and female partners, as well as to take the interplay of both partners' reports into account in statistical analyses.

Overall quality. As noted in our previous work (Van Schoors et al., 2015, 2017), certain characteristics of the existing research base make it particularly difficult to draw strong conclusions. For instance, heterogeneity across and within studies with regard to sample characteristics and different operationalizations of marital constructs poses barriers to conduct meaningful meta-analysis. The reviewed studies tended to have small heterogeneous samples and to rely upon cross-sectional designs, precluding identification of factors that may reliably predict which couples experience the greatest difficulties after being confronted with the challenges posed by pediatric cancer. Further limitations of the reviewed studies include the following: (a) most studies described only the experience of partners using qualitative quotes; (b) only seven studies (22%) used adequately demographically matched control groups or norms; and (c) only nine studies (28%) statistically assessed changes in couple functioning over time. Furthermore, all but two studies exclusively relied on participant's self-reports of couple functioning, despite the known drawbacks associated with this method (e.g., social desirability), especially when dyadic processes (e.g., communication, supportive exchanges, and conflict) are under investigation (Schwarz, Groves, & Schuman, 1998).

Discussion

The results of this systematic review generally indicate that most couples adapt well after being challenged by pediatric cancer in domains such as emotional closeness, marital support, marital satisfaction, and general marital adjustment. Thus, r*esilience,* defined as a return to, sustainment, or achievement of competent levels of functioning after being confronted with a stressor such as pediatric cancer (Hilliard et al.,, 2012), seems to characterize couples with children with cancer across most of the identified couple constructs. However, conflict and lack of sexual intimacy may occur for some of these couples too.

These conclusions, however, need to be considered in the context of the following precautions. First, data regarding the functioning of the couple prior to the illness, longitudinal data examining changes in couple functioning over time since diagnosis, and criteria for judging whether the functioning of the couple is "adaptive" were rarely available. Instead, we frequently relied on qualitative quotes of partners regarding perceived changes in the couple's functioning. While comparisons with healthy controls or norms were sometimes available, it is unknown whether adaptive (couple) functioning in the context of pediatric cancer is the same as typical (couple) functioning within families of healthy children (see Alderfer & Stanley, 2012; Van Schoors et al., 2015).

Second, strong conclusions are also hampered by the relative lack of studies using a theoretical framework. In addition, when theoretical frameworks were used, they were not specifically tailored to the couple coping with stress. As a consequence, there was a lack of clarity in the differences and similarities between the marital concepts used in the reviewed studies. For example, marital satisfaction, marital quality, and marital adjustment were used interchangeably in the reviewed studies, although theoretical frameworks and measurement guidelines for couple research encourage clear distinctions between the three constructs (Lawrence et al., 2009). More specifically,

marital satisfaction refers to *"global marital sentiment or happiness as a unitary construct"*; marital quality refers to *"marital processes, such as quality of a couple's conflict management skills, supportive transactions, sexual relations, or emotional intimacy"* and marital adjustment can be defined as *"a consideration of marital processes such as conflict management skills and marital outcomes such as marital satisfaction"* (Lawrence et al., 2009, p. 1028-1030).

Finally, most included studies utilized small, heterogeneous samples, and few studies are available within each dimension of couple functioning (ranging from 3 to 12 studies per dimension), thereby precluding robust conclusions. One possible reason for the limited amount of empirical work on the impact of a pediatric cancer diagnosis on *couple functioning* might be the lack of differentiation between partnership and parenthood. Indeed, in the context of pediatric cancer (in both research and clinical practice), partners are often addressed in their parenting role, which is also reflected in the large number of studies on parenting and pediatric cancer. However, the differentiation between partnership and parenthood *is* important, as both imply different responsibilities, roles, and behaviors (Berger & Bzostek, 2014; Karney & Bradbury, 2014). Moreover, men and women need to divide their energy and time between parenthood and partnership, as children (i.e., parenthood) make demands on couples that take time away from activities that promote and maintain their couple relationship (i.e., partnership), thereby pointing at the mutual influence between both (Bradbury & Karney, 2010; Kluwer, 2010).

Suggestions for Future Research

Future work, particularly studies adopting narrative techniques, should ideally rely on theoretical frameworks that incorporate partners' individual strengths and vulnerabilities (e.g., personality and family of origin experiences), external stressful events (e.g., low socio-economic status and

previous life events), as well as the underlying dyadic processes in couples (e.g., support provision and conflict management) in order to understand and predict variations in marital outcomes of couples facing pediatric cancer (e.g., vulnerability-stress-adaptation model; Karney & Bradbury, 1995). In addition, matching the unit of interest with the unit of measurement requires research involving both partners of the couple and taking into account their interdependence. This practice was lacking in most of the included studies but would allow for a more detailed analysis of couple functioning. One way to do this, as well as to go beyond the well-known disadvantages of global self-reports, is the use of observational methods or diary methods. Lastly, only about half of the included studies were quantitative. More quantitative research utilizing longitudinal designs with large, representative samples would benefit this field.

Clinical implications

On the basis of our review, we can conclude that problems within the couple subsystem only seem to occur for a subset of families and that most couples adapt well after a pediatric cancer diagnosis. As we cannot be sure, couples experiencing those problems likely comprise those with preexisting problems *as well as* those having difficulty specifically because of the stressor of childhood cancer.

Because these difficulties in the couple relationship may seem secondary to the more pressing need of ensuring adequate cancer and psychosocial care for the child, such issues may be overlooked by psychosocial providers in oncology or even seen as outside their purview of care. In addition, these problems may also be downplayed by the couple themselves (put on a back burner), as they as well primarily focus on their sick child and his/her treatment process. However, as these problems might negatively impact the adjustment of the child and his/her treatment, it is important to screen and remedy those problems, taking into account evidence-

based standards for psychosocial care in pediatric oncology (Wiener et al., 2015). For example, one can imagine that couples with different coping styles might experience elevated distress, anxiety, or depression (Manne et al., 2003; Hoekstra-Weebers et al., 1998b), which may be linked –in turn- with child distress and/or behavior problems. Studies assessing the direct influence of marital quality on psychosocial outcomes in children with cancer, however, are missing (Long & Marsland, 2011).

Interventions aimed at dealing with couple problems that get in the way of cancer care or hamper the adjustment of the child and family would ideally involve both members of the couple. However, efforts to provide such intervention formats may be difficult to achieve in practice (Stehl et al., 2009) and practitioners may need to rely on technology (e.g., telemedicine) to conduct conjoint sessions or work with members of the couple individually. In addition, the goals of such interventions may need to focus on finding ways for the couple to work together effectively to meet the needs of their child and family during cancer care, rather than making progress on long-standing difficulties within the couple. Once the couple is working together more effectively and capably managing the stress of cancer, then they should receive referral to community providers to address relational issues outside of cancer.

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CHAPTER 10

COUPLES DEALING WITH PEDIATRIC CANCER: A STUDY ON THE ROLE OF DYADIC COPING[1]

Pediatric cancer is a life-threatening disease that poses significant challenges to the ill child and his/her parents. Among the studies investigating risk and protective factors for the individual and relationship adjustment of parents being confronted with pediatric cancer, couple factors, such as dyadic coping, gained little research attention. Therefore, the aim of the current study was to explore the association between dyadic coping and individual/relationship outcomes of parents in the context of pediatric cancer. Participants were 59 couples of children diagnosed with leukemia or Non-Hodgkin lymphoma. Time since diagnosis varied from diagnosis to 20 months. Positive dyadic coping (i.e., supportive and common dyadic coping) and negative dyadic coping proved to be related to individual and relational outcomes of parents facing cancer in their child. In addition, while men and women reported to be equally satisfied with their partner and their sexual relationship, women reported higher levels of individual maladjustment. Our findings led to the conclusion that dyadic coping is important for both individual as well as relationship outcomes of parents when facing a diagnosis of cancer in their child. When meeting with families, both partners should be invited as a unit in order to best capture couple level experiences.

[1]Van Schoors, M., Loeys, T., Goubert, L., Berghmans, G., Ooms, B., Lemiere, J., Norga, K., & Verhofstadt, L. (2019). Couples dealing with pediatric blood cancer: A study on the role of dyadic coping. *Frontiers, 10*, 402. doi: 10.3389/fpsyg.2019.00402

Introduction

Pediatric cancer is an unpredictable and uncontrollable stressor that puts the diagnosed child at risk for adjustment difficulties (Alderfer & Kazak, 2006). There are a number of pediatric cancers, with blood cancer, including leukemia and lymphoma, as the most common type. Leukemia and lymphoma account for about 30 and 8% of all cancers in children, respectively (American Cancer Society, 2016). Due to advances in chemotherapy and stem cell transplantation, long-term survival of children with blood cancer can be achieved (Silverman & Weinstein, 1997). However, although many function well, some children with blood cancer (Rao et al., 1992; van der Does-van den Berg et al., 1995) or pediatric cancer in general (Kazak et al., 2001; Kestler & LoBiondo-Wood, 2012) experience social or emotional problems during or after treatment. In addition, the impact of a pediatric cancer diagnosis on the ill child's parents is undeniable. Every child is embedded in a broader social context, and therefore, a stressor (like pediatric cancer) influences not only the development and adaptation of that child, but also the context in which s/he lives and the subsystems with which s/he interacts (Social Ecology Model: Bronfenbrenner, 1977; Cipolletta et al., 2015). Indeed, in the context of pediatric cancer, there is abundant empirical evidence for the impact of the diagnosis and its treatment on the parents, both at the level of their individual functioning and couple functioning.

Concerning the impact of pediatric cancer on *parents' individual outcomes*, existing research revealed that a significant subset of parents report emotional distress, anxiety and acute or posttraumatic stress symptoms shortly after diagnosis (Grootenhuis & Last, 1997; Ljungman et al., 2014; Vrijmoet-Wiersma et al., 2008). Moreover, especially mothers seem to be impacted: they report more psychological distress than mothers of healthy children and fathers of children with cancer (Pai et al., 2007). In addition to the impact on parents' individual functioning, some studies have documented the impact of pediatric cancer on *parents' intimate relationship* (e.g., Hoekstra-Weebers et

al., 1998; Patistea et al., 2000). A recently conducted systematic review (Van Schoors et al., 2017) revealed that although most couples adjust well to the crisis of a pediatric cancer diagnosis in domains such as emotional closeness, couple support and marital satisfaction, most couples do experience difficulties in the domains of sexual intimacy and conflict, both on and off treatment.

It should be noted, however, that the research described above also revealed a considerable variability – both across and within studies – in individual outcomes as well as relationship outcomes for parents facing pediatric cancer. Given this great variability, a growing number of studies has tried to explain why some parents adjust better than others. Among these studies investigating risk and protective factors for individual and relationship functioning of parents being confronted with pediatric cancer, especially individual characteristics (e.g., catastrophic thoughts in parents; Caes et al., 2014) and family characteristics (e.g., family support; Fuemmeler et al., 2003) have been the topic of investigation. In contrast, so-called couple factors – characteristics of the intimate relationship of the child's parents– that may foster or inhibit parental individual and relationship outcomes gained less research attention. The current study aimed to address this gap by focusing on a couple-level variable that could be expected to moderate the impact of pediatric cancer on parents' individual and relationship outcomes, namely, the extent to which parents deal with the stressor of pediatric cancer as a dyad ("dyadic coping"; see Bodenmann, 1995). Dyadic coping has been identified in the couple research literature as well as the stress and coping literature as playing a cardinal role in individual and relationship functioning within couples facing severe stressors (e.g., Bodenmann 2005; Kayser et al., 1999)

"Dyadic coping" should be distinguished from other ways of coping with stress within intimate relationships, such as partners' individual coping (e.g., Garro, 2004; LaMontagne et al., 2003; Wong & Heriot, 2008) and their attempts at seeking social support from friends or relatives (e.g., Fife et al., 1987). In particular, in situations where there is the crossover of individual

stress from one partner to the other (e.g., work stress) or in cases of partners' shared stress from common sources (e.g., stress related to pediatric cancer), a joint appraisal of the stressful situation is required, which triggers dyadic coping, in addition to partners' individual coping. Within the dyadic coping literature, positive as well as more negative forms of coping as a dyad are described. Positive forms of dyadic coping include supportive dyadic coping (i.e., one partner assists the other in his/her coping efforts) and common dyadic coping (i.e., both partners participate in the coping process together). Negative forms of dyadic coping include hostile (i.e., support accompanied by distancing or sarcasm), ambivalent (i.e., support that is unwillingly) or superficial (i.e., support that is insincere) dyadic coping (Bodenmann, 1995; 1997; 2005).

Both theoretical and empirical arguments speak to the need of investigating (the role of) dyadic coping in the context of pediatric cancer. First, according to the Systemic Transactional Model (STM) of Stress and Coping in Couples, stressors always affect (directly or indirectly) both partners in an intimate relationship. This is true if the situation concerns primarily one partner - then his/her stress reactions and coping affects the other and turn into dyadic issues, representing the cross-over of stress and coping from one partner to the other (i.e., stressor of the self/partner) - and if the situation concerns both partners (i.e., shared stressors), both with regard to stress from daily hassles and more severe stressors (Bodenmann et al., 2016). So, stress *and* coping need to be understood as a systemic issue, a social process rooted in intimate relationships, with special attention to the interdependence and the mutual influence between romantic partners (Bodenmann et al., 2016). According to this theory, a pediatric cancer diagnosis needs to be considered as a shared and "dyadic stressor", as it is indeed a stressful event or encounter that concerns both partners, either directly or indirectly (Bodenmann, 1995; 1997). Both parents are *directly* involved in their child's illness, as shown by the finding that mostly one parent (temporarily) quits his/her job in order to accompany the diagnosed child day

and night (Van Schoors et al., 2018) or by the parents' individual emotional consequences described earlier (e.g., Pai et al., 2007). Also in line with this theory is that a dyadic stressor requires dyadic coping, conceptualized as the way couples cope with stress together in sharing appraisals of demands and planning together how to deal with the stressors. The importance of studying dyadic coping within the context of pediatric cancer can be derived from studies underscoring the positive role of coping-related activities, such as individual coping (e.g., Grootenhuis & Last, 1997) and social support (e.g., Fife et al., 1987) for the adjustment of parents and their ill child. Second, the importance of dyadic coping within the context of couples facing health and illness-related issues has been equally documented. For instance, the positive effect of dyadic coping on individual outcomes like *health* is largely documented (e.g., Berg & Upchurch, 2007; Meier et al., 2011), also in adult cancer studies (e.g., Badr et al., 2008; Kayser et al., 1999). Previous studies furthermore show robust and consistent associations between dyadic coping and relationship outcomes (Falconier et al., 2015). More specifically, a recent systematic review that focuses on couples coping with adult cancer illustrates that positive dyadic coping (i.e., supportive dyadic coping and common dyadic coping) improves relationship functioning, while negative dyadic coping impedes relationship functioning (Traa et al., 2015).

Taken together, based on theory (STM) and previous research on chronic illnesses in adulthood, we expect that dyadic coping may also be of importance in the context of pediatric cancer. More specifically, we expect that adequate dyadic coping (i.e., more supportive dyadic coping, more common dyadic coping, and less negative dyadic coping) is associated with better individual outcomes (i.e., less negative emotions: less stress, anxiety and depression, and lower levels of childhood illness-related parenting stress) and better relationship outcomes (i.e., higher marital and sexual adjustment) within parents being confronted with cancer in their child.

Method

Participants

The sample consisted of 59 heterosexual couples; all biological parents of children diagnosed with leukemia or non-Hodgkin lymphoma. They were all Caucasian and living in the Flemish part of Belgium. Mothers' mean age was 38.5 (Range 29–52); fathers' mean age was 40.5 (Range 30–56). Time since diagnosis varied from 0 to 20 months (M = 6.9, SD = 6.6). Forty-three women and thirty-seven men had a Bachelor or Master degree. In eight families, the diagnosed child was the only child. The remaining families had either two (28 families), three (20 families) or four (3 families) children. More details on the sample are listed in Table 1. Ethical approval from the University Hospitals of Ghent, Brussels, Antwerp, and Louvain had been secured for the study and the appropriate written informed consent forms were obtained for all participants.

Procedure

The present study is part of a larger study examining the impact of pediatric cancer on families, that is, the "UGhent Families and Childhood Cancer study". For this large-scale study, children diagnosed with leukemia or non-Hodgkin lymphoma between the age of zero and 18 years, their biological parents and any siblings were invited to take part in a survey study. Exclusion criteria were: (a) not speaking Dutch, (b) expression of a developmental disorder in the diagnosed child, and (c) relapse. Over a period of 3 years, 129 families participated; i.e., 65% of the eligible families. In 65 of these families, both parents filled out the questionnaires (50%), 59 of whom were married/co-habiting (91%) and 6 were divorced (9%). As this study focuses on the

Table 1

Background Characteristics of Couples of Children with Leukemia or non-Hodgkin lymphoma

	Demographic variable		Men | Women
Parents	*N* (couples)		59
	Age, mean (*SD*)		40.5 (6.7) | 38.5 (6.2)
	Education, *n*	Primary school	1| 0
		High School	21 | 16
		Bachelor/ Master	37 | 43
Ill child	*N*		59
	Sex, boys, *n*		36
	Age, mean (*SD*)		7.7 (5.1)
	Diagnosis, *n*	Acute lymphoblastic leukemia (ALL)	43
		Acute myeloid leukemia (AML)	3
		Non-Hodgkin Lymphoma	13
	Time since diagnosis in months (*SD*; Range)		6.9 (6.6; 0-20)

intimate relationship, the final sample only included the married or co-habiting couples (N = 59).

Measures

Dyadic Coping. A short version of the Dyadic Coping Inventory (DCI; Bodenmann, 2008) was used to measure several forms of dyadic coping. The questionnaire consists of 17 items, grouped into 6 subscales: (1) Supportive Dyadic Coping (e.g., "S/he makes me feel that s/he understands me and is committed to me"), (2) Common Dyadic Coping (e.g., "We try to tackle the problem together and work together"), (3) Negative Dyadic Coping (e.g., "S/he does not take my stress seriously"), (4) Own Stress Communication (e.g., "When I feel overwrought, I show my partner that I feel bad and that I need his/her emotional support"), (5) WE-Stress Appraisal and (6) Individual Stress-Appraisal. In this study, only the subscales supportive dyadic coping, common dyadic coping and negative dyadic coping were included given our focus on dyadic coping. Response options for each item ranged from 1 to 5 (*very rarely* to *almost always*). Scores for each subscale were obtained by summing the relevant items. The DCI has good reliability and validity (Ledermann et al., 2010). In the present study, Cronbach's alpha coefficients were .53/.83 (supportive dyadic coping), .67/.95 (common dyadic coping) and .75/.70 (negative dyadic coping) for men and women, respectively. The low Cronbach's alpha for the male supportive dyadic coping subscale could not be improved by dropping one or more items.

Depression, Anxiety, Stress. The Depression Anxiety Stress Scale (DASS-21; Lovibond & Lovibond, 1995) is a brief version of the 42-item DASS and consists of 21 items exploring negative emotions experienced over the last week. Participants rate the extent to which feelings of depression (e.g., "I felt that I had nothing to look forward to"), anxiety (e.g., "I experience trembling") and stress (e.g., "I found it hard to wind down") apply to them on a four-point scale from 0 (*never*) to 3 (*almost always*). Scores for depression,

anxiety and stress were obtained by summing the relevant seven items. The DASS-21 proved to be reliable in both clinical and community samples (Antony et al., 1998). In the present study, Cronbach's alpha coefficients were (for men and women, respectively) .88/.91 for depression, .77/.79 for anxiety and .85/.89 for stress.

Childhood Illness-Related Parenting Stress. The Pediatric Inventory for Parents (PIP; Streisand et al., 2001) measures childhood illness-related parenting stress. The questionnaire consists of 42 items grouped into four domain scales indicating the type of stressors parents are experiencing related to caring for their ill child: (1) medical care (e.g., "helping my child with medical procedures"), (2) communication (e.g., "speaking with child about his/her illness"), (3) role functioning (e.g., "being unable to go to work/job"), and (4) emotional functioning (e.g., "feeling numb inside"). Given the overlap between the DASS-21 and the emotional functioning subscale, the latter subscale was not included. In addition, both the frequency over the last week and the level of difficulty of each item is assessed on a five-point scale (frequency: 1 = *never* to 5 = *very often*; difficulty: 1 = *not at all* to 5 = *extremely*). Frequency and difficulty scores are summed for each of the three domain scales; these scale scores are then summed into an overall total frequency score (PIP-F) and total difficulty score (PIP-D) with higher scores indicating greater frequency and difficulty of illness-related stress. The PIP has good reliability and validity (Streisand et al., 2001). In the present study, Cronbach's alpha coefficients were .92/.92 for the total frequency score and .91/.90 total difficulty score, men and women, respectively.

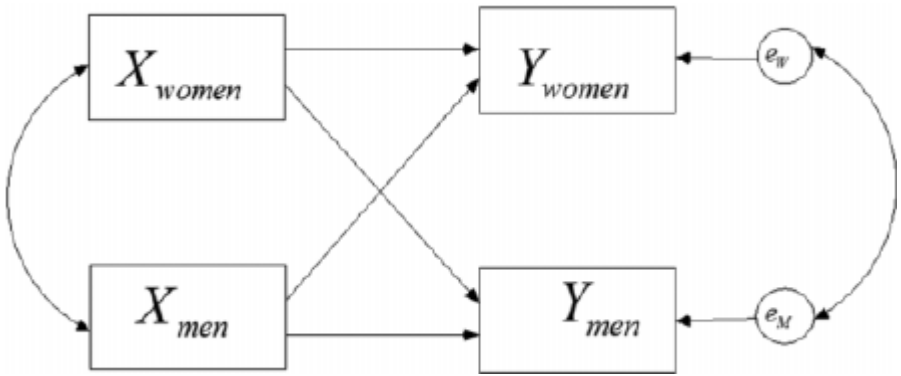
Marital Adjustment. The Maudsley Marital Questionnaire (MMQ; Arrindell et al., 1983) evaluates the marital relationship in general (e.g., "How much are you committed to this marriage?"), the sexual relationship (e.g., "Are you satisfied with the present frequency of sexual intercourse'?") and life in general (e.g., "Are you competent and successful at your job and your housework'?"). The questionnaire contains 20 items, each of which is rated on a 0 – 8 scale, with 0 representing the optimum response. A cutoff score >20

on the marital adjustment scale can be used to identify individuals who experience marital dissatisfaction (a level of marital dissatisfaction equal to the one reported by couples referred for marital counseling; Tuinman et al., 2005). In our study, 18 men and 19 women reported a score above 20 on the marital adjustment scale. When comparing the means on the MMQ marital adjustment scale of our study with a recent, Belgian, community sample (Hellemans, 2014), the current sample reported significantly higher levels of marital dissatisfaction (D = 3.88, t = 3.63, p < .001). The MMQ has good reliability and validity and the psychometric qualities of the Dutch version were also found to be satisfactory (Arrindell et al., 1983; Orathinkel et al., 2007). In the present study, only the two relationship subscales were taken into account, with a Cronbach's alpha of .91/.67 for marital adjustment and .84/.89 for sexual adjustment, men and women, respectively. For both subscales, a higher score indicates more maladjustment.

Data Analytic Strategy

We first describe means (with standard deviation and range) for all study variables and assess differences between men and women using a paired t-test. We further present correlations between study variables for men and women separately. The correlations for each study variable between men and women illustrate the non-independence within couples. To assess the association between the perception of supportive, common and negative dyadic coping (DCI) on the one hand and the frequency and difficulty of childhood illness-related parenting stress (PIP), depression, anxiety and stress (DASS), and marital and sexual adjustment (MMQ) on the other hand, we relied on the Actor-Partner Interdependence Model (APIM; Cook & Kenny, 2005). As shown in Figure 1, the APIM allows to simultaneously assess the effect of one's own perception of dyadic coping and one's partner perception of dyadic coping on one's own (actor) and one's partner outcome, while accounting for the correlation of outcomes within couples. The residuals of

Figure 1. The Actor-Partner Interdependence Model (APIM).



Note. X represents one' perception of dyadic coping (i.e., one of the dyadic coping subscales), while Y represents parenting stress (PIP), depression, anxiety and stress (DASS) or marital and sexual adjustment (MMQ subscales).

Note. For supportive and negative dyadic coping: an actor effect for women (men) can be interpreted as the effect of female (male) perception of her (his) partner's supportive/negative coping efforts on the female (male) adjustment; a partner effect of women in men (of men in women) can be interpreted as the effect of female (male) perception of her (his) partner's supportive/negative coping efforts on the partner's adjustment. For common dyadic coping: an actor effect for women (men) can be interpreted as the effect of female (male) perception of the couple's common coping efforts on the female (male) adjustment; a partner effect of women in men (of men in women) can be interpreted as the effect of female (male) perception of the couple's common coping efforts on the partner's adjustment.

men and women were allowed to be correlated and to have a different variance (i.e., an unstructured residual covariance). A separate APIM was fitted for each combination of dyadic coping subscales and outcome allowing for differential effects for male and female partners. Only if the overall tests for actor and partner effects [that is, testing the goodness-of-fit of models without actor (partner) effects] turned significant, actor and partner effects were inspected. Note that in all analyses, the time since diagnosis was included as a covariate, and was allowed to have a different effect on the male and female outcomes. All analyses were performed in the Structural Equation Modeling (SEM) framework (Stas et al., 2018) using the R-package lavaan. Unstandardized regression coefficients for actor and partner effects are presented with corresponding standard error and p-value. To assess gender differences in actor and partner effects, the difference between the male and female actor effect (partner effect, respectively) was calculated (hereafter referred to as the difference test). All tests were performed at the .05 significance level. Given the exploratory nature of this study, no correction for multiple testing was performed.

Results

Table 2 shows the descriptive statistics and correlations of the variables in our study. For common and negative dyadic coping, no significant gender differences were found. However, women reported experiencing more supportive behavior (supportive dyadic coping) from their partner than their male partner (D^{M-W}= -1.00, t(56) = -2.03, p = .047). Furthermore, higher levels of childhood illness-related stress (frequency D^{M-W}= -7.72, t(57) = -.2.82, p = .007; difficulty D^{M-W}= -9.04, t(57)= -4.45, p < .001), anxiety (D^{M-W}= -1.85, t(58) = -3.14, p < .001), depression (D^{M-W}= -1.61, t(58)= -2.11, p =.04) and stress (D^{M-W}= -2.98, t(58)= -4.59, p < .001) were found in women, as compared to men. Finally, regarding marital and sexual adjustment, no gender

Table 2 *Descriptive Statistics and Correlations of the Study Variables*

		Men										
		1	2	3	4	5	6	7	8	9	10	11
		12.98 (2.27), 8-18	**12.25 (2.26), 7-15**	**5.64 (2.30), 3-12**	**70.32 (17.89), 31-108**	**47.80 (14.49), 27-81**	**1.88 (2.47), 0-11**	**4.46 (4.19), 0-14**	**5.34 (3.65), 0-14**	**16.19 (11.90), 0-54**	**12.93 (8.93), 0-35**	**6.92 (6.57);0-20**
1	**13.98* (3.25), 7-20**	.06	.41**	-.40**	-.01	.05	-.05	.02	.20	-.29*	-.21	-.21
2	**12.22 (2.85), 4-15**	.75**	.44**	-.58**	-.14	-.35**	-.32*	-.32*	-.17	-60**	-.47**	-.27*
3	**5.93 (2.37), 3-11**	-.66**	-.63**	.26	.003	.19	.22	.29*	.15	.62**	.41**	.26*
4	**78.04** (21.07), 38-120**	.009	.06	.12	.38**	.61**	.095	.38**	.38**	-0.05	-.01	-.31*
5	**56.84*** (17.81), 29-108**	.060	.07	.04	.64**	.46**	.38**	.51**	.66**	.15	.23	.001
6	**3.73*** (3.80), 0-18**	.002	.01	-.03	.41**	.50**	.28*	.59**	.62**	.32*	.32*	.21
7	**6.07* (5.20), 0-21**	.09	.08	.12	.48**	.57**	.72**	.23	.75**	.39**	.32*	.06

Women

Questionnaire study

8	**8.32*** (4.70), 0-20**	.06	.02	-.002	.39**	.55**	.68**	.72**	.30*	.23	.38**	.03
9	**16.32 (11.42), 0-47**	-.57**	-.71**	.64**	-.09	.00	-.09	-.06	-.03	.71**	.74**	.36**
10	**12.05 (7.42), 0-32**	-.23	-.30*	.29*	.02	.07	-.08	.10	.10	.47**	.76**	.26*
11	**6.92 (6.57); 0-20**	-.11	-.34**	.192	-.58**	-.28*	-.25	-.27	-.23	.31*	.06	1

Note. Bold = Mean (SD), Range with gender differences = *p <.05, **p <.01, ***p <.001. Underlined = inter gender correlations (between men and women). *Note 2*: *Correlation is significant at the .05 level; ** Correlation is significant at the .001 level. *Note 3:* 1 = supportive dyadic coping (DCI): One's perceptions of their partner's supportive coping efforts; 2 = Common dyadic coping (DCI); 3 = Negative dyadic coping (DCI): One's perceptions of their partner's negative coping efforts; 4 = Parenting stress_Frequency (PIP-F); 5 = Parenting stress_Difficulty (PIP-D); 6 = Anxiety (DASS); 7 = Depression (DASS); 8 = Stress (DASS); 9 = Marital (Mal)adjustment (MMQ); 10 = Sexual (Mal)adjustment (MMQ), 11= Time since Diagnosis. *Note 3*: DCI, PIP & DASS: higher scores indicate higher levels of coping, parenting stress and negative emotions; MMQ: higher scores indicate higher marital/sexual maladjustment

Table 3

APIM analyses

	Overall Test	Difference Test
Actor effect CDC on PIP-D	X2(2)=8.181, p=.017	z=-2.577, p=.010
Partner effect CDC on PIP-D	X2(2)=9.223, p=.010	z=3.120, p=.002
Partner effect SDC on PIP-D	X2(2)=10.052, p=.007	z=2.608, p=.009
Actor effect NDC on depression	X2(2)=6.220, p=.045	z=0.358, p=.720
Partner effect SDC on depression	X2(2)=8.789, p=.012	z=2.221, p=.026
Partner effect SDC on anxiety	X2(2)=18.892, p<.001	z=3.011, p=.003
Partner effect SDC on stress	X2(2)=12.092, p=.002	z=2.799, p=.005
Actor effect SDC on marital adjustment	X2(2)=25.433, p<.001	z=1.010, p=.312
Actor effect CDC on marital adjustment	X2(2)=50.539, p<.001	z=0.524, p=.600
Actor effect NDC on marital adjustment	X2(2)=49.765, p<.001	z=-0.165, p=.869
Partner effect NDC on marital adjustment	X2(2)=20.538, p<.001	z=0.393, p=.694
Actor effect CDC on sexual adjustment	X2(2)=14.308, p=.001	z=1.406, p=.160
Actor effect NDC on sexual adjustment	X2(2)=12.569, p=.002	z=0.222, p=.824

Note. The Overall Test assesses whether the actor (partner, respectively) effects in both males and females are zero or not. The Difference Test assesses whether the actor (partner) effect is equal in men and women, or not. *Note 2: CDC* = Common dyadic coping (DCI); PIP-D = Parenting stress_Difficulty (PIP-D); SDC = Supportive dyadic coping (DCI); NDC = Negative dyadic coping (DCI); depression, anxiety and stress (DASS); marital and sexual adjustment (MMQ)

Table 4

APIM-results: An overview

		Men							Women						
		1	**2**	**3**	**4**	**5**	**6**	**7**	**1**	**2**	**3**	**4**	**5**	**6**	**7**
Men	**SDC**						X			X	X	X	X		
	CDC		X				X	X		X					
	NDC			X			X	X						X	
Women	**SDC**													X	
	CDC													X	
	NDC						X							X	X

Note. SDC = supportive dyadic coping (DCI): One's perceptions of their partner's supportive coping efforts; CDC = Common dyadic coping (DCI); NDC = Negative dyadic coping (DCI): One's perceptions of their partner's negative coping efforts; 1 = Parenting stress_Frequency (PIP-F); 2 = Parenting stress_Difficulty (PIP-D); 3 = Depression (DASS); 4 = Anxiety (DASS); 5 = Stress (DASS); 6 = Marital (Mal)adjustment (MMQ); 7 = Sexual (Mal)adjustment (MMQ). *NOTE 2*: X = Statistically significant effect

differences were found. Next, we discuss the results of the APIM-analyses (see Supplementary Table 8; at the end of this dissertation). We limit our discussion below to the gender-specific actor and partner effects for whom the global actor and partner test, respectively, were significant at .05 level (Table 3). Table 4 shows an overview of the significant APIM-results.

Dyadic Coping and Individual Outcomes

Childhood Illness-related Parenting Stress. More common dyadic coping reported by men was associated with lower difficulty scores of childhood illness-related parenting stress in men (actor effect; B = -2.58, SE = .89, p = .004). In addition, two partner effects were found: higher levels of supportive dyadic coping as perceived by men in their partner and more common dyadic coping reported by men were both associated with lower difficulty scores of parenting stress in women when facing illness in a child (B = -3.07, SE = 0.93, p= .001 and B = -3.14, SE = 1.09, p = .004, respectively).

Negative Emotions. When assessing the association between dyadic coping and negative emotions, one actor effect was found in men: Higher levels of negative dyadic coping perceived by men were associated with higher levels of depression in men (B = .50, SE = .24, p = .034). Furthermore, 3 partner effects were present in women. Higher levels of supporting dyadic coping as perceived by men in their partner were associated with lower levels of depression in women (B = -.82, SE = .27, p = .003), lower levels of anxiety in women (B = -0.78, SE = .19, p < .001) and lower levels of stress in women (B = -0.84, SE = .23, p < .001).

Dyadic Coping and Relationship outcomes

Both in men and women separately, actor effects of dyadic coping emerged when considering marital adjustment as outcome. In men, we found

that higher levels of supportive dyadic coping as perceived by men in their partner and more common dyadic coping reported by men (B = -2.23, SE = .60, p < .001; B = -1.18, SE = 0.60, p = .050, respectively) were associated with higher levels of marital adjustment reported by men. Negative dyadic coping as perceived by men in their partner was found to be associated with lower levels of marital adjustment reported by men (B=2.50, SE=0.48, p<.001). In women, we found that higher levels of supportive dyadic coping as perceived by women in their partner and more common dyadic coping reported by women (B = -2.66, SE = 0.43, p < .001) were associated with higher levels of marital adjustment reported by women (B = -1.91, SE = 0.37, p < .001). Negative dyadic coping as perceived by women in their partner was found to be associated with lower levels of marital adjustment reported by women (B = 2.62, SE = .47, p < .001).

One partner effect was found for coping reported by women on relationship adjustment reported by men: lower levels of negative dyadic coping as perceived by women in their partner (B = 1.75, SE = 0.47, p < .001 for, respectively) were associated with higher levels of relationship adjustment as reported by their partner. Furthermore, negative dyadic coping as perceived by men in their partner was associated with lower levels of relationship adjustment reported by women (B = 1.37, SE = 0.48, p = .005).

When considering sexual adjustment as an outcome, only actor effects were observed in men and women for some dyadic coping subscales. More specifically, higher levels of common dyadic coping reported by men was associated with higher levels of sexual adjustment reported by men (B= -1.60 , SE = 0.53, p = .003). Furthermore, for both men and women, higher levels of perceived negative dyadic coping in the partner were linked to lower levels of sexual adjustment (actor effects; B = 1.29, SE = 0.47, p= .006 for men and B = 0.83, SE = 0.41, p = .046 women, respectively).

Discussion

Using an Actor-Partner Interdependence Model (APIM; Cook & Kenny, 2005), the present study sought to examine whether dyadic coping was related to individual outcomes (negative emotions: anxiety, depression & stress and childhood illness-related parenting stress) and relationship outcomes (marital adjustment and sexual adjustment) in parents of children diagnosed with blood cancer.

Summary of results

Dyadic Coping and Individual Outcomes. Our findings indicate that both *positive* (i.e., supportive and common dyadic coping) and negative forms of dyadic coping matter for individual outcomes within parents being confronted with a cancer diagnosis in their child. This is in line with our prediction and with previous quantitative research on adult chronic illnesses (e.g., Meier et al., 2011; Regan et al., 2014). However, different patterns of findings emerged for supportive, common and negative dyadic coping.

More specifically, we found that the more men perceived their partner as *supportive*, the less depression, anxiety and stress (both general stress and difficulty scores on childhood illness-related stress) their partner experienced. In other words, the more men perceived their spouse as supportive, understanding and helping, the better the female *partner's* individual adjustment when facing pediatric cancer. These associations are in line with existing evidence that couple support is a protective and helpful factor in the individual adjustment to pediatric cancer (e.g., Morrow et al., 1982; Tarr & Pickler, 1999). However, we did not find the expected actor effects; i.e. associations between perceived supportive dyadic coping in one's partner and one's own individual adjustment. These findings seem to suggest that the benefits of support are mostly associated with support *giving* rather than support receiving, a finding that has also been reported by other researchers

in the context of health outcomes (Brown et al., 2003). Furthermore, more *common dyadic coping* reported by men was associated with lower difficulty scores on illness-related parenting stress for men and for women. So, the more men had the experience that both partners participated in the coping process symmetrically or complementary, the less they and their partner struggled with the care of their ill child.

Finally, the more men perceived their partner as negative, the more depressive complaints they experienced. This is in line with previous studies investigating the association between negative dyadic coping and negative emotions in adult chronically ill populations (e.g., Meier et al., 2011). Looking at the differential effects of the different types of dyadic coping, negative dyadic coping seems to be of less importance for the individual well-being of parents facing pediatric cancer. This finding is not in line with the literature on adult chronic illness describing negative forms of dyadic coping to be frequently occurring (Meier et al., 2011). This contradiction can be understood in two possible ways. First, it is possible that partner effects between negative dyadic coping and individual adjustment were not found in this study due to the relative small sample size (mimicking the observed associations, the power to detect such effects with $N = 59$ couples ranged from 5 to 64%). Second, in the context of adult chronic illness, there is one partner undergoing the illness, and one experiencing the illness from a certain distance. In the context of a child's cancer diagnosis, however, the child is ill, and therefore both parents may experience the illness in a more similar way. As a consequence, it is possible that couples, after facing a cancer diagnosis in their child, tend to understand each other better than in the context of adult chronic illness, and therefore, possibly engage less in negative dyadic coping.

Dyadic Coping and Relational Outcomes. For relationship outcomes within parents being confronted with a cancer diagnosis in their child, our findings indicated that both positive (i.e., supportive and common dyadic coping) and negative forms of dyadic coping matter. This is in line with our prediction, published quantitative studies (e.g., Falconier et al., 2015)

and a recent systematic review (Traa et al., 2015) in the context of adult chronic illnesses. More specifically, the present study shows that positive dyadic coping (i.e., supportive and common dyadic coping) was associated with higher marital adjustment, both in men and women (actor effects). In other words, the more a man perceives his partner as supportive and helping and the more he has the idea that both partners participate in the coping process symmetrically or complementary, the more he is satisfied with his marital relationship. The same pattern of findings was found for women. Furthermore, for negative dyadic coping, the more a man experiences distancing, mocking or sarcasm in his partner when talking about the illness, the less satisfied he is with his marital relationship. Again, this finding was replicated in women. Next to these so-called actor effects, the following partner effects were also found for marital adjustment. The more men and women perceived their partner as negative when talking about the cancer, the less satisfied their partner was in the marital relationship.

With regard to sexual adjustment, the more men experienced managing the cancer situation together, and the less negative their partner reacts, the more satisfied they were with their sexual relationship. Furthermore, the more women perceived their partner as negative, hostile or not interested, the less satisfied they were with their sexual relationship. These findings extend existing research by demonstrating that dyadic coping is not only related to marital adjustment and marital satisfaction (e.g., Falconier et al., 2015) but also to couples' sexual satisfaction.

Remarkably, for relationship adjustment, both actor effects and certain partner effects of dyadic coping were found to be important, whereas for sexual adjustment, only actor effects proved to be significant. So, how a parent describes the way in which s/he and his/her partner, as a couple, cope with the stressor together (i.e., supportive, negative or common) was at least partially related to their own and their partner's evaluation of the relationship (actor and partner effect) but only to their own evaluation of the sexual relationship (actor effect). The absence of partner effects in explaining sexual

adjustment may be linked to the fact that sexuality is, in se, an intimate domain and a difficult topic to discuss. As a consequence, the assessment of one's sexual relationship may be primarily linked to one's own appraisal of dyadic coping.

Gender. Gender differences as well as important gender similarities emerged from our data. Although at the relationship level, men and women reported to be equally satisfied with their partner and their sexual relationship, men and women did differ with regard to their individual adjustment. Across all individual outcomes, women reported higher levels of maladjustment (i.e., child's illness-related stress, anxiety, depression, stress) when facing a cancer diagnosis in their child than their male partner. This is in line with previous studies in the context of pediatric cancer, showing that especially mothers are impacted by the illness of the child (Pai et al., 2007). This finding may be explained by the increased burden assumed to be experienced by mothers in the care of children with cancer, as they are for example more likely to accompany the child to medical procedures (Kazak et al., 1996) and to stay in the hospital day and night (Van Schoors et al., 2018). In terms of dyadic coping, men and women only seemed to differ in the amount of supportive coping they perceived in their partner, with women reporting higher levels of supportive coping in their partner than men. These findings are not in line with the so-called marital support gap hypothesis, assuming that women are better support providers in their relationship than men are (see Verhofstadt et al., 2007 for a critical discussion). Comparing this finding to existing research on gender differences and similarities in dyadic coping is hard, however, as previous research focused on populations in which one of the partners was ill and therefore in a more support seeking/receiving position. Furthermore, important similarities between men and women in the association between dyadic coping and the relational outcomes under study emerged, more specifically the actor effects of dyadic coping on marital adjustment. Indeed, no significant differences were found in the actor/partner effects on relational outcomes between males and females (Table 3). This means that the pattern

of findings found in our male subsample was fully replicated within our female subsample and that for both parents of children with cancer, dyadic coping and relationship functioning are intertwined. However, the absence of evidence for a difference might also be due to the low power to detect such interactions in small samples (Gistelinck et al., 2018). For the individual outcomes, the patterns for men versus women were more heterogeneous, thus less parallels could be drawn between them. Indeed, several of the observed actor and partner effects on individual outcomes were significantly different between men and women (Table 3). Finally, gender effects also emerged in terms of effects of the predictor (i.e., the perception of dyadic coping). More specifically, men and women only differed in the *partner effects* of supportive dyadic coping on the individual outcomes (i.e., anxiety, depression and stress), and not in the actor effects. For common dyadic coping, however, gender differences were found in both actor and partner effects (Table 3). These tentative findings deserve further exploration in future research.

It is important to note that since no Type-I error correction was performed in this exploratory study, caution is warranted with regard to the interpretation of the above findings. All these findings should be reproduced in future studies.

Strengths and Limitations

A strength of this study is that it is the first to explore the association between dyadic coping and parental adjustment (individual and relationship outcomes), both within and between partners, after being confronted with a cancer diagnosis in their child. Furthermore, although most studies in the childhood cancer literature make use of a single-family member participant (e.g., Van Schoors et al., 2015), we included the perspectives of both partners. Discrepancies in perceptions across family members/partners (e.g., Alderfer et al., 2009) speak to the need to collect data from both members (e.g., Van Schoors et al., 2018). Additionally, by making use of the Actor-Partner

Interdependence Model (APIM; Cook & Kenny, 2005), we were able to model the interdependence in the dyadic relationship.

Despite the strengths of this study, some important limitations should be noted. First, we used a sample of Caucasian, heterosexual couples, thereby limiting the generalizability of our results. Future research should attempt to replicate these findings with more heterogeneous samples, e.g., also homosexual couples. Second, only Dutch speaking parents were included for participation. Therefore, with respect to the current multicultural society, this language criterion might have been a barrier for ethnic minorities. Third, we only focused on children with leukemia or non-Hodgkin lymphoma. As a consequence, it is important to highlight that parents of children with other cancer diagnoses may have different experiences. Fourth, time since diagnosis varied between the couples, ranging from zero to 20 months. The potential biases inherent in retrospective methods like the one used in the current paper may have influenced their responses (e.g., forgetting, defensiveness). In addition, future (longitudinal) studies should also take into account the possible impact of time since diagnosis, as it is plausible to assume that the effect of dyadic coping on outcomes has a different impact depending on how long the parents face the illness of their child. Now, we simply adjusted for the effect of time since diagnosis on the outcomes, but future studies may look at the interaction of time since diagnosis and the actor and partner effects of dyadic coping. Fifth, as the associations described in this study are correlational in nature, the temporal order of the variables under investigation could not be tested with the present data. It is also possible, for instance, that better parental adjustment elicits more adaptive dyadic coping strategies, as described above.

Clinical Implications

Difficulties in the couple relationship may seem secondary to the more pressing need of ensuring adequate cancer and psychosocial care for the child.

Therefore, such issues may be overlooked by psychosocial care providers in oncology or may even be downplayed by the couples themselves. However, this study shows that dyadic coping matters for individual and relational functioning in parents when facing cancer in their child. As a consequence, it is important to screen and tackle relational issues besides individual issues, taking into account evidence-based standards for psychosocial care in pediatric oncology. Interventions aimed at dealing with couple problems that get in the way of cancer care or hamper the adjustment of the child and/or family should take into account three specific recommendations. First, in working with families being confronted with a cancer diagnosis in a child, clinicians should not only focus on the adjustment of the child diagnosed with cancer or educational issues that arise post-diagnosis, but also on the impact of the illness on the parents in general and the parents' intimate relationship in particular. Moreover, clinicians should invite the couple system as a whole. Only by taking into account the perspectives of both members, couple level variables – such as dyadic coping – can be fully understood and improved when needed. Second, as previous research demonstrated that sexual relationships appear to be affected most negatively when facing a cancer diagnosis in their child (Lavee & May-Dan, 2003), clinicians should overcome their potential reluctance to discuss such topics together with the couple. Third, clinical interventions should be tailored to gender differences and specific characteristics of men and women facing pediatric cancer. For example, our findings suggest that women might be more vulnerable than men (cf. women reporting higher levels of individual maladjustment compared to men) when facing cancer in their child, and might therefore be in greater need of professional support from psycho-social workers or clinicians.

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GENERAL DISCUSSION

This doctoral dissertation aimed to gain insight into the impact of childhood cancer on the ill child, his/her family members, the family system as a whole and the parents' intimate relationship. In this final section, the main findings of the preceding empirical chapters will be recapitulated and, where possible, integrated. Then, a general answer regarding the three research aims will be provided. Finally, we will conclude with theoretical implications, limitations, recommendations for future research, and clinical implications.

Overview of the Main Findings

Globally, 300,000 children are diagnosed with cancer every year (American Childhood Cancer Organization, nd.), 341 of whom are in Belgium (Belgian Cancer Registry, 2020). Given the increased survival rate of children with cancer, more research has started to focus on the psychosocial consequences of living through cancer. To date, however, how childhood cancer affects the *family as a whole*, and its subsystems, i.e., the *siblings* and the couple (i.e., *parents' intimate relationship)* has received inadequate research attention. Furthermore, there is limited consideration of the *long-term effects* of childhood cancer on individual and family well-being, as well as limited use of *theoretical frameworks* in guiding research questions and the selection of variables.

The *research aims* of the present dissertation were to examine: (a) the short-term and long-term effects of childhood cancer on families, its members (patient, parents and siblings), and the parents' intimate relationship; and (b) the resources – situated at the individual level, intrafamilial level and contextual level – that may help families and their members to recover from this crisis and to adapt to the illness. In addition, given the lack of research on

and knowledge about the adaptation of siblings, specific attention was given to (c) the siblings' experiences regarding cancer and its consequences for their everyday family life.

To address these three aims, the present dissertation was divided into three parts: The impact on the family as a whole, The impact on individual family members and the family as a resource, and The impact on the parents' intimate relationship. Across all parts of this dissertation, the *double ABCX model* (see Fig. 1; McCubbin & Patterson, 1983) served as the underlying theoretical framework. For example, in *Part 1: The impact on the family as a whole*, we focused on family adaptation (Chapters 1, 3, 4) and how it is associated with individual, intrafamilial and contextual resources, both cross-sectionally and prospectively (Chapter 2). In *Part 2: The impact on individual family members and the family as a resource*, we focused on the association between the individual adaptation of patients, parents and siblings on the one hand, and intrafamilial resources (Chapters 5, 6, 7), and the family's perception of the disease on the other hand (Chapter 6). In *Part 3: The impact on the parents' intimate relationship*, we focused on couple adaptation (Chapter 9) and its association with intrafamilial resources (Chapter 10). In the next section, every chapter will be summarized, and conclusions across chapters will be described.

Part 1: The Impact on the Family as a Whole

In **Chapter 1**, our systematic review on family resilience after a pediatric cancer diagnosis was described. Resilient families are families that return to, sustain, or achieve competent levels of family functioning in one or more domains (i.e., cohesion, adaptation, communication) after being challenged by childhood cancer.

Following the guidelines for systematic reviews, searches were performed using Web of Science, Pubmed, Cochrane, PsycInfo and Embase. After screening 5563 articles, 85 articles fulfilled inclusion criteria and were

extracted for review. Forty-three of these articles were quantitative studies, 35 were qualitative studies and 7 were mixed methods. Most studies collected cross-sectional data (n = 67), and sample size varied from 3 to 209 families (6 to 465 individuals). Findings indicated that most families are resilient, adapting well to the crisis of a pediatric cancer diagnosis. More specifically, although qualitative studies revealed that the impact of childhood cancer on the family is overwhelming and severe, quantitative studies showed levels of family functioning across samples that were comparable to control groups or norms for the domains of cohesion, conflict, adaptability, expressiveness, support and general family functioning; both on- and off-treatment. Some exceptions, however, were also found. For example, siblings may experience being at the periphery of the family (decreased family cohesion), mothers of brain tumor patients reported lower levels of perceived family support compared to controls, and 60% of the siblings in a Chinese study claimed not to have a chance to talk about the illness with their parents or ill brother/sister.

Chapter 2 described a longitudinal questionnaire study. More specifically, guided by our theoretical framework the double ABCX model (McCubbin & Patterson, 1983), this chapter focused on family adaptation as an outcome and on parental psychological flexibility (individual resource), dyadic coping (intrafamilial resource), and network support (contextual resources) as resources contributing to family adaptation. Family adaptation was operationalized as the economic consequences for the family (financial impact), disruption in the family's normal social interactions (social impact), disequilibrium experienced by the parents relating to the psychological burden of the illness (e.g., difficulty of planning for the future; general family impact), and parents' satisfaction with the family's current way of life. Psychological flexibility refers to the willingness of an individual to experience unwanted or aversive stressors while pursuing one's values and goals, instead of avoiding unwanted or aversive stressors, thoughts and feelings (Hayes et al., 1999). Dyadic coping refers to the extent to which partners deal with a stressor, like pediatric cancer, as a dyad (Bodenmann, 1995). We expected that higher

levels of psychological flexibility in mothers and fathers of children diagnosed with cancer, more adequate dyadic coping in their couple relationship and more (amount and satisfaction with) network support would be associated with better family adaptation, both cross-sectionally and prospectively.

These predictions were tested in a sample of 70 mothers and 53 fathers (80 families) who provided questionnaire data at two measurement points (mean time since diagnosis T1 = 5.26; T2 = 18.86 months). Results were in line with our predictions and previous studies (Burke et al., 2014; Hoekstra-Weebers et al., 2001; Meier et al., 2011): psychological flexibility, dyadic coping, and network support proved to be cross-sectionally and positively related to parents' perception of family adaptation post-diagnosis; psychological flexibility and dyadic coping proved to predict better family adaptation over time. Specifically, higher levels of psychological flexibility were related to a lower impact on the family in general and a higher level of satisfaction with the current family's way of life. More adequate dyadic coping (i.e., more stress communication) predicted a lower financial impact. More perceived network support, and more satisfaction with this perceived support, were related to a lower impact on the family in general. Regarding the prospective results, higher levels of psychological flexibility predicted a lower impact on the family in general, and a lower financial impact.

In sum, this chapter provides empirical support for the double ABCX model. More specifically, for the association between resources ('b') at the individual (psychological flexibility), intrafamilial (stress communication) and contextual (network support) level, and family adaptation, both short term ('x') and long term ('xX') (see Fig. 1).

In **Chapter 3,** a qualitative study investigating parents' perspectives of changes in family functioning after a pediatric cancer diagnosis was described. Specifically, the aim of this chapter was to provide insight into parents' personal accounts of their experiences, and to obtain in-depth descriptions of changes in their family functioning when facing childhood cancer.

One-on-one interviews were conducted with ten mothers and ten fathers (10 couples) separately. This allowed each parent to provide their own perspective on changes in their family life post-diagnosis (Eisikovits & Koren, 2010), without having to consider their partner's feelings (Morris, 2001). Verbatim transcripts of these interviews served as the raw data for this study. Multi Family Member Interview Analysis (MFMIA; Van Parys et al., 2017) was then used as the methodological framework to analyze the individual interviews, focusing on the couple as the unit of analysis. This analysis led to the emergence of three themes, in line with existing literature (Norberg & Steneby, 2009; Prchal & Landolt, 2012): (a) Family cohesion: strengthened versus fragmented; (b) Educational norms and values: overindulgence versus being stricter, and (c) Normality: loss versus preservation. The conflicting dynamics present in these emerging themes exemplify the complexity of this process of family adaptation.

Specifically, in the first theme, parents perceived their family as a stronger unit. However, at the same time, fragmentations in the family unit were also reported, including a shift in focus toward the ill child, at the cost of attention to the family as a whole, for the siblings, and for the parents' intimate relationship. In the second theme, parents identified the need for a new parenting approach during treatment, one that compensated for the suffering of the diagnosed child by overindulgence. Post-treatment, however, parents started to question this overindulgence and seemed to adopt an even stricter parenting approach than pre-diagnosis to compensate for the overindulgence. The third theme illustrates the overwhelming impact of the cancer diagnosis on the family, resulting in the feeling that "nothing is normal anymore." While, at the same time, families strive for normality and try to safeguard the normal life of all family members, including siblings.

Chapter 4 described a qualitative study regarding siblings' experiences of their everyday life in a family where one child is diagnosed with cancer. The central research question of this chapter was "How do

siblings of children with cancer describe their everyday family life post-diagnosis?"

Interviews with 10 siblings (aged 10-16 years) were conducted. The verbatim transcripts of the interviews served as the raw data for an interpretative phenomenological analysis (IPA; Smith & Osborn, 2015). IPA has been applied successfully in the context of health psychology (Smith, 2011) and, specifically, cancer experiences (Reynolds & Lim, 2007); and has been used successfully to understand the lived experiences of children (Kvale & Brinkmann, 2009). The analysis led to the emergence of two themes: (a) Continuity within family life and (b) Beyond the familiar: facing illness-related challenges. Specifically, in comparison to pre-diagnosis, the siblings in our study experienced continuity in many aspects of their family life (Theme 1): They still experienced their family as an important source of support and information/communication, as warm and loving, and as a safe harbor where family members aimed to protect each other. However, at the same time, the siblings also reported challenges due to the cancer diagnosis, expressing that some things felt unmistakably different (Theme 2): Many felt that the family as a whole had been ripped apart post-diagnosis, with a greater focus on the diagnosed child and changing responsibilities for each family member.

Conclusion Part 1: These four studies (one systematic review, one quantitative study and two qualitative studies) all focused on the impact of childhood cancer on the family system as a whole (*family adaptation*). They contribute to the existing childhood cancer research field in three ways. First, the majority of childhood cancer studies do not focus on family-level outcomes (Van Schoors et al., 2015), instead, focusing mainly on consequences of the illness for individuals within the family (i.e., patients, parents and siblings). However, given the conceptualization that a family is more than the sum of its parts (Von Bertalanffy, 1973), and that a cancer diagnosis not only affects the individuals within the family, but also their

relationships with one another and the way in which the family functions (Alderfer & Kazak, 2006), a family impact is undeniable.

Second, within the pediatric cancer literature, most studies include one single respondent (Van Schoors et al., 2015), rather than considering the perspectives of multiple family members. As a consequence, the interdependence between family members and their relationships are mostly neglected. In Chapter 1, this issue was examined in the existing childhood cancer literature. The review showed that although the unit of interest was the family ("family resilience"), the unit of measurement was in most (53%) studies one single family member. In only 6% of the studies, all family members were included. In Chapter 2 and 3, this issue was addressed by including the perspectives of both mothers and fathers, and by making use of appropriate statistical techniques to address interdependencies within the data (i.e., multilevel modeling approach; MFMIA). Similarly, in Chapter 4, all siblings in the family aged between 10-16 years were interviewed.

Third, there are important parallels between the results of Chapter 1 (our review), Chapter 3 (qualitative study with parents) and Chapter 4 (qualitative study with siblings). These parallels increase the credibility of our empirical results. For example, both the review and the interview studies reveal the loss of normal family life during cancer treatment and the struggle post-treatment to return to normality. Moreover, the parents (Chapter 3) and the siblings (Chapter 4) in our qualitative studies indicated that the loss of normal family life was particularly true for the siblings; these siblings suddenly had to handle their problems with less parental help/support, and were expected to show increased responsibility and maturity. For cohesion, both the review and the qualitative studies found that family cohesion was strengthened by the illness: the illness brought the family closer together. In addition, the review revealed that increased closeness was not always perceived as being inclusive of the siblings (Chesler et al., 1991), a finding that was also emphasized by the parents and siblings in our interviews. Finally, in regard to family support, our qualitative findings are in line with past

literature summarized in the review. The family is an important source of support when facing childhood cancer, as family members – more than others – understand and share the burden that cancer brings.

Part 2: The Impact on Individual Family Members and The Family as a Resource

In **Chapter 5**, a meta-analysis on the associations between family functioning and child adaptation (patient and sibling) after a pediatric cancer diagnosis was described. Literature searches in Web of Science, Pubmed, PsycInfo, Cochrane and Embase were undertaken. After screening 5563 articles, 35 were identified regarding this topic; 30 contributed data for meta-analyses. Of the 35 studies, most were cross-sectional (77%), and sample size varied from 30 to 389 families (30 to 778 individuals). The statistical information extracted from each study was entered into Comprehensive Meta-Analysis (CMA) 3.0 statistical software (Borenstein et al., 2015) for analysis; Pearson's *r* correlations were the effect of interest. We found a significant association between family functioning and child adaptation (patient and siblings) after a pediatric cancer diagnosis. More specifically, greater family cohesion, expressiveness, and support and less family conflict were each associated with better child adaptation outcomes. For most domains, however, there was significant heterogeneity in the effect across the included studies. In sum, this chapter supports the double ABCX model, in particular the association between intrafamilial resources ('b'; family functioning) and adaptation of patients and siblings ('x', see Fig. 1).

Chapter 6 described a cross-sectional questionnaire study. More specifically, guided by our theoretical framework – the double ABCX model – this chapter focused on the association between family functioning (intrafamilial resource), cancer appraisal (perception), and the individual adaptation of all family members (patients, parents, siblings; outcome).

Individual adaptation was operationalized as perceived quality of life and four cancer-related emotional reactions (loneliness, uncertainty, helplessness & positive feelings). Family functioning was operationalized as the affective nature of family relationships (emotional closeness) and the extent to which the family was structured and open to change (family structure). Cancer appraisal referred to the extent to which cancer was perceived as uncontrollable versus manageable. We expected that better family functioning, perceiving the illness to be more manageable and less uncontrollable, and the interaction between these two variables, would be associated with better individual outcomes in patients, parents, and siblings.

These predictions were tested in a sample of 115 families (60 patients, 172 parents, 78 siblings). Mean time since diagnosis was 6.9 months. Results were in line with our predictions and previous studies (Hamama et al., 2000; Van Schoors et al., 2017): family functioning and the appraisal of the cancer diagnosis (but not the interaction) proved to be related to patients', parents' and siblings' quality of life and cancer-related emotions post-diagnosis. More specifically, more emotional closeness within the family (i.e., more cohesion and expressiveness, less conflict) was related to better quality of life in parents and less loneliness in all family members. Perceiving the illness as more manageable and less uncontrollable was related to less loneliness, less uncertainty, less helplessness and better quality of life in all family members. To conclude, this chapter partially supports the double ABCX model: the main effects of the resources ('b', family functioning) and the perception ('c', the appraisal of the cancer diagnosis) were found to be important, however, they did not interact to influence adjustment.

In **Chapter 7**, a cross-sectional questionnaire study was described. More specifically, guided by our theoretical framework – the double ABCX model – this chapter focused on the association between family functioning (intrafamilial resource), family support (intrafamilial resource), network support (contextual resource) and the individual adaptation of siblings (outcome). In line with Chapter 6, individual adaptation was operationalized

as perceived quality of life and the four cancer-related emotional reactions listed above; family functioning was operationalized as the affective nature of the family relationships (emotional closeness) and the extent to which the family was structured and open to change (family structure). Family support and network support were operationalized as sibling-perceived social support from the family and the external network. We expected that better family functioning, more family support, and more network support would be associated with better individual outcomes in siblings.

These predictions were tested in a sample of 81 siblings (mean age 10.32 years). Results were partially in line with our predictions and previous studies (Brown et al., 2003; Dolgin et al., 1997; Van Schoors et al., 2017): family functioning, family support, and network support were related to siblings' cancer-related emotional reactions post-diagnosis but no significant associations were found with quality of life. Specifically, more emotional closeness (i.e., more cohesion and expressiveness, less conflict) within the family was related to less loneliness, more emotional involvement in the illness, and more positive cancer-related feelings. More structure within the family (more clear family organization and more parental control) was related to more emotional involvement in the illness by the siblings and more positive cancer-related feelings. More perceived social support from the family and the external network was associated with feeling less lonely, having more emotional involvement in the illness process, and having more positive cancer-related emotions. In sum, this chapter supports the double ABCX model, in particular the association between resources ('b') at the intrafamilial (family functioning, family support) and contextual (network support) level, and the individual sibling adaptation ('x') (see Fig. 1).

Chapter 8 described a qualitative study investigating how family members support each other when facing pediatric cancer. One-to-one interviews were conducted with four families of children with blood cancer (four mothers, three fathers, five siblings). Verbatim transcripts of the interviews served as raw data for this study. In line with Chapter 3, Multi

Family Member Interview Analysis (MFMIA; Van Parys et al., 2017) was used as a methodological framework to analyze the individual interviews, with a focus on the *family* as the unit of analysis (note: in Chapter 3, the *couple* was the unit of analysis). The analysis led to the emergence of three themes describing ways that family members support one another. First (Theme 1), the families identified the need of being physically together, both as a family and with the child with cancer. Second (Theme 2), the families reported being eager to talk about the illness and its impact because talking was experienced as a relief. However, talking about emotions was sometimes experienced as hard and some family members preferred not talking about difficult emotions. Third (theme 3), the families described working together as a team in order to get everything organized.

Conclusion Part 2. These four studies (one meta-analysis, two quantitative studies and one qualitative study) all focused on the impact of childhood cancer on the individual family members (*individual adaptation)*. They contribute to the existing childhood cancer literature in three ways. First, most of the previous research trying to explain why some family members adjust better than others after a diagnosis of childhood cancer has focused on individual and contextual resources, overlooking intrafamilial resources. Our research upholds that the way in which the family as a whole deals with and responds to childhood cancer ('family functioning') impacts the adjustment of all members within the family (Alderfer et al., 2009; Barrera et al., 2009). To address this gap in the existing literature, all chapters in this part examined family functioning as a resource contributing to the individual adaptation of patients, parents, and siblings facing childhood cancer. Second, consistent with part 1, all chapters in this part considered the perspectives of multiple family members. In Chapter 6, data from *all* family members (patients, parents, siblings) were included. In Chapter 7, all siblings within the participating families, aged 5 years and older, completed questionnaires and were included in the analysis. In Chapter 8, parents' and siblings' perspectives were considered to better understand the family practice of support giving. In

all of these chapters, appropriate statistical techniques (multilevel modeling approach and MFMIA) were used to handle the interdependence of the data. Third, similarities between the findings of the meta-analysis (Chapter 5) and the quantitative studies (Chapters 6 and 7) increase the trustworthiness of our results. For example, emotional closeness within the family seems to be an important resource in the individual adaptation of family members facing childhood cancer. In Chapter 5, the meta-analysis showed that more family cohesion, more expressiveness, and less conflict were associated with better child adaptation (patients and siblings). These three family functioning domains together form the concept *'emotional closeness'*, as used in Chapter 6 and 7. In these quantitative studies, similar findings were found: more emotional closeness within the family was associated with better child and parent adaptation. In addition, family and network support proved to help family members to better cope with the illness: both Chapter 5 and Chapter 7 indicated that greater perceived support (from the family and the external network) was associated with better patient and sibling adaptation post-diagnosis.

Part 3: The Impact on the Parents' Intimate Relationship

In **Chapter 9**, our systematic review of parental couple functioning after a pediatric cancer diagnosis was described. Searches of Web of Science, Pubmed, Cochrane, PsycInfo and Embase resulted in the inclusion of 32 articles (17 qualitative, 13 quantitative and 2 mixed method papers). About two-thirds of the studies were cross-sectional (n = 21), and sample size varied from 8 individuals to 328 partners/164 couples. Results indicated that most couples adapt well to the crisis of their child receiving a cancer diagnosis in domains such as emotional closeness, support, marital satisfaction, and general marital adaptation. To the contrary, most couples experience difficulties in the domain of sexual intimacy and results regarding conflict are

mixed across qualitative (i.e., increased conflict) and quantitative studies (i.e., similar to pre-diagnosis).

Chapter 10 described a cross-sectional questionnaire study. More specifically, guided by our theoretical framework – the double ABCX model – this chapter focused on the association between dyadic coping (intrafamilial resources), individual outcomes, and relationship outcomes in parents facing childhood cancer. Dyadic coping refers to the extent to which partners deal with a stressor, like pediatric cancer, as a dyad (cfr. Chapter 2; Bodenmann, 1995). Individual outcomes were operationalized as feelings of depression, anxiety, and stress (both general stress and illness-related parenting stress). Relationship outcomes referred to parents' marital and sexual adaptation. We hypothesized that adequate dyadic coping (i.e., more supportive dyadic coping, more common dyadic coping, and less negative dyadic coping) would be associated with better individual adaptation (i.e., less negative emotions: less stress, anxiety and depression, and lower levels of childhood illness-related parenting stress), better marital adaptation, and better sexual adaptation within parents being confronted with cancer in their child.

These predictions were tested in a sample of 59 couples of children diagnosed with blood cancer. Mean time since diagnosis was 6.90 months post-diagnosis. Results were in line with our predictions and previous studies (e.g., Meier et al., 2011; Regan et al., 2014; Traa et al., 2015): supportive dyadic coping (e.g., "S/he makes me feel that s/he understands me and is committed to me"), common dyadic coping (e.g., "We try to tackle the problem together and work together"), and negative dyadic coping (e.g., "S/he does not take my stress seriously") were related to individual and relational outcomes for parents of children with cancer. More specifically, higher levels of common dyadic coping as reported by men was related to lower childhood illness-related stress (men and women), better marital adaptation (men), and better sexual adaptation (men). Men perceiving their partner as expressing more supportive dyadic coping was related to less childhood illness-related stress (women), depression (women), anxiety (women), stress (women) and

better marital adaptation (men). More negative dyadic coping as perceived by men in their partner was related to higher levels of depression (men), worse marital adaptation (men and women), and worse sexual adaptation (men). More common dyadic coping as reported by women and more supportive dyadic coping as perceived by women in their partner were related to better marital adaptation (women). More negative dyadic coping as perceived by women in their partner was related to worse marital adaptation (men and women), and lower sexual adaptation (women). In sum, this chapter supports the double ABCX model. More specifically, it supports the association between resources ('b') at the intrafamilial level (dyadic coping) and individual and family adaptation of parents facing childhood cancer ('x'; see Fig. 1).

Conclusion Part 3: These two studies (one review and one quantitative study) both focused on the impact of childhood cancer on the parents' intimate relationship (*couple adaptation*). They contribute to the existing childhood cancer literature in two ways. First, the focus on the parents' intimate relationship is rather unusual in the childhood cancer literature. Difficulties in the couple relationship often seem secondary to the more pressing need of the medical and psychosocial care for the ill child, and the consequences for the siblings and the family system as a whole. Therefore, such couple issues may be overlooked by clinicians and researchers, or may even be downplayed by the couples themselves. Based on the assumption that within families all family members mutually influence each other, (difficulties within) the parents' intimate relationship will have an impact on the individual adaptation of all family members, as well as the adaptation of the family system as a whole. We addressed this gap in the previous literature by conducting a systematic review and a quantitative questionnaire study on couple adaptation post-diagnosis. Second, whereas in Chapter 9 the impact on the parents' intimate relationship is described, Chapter 10 investigated why some couples adapt better than others, thereby shedding light on mechanisms underlying relationship outcomes of the child's parents. More specifically, the

extent to which parents deal with the stressor of pediatric cancer as a dyad ("dyadic coping"; see Bodenmann, 1995) was entered as resource. This study was the first investigating *dyadic coping* in the context of childhood cancer, and might foster future research to better understand this concept in the context of childhood cancer (e.g., Clever et al., 2019).

General Conclusion

Taken together, this doctoral dissertation helps us to formulate answers to our three research questions: (a) What are the short-term and long-term effects of childhood cancer on families, its members, and the parents' intimate relationship, (b) What resources may help families and their members to recover from crisis and adapt to the stressful circumstances resulting from childhood cancer, and (c) How do siblings experience childhood cancer within their families, its treatment, and their everyday family life post-diagnosis.

As expected, we found consistent evidence across all qualitative chapters (Chapters 3, 4, 8) that childhood cancer impacts the individuals within the family (patients, parents, siblings, *individual adaptation*), the family system as a whole (*family adaptation)* and the parents' intimate relationship (*couple adaptation*).

Based on our reviews (Chapters 1, 5, 9) and quantitative chapters (Chapters 2, 6, 7, 10), however, we know that this impact differs across individuals, families, and couples. This variability in outcomes emphasizes the need to investigate resources to explain why some family members, families, and couples adapt better than others. Therefore, this doctoral dissertation included the investigation of resources at three levels: individual resources (psychological flexibility: Chapter 2), intrafamilial resources (family functioning: Chapter 6, 7; family support: Chapter 7, 8; dyadic coping: Chapters 2, 9), and contextual resources (network support: Chapter 2, 7). Across the chapters, we found evidence of an important role for all included resources, emphasizing the need to take all three levels into account when

investigating individual, family, and couple adaptation in the context of childhood cancer.

Three specific resources were found to help families and their members best cope with the illness. First, this doctoral dissertation demonstrated the importance of *cohesion/emotional closeness*, both within the family (Chapters 3, 4, 5, 6, 7) and within the parents' intimate relationship ('supportive dyadic coping', Chapter 10). The more family members/parents feel that they are not alone and that they are surrounded and loved by their family/partner, the better able they were to adapt to the cancer diagnosis. Notably, this finding ("the more, the better") is not in line the Circumplex Model of Marital and Family Systems (CMMFS; Olson et al., 1979), a widely used model to describe and analyze family functioning. According this model, balanced levels of cohesion are most conductive to healthy family functioning, whereas unbalanced levels of cohesion are associated with unhealthy family functioning and thus maladaptation (Olson, 2011). Moreover, the finding "the more cohesion, the better" shows that what is considered as maladaptive in non-crisis situations, might be adaptive when facing a severe, life threatening stressor like childhood cancer.

Second, this doctoral dissertation showed the importance of family (Chapters 3, 5, 6, 7) and couple (Chapter 2) *expressiveness*: being able to share cancer-related thoughts and emotions within the family or the couple relationship helped the family members to best cope with the illness. Importantly, it is the *ability* to share thoughts and emotions within the family or the couple relationship that is predictive for adaptation, not the amount of conversations. In other words, the idea "the more sharing, the better" is not correct: as shown in chapter 8, some families/family members prefer not to talk about the cancer experience or emotions evoked by such an intrusive situation, as they are too tired or too emotional at that time, or because they don't want to evoke the same negative emotions in the other family members.

Third, this doctoral dissertation demonstrated the importance of *network support* (Chapters 2, 7). The more support, and the more one is

satisfied with his/her perceived support, the better the individual and family adapts. In other words, being surrounded by a supportive network not only helps the individual, but also the family as a whole, to deal with the illness.

Taken together, when being confronted with a potentially traumatic event, like childhood cancer, it is important to not stand alone. Consistent with the Social Ecological Model of Bronfenbrenner (1977) that demonstrate the dynamic interrelations among various personal and environmental factors, an individual needs to *feel* connected to a broader social network. He or she needs to be loved and embraced (cohesion) and to be heard and understood (expressiveness; support). In other words, childhood cancer does not only *impact* the ill child and his/her social environment, this social environment is *needed* to best cope with the illness.

Regarding research aim 3, our findings (see Chapter 1, 4, 5, 6, 7) emphasized the unique position of the sibling in the family when facing childhood cancer: as for the ill child, the siblings' world is impacted. But in contrast to the ill child, siblings often feel that they are at the periphery of the family and need to handle their emotions with less parental help. These findings reinforce the need to attend to siblings in research endeavors and in clinical practice, as they need help, too, (from parents, their network and clinicians) to best cope with the illness.

Theoretical Implications

One strength of this doctoral dissertation, is its grounding in an underlying theoretical framework, i.e., the double ABCX model (McCubbin & Patterson, 1983). As described in Chapter 1, in the majority of existing studies on family adaptation when facing childhood cancer (n = 71, 84%), no theoretical framework is specified as guiding the research questions or selection of the variables. Such failure to use theoretical frameworks risks limiting progression of the field, as advances cannot be made if theories go

untested and unrevised. In this section, the usefulness of the double ABCX model as the underlying theoretical frame for our work will be evaluated, and the theoretical implications of our work will be outlined.

In line with the double ABCX model (McCubbin & Patterson, 1983), this doctoral dissertation supports the importance of resources and perception in the prediction of individual, family, and couple adaptation when facing childhood cancer. In other words, adjustment post-diagnosis was better, when more individual, intrafamilial and contextual resources were available and the illness was perceived as more manageable and less uncontrollable. As a consequence, we strongly recommend including these variables in future research, defining them in theory, and translating these findings into clinical recommendations (see below).

However, based on this doctoral dissertation, several remarks can be made. First, as one can see in figure 1, we mainly focused on pre-crisis variables ('b', 'c') and indicators of crisis ('x') in our chapters, with only one chapter including post-crisis variables ('xX'; Chapter 2). This cross-sectional focus was mainly due to difficulties related to conducting *longitudinal research* (e.g., Chapter 7: for only a limited number of siblings multiple measurements were available). As a consequence, this doctoral dissertation is rather a reflection of the ABCX model (Hill, 1958), and not the double ABCX model (McCubbin & Patterson, 1983). From a theoretical point of view, however, we do believe in the merit of the double ABCX model, as this model (1) tries to explain and predict how families *recover from crisis* and why some are better able to adapt than others *over time* (Patterson, 1988) and (2) makes a distinction between *individual, intrafamilial* and *contextual* resources (as supported by our data).

Second, what is currently missing – in the double ABCX model and the present dissertation – is an objective assessment of the *stressor*. While this stressor is the same for all families (i.e., childhood cancer diagnosis), there is great variability in how this illness manifests, its treatments, and its prognosis (National Cancer Institute, nd.). For example, for acute lymphatic leukemia

patients (ALL), prognosis is better than for acute myeloid leukemia patients (AML), but the treatment course is much longer for ALL (2 years vs. 6 months for AML; WebMD, 2019). Further, individual responses to the same diagnosis and treatment can vary widely. For example, while some patients experience extreme levels of nausea and pain during cancer treatment, others may not experience such severe side effects (National Cancer Institute, nd.). In addition, in line with other family stress models (e.g., the Resilience Model of Family Stress, Adjustment and Adaptation; McCubbin & McCubbin, 1991), the integration of the concept 'vulnerability' can be useful. Vulnerability can be described as the risk of an individual or family to be physically or emotionally wounded by an unexpected event; and includes the pileup of concurrent and prior stressors, strains and transitions and their demands (Weber, 2011). By including this concept as a pre-crisis variable, a person's or family's history can be taken into account.

Third, one of the major contributions of the double ABCX model to stress theory, is the inclusion of the concept *coping* (Weber, 2011). Remarkably, coping strategies are included only as post-crisis variables (see Fig. 1). However, in line with other family stress theories, such as the Family Adjustment and Adaptation Response model (FAAR; Patterson, 1989), and several empirical studies (Barrera et al., 2004; Dahlquist et al., 1993), coping also matters in the pre-crisis stage.

Fourth, this doctoral dissertation provides evidence for the importance of the family's established patterns of functioning (*family functioning)* as a buffer against individual, family, and couple maladaptation. This idea is one of the key principles of the Typology Model of Family Adjustment and Adaptation (McCubbin, 1995). According to this theory, some *family types* are related to better outcomes. Family type can be defined as the composition of several characteristics of the family system (e.g., coherence, flexibility, bonding) and explains how families *typically* appraise, operate and/or behave (McCubbin et al., 1988). Given our evidence on the role of family functioning in the prediction of adjustment, and supported by the Typology Model of

Family Adjustment and Adaptation (McCubbin, 1995), we propose a more pronounced position of this concept in theoretical models.

To conclude, this doctoral dissertation supports the use of the double ABCX model in the context of childhood cancer. However, based on our results, other family stress models, and evidence supporting them within the current childhood cancer literature, we also recommend including other variables, i.e., an objective measure of the stressor, consideration of perceptions of vulnerability, family type and pre-crisis coping strategies.

Strengths, Limitations and Suggestions for Future Research

The present dissertation has several strengths, such as (1) the focus on the family as whole (both as an outcome and as a resource), siblings, and the parents' intimate relationship; (2) the inclusion of all family members' perspectives, combined with appropriate statistical techniques to take into account the interdependence of the data; (3) the use of both qualitative and quantitative methods; (4) the investigation of long-term consequences when facing childhood cancer (i.e., Chapter 2); and (5) grounding in an underlying theoretical framework, i.e., the double ABCX model. However, the results should be interpreted in light of some limitations. In this next section, we aim to delineate some general limitations of this dissertation, and we will provide suggestions for future research.

Ethical issues

When receiving a cancer diagnosis, the family's world turns upside down. As a researcher, it is most interesting to investigate short-term individual, family, and couple consequences immediately after diagnosis. However, from an ethical and human perspective, it is questionable to disturb

and add to the burden of these families immediate after diagnosis. We decided to set our first measurement within the first three weeks post-diagnosis. Although this timeline seemed feasible in advance, we were confronted with a low response rate at T1 (40%). Recruitment and data collection from families further along in their treatment process (i.e., refreshment samples; Taylor et al., 2020), produced a higher response rate (64%). It seems that participating in research in the first month post-diagnosis is extremely challenging and demanding; and future researchers should carefully weigh the benefits of research with the burden for the families.

Generalizability

The samples of our studies mainly consisted of white, Dutch speaking, married, middle-class families. This can be an issue for the generalizability of our results. Prior research shows that low SES and single parent household are risk factors for poorer parent and child adaptation post-diagnosis (Mulhern et al., 1989; Van Dongen-Melman et al., 1995). Therefore, research with a more heterogeneous sample might show slightly different results. Second, all children were diagnosed with leukemia of non-Hodgkin lymphoma. Children with other cancer diagnoses and their families might have different experiences (e.g., severe behavioral changes in brain tumor patients). Third, despite our efforts to include all family members, it was especially difficult to convince fathers to participate (cfr. previous studies; Davison et al., 2017): with 87 participating fathers and 118 participating mothers, several of our findings are mainly based on the perspectives of mothers. We know, based upon the meaningful discrepancies we found in patterns of results for mothers and fathers (e.g., see Chapter 6), that findings derived from data from mothers cannot simply be generalized to fathers.

Taken together, future research should include more heterogeneous samples (e.g., different languages and ethnic origins; heterosexual and

homosexual couples; married and single parents; different cancer diagnoses), with special attention to the inclusion of fathers.

Causality

The associations described in most chapters (except for Chapter 2) are cross-sectional in nature. As a consequence, the temporal cause and effect relationships of the variables under investigation could not be tested and inverse associations (e.g., individual maladjustment predicting worse family functioning) are also possible. In Chapter 7, longitudinal analyses were considered, but were ultimately not done because only 13 siblings provided longitudinal data, and such a small sample would have led to severe power issues. As such, the findings regarding variables associated cross-sectionally with individual and couple adaptation to childhood cancer need to be evaluated and expanded upon in longitudinal designs (for example: Van Schoors et al., in preparation).

Measurements

This dissertation included a longitudinal survey study. For this survey study, families were asked to complete a set of questionnaires at five different time points. This set of questionnaires was composed based on the double ABCX model, with every component of the model corresponding with a specific questionnaire (e.g., perception = Perceived Stress Scale). In addition, for several variables (e.g., individual adaptation), both generic (e.g., Depression Anxiety and Stress Scale) and condition-specific measurements (e.g., Situation Specific Emotional Reaction Questionnaire) were included. This resulted in a test battery requiring, on average, 70 minutes for parents, and, at maximum, 50 minutes for children (depending on their age) to complete. We might assume that the length of the test battery is related to the high dropout percentages in the included studies: 32% of the families had one

measurement, 30% had two measurements, and 19% had three measurements; whereas only 7% and 12% of the families had four or five measurements, respectively.

Furthermore, family functioning and family adaptation were measured using the Family Environment Scale and the Impact on Family Scale, respectively. Two recent reviews, however, documented the difficulty of such questionnaires in capturing the unique and specific changes in family functioning when facing a life-threatening pediatric illness, like childhood cancer (Alderfer et al., 2008; Hildenbrand et al., in revision*)*. Indeed, according to these reviews, *population based family measures*, like the IOF and the FES, (a) have psychometric properties that are less than ideal or are simply not well documented in pediatric samples with chronic illnesses, which may lead to instability of findings or even erroneous conclusions; and (b) may not be sensitive to the types of processes and patterns that arise among families being confronted with severe life threatening pediatric illnesses. Furthermore, families of children with chronic health conditions may score in ‘‘unhealthy’’ or ‘‘dysfunctional’’ ranges using norms and cut-offs developed for such measures based on the general population; however, these patterns of functioning may actually be adaptive for these families at that specific point in time. For example, rigidity with regard to roles and rules might be problematic in the general population; yet, for families needing to adhere to a complex treatment regimen for their child, greater rigidity may be associated with better outcomes (Van Schoors et al., 2020).

Taken together, these issues suggest that future research should be designed to focus specifically on a few variables of interest to decrease participant burden (< 30 minutes). In addition, a deeper understanding of (changes in) family functioning after a childhood cancer diagnosis is warranted. What is needed to further this understanding is empirical research extending *beyond* the limited base of population-based family measures-studies on which existing knowledge currently rest.

Clinical Implications

This doctoral dissertation demonstrate that the life of all family members, the family system as a whole, and the parents' intimate relationship are affected by a childhood cancer diagnosis and its treatment. As a consequence, when facing childhood cancer, a holistic approach – including interventions at the individual, couple, and family level – is needed to best help families to cope with this severe stressor. Moreover, the finding that entire families are affected by the illness fully supports the recommendations of the Pediatric Psychosocial Preventative Health Model (PPPHM; Kazak, 2006). According this model, all families of children diagnosed with cancer should be screened for factors potentially predisposing them for maladjustment or distress, including individual, intrafamilial, and contextual risk factors. The Psychosocial Assessment Tool (PAT) is a screening instrument designed to assess such psychosocial risk in families of children newly diagnosed with cancer (Pai et al., 2008). Accordingly, in line with the PPPHM, clinical interventions should be tailored to these risk factors, the families' specific care needs, and the care expectancies of these families ranging from standard psychosocial care to more intensive individual or family therapy (see Kazak, 2006 for greater detail).

Furthermore, clinicians may foster adaptation of all family members, the family as a whole and the parents' intimate relationship by mobilizing *resources*. Based on this doctoral dissertation, resources at all three levels (individual, intrafamilial, and contextual resources) proved to be important for individual, family and couple adaptation. First, as psychological flexibility (individual resource) seemed to be important (Chapter 2), family members could benefit from interventions targeting the promotion of acceptance of unwanted negative thoughts and emotions, e.g., using Acceptance and Commitment Therapy (Hayes et al., 2012). Second, given the importance of *family functioning* in the prediction of adaptation post-diagnosis (intrafamilial resource, Chapters 5, 6, 7), family functioning should be routinely assessed in

this population. In case of dysfunctional family functioning, empirically based family-level intervention approaches can be used, as described in the literature (e.g., Rolland & Walsh, 2006; Saltzman et al.2013). In addition, across the treatment process, particular family functioning domains should be given special attention. For example, clinicians should (1) promote cancer-related communication within the family, taking into account the complexity of sharing emotions and the individual differences in expressiveness (see Chapter 8), and (2) enhance family cohesion/togetherness. The latter can be operationalized by supporting *rooming in* for parents or flexible visiting hours for siblings. Third, as dyadic coping matters for the individual and relational functioning of parents facing cancer in their child (Chapters 2, 10), it is important to screen and tackle relational issues besides individual issues. For example, clinicians should invite the couple system as a whole, as only by taking into account the perspectives of both members, couple level variables – such as dyadic coping – can be fully understood and improved when needed. Furthermore, couples could benefit from Couples Coping Enhancement Training (CCET; Widmer et al., 2005). This training aims to strengthen the coping competencies of both partners by strengthening dyadic communication and dyadic coping. Fourth, as network support is an important contextual resource (Chapters 2, 7), clinicians should map the existing social network of the families and help families to ask for (emotional or practical) help where/when needed.

Finally, clinical interventions should be tailored to some important individual characteristics of the family members. For example, this doctoral dissertation suggest that mothers might be more vulnerable than fathers (Chapters 3, 10), or that siblings have less positive cancer-related feelings than the other family members (Chapter 7). As a consequence, clinical cancer-related interventions should not only be tailored to family risk factors (PPPHM; Kazak, 2006), but also to the specific family members (mothers vs. fathers vs. siblings vs. patients) ánd the individuality of each person.

General Conclusion

Childhood cancer is a severe, life threatening disease that impacts the family as a whole, its members, and the parents' intimate relationship. Guided by the double ABCX model, the present dissertation aimed to gain insight into the short- and long-term impacts on individuals, families, and couples dealing with childhood cancer and the resources that may help in recovery from this crisis. As the results of our chapters (as discussed above) imply, resources at all three levels (individual, intrafamilial, contextual) and family members' perception are important to best understand individual, family, and couple adaptation when facing childhood cancer. More specifically, more psychological flexibility (individual resource), more adequate dyadic coping (intrafamilial resource) and more social support (contextual resource) were associated with better family adaptation; more emotional closeness within the family (intrafamilial resource), a more firm family structure (intrafamilial resource), more family support (intrafamilial resource), more network support (contextual resource), and perceiving the illness as manageable (perception) were associated with better individual adaptation, whereas more adequate dyadic coping (intrafamilial resource) was associated with better individual and couple adaptation of parents facing childhood cancer. Consequently, we recommend to shift focus in childhood cancer research and clinical practice away from a narrow individual perspective toward a more holistic approach, including an individual, intrafamilial, and contextual perspective.

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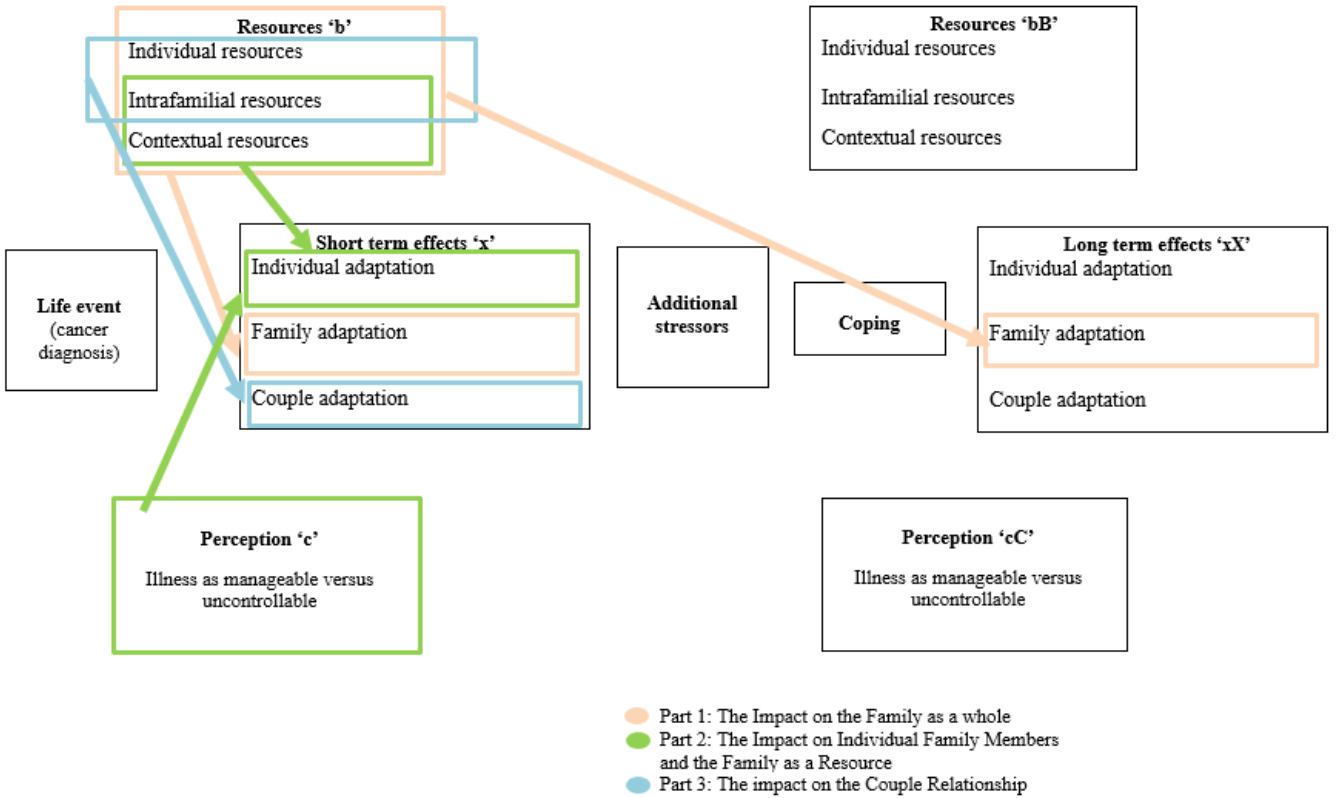

Figure 1. The double ABCX model: An overview of the current doctoral dissertation

ENGLISH SUMMARY

Every year, 300,000 children are diagnosed with cancer worldwide (American Childhood Cancer Organization, nd.). In Belgium, it concerns 341 children annually (Belgian Cancer Registry, 2020). The most common cancer diagnoses in children are leukemia (28% of all cancer diagnoses in children), brain and spinal cord tumors (26%), neuroblastoma (6%), lymphoma (including both Hodgkin (3%) and non-Hodgkin (5%)), Wilms tumor (5%), and rhabdomyosarcoma (3%) (American Cancer Society, 2019). In this dissertation, to increase homogeneity of the sample, we focused on leukemia and Non-Hodgkin lymphoma. These types of cancer have similar treatment protocols and together they represent one third of all cancer diagnoses in children. In *leukemia*, the production of the white blood cells (WBc) in the bone marrow is disturbed. Based on the type of the WBc (lymphocytes vs. granulocytes) and the speed of the disturbance (acute vs. chronic), leukemia can be divided in four subtypes: acute lymphatic leukemia (ALL), acute myeloid leukemia (AML), chronic lymphatic leukemia (CLL) and chronic myeloid leukemia (CML). *Non-Hodgkin lymphoma* starts in the lymphatic system, with an uncontrolled cell division of the lymphocytes (Lardon, 2011). Non-Hodgkin lymphoma has four stages, from stage 1 (i.e., the cancer is situated in a single region or organ) to 4 (i.e., the cancer has spread to other organs outside the lymph system) (Cancer Treatment Centers of America, 2020). To cure leukemia or non-Hodgkin lymphoma, there are three important treatment methods, that is radiotherapy, chemotherapy, and stem cell transplantation.

State of the Art: What is the Current Evidence on the Psychosocial Consequences of Childhood Cancer?

The current literature shows that childhood cancer not only impacts the ill child, but also the other family members (parents, siblings), the family system as a whole, and the parents' intimate relationship. More specifically, the *child with cancer* often experiences a range of difficulties, such as pain, fatigue, reduced immunity, anxiety, and uncertainty (Voûte et al., 1997). *Parents* of children diagnosed with cancer often report significantly higher levels of distress, posttraumatic stress symptoms, parental conflict, emotional problems (anxiety, feelings of depression), and physical complaints (insomnia, fatigue), compared to parents with healthy children (Pai et al., 2007). *Siblings* often report poor quality of life and negative emotional reactions (Alderfer et al., 2010).

Few studies have documented the impact of childhood cancer on the *family as a whole* (see Pai et al., 2007 for an overview). Overall, quantitative studies revealed that most families function within normative ranges (e.g., adaptability, Pai et al., 2007; family support, Brown et al., 2003) or even report improved functioning in some realms (e.g., cohesion, Cornman, 1993). Qualitative studies, however, indicated a loss of normal family life (Bjork et al., 2009; Clarke-Steffen, 1997) and troubles balancing multiple family needs including those of siblings (Bjork et al., 2009). Similarly few studies have examined the impacts of childhood cancer on family subsystems, like the *couple subsystem*. The few that are available (e.g., Hoekstra-Weebers et al., 1998; Patistea et al., 2000) reveal that although most couples adjust well to the crisis of childhood cancer in domains such as emotional closeness, couple support, and marital satisfaction, most couples do experience difficulties in the domains of sexual intimacy, both during and after treatment.

As the current literature documents a considerable *variability* in individual, family and couple outcomes to childhood cancer, a growing

number of studies has tried to explain why some family members, families and couples adapt better than others. Therefore, the latter studies investigated the role of potential resources situated at three levels: the individual level (e.g., personality; Erickson & Steiner, 2001), the intrafamilial level (e.g., family cohesion; Alderfer et al., 2009), and the contextual level (e.g., network support; Corey et al., 2008).

Limitations of Existing Literature and Research Aims of the Current Dissertation

To date, the issues of how childhood cancer affects the *family as a whole* and the *parents' intimate relationship* have received inadequate research attention. Also, there is little research on how childhood cancer affects family members other than the ill child and his or her parents, namely the *siblings* of the child with cancer. Furthermore, much of the existing research regarding the effects of childhood cancer has relied on cross-sectional rather than longitudinal designs, providing limited information about *patterns of adjustment across time*. Finally, in the majority of existing pediatric cancer studies, no *theoretical frameworks* are specified as guiding the research questions or the selection of variables. Failure to use theoretical frameworks jeopardizes progression of the field as advances cannot be made if theories go untested and unrevised.

To address these limitations, the *main aims* of the present dissertation were to examine (a) the short-term and long-term effects of childhood cancer on families, its members (patients, parents, and siblings) and the parents' intimate relationship, and (b) the resources – situated at the individual level, intrafamilial level, and contextual level – that may help families and their members to recover from the cancer related crisis and to adapt to the stressful circumstances resulting from it. In addition, given our focus on all family

members and the lack of research on and knowledge about siblings, the present dissertation will give special attention to (c) how siblings experience the illness and its treatment, and the consequences for their everyday family life.

As underlying conceptual framework, we used the *double ABCX model* (McCubbin & Patterson, 1983). The double ABCX model describes how a stressor (i.e., childhood cancer) impacts the adaptation of each family member (individual level), the family subsystems (family subsystem level), and the family as a whole (family level) over time and identifies variables that allow an understanding of why some family members, families and couples manage to adapt better than others (Weber, 2010). In addition, the model acknowledges that an individual's and a family (sub)system's response to a major stressor, like childhood cancer, develops over time and that family/individual adaptation is influenced by the family members' *resources* and the *perception* family members have of the stressful event (i.e., childhood cancer diagnosis). Based on the childhood cancer literature, this dissertation includes the following resources: (a) each family member's psychological flexibility (i.e., the skill to flexibly adapt to fluctuating situational demands, being open and accepting of emotional experiences, and being willing to engage in difficult activities to persist in the direction of important values; Kashdan & Rottenberg, 2010) as an *individual resource*, (b) family functioning (e.g., support, cohesion, communication, conflict in the family and its subsystems) and dyadic coping (i.e., the extent to which parents deal with the stressor of pediatric cancer as a dyad; Bodenmann, 1995) as *intrafamilial* resources, and (c) the family's social network (e.g., friends, relatives) and the support received from them as *contextual* resources. So, we predict better adaptation at the individual and family (subsystem) level for families with higher levels of individual, intrafamilial, and contextual *resources*, and when the illness is perceived as more manageable and less uncontrollable (*perception).*

Findings

In order to address the research aims addressed above, the present dissertation was divided in three parts and includes 10 chapters (i.e., 2 systematic reviews, 1 meta-analysis, 3 qualitative studies and 4 quantitative studies). The first part, "*The impact on the family as a whole*", focuses on the consequences of childhood cancer for the family system (i.e., family adaptation). The second part, "The *impact on individual family members and the family as a resource*", focuses on the individual adaptation of patients, parents, and siblings and on potential resources that can explain why some family members adapt better than others. The thread running through the chapters in this part is the inclusion of a key intrafamilial resource, that is "family functioning" (= the way in which the family as a whole deals with and responds to childhood cancer). So, in part 1 family functioning is conceptualized as an outcome variable; in part 2 family functioning is conceptualized as a resource that may contribute to individual adaptation of a patient, his or her parents and siblings when facing childhood cancer. The third part, "*The impact on the parents' intimate relationship*", focuses on the consequences of childhood cancer for the parental couple subsystem.

As expected, we found consistent evidence across all qualitative chapters (Chapters 3, 4, 8) that childhood cancer impacts the individuals within the family (patients, parents, siblings; *individual adaptation*), the family system as a whole (*family adaptation)* and the parents' intimate relationship (*couple adaptation*). Based on our reviews (Chapters 1, 5, 9) and quantitative chapters (Chapters 2, 6, 7, 10), however, we know that this impact is different across individuals, families and couples. This variability in outcomes emphasized the need to investigate resources and urged to explain why some family members, families, and couples adapt better than others. Therefore, this doctoral dissertation included individual resources (psychological flexibility: Chapter 2), intrafamilial resources (family

functioning: Chapter 6, 7; family support: Chapter 7, 8; dyadic coping: Chapters 2, 9), and contextual resources (network support: Chapter 2, 7).

Across the chapters, we found evidence for all included resources, emphasizing the need to take all three levels into account when investigating individual, family and couple adaptation in the context of childhood cancer. Moreover, three resources in particular helped families and its members to best cope with the illness: *cohesion/emotional closeness*, *expressiveness*, and *network support*. In other words, the more one feels connected within and loved by the family, the more one can share cancer-related thoughts and emotions within the family, and the more support from the external network, the better the adaptation post-diagnosis. In addition, we found that the *perception* family members have of the illness, impacts the adaptation as well: the more one perceived the illness as manageable and the less as uncontrollable, the better the individual adaptation.

Regarding research aim 3, our findings (see Chapter 1, 4, 5, 6, 7) emphasized the unique position of the sibling in the family when facing childhood cancer: as for the ill child, the siblings' world is impacted. But in contrast to the ill child, siblings often feel that they are at the periphery of the family and need to handle their emotions with less parental help. These findings reinforce the need to attend to siblings in research endeavors and in clinical practice, as they need help too (from parents, their network and clinicians) to best cope with the illness.

Limitations and Suggestions for Future Research

The results of the present dissertation should be interpreted in the light of some limitations. A first limitation concerns the sample characteristics. The samples of our studies mainly consisted of white, Dutch speaking, married, middle-class families. As prior research showed that low SES and single parent household are risk factors for parent and child adaptation (Mulhern et

al., 1989; Van Dongen-Melman et al., 1995), research with a more heterogeneous sample might show slightly different results. Second, all children were diagnosed with leukemia of non-Hodgkin lymphoma. Children with other cancer diagnoses and their families might have different experiences (e.g., severe behavioral changes in brain tumor patients). Third, the associations described in most chapters (except for Chapter 2) are cross-sectional in nature. As a consequence, the temporal cause and effect relationships of the variables under investigation could not be tested and inverse associations (e.g., individual maladjustment predicting worse family functioning) are also possible. Fourth, all family measures used in this doctoral dissertation are population based family measures. However, most population based family measures have psychometric properties that are less than ideal or are simply not well documented in pediatric samples with chronic illnesses. This might have led to instability of findings or even erroneous conclusions. In addition, by applying population based family measures, researchers ignore the specificity of families being confronted with severe life threatening pediatric illnesses. Indeed, what is ‘‘dysfunctional’’ in the general population (i.e., high family cohesion), may actually be adaptive for families facing a severe stressor, like childhood cancer, at that specific point in time.

Implications

The aforementioned contributions of the reviews, qualitative and quantitative studies in the present dissertation can be translated into several relevant theoretical and clinical implications.

Theoretical Implications

This doctoral dissertation supports the double ABCX model by emphasizing the importance of resources (individual, intrafamilial, and

contextual resources) and perception in the prediction of individual, family and couple adaptation when facing childhood cancer. However, also based on this doctoral dissertation, several critical remarks can be made. First, what is currently missing – in the double ABCX model and the present dissertation – is an objective assessment of the *stressor*. While this stressor is the same for all families (i.e., childhood cancer diagnosis), there is great variability in how this illness manifests, its treatments, and its prognosis (National Cancer Institute, nd.). Second, in line with other family stress models (e.g., the Resilience Model of Family Stress, Adjustment and Adaptation; McCubbin & McCubbin, 1991), the integration of the concept 'vulnerability' can be useful. Vulnerability can be described as the risk of an individual or family to be physically or emotionally wounded by an unexpected event; and includes the pileup of concurrent and prior stressors, strains and transitions and their demands (Weber, 2011). By including this concept as a pre-crisis variable, a person's or family's history can be taken into account. Third, one of the major contributions of the double ABCX model to stress theory, is the inclusion of the concept *coping* (Weber, 2011). Remarkably, these coping strategies were included only as post-crisis variables. However, in line with other family stress theories, such as the Family Adjustment and Adaptation Response model (FAAR; Patterson, 1989), and several empirical studies (Barrera et al., 2004; Dahlquist et al., 1993), coping also matters in the pre-crisis stage. Fourth, this doctoral dissertation provides evidence for the importance of the family's established patterns of functioning (*family functioning)* as buffer against individual, family, and couple maladaptation. This idea is one of the key principles of the Typology Model of Family Adjustment and Adaptation (McCubbin, 1995). According to this theory, some *family types* are related to better outcomes. Family type can be defined as the composition of several characteristics of the family system (e.g., coherence, flexibility, bonding) and explains how families *typically* appraise, operate and/or behave (McCubbin et al., 1988). Given our evidence on the role of family functioning in the prediction of adjustment, and supported by the Typology Model of Family

Adjustment and Adaptation (McCubbin, 1995), we propose a more pronounced position of this concept in theoretical models.

Clinical Implications

This doctoral dissertation demonstrated that the life of all family members, the family system as a whole, and the parents' intimate relationship are affected by a childhood cancer diagnosis and its treatment. As a consequence, when facing childhood cancer, a holistic approach – including interventions at the individual, couple, and family level – is needed to best help families to cope with this severe stressor. Furthermore, clinicians may foster adaptation of all family members, the family as a whole and the parents' intimate relationship by mobilizing *resources*. Based on this doctoral dissertation, resources at all three levels (individual, intrafamilial, and contextual resources) proved to be important for individual, family, and couple adaptation. First, as psychological flexibility (individual resource) seemed to be important (Chapter 2), family members could benefit from interventions targeting the promotion of acceptance of unwanted negative thoughts and emotions, e.g., using Acceptance and Commitment Therapy (Hayes et al., 2012). Second, given the importance of *family functioning* in the prediction of adaptation post-diagnosis (intrafamilial resource, Chapters 5, 6, 7), family functioning should be routinely assessed in this population. In case of dysfunctional family functioning, empirically based family-level intervention approaches can be used, as described in the literature (e.g., Rolland & Walsh, 2006; Saltzman et al.2013). Third, as dyadic coping matters for the individual and relational functioning of parents facing cancer in their child (Chapters 2, 10), it is important to screen and tackle relational issues besides individual issues. For example, clinicians should invite the couple system as a whole, as only by taking into account the perspectives of both members, couple level variables – such as dyadic coping – can be fully understood and improved when needed. Furthermore, couples could benefit

from Couples Coping Enhancement Training (CCET; Widmer et al., 2005). This training aims to strengthen the coping competencies of both partners by strengthening dyadic communication and dyadic coping. Fourth, as network support is an important contextual resource (Chapters 2, 7), clinicians should map the existing social network of the families and help families to ask for (emotional or practical) help where/when needed. Finally, clinical interventions should be tailored to some important individual characteristics of the family members. For example, this doctoral dissertation suggest that mothers might be more vulnerable than fathers (Chapters 3, 10), or that siblings have less positive cancer-related feelings than the other family members (Chapter 7). As a consequence, clinical cancer-related interventions should not only be tailored to family risk factors, but also to the specific family members (mothers vs. fathers vs. siblings vs. patients) ánd the individuality of each person.

General Conclusion

Childhood cancer is a severe, life threatening disease that impacts the family as a whole, its members, and the parents' intimate relationship. Guided by the double ABCX model, the present dissertation aimed to gain insight into the short- and long-term impacts on individuals, families, and couples dealing with childhood cancer and the resources that may help in recovery from this crisis. As the results of our chapters (as discussed above) imply, resources at all three levels (individual, intrafamilial, contextual) and family members' perception are important to best understand individual, family, and couple adaptation when facing childhood cancer. More specifically, more psychological flexibility (individual resource), more adequate dyadic coping (intrafamilial resource) and more social support (contextual resource) were associated with better family adaptation; more emotional closeness within the family (intrafamilial resource), a more firm family structure (intrafamilial

resource), more family support (intrafamilial resource), more network support (contextual resource), and perceiving the illness as manageable (perception) were associated with better individual adaptation, whereas more adequate dyadic coping (intrafamilial resource) was associated with better individual and couple adaptation of parents facing childhood cancer. Consequently, we recommend to shift focus in childhood cancer research and clinical practice away from a narrow individual perspective toward a more holistic approach, including an individual, intrafamilial, and contextual perspective.

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NEDERLANDSTALIGE SAMENVATTING

Wereldwijd krijgen elk jaar 300 000 kinderen de diagnose kanker (American Childhood Cancer Organization, nd.), met 341 kinderen per jaar in België (Belgian Cancer Registry, 2020). De meest voorkomende kankerdiagnoses bij kinderen zijn leukemie (28%), hersen- en ruggenmergtumoren (26%), neuroblastomen (6%), lymfomen (zowel Hodgkin (3%) als non-Hodgkin (5%) lymfomen), Wilms tumoren (5%) en rhabdomyosarcomen (3%) (American Cancer Society, 2019). Om de homogeniteit van de steekproef te verhogen, focussen we in dit proefschrift op kinderen met leukemie en non-Hodgkin lymfoom. Beide kankersoorten hebben een vergelijkbare behandeling en samen vormen zij één derde van alle kankerdiagnoses bij kinderen. Bij *leukemie* is de productie van de witte bloedcellen (WBc) in het beenmerg verstoord. Op basis van het type WBc (lymfocyten vs. granulocyten) en de snelheid waarmee de ziekte zich ontwikkelt (acuut vs. chronisch), onderscheiden we vier subtypes: acute lymfatische leukemie (ALL), acute myeloïde leukemie (AML), chronische lymfatische leukemie (CLL) en chronische myeloïde leukemie (CML). *Non-Hodgkin lymfomen* starten in het lymfesysteem en worden gekenmerkt door een ongecontroleerde celdeling van de lymfocyten (Lardon, 2011). Bij non-Hodgkin lymfomen maakt men een onderscheid tussen vier stadia, gaande van stadium 1 (d.i. kanker in één plaats of orgaan) tot stadium 4 (d.i. uitgezaaide kanker buiten het lymfesysteem) (Cancer Treatment Centers of America, 2020). De behandeling van zowel leukemie als non-Hodgkin lymfoom bestaat uit drie grote behandelmethodieken, namelijk radiotherapie, chemotherapie en stamceltransplantaties.

Stand van Zaken: Wat Weten we over de Psychosociale Gevolgen van Kinderkanker?

De huidige literatuur omtrent de individuele, gezins- en relationele gevolgen van kinderkanker toont aan dat een diagnose kinderkanker niet enkel een impact heeft op het zieke kind, maar ook op de andere gezinsleden (ouders, broers/zussen; individuele adaptatie), het gezin in zijn geheel (gezinsadaptatie) en de partnerrelatie van de ouders (relationele adaptatie). Met name, *kinderen met kanker* krijgen vaak te maken met verschillende moeilijkheden, zoals pijn, vermoeidheid, angst en onzekerheid (Voûte et al., 1997). *Ouders* van kinderen met kanker kunnen significant meer distress, posttraumatische stresssymptomen, ouderlijk conflict, emotionele problemen (zoals gevoelens van angst en depressie) en fysieke klachten (zoals slapeloosheid en vermoeidheid) rapporteren in vergelijking met ouders van gezonde kinderen (Pai et al., 2007). *Broers/zussen*, tenslotte, rapporteren naar aanleiding van de ziekte van hun broer/zus vaak een verminderde levenskwaliteit en negatieve emotionele reacties, zoals verdriet, jaloezie of angst (Alderfer et al., 2010).

Een aantal studies brachten de impact van kinderkanker op het *gezin in zijn geheel* in kaart (zie Pai en collega's (2007) voor een overzicht). Kwantitatieve studies toonden aan dat voor de meeste gezinnen het gezinsfunctioneren vergelijkbaar is met normen (Pai et al., 2007; Brown et al., 2003) of zelfs verbeterde in vergelijking met vóór de diagnose (e.g. cohesie, Cornman, 1993). Kwalitatieve studies daarentegen, onthulden een verlies van een "normaal" gezinsleven (Bjork et al., 2009; Clarke-Steffen, 1997) en moeilijkheden om aan de noden van alle gezinsleden tegemoet te komen, inclusief de noden van de gezonde broers/zussen (Bjork et al., 2009). Betreffende de partnerrelatie van de ouders toonden een aantal studies (o.a. Hoekstra-Weebers et al., 1998; Patistea et al., 2000) aan dat – hoewel het

merendeel zich goed aanpast – sommige koppels problemen ervaren op vlak van seksuele intimiteit, zowel tijdens als na de kankerbehandeling.

Tenslotte, de huidige literatuur omtrent de individuele, gezins- en relationele gevolgen van kinderkanker toont aanzienlijke variabiliteit in uitkomsten: hoewel de meeste gezinsleden/gezinnen zich goed aanpassen na een diagnose kinderkanker, ontwikkelen anderen individuele, gezins- en/of relationele problemen tijdens of na de behandeling. Meer en meer onderzoekers probeerden dan ook deze variabiliteit te verklaren door zicht te krijgen op mogelijke hulpbronnen: wat maakt dat sommige gezinsleden, gezinnen of koppels zich beter aanpassen dan anderen? Deze hulpbronnen kunnen zich situeren op drie niveaus: individueel niveau (o.a. persoonlijkheid; Erickson & Steiner, 2001), intrafamiliaal niveau (o.a. cohesie; Alderfer et al., 2009) en contextueel niveau (o.a. sociale steun; Corey et al., 2008).

Beperkingen binnen Bestaand Literatuur en Huidige Onderzoeksdoelen

Tot op heden is er weinig onderzoek naar de impact van kinderkanker op het *gezin in zijn geheel* en de *partnerrelatie van de ouders*. Eveneens is er weinig onderzoek omtrent de gevolgen van kinderkanker voor de *broers en zussen* van het kind met kanker. Daarnaast maakt bestaand onderzoek naar de gevolgen van kinderkanker voornamelijk gebruik van cross-sectionele designs, waardoor kennis over de *langetermijngevolgen* beperkt blijft. Tenslotte is er in het merendeel van de bestaande kinderkankerstudies geen onderliggend *theoretisch kader*, waardoor de selectie van de variabelen en de interpretatie van de resultaten eerder arbitrair verloopt.

Om tegemoet te komen aan deze beperkingen, stelden wij in dit proefschrift volgende onderzoeksdoelstellingen op: onderzoek naar (a) de korte- en langetermijngevolgen van kinderkanker voor gezinnen, gezinsleden

(patiënt, ouders, broers/zussen) en de partnerrelatie van de ouders en (b) hulpbronnen – zowel individuele, intrafamiliale als contextuele hulpbronnen – die gezinnen kunnen helpen om zich aan te passen aan de stressvolle omstandigheden na een diagnose kinderkanker. Gezien onze focus op *alle* gezinsleden en het gebrek aan onderzoek naar en kennis over broers/zussen, focust dit proefschrift in het bijzonder op (c) hoe broers/zussen de kankerdiagnose en de behandeling ervaren, alsook de gevolgen voor hun dagdagelijks gezinsleven.

Als onderliggend conceptueel model maakten wij gebruik van het *dubbele ABCX model* (McCubbin & Patterson, 1983). Het dubbele ABCX model beschrijft hoe een stressor (d.i. kinderkanker) een impact heeft op de adaptatie van elk gezinslid, de gezinssubsystemen en het gezin in zijn geheel, en identificeert variabelen die helpen te begrijpen waarom sommige gezinsleden, gezinnen en koppels zich beter aanpassen dan anderen (Weber, 2010). Daarnaast erkent het model dat de individuele en gezinsrespons na een stressor, zoals kinderkanker, verandert over de tijd heen en dat de adaptatie (zowel individueel, familiaal als relationeel) beïnvloed wordt door de *hulpbronnen* die de gezinsleden bezitten en de *perceptie* die de gezinsleden hebben over de stressvolle gebeurtenis (d.i. kinderkanker).

Vertrekkend vanuit de bestaande kinderkankerliteratuur includeerde dit proefschrift de volgende hulpbronnen: (a) de psychologische flexibiliteit van elk gezinslid (d.i. de vaardigheid om zich flexibel aan te passen aan veranderende omstandigheden, open te staan voor en het aanvaarden van emotionele, ingrijpende gebeurtenissen; Kashdan & Rottenberg, 2010) als *individuele hulpbron*, (b) gezinsfunctioneren (o.a. steun, cohesie, communicatie, conflict binnen het gezin en de -subsystemen) en dyadische coping (d.i. de mate waarin partners als een koppel omgaan met de stressor; Bodenmann, 1995) als *intrafamiliale hulpbronnen* en (c) het sociaal netwerk van het gezin (o.a. vrienden, kennissen) en de steun die zij ervaren van dit netwerk als *contextuele hulpbronnen*. Samengevat, wij voorspelden een betere adaptatie op individueel, familiaal en relationeel niveau wanneer een gezin

meer individuele, intrafamiliale en contextuele *hulpbronnen* bezat, en wanneer men de ziekte als meer hanteerbaar en minder oncontroleerbaar beoordeelde (*perceptie).*

Resultaten

Om een antwoord te kunnen formuleren op de onderzoeksdoelstellingen – zoals hierboven beschreven –, includeerden we tien hoofdstukken (d.i. twee systematische reviews, één meta-analyse, drie kwalitatieve studies en vier kwantitatieve studies), opgedeeld in drie delen. Het eerste deel, *"De impact op het gezin in zijn geheel"* focust op de gevolgen van kinderkanker voor het gezin (d.i. gezinsadaptatie). Het tweede deel, *"De impact op de individuele gezinsleden en het gezin als hulpbron"* focust op de individuele adaptatie van patiënten, ouders en broers/zussen en op mogelijke hulpbronnen die kunnen verklaren waarom sommige gezinsleden zich beter aanpassen dan anderen. De rode draad doorheen de hoofdstukken in dit deel is de inclusie van het *gezinsfunctioneren*, een belangrijke intrafamiliale hulpbron. Gezinsfunctioneren is de manier waarop een gezin omgaat met en zich aanpast aan de kankerdiagnose. In het eerste deel wordt gezinsfunctioneren dus beschouwd als uitkomstvariabele, terwijl gezinsfunctioneren in het tweede deel gezien wordt als een predictor voor de individuele adaptatie van de verschillende gezinsleden. Het derde deel, *"De impact op de partnerrelatie van de ouders"* focust op de gevolgen van kinderkanker voor de partnerrelatie van de ouders.

In lijn met onze verwachtingen toonden de kwalitatieve hoofdstukken (Hoofdstukken 3, 4, 8) aan dat een diagnose kinderkanker een grote impact heeft op de verschillende gezinsleden (patiënten, ouders, broers/zussen; individuele adaptatie), het gezin in zijn geheel (gezinsadaptatie) en de partnerrelatie van de ouders (relationele adaptatie). Op basis van onze reviews (Hoofdstukken 1, 5, 9) en kwantitatieve hoofdstukken (Hoofdstukken 2, 6, 7,

10) echter, weten we dat er een grote variabiliteit is in uitkomsten. Om deze variabiliteit te verklaren, werden in dit proefschrift zowel individuele hulpbronnen (psychologische flexibiliteit: Hoofdstuk 2), intrafamiliale hulpbronnen (gezinsfunctioneren: Hoofdstukken 5, 6, 7; gezinssteun: Hoofdstukken 7 & 8) als contextuele hulpbronnen (steun uit het netwerk: Hoofdstukken 2 & 7) opgenomen.

Over de verschillende hoofdstukken heen vonden we evidentie voor alle geïncludeerde hulpbronnen. Deze bevinding benadrukt het belang om zowel individuele, intrafamiliale als contextuele hulpbronnen op te nemen bij onderzoek naar de gevolgen van kinderkanker. Drie hulpbronnen bleken in het bijzonder van belang te zijn: cohesie/emotionele verbinding binnen het gezin, expressiviteit binnen het gezin en steun uit het netwerk. Met andere woorden, hoe meer gezinsleden zich verbonden voelen met en geliefd voelen door hun gezin, hoe meer ze kanker-gerelateerde gedachten en gevoelens kunnen delen binnen het gezin en hoe meer steun ze ervaren vanuit het netwerk, hoe beter de adaptatie na een diagnose kinderkanker. Daarnaast vonden we evidentie voor het belang van perceptie: hoe meer men de ziekte ervaart als hanteerbaar en hoe minder als oncontroleerbaar, hoe beter de individuele adaptatie van de verschillende gezinsleden.

Dit proefschrift benadrukt tenslotte de unieke positie van de broers/zussen binnen het gezin na een diagnose kinderkanker (zie Hoofdstukken 1, 4, 5, 7). Net zoals het zieke kind komt de wereld van de siblings op zijn kop te staan. Echter, anders dan het zieke kind, ervaren ze zichzelf vaak aan de zijlijn van het gezin en moeten ze hun negatieve gedachten en gevoelens vaker alleen verwerken. Bijgevolg verdienen deze broers/zussen meer aandacht in onderzoek en klinische praktijk, aangezien ook zij hulp (van ouders, hun netwerk én hulpverleners) nodig hebben om deze ziekte een plaats te geven.

Beperkingen en Suggesties voor Verder Onderzoek

De resultaten van dit proefschrift moeten geïnterpreteerd worden rekening houdend met een aantal beperkingen. Een eerste beperking betreft de steekproef. In onze studies namen voornamelijk blanke, Nederlandstalige, getrouwde gezinnen deel, komende uit de middenklasse. Aangezien voorgaand onderzoek heeft aangetoond dat een lage socio-economische status en éénoudergezinnen risicofactoren zijn voor ouder- en kind-adaptatie na een diagnose kinderkanker (Mulhern et al., 1989; Van Dongen-Melman et al., 1995), kunnen we veronderstellen dat onderzoek met een meer heterogene steekproef andere resultaten biedt. Ten tweede, alle kinderen in dit proefschrift kregen de diagnose leukemie of non-Hodgkin lymfoom. Kinderen met een andere kankerdiagnose en diens gezinnen kunnen andere ervaringen hebben (bv. kinderen met hersentumoren kunnen ernstige gedragswijzigingen vertonen). Ten derde, in de meeste hoofdstukken (met uitzondering van Hoofdstuk 2) werden cross-sectionele verbanden onderzocht. Hierdoor kon de temporele volgorde van de onderzochte verbanden niet getest worden en zijn omgekeerde verbanden eveneens mogelijk (bv. individuele maladaptatie voorspelt een slechter gezinsfunctioneren). Ten vierde, alle gebruikte gezinsvragenlijsten zijn populatie-vragenlijsten (d.i. ontwikkeld voor de algemene bevolking en niet specifiek voor deze context). Echter, deze instrumenten bevatten zwakke psychometrische eigenschappen en werden (nog) niet gevalideerd in medisch pediatrische steekproeven. Het gebruik van deze vragenlijsten kan dus leiden tot onstabiele resultaten of zelfs foutieve conclusies. Bovendien kan door het gebruik van algemene vragenlijsten de specificiteit van gezinnen die geconfronteerd worden met een ernstige, levensbedreigende pediatrische ziekte over het hoofd gezien worden. Immers, wat “dysfunctioneel” is in de algemene populatie (bv. zeer hoge cohesie), kan voor deze gezinnen op dat specifiek moment functioneel zijn.

Implicaties

Op basis van de hierboven beschreven bevindingen kunnen we enkele theoretische en klinische implicaties naar voor schuiven.

Theoretische Implicaties

Dit proefschrift bevestigt het dubbel ABCX model, met name het belang van *hulpbronnen* en *perceptie* in het voorspellen van individuele, gezins- en relationele adaptatie na een diagnose kinderkanker. Echter, op basis van dit proefschrift kunnen we ook enkele kritische reflecties formuleren. Ten eerste, wat momenteel ontbreekt – in huidig proefschrift en het dubbel ABCX model – is een objectieve maat voor de stressor. Ook al is deze stressor voor alle geïncludeerde gezinnen dezelfde (d.i. een diagnose kinderkanker), toch toont huidig onderzoek aan dat er grote verscheidenheid bestaat in de manier waarom deze stressor zich uit, in behandeling en in prognose (National Cancer Institute, nd.). Ten tweede, in lijn met andere theoretische modellen binnen de gezins- en stressliteratuur (o.a. Resilience Model of Family Stress, Adjustment and Adaptation; McCubbin & McCubbin, 1991) kan de integratie van het concept "kwetsbaarheid" nuttig zijn. Kwetsbaarheid kan gezien worden als het risico van een individu of gezin om fysisch of emotioneel geraakt te worden door onvoorspelbare gebeurtenissen en omvat de opeenstapeling van huidige en vroegere stressoren, lasten en veranderingen, alsook de eisen die hiermee gepaard gaan (Weber, 2011). Door dit concept toe te voegen aan de pre-crisis fase, kan de geschiedenis van een persoon en/of een gezin mee in acht genomen worden. Ten derde, één van de belangrijkste bijdragen van het dubbel ABCX model aan de huidige stress-modellen is de inclusie van het concept *coping* (Weber, 2011). Deze coping strategieën werden echter enkel als post-crisis variabelen geïncludeerd. In lijn met andere modellen binnen de gezins- en stressliteratuur (o.a. Family Adjustment and Adaptation Response model; Patterson, 1989) en verschillende empirische

studies (Barrera et al., 2004; Dahlquist et al., 1993), echter, weten we dat coping ook tijdens de pre-crisis fase van belang is. Ten vierde, dit proefschrift onderstreept het belang van gezinsfunctioneren als buffer voor individuele, gezins- en relationele maladaptatie. Dit idee is één van de basisprincipes van het Typology Model of Family Adjustment and Adaptation (McCubbin, 1995). Volgens dit model zijn bepaalde gezinstypes gerelateerd aan betere uitkomsten. Een gezinstype verklaart hoe gezinnen zich gebruikelijk gedragen en bestaat uit een samenspel van verschillende gezinskarakteristieken (bv. verbondenheid, flexibiliteit, steun) (McCubbin et al., 1988). Gegeven de evidentie van dit proefschrift omtrent de rol van gezinsfunctioneren in het voorspellen van adaptatie, en ondersteund door het Typology Model of Family Adjustment and Adaptation (McCubbin, 1995) achten wij een meer centrale plaats van dit concept binnen theoretische modellen als waardevol.

Klinische Implicaties

Dit proefschrift toont aan dat het leven van alle individuele gezinsleden, het gezin in zijn geheel en de partnerrelatie van de ouders beïnvloed wordt door de diagnose en behandeling van kinderkanker. Wanneer gezinnen in aanraking komen met kinderkanker, is een holistische benadering – bestaande uit individuele, koppel- en gezins-interventies – bijgevolg noodzakelijk om gezinnen optimaal te kunnen begeleiden. Daarnaast kunnen hulpverleners adaptatie van alle gezinsleden, het gezin in zijn geheel en de partnerrelatie van de ouders bevorderen door in te spelen op de bestaande en/of beschikbare *hulpbronnen.* Relevante hulpbronnen situeren zich op drie niveaus: op individueel, intrafamiliaal en contextueel niveau. Ten eerste, aangezien psychologische flexibiliteit (individuele hulpbron; Hoofdstuk 2) relevant bleek binnen dit proefschrift, kunnen gezinnen baat hebben bij interventies die gericht zijn op het bevorderen van de acceptatie van ongewenste, negatieve gevoelens en gedachten, zoals Acceptance and Commitment Therapy (Hayes et al., 2012). Ten tweede, gegeven de centrale

rol van het gezinsfunctioneren (intrafamiliale hulpbron; Hoofdstukken 5, 6, 7) binnen dit proefschrift, raden wij aan om het gezinsfunctioneren routinematig te screenen in deze populatie. Bij detectie van een disfunctioneel gezinsfunctioneren kan vervolgens gebruik gemaakt worden van empirisch ondersteunde gezinsinterventies (zie Rolland & Walsh, 2006 en Saltzman et al., 2013 voor enkele voorbeelden). Ten derde, aangezien dyadische coping (intrafamiliale hulpbron; Hoofdstukken 2 & 10) een rol speelt voor de individuele en relationele adaptatie van ouders van kinderen met kanker, is het belangrijk om *ook* relationele aspecten te bevragen en te begeleiden, naast de meer voor de hand liggende individuele en familiale aspecten. Bijvoorbeeld, ouders kunnen *als koppel* uitgenodigd worden door de hulpverlener, aangezien perspectieven van beide partners noodzakelijk zijn om bepaalde relationele aspecten, zoals dyadische coping, te kunnen begrijpen en te verbeteren indien nodig. Daarnaast kunnen sommige koppels baat hebben bij Couples Coping Enhancement Training (CCET; Widmer et al., 2005). Het doel van deze training is om de coping competenties van beide individuele partners te versterken door in te spelen op de dyadische communicatie en dyadische coping. Ten vierde, gezien het belang van sociale steun (contextuele hulpbron; Hoofdstukken 2 & 7) kunnen hulpverleners gezinnen helpen bij het in kaart brengen van het sociaal netwerk van het gezin, alsook bij het (leren) vragen van steun indien nodig. Tenslotte dienen hulpverleners rekening te houden met enkele belangrijke individuele kenmerken van de gezinsleden. Dit proefschrift toont bijvoorbeeld aan dat moeders kwetsbaarder zijn dan vaders (Hoofdstukken 3 & 10), en dat siblings minder positieve kanker-gerelateerde gevoelens ervaren dan de andere gezinsleden (Hoofdstuk 7). Interventies dienen dus niet enkel afgestemd te worden op de risicofactoren van het gezin, maar ook op de verschillende gezinsleden (patiënt vs. moeder vs. vader vs. broer/zus) én de individualiteit van elke persoon.

Conclusie

Kinderkanker is een ernstige, levensbedreigende ziekte die een grote impact heeft op het zieke kind, maar ook op zijn/haar gezinsleden (ouders, broers/zussen), het gezin in zijn geheel en de partnerrelatie van de ouders. Ondersteund door het dubbel ABCX model beoogt dit doctoraat inzicht te verwerven in de korte- en langetermijngevolgen van kinderkanker voor de individuele gezinsleden, de gezinnen en de koppels, alsook de hulpbronnen die gezinsleden/gezinnen/koppels kunnen helpen om zich aan te passen na diagnose. De resultaten van dit doctoraat tonen aan dat zowel hulpbronnen (individuele, intrafamiliale en contextuele hulpbronnen) en perceptie belangrijk zijn in het begrijpen van individuele, gezins- en relationale adaptatie na een diagnose kinderkanker. Meer specifiek, meer psychologische flexibiliteit (individuele hulpbron), meer adequate dyadische coping (intrafamiliale hulpbron), en meer netwerksteun (contextuele hulpbron) hangen samen met een betere gezinsadaptatie. Meer emotionele betrokkenheid binnen het gezin (intrafamiliale hulpbron), een duidelijkere gezinsstructuur (intrafamiliale hulpbron), meer gezinssteun (intrafamiliale hulpbron), meer netwerksteun (contextuele hulpbron) en de ziekte beoordelen als hanteerbaar in plaats van oncontroleerbaar, hangen samen met een betere individuele adaptatie; en meer adequate dyadische coping (intrafamiliale hulpbron) hangt samen met een betere individuele en relationele adaptatie van ouders na confrontatie met kanker bij hun kind. Ten gevolge raden we aan om de focus in psycho-oncologisch onderzoek en praktijk te verleggen van een beperkt individueel perspectief naar een meer holistische benadering, inclusief een individueel, intrafamiliaal en contextueel perspectief.

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SUPPLEMENTARY TABLES

Supplementary Table 1 – Chapter 1

Summary of design characteristics and findings of all reviewed studies included in chapter 1

Authors	Country of origin	Design	Sample Size: Families (individuals)[1]	Treatment Status[2]	Family Functioning Measure(s)[3]	Findings[4]
Ach et al., 2013	USA	Cross-sectional	*N* = 164 (153 mothers, 101 fathers); *N* = 164 controls (158 mothers, 92 fathers)	1 – 5 years (*M* = 3.1) post-treatment	FES	Mothers of children with cancer reported lower levels of family **support** than controls, but equal levels of family **conflict**. Fathers' reports of support and conflict were no different between the two groups.
Adduci et al., 2012	Italy	Cross-sectional	*N* = 64 (64 survivors, 64 parents)	≥ 1 year post-treatment	Qualitative interviews (with categorical coding)	31% of the families (*n* = 20) evidenced effective (complete, truthful, consistent, comprehensible, continuous, gradual, and personalized) **communication** patterns; 19% (*n* = 12) avoided communication; the rest were "ineffective."
Alderfer et al., 2009	USA	Cross-sectional	*N* = 150 (144 survivors,	1 – 12 years	FAD	47% of survivors, 25% of mothers and 30% of fathers reported poor family functioning across four or more FAD

			144 mothers, 104 fathers)	(*M* = 5.3) post-treatment		subscales; 35% of families fell in the 'unhealthy' range for **general family functioning** when using a family mean. 62% of survivors, 32 % of mothers and 37% of fathers reported 'unhealthy' **communication** patterns with 39% of families in this range when using the family mean.
Alderfer & Hodges, 2010	USA	Cross-sectional	*N* = 161 (161 siblings)	3.7 – 38 months (*M* = 16.7) post-diagnosis	CASSS	Per sibling report, parent **support** was less plentiful and important than support from friends and equal in amount and importance to support from teachers.
Arabiat et al., 2013	Jordan	Cross-sectional	*N* = 51 (51 mothers)	Hospitalized children with cancer	Qualitative interviews	Mothers (43%) indicated that family **cohesion** was strengthened by the illness. More than half (56%) described a state of disequilibrium in their families (**general family functioning**) and commented on a number of issues, such as trouble balancing multiple family needs
Barrera et al., 2007	USA	Longitudinal	*N* = 36 (36 survivors, 22 siblings)	pre HPCT, 6 months, 1 year, and 2 years post-HPCT	SSSC	Across time points, survivors generally reported more parental **support** than the normative values. Sibling reports of parental support (only assessed at the final time point) were no different from norms.
Beek et al., 2014	The Netherlands	Cross-sectional	*N* = 51 (45 survivors, 42 mothers, 8 fathers)	$\geq$ 2 years post-diagnosis, all off-treatment	FES	Survivors reported more **cohesion** and expressiveness (**communication**) and less **conflict** than norms. Parents reported less conflict. Parent-reported cohesion

						and expressiveness were no different from norms.
Beltrao et al., 2007	Brazil	Cross-sectional	*N* = 10 (10 mothers)	?	Qualitative interviews	The family is an important source of **support**.
Bjork et al., 2009	Sweden	Cross-sectional (from a longitudinal study)	*N* = 11 (4 patients, 9 mothers, 9 fathers, 4 siblings)	On-treatment	Qualitative interviews	Family life and **general family functioning** was disrupted. Attention was focused on the patient: only when his/her needs were satisfied could the focus move to other family members. Siblings reported feeling separate from the rest of the family.
Bjork et al., 2011	Sweden	Cross-sectional (from a longitudinal study)	*N* = 10 (4 patients, 10 mothers, 8 fathers, 2 siblings)	2 – 11 months post-treatment	Qualitative interviews	The cancer experience affected the entire family (**general family functioning**) and family members felt changed. Life was different than before.
Brody & Simmons, 2007	USA	Cross-sectional	*N* = 8 (8 fathers)	3 – 30 months (*M* = 18) post-diagnosis	Qualitative interviews	Fathers reported difficult changes in their family lives (**general family functioning**), that family **cohesion** was strengthened by the illness and that family **support** was important.
Brown et al., 2003	USA	Cross-sectional	*N* = 52 (52 survivors, 52 mothers); *N* = 42 controls (42 children, 42 mothers)	1 – 14 years (*M* = 5.8) post-treatment	FES, PSS-Fa	Survivors reported more social emotional **support** from their families than did controls; no differences emerged for survivors' reports of **conflict** or mothers' reports of support and conflict compared to controls.

Carlson-Green et al., 1995	USA	Cross-sectional	*N* = 63 (63 mothers)	1 – 123 months (*M* = 44) post-diagnosis	FES	Mothers' reports of **cohesion** were no different from norms.
Chesler et al., 1991	USA	Cross-sectional	*N* = 17 (21 siblings)	?	Qualitative interviews	Siblings indicated that family life (**general family functioning**) was disrupted, nearly all (95%) reported that family **cohesion** was strengthened by the illness, however, that increased closeness was not always perceived as being inclusive of the siblings (i.e., siblings felt overlooked and left out).
Clarke et al., 2005	UK	Cross-sectional	*N* = 55 (55 mothers)	~4 months post-diagnosis ("newly diagnosed")	Qualitative interviews	Four **communication** styles emerged across interviews: minimal information (40%); ambiguous information (19%); factual information (35%); and full information (7%).
Clarke et al., 2008	UK	Cross-sectional	*N* = 39 (39 mothers)	At least 5 years post-treatment	Qualitative interviews	69% (*n* = 27) of mothers reported open **communication** with their children regarding diagnosis and treatment; the rest provided minimal information.
Clarke-Steffen, 1997	USA	Longitudinal	*N* = 7 (6 patients, 7 mothers, 7 fathers, 12 siblings)	7-30 days post-diagnosis, 1 week after remission, 3 months later	Qualitative interviews	Although families revealed a loss of normal family life during treatment (**general family functioning**), most of them also reported increased **cohesion**.

Cohen et al.,1994	USA	Cross-sectional	*N* = 129 (125 mothers, 4 fathers)	0 – 4 years post-diagnosis	FACES II	The mean scores for **cohesion** and **adaptability** were no diferent from norms, however, a greater percentage of families of children with cancer scored within the enmeshed range (21% vs. 14%) than expected based upon norms. Percentages falling in the disengaged, rigid and chaotic categories were no different from that expected.
Cornman, 1993	USA	Cross-sectional	*N* = 20 (20 patients, 19 mothers, 19 fathers, 20 siblings)	At least 2 months into maintenance	FES, C-FES	Patients, parents and siblings reported significantly higher levels of **cohesion,** expressiveness (**communication**) and **conflict** compared to norms.
Enskär et al., 1997a	Sweden	Cross-sectional	*N* = 10 (10 patients)	?	Qualitative interviews	These teens reported that during the disease period **cohesion** within the family increased and that the greatest **support** they received came from their family, especially their mother
Enskär et al., 1997b	Sweden	Cross-sectional	*N* = 5 (5 patients, 4 mothers, 1 father)	0.9 – 4.5 years post-diagnosis	Qualitative interviews	Togetherness (**cohesion**) and **support** are important within the family.
Enskär et al., 1997c	Sweden	Cross-sectional	*N* = 15 (12 mothers, 4 fathers)	<1 to >5 years post-diagnosis	Qualitative interviews	The cancer governs the whole family's everyday life (**general family functioning**). Family is the best source of **support**.
Ferrell et al., 1994	USA	Cross-sectional	*N* = 21 (21 mothers; 10 fathers)	1 - 67 months	Qualitative interviews	Parents reported a struggle for normalcy and a disruption of family system (**general family functioning**) caused by cancer pain.

				(*M* = 17.4) post-diagnosis		
Fife et al., 1987	USA	Longitudinal	*N* = 34 (33 mothers, 27 fathers)	~10 days, 2, 4, 7, and 10 months post-diagnosis	FES	**General family functioning** (including cohesion, expressiveness and conflict) showed relative stability across the first year post-diagnosis.
Foley et al., 2000	USA	Cross-sectional	*N* = 29 (24 patients, 29 mothers, 21 fathers)	> 6 months post-treatment	FAD	Per survivor, mother and father report, **general family functioning** among these families of children with hypothalamic brain tumors was no different from non-clinical family norms.
Gerhardt et al., 2007	USA	Cross-sectional	*N* = 49 (48 mothers, 33 fathers); *N* = 49 controls (49 mothers, 29 fathers)	On treatment (*M* = 18 months post-diagnosis)	FES	There were no differences for mothers' or fathers' reports of family **support** between the two groups. Mothers reported significantly lower levels of **conflict** than the control group; fathers' reports of conflict were similar across the two groups.
Greenberg et al., 1989	USA	Cross-sectional	*N* = 138 (138 mothers); *N* = 92 controls (92 mothers)	5 – 16.3 years (*M* = 8.8) post-diagnosis	FES	Mothers' reports of **cohesion**, expressiveness (**communication**) and **conflict** within their families were no different across the two groups and were within 1 standard deviation of norms.
Greenberg & Meadows, 1992	USA	Cross-sectional	*N* = 118 (118 survivors; 120 parents)	Off treatment	Qualitative interviews	When asked about the impact of surviving cancer, 10% of survivors reported **general family functioning** difficulties. Parents reported that family

				(*M* = 8.8 years post-diagnosis)		**support** was important in the context of cancer.
Haluska et al., 2002	USA	Cross-sectional	*N* = 64 (64 patients); *N* = 115 controls	"varying stages in cancer treatment"	PSS-Fa	Patients reported greater parental **support** than healthy controls.
Havermans & Eiser, 1994	UK	Cross-sectional	*N* = 21 (21 siblings)	Off treatment ≤ 2 years	Qualitative interviews	In regard to **communication**, most were satisfied; 24% (*n* = 5) said they would have liked to know more and 5% (*n* = 1) said he had been told too much. Many (62%, *n* = 13) said that family **support** helped them cope.
Horwitz & Kazak, 1990	USA	Cross-sectional	*N* = 25 (25 mothers); plus *N* = 25 controls	6 – 41 months (*M* = 16.4) post-diagnosis	FACES II	A greater percentage of families of children with cancer fell in the extreme ranges for **adaptability**, than did the controls (56% vs. 20%). No differences were found for **cohesion**, either in mean scores or the percentages at the extremes.
Jackson et al., 2008	Australia	Longitudinal	*N* = 88 (53 mothers, 35 fathers)	Diagnosis, 6, 12 & 24 months post-diagnosis	Qualitative interviews; FACES II	In interviews at diagnosis, the majority of families reported that **support** mainly came from family. (FACES II scores are not reported.)
Kazak & Meadows, 1989	USA	Longitudinal	*N* = 35 (35 survivors, 35 mothers, 35 fathers; 25 of each at	> 5 years post-treatment (T2 was 6 months after T1)	FACES II, SSRS	Survivors', mothers' and fathers' ratings of **adaptability** and **cohesion** were not different from controls or norms with two exceptions: survivors' ratings of adaptability at T1 and mothers' ratings of adaptability at T2 were lower than controls (but not norms). Survivors'

			T2); *N* = 13 controls (9 each at T2)			ratings of family **support** were no different from controls at either time point
Kazak et al., 1994	USA	Longitudinal	*N* = 74 at T1 (74 survivors, 71 mothers, 58 fathers); *N* = 59 at T2 (individuals not reported)	> 5 years (*M* = 5.9) post-treatment (T2 was 1 year after T1)	FACES III, SSRS	Survivors', mothers' and fathers' ratings of **cohesion** and **adaptability** were no different from norms at either time point. Survivors rated s**upport** from family as being greater than support from any other source at both time points.
Kazak et al., 1997	USA	Cross-sectional	*N* = 130 (130 mothers, 96 fathers); *N* = 155 controls (148 mothers, 80 fathers)	≥ 1 years (*M* = 5.8) post-treatment	FACES IIIA	No differences were found between the groups for mothers' or fathers' reports of **general family functioning** or **communication**.
Koch, 1985	USA	Cross-sectional	*N* = 32 (26 mothers, 2 fathers, 33 siblings)	6 – 36 months post-diagnosis; on treatment	Qualitative interviews	22% (*n* = 7) of families indicated that family **cohesion** was strengthened by the illness. All attention and priority was focused on the patient.
Kronenberger et al., 1998	USA	Cross-sectional	*N* = 24 (24 mothers)	1 – 116 (*M* = 22) months post-diagnosis	FES	No significant differences were found between mothers' reports and standardized norms for ratings of family **support** or **conflict** during the pre-transplant period.

Kvist et al., 1991	Finland	Cross-sectional	*N* = 53 (53 patients; 52 parents)	> 1 month after ending chemo-therapy	Survey created for the purposes of this study	Parents (92%, 85%) and patients (68%, 40%) reported increased mother-patient and father-patient **cohesion** (respectively). The remainder reported no changes. Parents (39%) and patients (26%) reported that parent-sibling relationships became stronger. 11% of parents and 6% of patients reported that parent-sibling relationships became weaker. The rest reported no changes.
Kyngas et al., 2001	Finland	Cross-sectional	*N* = 14 (14 patients)	< 1 to > 5 years post-diagnosis; on- and off-treatment	Qualitative interviews	The family is an important source of **support**.
Long et al., 2013	USA	Cross-sectional	*N* = 209 (186 mothers, 70 fathers, 209 siblings)	1 – 38 months (*M* = 17.5) post-diagnosis	FAD	47% of siblings, 26% of mothers and 38% of fathers reported 'unhealthy' levels of **general family functioning.**
Madan-Swain et al., 1993	USA	Cross-sectional	*N* = 32 (19 patients, 32 siblings); *N* = 10 controls	2 – 24 months post-diagnosis	C-FES	Per patient and sibling report, no differences were observed between families of children with cancer and controls on measures of **general family functioning**.
Madan-Swain et al., 1994	USA	Cross-sectional	*N* = 25 (25 survivors, 25 mothers,	5 – 13 years (*M* = 8.4) post-treatment	FACES-III, IPAC	In regard to **adaptability**, mothers of children with cancer characterized their families as more rigid and less flexible than controls. No differences were found

			25 fathers); *N* = 16 controls (16 of each)			for mother-reported **cohesion** or survivor-reported adaptability and cohesion. Survivors, mothers, and fathers rated family **communication** similar to controls.
Manne et al., 1995	USA	Cross-sectional (data from a longitudinal study)	*N* = 59 (55 mothers, 4 fathers)	*M* = 51 days after diagnosis	FACES III	Parent reports of family **cohesion** and **adaptability** were similar to means reported in a normative sample.
Manne & Miller, 1998	USA	Cross-sectional	*N* = 50 (50 patients)	2 – 36 months (*M* = 6) post-diagnosis	NRI	Patients reported levels of **support** from mothers, fathers and siblings that were similar to those of a healthy comparison group. Patients reported more **conflict** with mothers and fathers than controls.
Martin et al., 2012	USA	Cross-sectional	*N* = 44 (19 mothers, 25 fathers)	$\geq$ 6 months (*M* = 42) post-diagnosis	FAD	20% (n = 9) of the parents reported "unhealthy" **general family functioning**.
McGrath et al., 2005	Australia	Cross-sectional (data from a longitudinal study)	*N* = 3 (3 patients, 3 mothers, 1 father)	2-3 months post-diagnosis	Qualitative interviews	The cancer diagnosis caused disruption of normalcy and daily family life (**general family functioning**). Family **support** made it easier for them to cope.
Morris et al., 1997	USA	Cross-sectional	*N* = 33 (33 parents); *N* = 32 controls (32 parents)	?	FES	Parents of children with cancer reported less **cohesion,** similar levels of expressiveness **(communication)** and more **conflict** than parents of healthy children.

Neil-Urban & Jones, 2002	USA	Cross-sectional	*N* = 10 (10 fathers)	?	Qualitative focus groups	Fathers reported that family **cohesion** was strengthened by the illness.
Nicholas et al., 2009	USA	Cross-sectional	*N* = 16 (16 fathers)	On-treatment	Qualitative interviews	Fathers indicated that family **cohesion** was strengthened by the illness, in particular the bond between parents and patient. They also remarked that family **support** is important and helpful.
Nichols, 1995	USA	Cross-sectional	*N* = 20 (20 patients)	?	NSSQ	All adolescents listed at least one parent in their social network. Satisfaction with family **support** was greater than that from any other source (i.e., other relatives, friends)
Noll et al., 1995 Study 1	USA	Cross-sectional	*N* = 25 (25 mothers, 20 fathers); *N* = 25 controls (25 mothers, 22 fathers)	*M* = 52 months post-diagnosis; on- and off-treatment	FES	No differences between groups or between the families of children with cancer and norms were found for mothers' or fathers' ratings of **general family functioning** (FRI), family **support** or **conflict**.
Noll et al., 1995 Study 2	USA	Cross-sectional	*N* = 42 (42 mothers, 33 fathers); *N* = 42 controls (42 mothers, 38 fathers)	*M* = 13 months post-diagnosis; on-treatment	FES	No differences between groups or between the families of children with cancer and norms were found for mothers' or fathers' ratings of **general family functioning** (FRI), family **support** or **conflict**.
Norberg & Steneby, 2009	Sweden	Cross-sectional	*N* = 7 (7 mothers, 4 fathers)	20 – 38 months	Qualitative interviews	Parents indicated that family **cohesion** was strengthened by the illness, especially the bond between parents and

				post-treatment		survivors; siblings were sometimes on the periphery.
Ozono et al., 2007	Japan	Cross-sectional	*N* = 89 (88 survivors, 87 mothers, 72 fathers)	≥ 5 years (*Md* = 10.8) post-diagnosis	FRI (short-form of FES)	41% of individuals reported good **general family functioning** (high cohesion, high expressive, low conflict), 46% reported moderate functioning, and 13% reported poor functioning. 26% of families had ≥ 1 member reporting poor functioning.
Patterson et al., 2004	USA	Cross-sectional	*N* = 26 (26 mothers; 19 fathers)	1 – 9 years (*M* = 4) post-treatment	Qualitativefocus groups	100% of the families (96% of parents) described family strain; 40% of the parents reported that they had trouble, during treatment, balancing multiple family needs and 22% reported a loss of a normal family life (**general family functioning**)
Pelcovitz et al., 1998	USA	Cross-sectional	*N* = 23 (23 patients) *N* = 23 controls	0 – 11 years (*M* = 3.3) post active treatment	FACES III	Survivors' ratings of family **cohesion** and **adaptability** were no different from those of healthy comparisons.
Perricone, 2012	Italy	Cross-sectional	*N* = 34 (34 mothers)	1 – 3 months (*M* = 1.6) post-diagnosis	FACES III	In regard to **adaptability**, mothers reported a tendency to function chaotically. (Findings regarding cohesion are reported inconsistently across results and discussion; no comparison to norms).
Peterson et al., 2012	USA	Cross-sectional	*N* = 135 (129 mothers, 80 fathers)	*M* = 11 years post-diagnosis	FAD	24% of mothers and 24% of fathers reported "unhealthy" **general family functioning.**

Prchal & Landolt, 2012	Switzerland	Cross-sectional	*N* = 7 (7 siblings)	8 – 23 months (*M* = 16.8) post-diagnosis	Qualitative interviews	Siblings reported disrupted family routines, being separated from the family due to treatment, and a general loss of family life (**general family functioning**); most (6 of 7) reported that family **cohesion** was strengthened by the illness. Regarding **communication**, siblings appreciated being well informed, though some were tired about hearing about cancer
Quin, 2004	Ireland	Cross-sectional	*N* = 77 (74 mothers, 46 fathers)	≥ 2 years post-treatment	Qualitative interviews	One third of the parents reported that pediatric cancer had a positive effect on **cohesion** (i.e., strengthened family bonds); 25% reported a negative effect (i.e., attention focused towards patient at detriment to siblings); the remainder reported no effect.
Rait et al., 1992	USA	Cross-sectional	*N* = 88 (88 survivors)	≥ 3 months (*M* = 37.4) post-treatment completion	FACES III	Survivors' reports of family **adaptability** were no different from norms. Survivors' reports of **cohesion**, however, were lower than norms; nearly 40% characterized their family as disengaged.
Ritchie, 2001	USA	Cross-sectional	*N* = 45 (45 patients)	On and off treatment	Qualitative interviews	All adolescents reported at least one parent as important providers of emotional **support**; 76% (*n* = 34) endorsed mothers and 49% (*n* = 22) endorsed fathers.
Rocha-Garcia, et al., 2003	Mexico	Cross-sectional	*N* = 51 (51 mothers, 51 fathers)	During first hospital stay post-diagnosis	Qualitative interviews	Most families (82%) reported that family **cohesion** was strengthened by the illness.

Rosenberg et al., 2014	USA	Cross-sectional	*N* = 96 (75 mothers, 17 fathers, 3 others)	≥ 6 months (*M* = 34) off-treatment	FACES II	Parents reported lower levels of **cohesion**, but higher levels of **adaptability** than norms.
Sargent et al., 1995	USA	Cross-sectional	*N* = 179 (254 siblings)	6 – 42 months post-diagnosis	Qualitative interviews	18% reported increased family separations and disruptions (**general family functioning**), 16% reported increased family **cohesion** when asked about biggest family change since diagnosis
Sawyer et al., 1986	Australia	Cross-sectional	*N* = 42 (42 patients, 42 parents, 56 siblings) *N* = 42 controls (42 children, 42 parents, 54 siblings)	≥ 2 years (*M* = 5.7) post-diagnosis	FCI (only completed by parents and children aged 10 and older)	No differences were found between families of children with cancer and the control group in regard to **general family functioning.**
Sawyer et al., 1997	Australia	Longitudinal	*N* = 38 (38 mothers); *N* = 39 controls (39 mothers)	At diagnosis (*M* = 5.3 weeks), 1, and 2 years post-diagnosis	FAD	No differences were found between mothers' report of **general family functioning** across the two groups or across time (also no interactions).
Sawyer et al., 2000	Australia	Longitudinal (extension of study above)	*N* = 38 (38 mothers); *N* = 39 controls (39 mothers)	Diagnosis, 1, 2, 3, and 4 years post-diagnosis	FAD	No differences were found between mothers' report of **general family functioning** across the two groups or across time (also no interactions).

Seaver et al., 1994	USA	Cross-sectional	*N* = 18 (18 parents)	4.75 – 18.6 (*M* = 9.33) years post-diagnosis	QRS	Family adjustment problems were common in this sample; 11% specifically indicating problems with family disharmony **(general family functioning)**.
Shortman et al., 2013	UK	Cross-sectional	*N* = 6 (6 mothers)	17 – 35 months (*M* = 27) post-diagnosis	Qualitative interviews	Mothers reported on the important of family **support,** but that family interactions were not always positive, and that **conflict** arose during treatment course.
Sidhu et al., 2005	Australia	Cross-sectional	*N* = 8 (8 mothers, 1 father)	On active treatment	Qualitative focus groups	Organizing family life around treatment was reported as challenging by all parents. Five of the 8 parents described having only limited time and energy and therefore difficulty trying to meet everybody's needs (**general family functioning**).
Sloper, 1996	UK	Cross-sectional	*N* = 98 (80 mothers, 40 fathers)	5 – 10 months (*M* = 6.6) post-diagnosis	Qualitative interviews	52% reported that separations and disruptions of family life had a negative impact on the family (**general family functioning**). 70% reported increased family **cohesion** was strengthened by the illness, 19% indicated no change and 11% felt that family members had drawn apart.
Sloper, 2000	UK	Longitudinal	T1: *N* = 94 (94 siblings); T2: *N* = 64 (64 siblings)	5 – 10 months (*M* = 6.6) post-diagnosis	Qualitative interviews	Half of the siblings reported increased family **cohesion** at each time point, though these were not necessarily the same siblings (some increased, some decreased over time). By the time of the interviews, most siblings (83%) felt that

				and 9 – 15 months (*M* = 12.4) later		they had received enough information about the illness and treatment. However, nearly two thirds said they had wanted more information in the early days after diagnosis (**communication**).
Streisand et al., 2003	USA	Cross-sectional	*N* = 116 (96 mothers, 20 fathers)	*M* = 38 (*Mdn* = 18) months post-diagnosis	FAD	No differences with norms were found for parent reports of **communication** and **general family functioning.**
Trask et al., 2003	UK	Cross-sectional	*N* = 28 (28 children, 28 mothers, 1 father)	$\geq$ 1 month (*M* = 18) post-diagnosis, $\leq$ 1 year post-treatment	FACES II, SSSC, CSI	Patients reported significantly greater levels of **cohesion** and **adaptability** within their families than standardized norms. Parents were viewed as the patient biggest source of **support**.
Varni et al., 1996	USA	Longitudinal	*N* = 62 (59 mothers, 3 fathers)	1 month, 6 months, 9 months post-diagnosis	FES	Parents reported greater levels **cohesion** and expressivity (**communication**), and equal levels of **conflict** compared to norms across time points (standard scores in this paper were statistically compared to norms by review authors).
Velasco et al., 1983	Mexico	Cross-sectional	*N* = 10 (10 patients, one or both parents)	? (in remission)	Qualitative interviews	The majority of the families (80 %) reported that family **cohesion** was strengthened by the illness, sometimes with a tendency toward enmeshment.
Wang & Martinson, 1996	Taiwan	Longitudinal	At T1: *N* = 45 (45 mothers, 45 fathers,	T1: $\geq$ 6 months on treatment;	Qualitative interviews, FES (completed by parents)	Most siblings (60%) claimed not to have a chance to talk about the illness with their parents or sick brother/sister (**communication**). (FES scores not

			45 siblings) At T2: *N* = 30 (30 of each)	T2: 12 months later		compared to norms or examined over time)
Ward-Smith et al., 2005	USA	Longitudinal	*N* = 9 (9 mothers)	1 month and 7 months post-diagnosis	Qualitative interviews	Mothers reported that having a child with cancer impacted the rest of the family and that they had difficulty juggling family responsibilities (**general family functioning**).
Wesley et al., 2013	USA	Cross-sectional	*N* = 102 (102 patients)	1 – 196 (*M* = 21) months post-diagnosis; on treatment)	PSS-Fa; FAD	Relative to normative scores, adolescents reported average levels of **general family functioning** and perceived family **support**.
Wiener et al., 2008	USA	Cross-sectional	*N* = 14 (14 siblings)	4 – 48 (*M* = 22) months post-donation	Qualitative interviews	36% (n = 5) of the siblings reported a closer relationship (increased **cohesion**) with the patient and/or other family members.
Woodgate & Degner, 2003	Canada	Longitudinal	*N* = 39 (39 complete families, i.e. parent(s), sibling, child with cancer)	On and off treatment	Qualitative interviews/ focus groups, observation	Most families reported that family **cohesion** was strengthened by the illness. The family was seen as the most important form of **support**.
Woodgate, 2006a	Canada	Longitudinal	*N* = 22 (30 siblings)	On and off treatment	Qualitative interviews/ focus groups, observation	Siblings reported a loss of a family way of life, how they and their family members related to one another and how their family functioned (**general family**

						functioning); most also reported that family **cohesion** was strengthened by the illness.
Woodgate, 2006b	Canada	Longitudinal	*N* = 15 (15 patients)	?	Qualitative interviews, focus groups, observation	Family was identified as the most important source of **support** throughout the cancer treatment trajectory.
Yonemoto et al., 2009	Japan	Cross-sectional	*N* = 30 (30 survivors)	60 – 368 months (*M* = 202) post-treatment	APGAR	No differences were found between survivors' reports of **general family functioning** and controls (control data from a different study).

Note. [1]Only the participants completing the family functioning measures are listed. [2]Time since diagnosis or time since the end of treatment, with means or medians, is listed if this information was available in the manuscript; if not available, other descriptors of the treatment status of the sample (i.e., on-treatment, off-treatment) are provided if available. [3]Only the measures assessing family functioning are listed; abbreviations are defined below. [4]Only findings relevant to this systematic review are reported here; the relevant domains of family functioning are provided in bold.

FES – Family Environment Scale; FAD – Family Assessment Device; CASSS – Child and Adolescent Social Support Scale; SSSC – Social Support Scale for Children; PSS-Fa – Perceived Social Support from Family; FACES II (also III, IIIa) – Family Adaptability and Cohesion Evaluation Scale II (III or IIIa); C-FES – Children's Version of the Family Environment Scale; SSRS – Social Support Rating Scale; IPAC – Inventory of Parent-Adolescent Communication; NRI – Network of Relationships Inventory; NSSQ – Norbeck Social Support Questionnaire; FRI – Family Relationship Index; FCI – Family Concept Inventory; QRS – Holroyd Questionnaire on Resources and Stress-Short Scale; CSI – Coping Strategies Inventory; APGAR – Adaptation, Partnership, Growth, Affection, Resolve index.

Supplementary Table 2 – Chapter 2

Models fit for the prospective analyses of the dependent variable General family impact

	Block 1: Control for initial status	Block 2: Adding variables of interest
Predictor	General family impact T2 (N = 111, 74 families) Coefficient B [CI]	General family impact T2 (N = 111, 74 families) Coefficient B [CI]
Variables of interest		
Psychological flexibility T1	-	-.16 [-.26, -.06]**
Stress communication T1	-	.03 [-.40, .46]
Supportive DC T1	-	.04 [-.30, .38]
Common DC T1	-	.24 [-.23, .71]
Negative DC T1	-	-.04 [-.43, .34]
Total network support T1	-	-.07 [-.23, .08]
Satisfaction with network support (too few vs. enough) T1	-	.94 [-1.27, 3.15]
Satisfaction with network support (too much vs. enough) T1	-	.43 [-1.83, 2.69]
Covariates		
Time since diagnosis	-.07 [-.14, .006]	-.07 [-.14, -.003]*
Age ill child	-.09 [-.33, .16]	-.07 [-.32, .18]
Diagnosis (AML vs. ALL)	.74 [-2.08, 3.55]	.80 [-2.02, 3.64]
Diagnosis (CML vs. ALL)	-3.78 [-11.27, 3.71]	-4.96 [-12.50, 2.58]
Diagnosis (Non Hodgkin vs. ALL)	.40 [-2.17, 2.98]	.30 [-2.27, 2.87]
Sex parent (women vs. men)	.91 [-.35, 2.17]	.55 [-.91, 2.01]
Age parent	.002 [-.17, .17]	-.01 [-.19, .16]
Family status (Divorced vs. Married)	2.69 [-.84, 6.21]	2.47 [-1.19, 6.12]
T2 minus T1	-.07 [-.14, .005]	-.06 [-.14, .01]
Outcome variables at previous time		
Financial impact T1	-	-
General family impact T1	.52 [.33, .70]***	.38 [.17, .59]***

Social impact T1	-	-
Satisfaction with internal family fit T1	-	-
Δ Deviance[1]	29.06***	15.61*

Note. ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia; [1]For the control model (block 1), the deviance is relative to the model with only covariates. For the prediction model (block 2) , the deviance is relative to the control model; * p < .05, ** p < .01, *** p < .001

Supplementary Table 3 – Chapter 2

Models fit for the prospective analyses of the dependent variable Financial impact.

	Block 1: Control for initial status	Block 2: Adding variables of interest
Predictor	Financial impact T2 (N = 111, 74 families) Coefficient B [CI]	Financial impact T2 (N = 111, 74 families) Coefficient B [CI]
Variables of interest		
Psychological flexibility T1	-	-.08 [-.13, -.03]**
Stress communication T1	-	-.26 [-.46, -.05]*
Supportive DC T1	-	.16 [-.02, .33]
Common DC T1	-	.11 [-.12, .35]
Negative DC T1	-	-.04 [-.23, .16]
Total network support T1	-	-.03 [-.10, .04]
Satisfaction with network support (too few vs. enough) T1	-	.25 [-.78, 1.29
Satisfaction with network support (too much vs. enough) T1	-	.44 [-.61, 1.49]
Covariates		
Time since diagnosis	-.004 [-.04, .03]	-.007 [-.04, .02]
Age ill child	.02 [-.09, .13]	.01 [-.10, .12]
Diagnosis (AML vs. ALL)	.16 [-1.09, 1.41]	.16 [-.1.02, 1.33]
Diagnosis (CML vs. ALL)	-2.80 [-6.04, .44]	-3.82 [-6.86, -.79]*

Diagnosis (Non Hodgkin vs. ALL)	-.58 [-1.73, .57]	-.52 [-1.58, .55]
Sex parent (women vs. men)	-.02 [-.72, .68]	.07 [-.73, .88]
Age parent	.02 [-.06, .10]	.03 [-.05, .11]
Family status (Divorced vs. Married)	.31 [-1.36, 1.97]	.91 [-.70, 2.51]
T2 minus T1	-.002 [-.04, .04]	-.01 [-.05, .03]
Outcome variables at previous time		
Financial impact T1	.62 [.43, .80]***	.57 [.39, .75]***
General family impact T1	-	-
Social impact T1	-	-
Satisfaction with internal family fit T1	-	-
Δ Deviance[1]	39.25***	24.83**

Note. ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia; [1]For the control model (block 1), the deviance is relative to the model with only covariates. For the prediction model (block 2) , the deviance is relative to the control model; * p < .05, ** p < .01, *** p < .001

Supplementary Table 4 – Chapter 2

Models fit for the prospective analysis of the dependent variable Social impact.

	Block 1: Control for initial status	Block 2: Adding variables of interest
Predictor	Social impact T2 (N = 111, 74 families) Coefficient B [CI]	Social impact T2 (N = 111, 74 families) Coefficient B [CI]
Variables of interest		
Psychological flexibility T1	-	-.10 [-.19, .001]
Stress communication T1	-	.01 [-.40, .43]
Supportive DC T1	-	.12 [-.20, .45]
Common DC T1	-	.31 [-.15, .77]
Negative DC T1	-	-.01 [-.38, .36]

Total network support T1	-	-.08 [-.23, .06]
Satisfaction with network support (too few vs. enough) T1	-	1.05 [-1.05, 3.15]
Satisfaction with network support (too much vs. enough) T1	-	1.03 [-1.13, 3.19]
Covariates		
Time since diagnosis	-.06 [-.13, .008]	-.06 [-.13, .01]
Age ill child	-.07 [-.31, .17]	-.05 [-.30, .20]
Diagnosis (AML vs. ALL)	2.03 [-.69, 4.75]	2.00 [-.80, 4.80]
Diagnosis (CML vs. ALL)	-5.14 [-12.45, 2.18]	-6.56 [-14.17, 1.04]
Diagnosis (Non Hodgkin vs. ALL)	-.31 [-2.81, 2.20]	-.63 [-3.19, 1.94]
Sex parent (women vs. men)	.14 [-1.11, 1.38]	.05 [-1.40, 1.51]
Age parent	-.005 [-.17, .16]	-.01 [-.18, .16]
Family status (Divorced vs. Married)	2.15 [-1.29, 5.58]	2.40 [-1.20, 6.00]
T2 minus T1	-.05 [-.13, .02]	-.04 [-.11, .04]
Outcome variables at previous time		
Financial impact T1	-	
General family impact T1	-	
Social impact T1	.41 [.21, .61]***	.34 [.14, .54]**
Satisfaction with internal family fit T1	-	
Δ Deviance[1]	15.81***	13.84

Note. ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia; [1]For the control model (block 1), the deviance is relative to the model with only covariates. For the prediction model (block 2) , the deviance is relative to the control model; * $p < .05$, ** $p < .01$, *** $p < .001$

Supplementary Table 5 – Chapter 2

Models fit for the prospective analysis of the dependent variable Satisfaction with internal family fit.

	Block 1: Control for initial status	Block 2: Adding variables of interest
Predictor	Satisfaction with internal family fit T2 (N = 109, 73 families) Coefficient B [CI]	Satisfaction with internal family fit T2 (N = 109, 73 families) Coefficient B [CI]
Variables of interest		
Psychological flexibility T1	-	.16 [-.07, .39]
Stress communication T1	-	-.15 [-1.08, .79]
Supportive DC T1	-	-.24 [-1.02, .54]
Common DC T1	-	.46 [-.59, 1.52]
Negative DC T1	-	-.39 [-1.25, .48]
Total network support T1	-	.21 [-.13, .54]
Satisfaction with network support (too few vs. enough) T1	-	-1.38 [-6.10, 3.34]
Satisfaction with network support (too much vs. enough) T1	-	.20 [-4.62, 5.02]
Covariates		
Time since diagnosis	.09 [-.06, .25]	.10 [-.06, .25]
Age ill child	.37 [-.13, .87]	.22 [-.31, .76]
Diagnosis (AML vs. ALL)	2.55 [-3.37, 8.47]	2.85 [-3.13, 8.82]
Diagnosis (CML vs. ALL)	12.64 [-3.51, 28.79]	17.80 [1.11, 34.49]*
Diagnosis (Non Hodgkin vs. ALL)	.39 [-4.98, 5.76]	.31 [-5.09, 5.72]
Sex parent (women vs. men)	.98 [-1.65, 3.61]	2.47 [-.79, 5.73]
Age parent	-.35 [-.70, .01]	-.24 [-.62, .14]
Family status (Divorced vs. Married)	-6.92 [-15.68, 1.84]	-7.43 [-16.45, 1.60]

T2 minus T1	.06 [-.10, .23]	.08 [-.10, .26]
Outcome variables at previous time		
Financial impact T1	-	-
General family impact T1	-	-
Social impact T1	-	-
Satisfaction with internal family fit T1	.50 [.29, .72]***	.30 [.01, .59]*
Δ Deviance[1]	21.25***	9.75

Note. ALL = Acute lymphoblastic leukemia, AML = Acute myeloid leukemia, CML = Chronic myeloid leukemia; [1]For the control model (block 1), the deviance is relative to the model with only covariates. For the prediction model (block 2) , the deviance is relative to the control model; * $p < .05$, ** $p < .01$, *** $p < .001$

Supplementary Table 6 – Chapter 5

Methodological characteristics and summary of results for studies identified for review and included in chapter 5

Authors and Scientific Merit Rating	*N* Families (individuals)	Age range of Children	Cancer Diagnoses and Time Frame	Family Construct(s), Measure(s) Used and Reporter	Child Outcome(s), Measure(s) Used and Reporter	Findings
Adduci et al., 2012 2.44	64 (64 survivors, 64 mothers; 64 fathers)	4-18 years (*M* = 9.5; *SD* = 3.4)	Brain tumors; more than 1 year post-treatment	Communication (Qualitative classification based on interview)	Problem Behavior (CBCL, one parent report)	- Families classified as displaying avoidant or ineffective **communication** had children with more Internalizing problems (*r* = -0.40, 95% CI: -0.60 – -0.19) - The groups did not differ significantly on Externalizing problems (*r* = -0.14, 95% CI: -0.39 – 0.10)
Alderfer & Hodges, 2010 2.00	161 (161 siblings, 145 mothers, 16 fathers) Focus: Sibling Adjustment	8-18 years (*M* = 12.6; *SD* = 2.9)	All diagnoses; 3 – 38 months post-diagnosis (*M* = 16.7, *SD* = 6.9)	Support (CASSS, sibling-report)	Problem Behavior, Social Competence (CBCL, parent report); Anxiety (RCMAS, sibling report);	Note: Bivariate associations calculated Greater **support** from parents was significantly related to: fewer child-reported depression symptoms (*r* = -0.31, 95% CI: -0.44 – -0.16); fewer externalizing problems (*r* = -0.22, 95% CI: -0.36 – -0.07); fewer total behavior problems (*r* = -0.21, 95% CI: -0.36 – -0.06) and greater social competence (*r* = 0.24, 95% CI: 0.09 – 0.38) Parental **support** was not significantly related to: child-reported anxiety (*r* = -

					Depression (CDI, sibling report); PTSS (CPSS, sibling report)	0.15, 95% CI: -0.30 – 0.01); PTSS (*r* = -0.12, 95% CI: -0.27 – 0.04), or internalizing problems (*r* = 0.09, 95% CI: -0.24 – 0.07)
Alderfer et al., 2009 2.67	150 (144 survivors, 144 mothers & 104 fathers)	11-19 years (*M* = 14.7; *SD* = 2.4)	All diagnoses; 1-12 years (*M* = 5.3) post-treatment	Communication, Affective Responsiveness, Affective Involvement, Problem Solving, Behavioral Control, Roles, General Family Functioning (FAD, survivor, mother, father report)	PTSS (SCID, child interview)	- **Communication** was not significantly associated with PTSS based on survivor (*r* = 0.14, 95% CI: -0.02 – 0.30) mother (*r* = 0.15, 95% CI: -0.01 – 0.31), or father (*r* = 0.02, 95% CI: -0.17 – 0.21) report - **Affective Responsiveness** was significantly associated with PTSS based on survivor (*r* = 0.24, 95% CI: 0.08 – 0.39) and mother (*r* = 0.22, 95% CI: 0.06 – 0.37), but not father (*r* = 0.11, 95% CI: -0.08 – 0.30) report - **Affective Involvement** was significantly associated with PTSS based on survivor (*r* = 0.28, 95% CI: 0.12 – 0.42), but not mother (*r* = 0.16; 95% CI: -0.004 – 0.32) or father (*r* = 0.14, 95% CI: -0.05 – 0.32) report - **Roles** was significantly associated with PTSS based on survivor (*r* = 0.18, 95% CI: 0.02 – 0.33) and mother (*r* = 0.26, 95% CI: 0.10 – 0.41), but not father (*r* = 0.05, 95% CI: -0.14 – 0.24) report

						- **Problem Solving** was significantly associated with PTSS based on survivor (*r* = 0.21, 95% CI: 0.05 – 0.36) and mother (*r* = 0.19, 95% CI: 0.03 – 0.34), but not father (*r* = 0.08, 95% CI: -0.11 – 0.27) report - **Behavioral Control** was not significantly associated with PTSS based on survivor (*r* = 0.04, 95% CI: -0.12 – 0.20), mother (*r* = -0.01, 95% CI: -0.17 – 0.15), or father (*r* = -0.05, 95% CI: -0.14 – 0.24) report - **General Family Functioning** was significantly associated with PTSS based on survivor (*r* = 0.22, 95% CI: 0.06 – 0.37) and mother (*r* = 0.18, 95% CI: 0.02 – .33), but not father (*r* = 0.08, 95% CI: -0.11 – 0.27) report Note: Bivariate correlations provided by author
Barakat et al., 2010 2.44	102 (102 patients, 102 parents)	13-19 years (*M* =15.8; *SD* =1.8)	All diagnoses; 1-193 months (*M* = 20.5, SD = 38.6) post diagnosis	Roles (FAD, patient and parent report)	Quality of Life (PedsQL, patient and parent report)	- Better defined **family roles** (patient report) were associated with better patient-reported psychosocial QOL (*r* = 0.27, 95% CI: 0.08 – 0.44) but were unrelated to physical QOL (*r* = 0.09, 95% CI: -.11 – .28) - Better defined **family roles** (parent report) were significantly correlated with better parent-reported patient psychosocial QOL (*r* = 0.32, 95% CI: 0.13 – 0.48) and

						physical QOL (r = 0.19, 95% CI: -.005 – .37) Note: Cross-informant bivariate associations were not examined; sign of correlations reversed to aid interpretation.
Barakat et al., 1997 2.22	309 (309 survivors, 309 mothers, 213 fathers); 219 controls (219 children, 211 mothers, 114 fathers)	8-20 years (M =13.5; SD = 3.4 for survivors, M = 12.3; SD = 2.7 for controls)	All diagnoses except brain tumors; > 1 year post-treatment (M = 5.9; SD = 3.5)	Cohesion, Adaptability (FACES IIIa, parent report);	PTSS (IES, TSC, PTSD-RI; child report); Anxiety (RCMAS, child report)	In regression equations including child age, gender, and race, cancer treatment intensity, years off treatment, child age at diagnosis, past perceived life threat, **mother reported cohesion, satisfaction and adaptability and mother social support resources, cohesion** (Beta = -0.02) and **adaptability** (Beta = 0.06) were not significant contributors to survivor PTSS (PTSD-RI score) Note: Bivariate associations unavailable
Barrera et al., 2009 2.33	99 at T1 (99 mothers); 49 at T2 (49 mothers); and 48 at T3 (48 mothers)	1-17 years (M = 8.3; SD = 4.4)	Cancer and blood disorders, not brain tumor; T1: pre-SCT, T2: 1 year post, T3: 2 years post	Cohesion (FACES III, mother report)	QOL (CHQ, POQOL; mother report)	- **Cohesion** was not significantly correlated with any of the global or subscale QOL scores (6 indicators; 2 of which were psychosocial) cross-sectionally at T1, T2 or T3 - **Cohesion** did not reach significance in regression equations predicting change in QOL from pre-SCT to 2 years post-SCT Note: Bivariate correlations not provided but estimation possible (r=0; one-tailed p = .50)

Beek, 2014 2.22	51 (45 patients, 42 mothers, 8 fathers)	12-18 years (*M* = 14.8; *SD* = 1.9)	Brain tumor; ≥ 2 years post-diagnosis, (*M* = 7.4, *SD* = 3.3); off-treatment	Cohesion, Expressiveness, Conflict, Organization, Control, Family Values, Social Orientation (FES, patient and parent report)	Problem behavior (CBCL, parent report; YSR, patient report)	MANOVAs were used to compare patients with behavior problems to those without behavior problems across the 7 **family functioning** scales: - no differences for patient-reported **family functioning** as a function of self-reported internalizing or externalizing problems - no difference for parent-reported **family functioning** as a function of parent-report internalizing or externalizing problems Note: Bivariate associations unavailable
Brown, Madan-Swain & Lambert, 2003 2.00	52 (52 survivors, 52 mothers); 42 controls (42 children without cancer; 42 mothers)	12 – 23 years (*M* = 17 years; *SD* = 3.44)	Leukemia & solid tumors; 1 – 14 years post-treatment (*M* = 5.8)	Conflict (FES, mother and child report); Support (FES, mother and child report; Perceived Social Support-Family, child report)	PTSS (PTSD-RI, child report)	- Survivor-reported **conflict** was not significantly correlated with survivor PTSS (*r* = 0.16, 95% CI: -0.12 – 0.42) - Survivor-reported **support** was not significantly correlated with survivor PTSS (FES-Support: *r* = -0.09, 95% CI: -0.38 – 0.16; PSS-Fa: *r* = -0.12, 95% CI: -0.38 – 0.16) Note: Cross-informant bivariate associations not reported
Bruce et al., 2010 2.39	52 (52 survivors, 46 mothers, 6 fathers)	8-16 years (*M* not reported)	Brain tumors; 0.5 – 7 years post-treatment (*M* not reported)	Conflict (PCIQ-R, parent and child report)	PTSS (IES-8, child report)	- More survivor-reported **conflict resolution skill** within the family was significantly associated with less survivor PTSS (*r* = -0.34, 95% CI: -0.56 – -0.07) Note: Cross-informant bivariate associations not reported

Carlson-Green et al., 1995 2.22	63 (63 patients; 63 mothers)	2-16 years (*M* = 7.0; *SD* = 4.1)	Brain tumors; T1: 1-123 months (*M* = 44) post-diagnosis; T2: 3 – 56 months later (*M* = 24; *SD* = 13.4)	Cohesion, Control (FES, parent report)	Problem behavior (CBCL, parent report)	- **Cohesion** at T1 was not significantly associated with total behavioral problems at T2 (*r* = -0.12, 95% CI: -0.36 – 0.13) - **Control** at T1 was not significantly associated with total behavioral problems at T2 (*r* = 0.14, 95% CI: -.11 – .38)
Cohen et al. 1994 2.11	129 (125 mothers, 4 fathers) Focus: Sibling Adjustment	4-16 years (*M* = 10.0; *SD* = 3.6)	Leukemia, lymphoma, solid tumors; 0-4 years post-diagnosis	Cohesion, Adaptability (FACES II, parent report)	Problem behavior; Social competence (CBCL, parent report)	Comparisons between **disengaged, connected and enmeshed families** revealed: - no differences for internalizing - the disengaged group (low cohesion) had higher externalizing and lower social competence scores than the enmeshed group (high cohesion) - the connected group (moderate cohesion) fell between disengaged and enmeshed groups for externalizing and social competence scores Comparisons between **rigid, flexible and chaotic groups** revealed: - no difference for internalizing - the rigid (low adaptability) and flexible (moderate adaptability) groups had higher externalizing and lower social competence scores than the chaotic (high adaptability) group

						- **rigid and flexible groups** were not significantly different
Dolgin et al., 1997 - Study 2 2.00	70 (70 parents) Focus: Sibling Adjustment	6 – 18 years (*M* = 12.2, *SD* = 3.8)	Leukemia, lymphoma, solid tumors; 14 – 42 months (*M* = 26.7) post-diagnosis	Support, Expressiveness, Conflict (Family Relations Scale, parent report)	Problem behavior (CBCL, parent report)	Note: Bivariate associations calculated - Greater **expressiveness** was associated with fewer Internalizing (*r* = -0.26, 95% CI: -0.47 – -0.03), Externalizing (*r* = -0.20, 95% CI: -0.42 – 0.04) and Total Problems (*r* = -0.22, 95% CI: -0.43 – 0.02) - Greater **support** was associated with fewer Internalizing (*r* = -0.59, 95% CI: -0.69 – -0.35), Externalizing (*r* = -0.54, 95% CI: -0.73 – -0.41) and Total Problems (*r* = -0.67, 95% CI: -0.78 – 0.52) - Greater **conflict** was associated with more Internalizing (*r* = 0.31, 95% CI: 0.08 – 0.51), Externalizing (*r* = 0.47, 95% CI: 0.26 – 0.64) and Total Problems (*r* = 0.47, 95% CI: 0.26 – 0.64)
Horwitz & Kazak, 1990 2.00	25 (25 mothers); 25 controls (25 mothers) Focus: Sibling Adjustment	3-5 years (*M* = 4.7; *SD* = 1.0)	All diagnoses; 6-41 months post-diagnosis (*M* = 16.4, *SD* = 8.5)	Cohesion, Adaptability (FACES II, parent report)	Problem behavior (CBCL, parent report)	Note: Signs reversed to aid interpretation - Greater **cohesion** was significantly associated with fewer total behavior problems (*r* = -0.65, 95% CI: -0.83 – -0.34), internalizing (*r* = -0.63, 95% CI: -0.82 – -0.31) and externalizing (*r* = -0.56, 95% CI: -0.78 – -0.21) symptoms - Greater **adaptability** was significantly associated with fewer total behavior problems (*r* = -0.41, 95% CI: -0.69 – -0.02) and internalizing symptoms (*r* = -0.49,

						95% CI: -0.74 – -0.12) ; externalizing results not reported
Houtzager et al., 2004 2.56	56 at T1 (83 siblings, 56 parents); 45 at T2 (66 siblings; 45 parents); 40 at T3 (60 siblings; 40 parents); 38 at T4 (57 siblings, 38 parents) Focus: Sibling Adjustment	7-19 years (*M* = 11.0; *SD* = 2.8) at T1	All diagnoses; 1, 6, 12 & 24 months post-diagnosis	Cohesion, Adaptability (FACES, sibling report)	Problem behavior (CBCL, parent report; YSR, child report); Anxiety (STAI-C, child report); Quality of life (DuCATQoL, child report); Emotional Reactions (SSERQ-s, child report)	When entered into multiple regression equations including child gender, age, cancer diagnosis, number of days hospitalized, death of child with cancer, coping, parent mental health, **cohesion and adaptability**: - greater **cohesion** was associated with more anxiety and insecurity but was unrelated to QOL, self- and parent-reported behavior problems, loneliness and positive emotions - greater **adaptability** was associated with more anxiety, poorer QOL, more self-reported behavioral problems, more insecurity and loneliness, but was unrelated to parent-reported behavioral problems and positive emotions Note: Bivariate associations unavailable
Jobe-Shields et al., (2009) 2.33	146 (146 patients, 146 parents)	6 – 18 years (*M* = 13.2, *SD* = 3.7)	Stem cell or Bone marrow transplant (85% had cancer diagnoses); At time of transplant	Cohesion, Expressiveness, Conflict (FES, parent report)	PTSS (PTSD-RI, child report)	- **Cohesion** (*r* = -0.12, 95% CI: -0.28 – 0.04), and **conflict** (*r* = .09, 95% CI: -0.74 – 0.25) were not significantly associated with child illness-related PTSS; however, greater **expressiveness** was related to less PTSS (*r* = -0.17, 95% CI: -0.32 – -0.01) - **Cohesion** and parental depression interacted such that cohesion only predicted child PTSS when parental depression was low

Kazak et al., 1997 2.56	130 (130 survivors, 130 mothers, 96 fathers),	8-19 years (*M* = 13.5; *SD* = 3.4)	Leukemia; > 1 year post-treatment (*M* = 5.8; *SD* = 3.1)	Communication, General Family Functioning (FACES IIIa, mother and father report)	Anxiety (RCMAS, child report); PTSS (PTSD Index, IES, TSC, child report)	- Neither **Communication** nor **General Family Functioning** as reported by either mothers or fathers were associated with survivor Anxiety or PTSS (three scales) Note: Bivariate correlations not provided, but estimation possible (*r*s = 0)
Kim & Yoo, 2010 2.06	74 (74 patients)	10-15 years (*M* = 13.1; *SD* = 2.2)	All diagnoses, no CNS involvement; .5 – 14 years (*M* = 4.2, *SD* = 3.8) post-diagnosis	Cohesion, Adaptability (FACES III, child report)	Resilience (Resilience Scale, child report)	- Greater **cohesion** was associated with more resilience (*r* = 0.51, 95% CI: 0.32 – 0.66) - Greater **adaptability** was associated with more resilience (*r* = 0.47, 95% CI: 0.27 – 0.63)
Long et al., 2013 2.39	209 (209 siblings, 186 mothers, 70 fathers) Focus: Sibling Adjustment	8-18 years (*M* = 12.5; *SD* = 2.7)	All diagnoses; 1-38 months post-diagnosis (M = 17.5, SD = 7.7)	General Family Functioning (FAD, sibling and parent report)	Depression (CDI, sibling report), Anxiety (RCMAS, sibling report), PTSS (CPSS, sibling report)	- Better **general family functioning** (child report) was significantly associated with fewer depression (*r* = -0.50, 95% CI:-.60 – -.39), anxiety (*r* = -0.33, 95% CI: -.45 – -.20) and post-traumatic stress symptoms (*r* = -0.39, 95% CI: -.50 – -.27) - **General family functioning** (mother report) was unrelated to child depression (*r* = -0.08, 95% CI: -.22 – .07), anxiety (*r* = -0.02, 95% CI: -.16 – .12) and post-traumatic stress symptoms (*r* = -0.01, 95% CI: -.15 – .13) - **General family functioning** (father report) was unrelated to child depression (*r* = -0.13, 95% CI: -.35 – .11), anxiety (*r* = -0.12, 95% CI: -.35 – .12) and post-

						traumatic stress symptoms (r = -0.11, 95% CI: -.34 – .13) Note: Bivariate correlations provided by author; signs reversed to aid interpretation
Maurice-Stam et al., 2007 2.22	106 (52 patients; 54 parents)	1-5 years and 8-15 years (M = 7.9, SD = 4.5)	All diagnoses; 2.0 – 29.7 months post-diagnosis (M = 13.7, SD = 8.2); all off treatment at least 2 months	Cohesion, Adaptability, (FACES, parent report), Communication (Exchange of Emotions Questionnaire, child report)	Quality of Life (TAPQOL, parent report; TACQOL, DUCATQoL, child report)	For 1-5 year olds (TAPQOL): - Greater **cohesion** was associated with more anxiety related to health status (r = 0.41, 95% CI: 0.15 – 0.62) but was unrelated to problem behavior related to health status (r = 0.10, 95% CI: -0.18 – 0.36) and motor problems (r = 0.12, 95% CI: -0.18 – 0.39) - **Adaptability** was not associated with anxiety related to health status (r = -0.10, 95% CI: -0.37 – 0.18), problem behavior related to health status (r = 0.11, 95% CI: -0.17 – 0.38) or motor problems (r = 0.12, 95% CI: -0.18 – 0.39) For 8-15 year olds (self-reported QOL): - **Cohesion** was not associated with overall QOL (r = 0.29, 95% CI: -0.00 – 0.53) - **Adaptability** was not associated with overall QOL (r = -0.24, 95% CI: -0.49 – 0.06) - Greater **expressiveness** was unrelated to overall QOL (M1: r = -0.15, 95% CI: -0.42 – 0.14; M2: r = 0.17, 95% CI: -0.12 – 0.43)

						Note: Bivariate correlations provided by author; sign reversed for TAPQOL to aid interpretation
Morris et al, 1997 2.11	65 (33 parents of children with cancer; 32 parents of controls)	Children with cancer: 2 – 16 years (*M* = 6.0); Controls: 2 – 11 years (*M* = 5.4 years)	ALL & children visiting pediatrician; Time frame from cancer diagnosis not reported	Cohesion, Expressiveness, Conflict (FES, parent report)	Problem behavior (CBCL, parent report)	For children with cancer: - Greater **cohesion** was associated with fewer internalizing problems (*r* = -0.37, 95% CI: -0.63 – -0.03) but was not associated with externalizing problems (*r* = -0.14, 95% CI: -0.46 – 0.21) - More **expressiveness** was associated with fewer internalizing (*r* = -0.34, 95% CI: -0.61 – 0.00) but was not associated with externalizing problems (*r* = -0.15, 95% CI: -0.47 – 0.20) - More **conflict** was associated with more externalizing (*r* = 0.42, 95% CI: 0.09 – 0.67) but was not associated with internalizing problems (*r* = 0.18, 95% CI: -0.17 – 0.49)
Newby, 2000 1.89	42 (42 mothers, 42 fathers)	6-18 years (*M* = 13.1; *SD* = 2.8)	All diagnoses except brain tumors; 2 – 17 years post-treatment (*M* = 6.8; *SD* = 3.2)	Cohesion, Expressiveness, Organization (FES, parent report)	Problem behavior (CBCL, parent report)	- More **cohesion** was associated with fewer total behavior problems (*r* = -.33, 95% CI: -0.58 – -0.03) - **Expressiveness** (*r* = .04, 95% CI: -0.27 – 0.34) and **Organization** (*r* = .14, 95% CI: -0.17 – 0.43) were not associated with total behavior problems
Ozono, 2010 2.17	89 (88 survivors, 87 mothers, 72 fathers)	12-20 years (*M* = 16.2; *SD* = 2.2)	All diagnoses except brain tumors; > 5 years post-diagnosis	General Family Functioning (FRI, child, mother and father report)	Anxiety (STAI, child report); Depression (CDI,	Three **family functioning clusters** were identified: **supportive** (high cohesion & expressiveness; low conflict); **intermediate** (moderate cohesion, expressiveness & conflict) and **conflictive**

			(*M* = 10.8; *SD* = 3.4)		child report); PTSS (IES-R, child report)	(low cohesion & expressiveness, high conflict) - Survivors in "**conflictive families**" reported more PTSS, more depressive symptoms and more anxiety than those in "**supportive families**." Note: Bivariate associations calculated
Ozono, 2007 2.22	89 (88 survivors, 87 mothers, 72 fathers)	12-20 years (*M* = 16.2; *SD* = 2.3)	All diagnoses except brain tumors; 5-19 years (M = 10.8) post-diagnosis	Communication, Roles, Problem Solving, Affective Involvement, Affective Responsiveness, Behavioral Control General Family Functioning (FAD; child report)	PTSS (IES-R, child report)	- Survivors with severe PTSS had families with **poorer roles and affective responsiveness** than survivors without PTSS - No differences between groups for **communication, problem solving, affective involvement, behavioral control** or **general family functioning** Note: Bivariate associations calculated/estimated
Pelcovitz, 1998 1.89	23 (23 patients, 23 mothers); 27 abused adolescents; 23 healthy, nonabused adolescents	14-23 years (*M* = 16; *SD* not reported)	Leukemia, lymphoma, carcinoma, Wilm's tumor; 0 – 11 years (*M* = 3.3) post active treatment	Cohesion, Adaptability (FACES III, child report)	PTSD (Structured Clinical Interview for DSM, child report)	Those meeting criteria for PTSD were compared to those not meeting criteria on **family functioning**: - **cohesion** results not reported - adolescents with PTSD saw their families as more **chaotic** (high in adaptability) than those without PTSD Note: Bivariate associations unavailable

Penn, 2009 2.17	35 (35 patients, 35 parents)	1-16 years (*M* = 9.1; *SD* not reported)	Brain tumors; 1 (0.8-5.0), 6 & 12 (11.2-18.7) months post-diagnosis	General family functioning (FAD, parent report)	Quality of life (PedsQL, parent and child report)	- **General Family Functioning** was not associated with parent-reported child QOL at T1 (*r* = 0.09, 95% CI: -0.27 – 0.43), T2 (*r* = -0.02, 95% CI: -0.37 – 0.33) or T3 (*r* = -0.13, 95% CI: -0.45 – 0.22) - **General Family Functioning** was not associated with child-reported QOL at T1 (*r* = 0.07, 95% CI: -0.35 – 0.47), T2 (*r* = 0.15, 95% CI: -0.26 – 0.52) or T3 (*r* = -0.20, 95% CI: -0.54 – 0.20) - **General Family Functioning** at T1 was not associated with parent-reported child QOL at T3 (*r* = 0.04, 95% CI: -0.31 – 0.38) or child-reported QOL at T3 (*r* = -0.07, 95% CI: -0.45 – 0.33)
Phipps & Mulhern, 1995 2.22	N = 34 – 41 families at T1 and 13 – 15 families at T2 (equal numbers of patients and parents)	4 – 16 years (M = 10.6, SD = 5.7)	SCT or BMT for oncological or hematologic diagnoses, not including brain tumors or severe combined immune deficiency; 1 week before transplant and 6 – 12 months (*M* = 8.2) post-transplant	Cohesion, Expressiveness, Conflict (FES, parent report)	Problem behavior, Social competence (CBCL, parent report); Anxiety, (Piers Harris Self-Concept Scale, child report)	Cross-sectional results pre-transplant: - Correlations between **cohesion** and internalizing (*r* = -0.27, 95% CI: -0.53 – 0.04), externalizing (*r* = -0.28, 95% CI: -0.54 – 0.03), total behavior problems (*r* = -0.20, 95% CI: -0.48 – 0.12), social competence (*r* = 0.09, 95% CI: -0.22 – 0.39) and child-reported anxiety (*r* = -0.16, 95% CI: -0.47 – 0.19) sign reversed to aid interpretation were non-significant. - Greater **expressiveness** was associated with fewer internalizing symptoms (*r* = -0.32, 95% CI: -0.57 – -0.01), but was unrelated to externalizing (*r* = -0.23, 95% CI: -0.50 – 0.08), total behavior problems (*r* = -0.23, 95% CI: -0.50 – 0.08), social

competence (r = 0.06, 95% CI: -0.25 – 0.36) and child-reported anxiety (r = -0.25, 95% CI: -0.47 – 0.19) sign reversed

- More **conflict** was associated with more internalizing (r = 0.49, 95% CI: 0.22 – 0.69), externalizing (r = 0.57, 95% CI: 0.32 – 0.75) and total behavior problems (r = 0.48, 95% CI: 0.20 – 0.69), but was not associated with social competence (r = 0.06, 95% CI: -0.25 – 0.36) or anxiety (r = 0.24, 95% CI: -0.11 – 0.54) sign reversed

Prospective results:

- **Cohesion** pre-transplant was not associated with internalizing (r = -0.45, 95% CI: -0.78 – 0.08), externalizing (r = -0.32, 95% CI: -0.72 – 0.23), or total behavior problems (r = -0.40, 95% CI: -0.76 – 0.14), but was associated with greater social competence (r = 0.60, 95% CI: -.13 – 0.85), and less anxiety post-transplant (r = -0.54, 95% CI: -0.84 – 0.02) sign reversed

- More **expressiveness** pre-transplant was associated with fewer internalizing (r = -0.71, 95% CI: -0.90 – -0.31) and externalizing problems (r = -0.57, 95% CI: -0.84 – -0.08), better social competence (r = 0.61, 95% CI: 0.14 – 0.86), and less child-reported anxiety post-transplant (r =

						-0.92, 95% CI: -0.98 – -0.75)$_{\text{sign reversed}}$; the correlation with total behavior problems did not reach statistical significance (*r* = -0.35, 95% CI: -0.73 – 0.20) - More **conflict** pre-transplant was associated with more internalizing (*r* = 0.73, 95% CI: 0.35 – 0.90) and total behavior problems (*r* = 0.60, 95% CI: 0.13 – 0.85), less social competence (*r* = -0.61, 95% CI: -0.86 – 0.14), and more child-reported anxiety post-transplant (*r* = 0.57, 95% CI: 0.03 – 0.85)$_{\text{sign reversed}}$; correlations with externalizing symptoms did not reach statistical significance (*r* = 0.44, 95% CI: -0.09 – 0.78)
Rait et al., 1992 2.0	88 (88 survivors)	12-19 years (*M* = 15.6; *SD* = 1.8)	Hematological malignancies; At least 3 months post-treatment (*M* = 37.4; SD = 29.7)	Cohesion / Adaptability (FACES III, child report)	Mental Health (Rand MHI, child report); Self-esteem (Rosenberg Self-Esteem Scale, child report); Problem behavior, Social competence (YSR, child report)	After controlling for sex, age, age at diagnosis and time since treatment, and when entering **cohesion** and **adaptability** together in regression equations: - More **cohesion** (Beta - .42) and less **adaptability** (Beta = -.27) were was associated with better mental health - More **cohesion** (Beta = .34) and less **adaptability** (Beta = -.31) were associated with better self-esteem - More **cohesion** was associated with fewer total behavioral problems (Beta = -.23); **adaptability** was non-significant (Beta = .11) - More **cohesion** was associate with better social competence (Beta = .26);

						adaptability was no-significant (Beta = .15) Note: Bivariate associations unavailable
Santos et al., 2015 2.56	389 (389 patients; 389 parents)	8-20 years (M = 13.3; SD = 3.5)	All diagnoses; 3-132 months (M = 28.2; SD = 27.0) post-diagnosis	Cohesion (FES, patient, parent report)	QOL (PedsQL Cancer Module, child report)	- Parent reports of **cohesion** were not significantly associated with child QOL (*r* = 0.04, 95% CI: -.06 – .14) - Child reports of greater **cohesion** were significantly associated with greater child QOL (*r* = 0.19, 95% CI: .09 – .28)
Sawyer, 1998 1.89	38 (38 mothers)	2-5 years (*M* =3.5; *SD* = 1.1)	All diagnoses except brain tumors; T1: at diagnosis (M = 5.3 weeks; SD = 3.5); T2: ~2 years post-diagnosis (details not reported)	General Family Functioning (FAD, parent report)	Problem behavior (CBCL, parent report)	Better **family functioning** at T1 was significantly associated with fewer Externalizing problems (*r* = -0.34, 95% CI: -0.60 – -0.02) and Total behavior problems (*r* = -0.37, 95% CI: -0.62 – -0.06) at T2, but was not associated with Internalizing problems (*r* = -0.24, 95% CI: -0.52 – -0.09) Note: Signs reversed to aid interpretation
Trask et al., 2003 2.44	28 (28 patients, 28 parents)	11-18 years (*M* = 13.6; *SD* = 1.9)	All diagnoses; 1 month post-diagnosis to 12 months post-treatment (*M* = 18 months post-diagnosis; *SD* = 20)	Cohesion, Adaptability (FACES II, child report) Support (SSSC, child report)	Problem behavior (YSR, child report)	**Cohesion, Adaptability** and **Support** were not significantly associated with internalizing or externalizing symptoms; correlations not provided Note: Bivariate correlations unavailable, but could be estimated (*r*s = 0)

Varni et al., 1996 2.44	62 at T1 (59 mothers, 3 fathers); 42 at T2; 47 at T3	5 – 13 years (*M* = 8.0, *SD* = 2.3)	All cancer diagnoses; 1 month, 6 months, and 9 months post-diagnosis	Cohesion, Expressiveness, Conflict (FES, parent report)	Problem behavior, Social competence (CBCL, parent report)	Cross-sectional Results: - Greater **cohesion** was associated with fewer internalizing problems at T1(*r* = -0.33, 95% CI: -0.54 – -0.09), T2 (*r* = -0.51, 95% CI: -0.71 – -0.24) and T3 (*r* = -0.30, 95% CI: -0.54 – -0.01); fewer externalizing problems at T1 (*r* = -0.50, 95% CI: -0.67 – -0.29) and T2 (*r* = -0.57, 95% CI: -0.75 – -0.32), but not T3 (*r* = -0.20, 95% CI: -0.46 – 0.09); and greater social competence at T1 (*r* = 0.36, 95% CI: 0.12 – 0.56) and T2 (*r* = 0.52, 95% CI: 0.26 – 0.71), but not T3 (*r* = 0.23, 95% CI: -0.06 – 0.49) - Greater **expressiveness** was associated with fewer internalizing problems at T1(*r* = -0.38, 95% CI: -0.58 – -0.14), T2 (*r* = -0.38, 95% CI: -0.61 – -0.09) and T3 (*r* = -0.24, 95% CI: -0.49 – 0.05); fewer externalizing problems at T1 (*r* = -0.34, 95% CI: -0.54 – -0.10), T2 (*r* = -0.44, 95% CI: -0.66 – -0.16), and T3 (*r* = -0.25, 95% CI: -0.50 – 0.04); and greater social competence at T1 (*r* = 0.24, 95% CI: -0.01 – 0.46), T2 (*r* = 0.49, 95% CI: 0.22 – 0.69), and T3 (*r* = 0.26, 95% CI: 0.08 – 0.59) - More **conflict** was associated with more internalizing problems at T1 (*r* = 0.21, 95% CI: -0.04 – 0.44), but not at T2 (*r* = 0.14, 95% CI: -0.17 – 0.43) or T3 (*r* = 0.06, 95% CI: -0.23 – 0.34); more externalizing problems at T1 (*r* = 0.23, 95% CI: -0.21 –

		0.45), but not at T2 (r = 0.21, 95% CI: -0.10 – 0.48) or T3 (r = 0.24, 95% CI: -0.07 – 0.51) and was not associated with social competence at T1 (r = -0.08, 95% CI: -0.32 – 0.17), T2 (r = 0, 95% CI: -0.30 – 0.30) or T3 (r = 0, 95% CI: -0.29 – 0.29). Prospective Results: - Greater **cohesion** at T1 was significantly associated with fewer internalizing problems at T2 (r = -0.35, 95% CI: -0.59 – -0.05) and T3 (r = -0.39, 95% CI: -0.62 – -0.11); externalizing problems at T2 (r = -0.56, 95% CI: -0.74 – 0.31) and T3 (r = -0.44, 95% CI: -0.65 – -0.17), and social competence at T2 (r = 0.39, 95% CI: 0.10 – 0.62) and T3 (r = 0.32, 95% CI: 0.03 – 0.56) - Greater **expressiveness** at T1 was significantly associated with fewer internalizing problems at T2 (r = -0.29, 95% CI: -0.55 – 0.02) and T3 (r = -0.40, 95% CI: -0.62 – -0.12) and fewer externalizing problems at T2 (r = -0.36, 95% CI: -0.60 – -0.06) and T3 (r = -0.32, 95% CI: -0.56 – -0.03); expressiveness at T1 was not significantly associated with social competence at T2 (r = 0.29, 95% CI: -0.02 – 0.55) but it was associated with greater social competence at T3 (r = 0.26, 95% CI: -0.04 – 0.52)

						- **Conflict** at T1 was not significantly associated with internalizing problems at T2 (r = 0.22, 95% CI: -0.09 – 0.49) or T3 (r = 0.08, 95% CI: -0.22 – 0.37); externalizing problems at T2 (r = 0.24, 95% CI: -0.07 – 0.51) or T3 (r = 0.14, 95% CI: -0.16 – 0.42), or social competence at T2 (r = -0.19, 95% CI: -0.47 – 0.12) or T3 (r = -0.24, 95% CI: -0.57 – 0.06)
Varni, 1994 2.33	30 (30 patients, 30 parents)	8-13 years (M = 10.7; SD = 1.7)	All diagnoses; 9 months post-diagnosis (range, etc not provided)	Support (SSSC-parents, child report)	Problem behavior (CBCL, parent report); Depression (CDI, child report); Anxiety (STAIC, child report); Self-esteem (SPPC, child report)	Greater **support** from parents was associated with fewer depressive symptoms (r = -0.34, 95% CI: -0.62 – 0.02) and externalizing problems (r = 0.32, 95% CI: -0.61 – 0.05), but was unrelated to internalizing problems (r = 0.01, 95% CI: -0.35 – 0.37), anxiety (r = -0.28, 95% CI: -0.58 – 0.09), and self-esteem (r = 0.25, 95% CI: -0.12 – 0.56)
Wang & Martinson, 1996 2.22	45 at Time 1 (90 parents, 45 siblings); 30 at Time 2 Focus: Siblings	7-16 years (M, SD not reported)	Leukemia, solid tumors; T1: > 6 months post-diagnosis, T2: 12 months later	Cohesion (FES, parent report)	Social competence (CBCL, parent report)	- Greater **cohesion** was associated with greater social competence at both T1 and T2 Note: Prospective associations were not investigated; bivariate correlations were not provided but could be estimated

Wesley, 2013 2.44	102 (102 patients)	13-19 years (*M* = 15.6; *SD* = 1.8)	All diagnoses; 1-196 months (*M* = 20.7, *SD* = 37.4) post-diagnosis and on treatment	Support (PSS-Fa, child report); General Family Functioning (FAD, child report)	Positive and Negative affect (PANAS, child report)	- Family **support** was not significantly correlated with positive (*r* = 0.10, 95% CI: -0.10 – 0.29) or negative (*r* = -0.09, 95% CI: -0.28 – 0.11) affect - Better **family functioning** was significantly correlated with more positive affect (*r* = 0.17, 95% CI: -0.03 – 0.35); but was unrelated to negative affect (*r* = -0.12, 95% CI: -0.31 – 0.08)$_{\text{sign reversed for interpretation}}$
Yonemoto, 2009 1.39	30 (30 survivors)	$\geq$ 20 years (*M*, *SD* not reported)	Osteosarcoma; 5-30 years (*M* = 16.8) post-treatment	General Family Functioning (APGAR, child report)	PTSS (IES-R, child report); Post-traumatic growth (PTGI, child report)	Better **general family functioning** was significantly correlated with less survivor PTSS (r = -.50, 95% CI: -.73 – -.17), but was unrelated to post-traumatic growth (r = .30, 95% CI: -.07 – .60)

Note. The columns shaded did not provide data for meta-analysis

Supplementary Table 7 – Chapter 9

Summary of design characteristics and findings of all reviewed studies included in chapter 9

Authors	Country of origin	Design	Sample size: Families (individuals)[1]	Marital Status (couples)	Treatment Status[2]	Couple Functioning Measure(s)[3]	Findings[4]

Barbarin, Hughes & Chesler, 1985	USA	Cross-sectional	N = 32 (32 female partners, 32 male partners)	$N_{married}$ = 32	On-treatment	Qualitative interviews & self-developed questionnaires (marital quality, social support)	-60% of the participants indicated an increase in **couple connectedness** since diagnosis, 34% reported no change, 5% reported a decrease in couple connectedness -65% of the participants described their partner as an important source of **support**; 14% as not helpful at all -72% of the participants reported that the marital relationship presented no problem during treatment; 5% indicated **marital adjustment** to be a major concern -Wives' and husbands' ratings of **marital quality** agreed closely with one another
Beltrao et al., 2007	Brazil	Cross-sectional	N = 10 (10 mothers)	$N_{married}$ = 4; $N_{cohabiting}$ = 4; $N_{divorced}$ = 2	?	Qualitative interviews	Most female participants indicated that couple **closeness** was strengthened by the illness, and that marital relations are an important source of

							(emotional and practical) **support**
Brody & Simmons, 2007	USA	Cross-sectional	N = 8 (8 fathers)	?	3 – 30 months (M = 18) post-diagnosis	Qualitative interviews	The relationships with their wives was **strengthened** during the illness process
Brown et al., 1992	Georgia	Cross-sectional	N = 55 (55 parents)	$N_{married/cohabiting}$ = 32; $N_{divorced/seperated}$ = 18; N_{single} = 5	New diagnoses, 1 year post-diagnosis, 1 year after treatment-completion	MAT	Off-therapy group (T3) reported more **marital satisfaction** than did parents of children in the newly diagnosed (T1) or 1 year post-diagnosis groups (T2)
Cornman, 1993	USA	Cross-sectional	N = 20 (19 mothers, 18 fathers)	$N_{married}$ = 18; $N_{divorced}$ = 2	At least 2 months into maintenance	DAS	The marital dyads were significantly less well-adjusted than the married norms for **dyadic adjustment**, yet they were significantly more adjusted than the comparative divorced sample -No significant differences between reported **marital adjustment** by wives and husbands

Dahlquist et al., 1996	Texas	Longitudinal	N = 42 (42 mothers, 42 fathers)	$N_{married}$ = 42	2 months & 20 months post-diagnosis	DAS	19% of the female participants and 24% of the male participants showed clinically elevated **marital distress** scores 20 months post-diagnosis No changes on **marital distress** across time
Dahlquist et al., 1993	USA	Cross-sectional	N = 67 (67 mothers, 67 fathers)	$N_{married}$ = 67	6-10 weeks post-diagnosis	DAS	25% of the female participants and 28% of the showed clinically elevated **marital distress** scores at diagnosis
Enskär et al., 1997	Sweden	Cross-sectional	N = 15 (12 mothers, 4 fathers)	$N_{married/cohabiting}$ = 6; $N_{divorced}$ = 4; N_{single} = 2, $N_{stepfamily}$ = 3	<1 to > 5 year post-diagnosis	Qualitative interviews	**Couple closeness** changed during the illness trajectory, with spouses sticking together during strenuous periods and not having the strength to stick together in more restful periods -Best **support** was given by the spouse -Participants found it nearly impossible to have any **privacy** and **integrity** -Some got **divorced** during disease, but they were not

							sure if the child's disease had been the cause
Ferrell et al., 1994	USA	Cross-sectional	N =21 (21 mothers, 10 fathers)	?	1 – 67 months (*M* = 17.4 months)	Qualitative interviews	Cancer diagnosis had a negative impact on the **sexual relationship** of the parents: no time together for parents, lack of privacy because the sick child moved into parents' bedroom or because the sick child demanded constant attention and care
Fife et al., 1987	USA	Longitudinal	N = 34 (33 mothers, 27 fathers)	$N_{married}$ = 23; $N_{divorced}$ = 5; N_{single} = 6	10 days, 2, 4, 7, and 10 months post-diagnosis	MAT	The level of **marital satisfaction**, for both female and male participants, were considerably lower than scores for well-adjusted couples, but above the mean for couples seeking therapy
Fletcher & Clarke, 2003	Canada	Cross-sectional	N = 25 (25 mothers)	?	Post-treatment and < 5 years post-diagnosis	Qualitative interviews	Not all **marital relationships** were in jeopardy due to diagnosis and treatment

Greenberg & Meadows, 1992	USA	Cross-sectional	N = 118 (120 parents)	$N_{married/cohabiting}$ = 120	Off-treatment (*M* = 8.8 years)	Qualitative interviews	-25% of the participants reported **marital conflict** -The spouse was the most important source of **support** -Participants reported a negative impact of the illness on the **sexual relationship**, with worry being the main reason, not the lack of love -According their **general marital adjustment**, 23% of the participants reported a positive change, whereas 25% reported marital difficulties, with 46% of those reporting marital difficulties still being married, 33% being divorced as a direct result of the child's illness and 21% being **divorced** due to problems prior to the illness
Hoekstra-Weebers, Jaspers et al., 1998	The Netherlands	Longitudinal	N = 62 (62 mothers, 62 fathers)	$N_{married}$ = 61	Diagnosis, 6 months post-diagnosis,12 months	MMQ	Significant increase in reported levels of **marital dissatisfaction** over time for both female and male participants -The level of **marital**

					post-diagnosis		**satisfaction** was comparable with a comparison group one year post-diagnosis, but lower than couples who were referred for treatment of their marital problems -No significant differences between male and female partners regarding their reported levels of **marital dissatisfaction**
Khoury et al., 2013	Lebanon	Cross-sectional	N = 12 (10 mothers; 2 fathers)	$N_{married}$ = 11; $N_{divorced}$ = 1	3 months – 6 year post-diagnosis	Qualitative interviews & observation field notes	Different experiences among couples: some reported an increase in **couple connectedness**; others become very **nervous** and started **shouting** at each other
Lahteenmaki et al., 2004	Finland	Longitudinal	N = 26 (24 mothers, 19 fathers at T1; 19 mothers, 15 fathers at T2) N = 46 controls (46	$N_{married}$ = 14; $N_{cohabiting}$ = 5; $N_{divorced}$ = 4	3 months post-diagnosis, 12 months post-diagnosis	Self-developed questionnaires	-Only 8% of female participants and 5% of male participants experienced regularly **difficulties** with their partner within three months after diagnosis. Nine months later, 21% of the female participants and 7% of the male participants reported regular

			mothers, 24 fathers)				difficulties, a non-significant difference across time -The level of **marital satisfaction** was comparable with a population based control group
Lansky et al., 1978	USA	Cross-sectional	N = 191 (191 parents)	$N_{married}$ = 180; $N_{divorced}$ = 8	On- and off-treatment	MMPI	-The yearly **divorce rate** of families with pediatric cancer did not differ from the rate in the general population -Parents of children with cancer experienced more **marital distress** than a norm group, but less than whose marital problems tend them to participate in marital counseling, with marital distress hardly ever resulting in divorce
Lavee, 2005	Israel	Cross-sectional	N = 35 (35 mothers, 35 fathers)	$N_{married}$ = 35	1 – 7 years post-diagnosis (M = 3.5)	ENRICH	Both positive and negative changes (caused by cancer experience) in **marital quality** emerged -Marital partners tend to have a similar perception of change in their **marital**

							quality after pediatric cancer diagnosis
Lavee & Mey-Dan, 2003	Israel	Cross-sectional	35 (35 mothers, 35 fathers)	$N_{married}$ = 35	1 – 7 years post diagnosis (M = 3.5; SD = 2.2)	ENRICH	-Participants reported improved **conflict resolution** between spouses (both reported by female participants and male participants) -29% of the couples reported positive changes in their **communication patterns**, 20% reported no changes and none reported negative changes -Negative effect of child's illness on the **sexual relationship**: in 45.7% both spouses reported a deterioration in their sexual relationship and only 1 couple agreed that their sexual relationship had actually improved -Regarding their **general marital adjustment**: deterioration during the first year and in cases of long-term illness, but a positive change among participants whose children had been ill for two or three

							years -Husbands tended to perceive a more positive **change** in relationship than did wives
Manne et al., 2003	USA	Longitudinal	N = 148 (148 mothers)	$N_{married}$ = 135; $N_{cohabiting}$ = 13	HSCT, 3 months and 6 months post-HSCT	Perceived partner criticism and avoidance	Descriptive information regarding perceived partner **criticism** and **avoidance** -**Criticism** T1: M=18.25; SD = 7.80 T2: 18.17; SD = 7.32 T3: 18.21; SD = 7.90 -**Avoidance** T1: 5.32; SD = 2.51 T2: 5.13; SD = 2.24 T3: 5.16; SD = 2.17 -No norms / no further information
McGrath, 2001	Australia	Cross-sectional	N= 12 (12 mothers, 4 fathers)	$N_{married/cohabiting}$ = 12	On-treatment	Qualitative interviews	-Majority (N=14) talked about the importance of **support** they were receiving from their partner (both emotional and practical support) -Not all parents were capable of proving the extent of emotional

							support needed for such a difficult situation
Mercer & Ritchie, 1997	Canada	Cross-sectional	N = 20 (? mothers, ? fathers)	$N_{married/cohabiting}$ = 17; $N_{separated}$ = 1; N_{single} = 2	?	Qualitative interview	Spouse is the most frequently reported source of **support**
Morrow et al., 1982	USA	Cross-sectional	N = 107 (65 mothers, 42 fathers)	$N_{married}$ = 102; $N_{divorced}$ = 5	On-treatment and off-treatment	Rating 11 potential sources of social support	Spouse is one of the most frequently reported sources of **support** -**Support** from spouse was one of the greatest and most helpful sources of support
Patistea et al., 2000	Greece	Cross-sectional	N = 42 (41 mothers; 30 fathers)	$N_{married}$ = 41	0-3 months post-diagnosis	Qualitative interviews	-45% of the participants reported an increase in **marital closeness**, whereas 17% reported no change and 38% reported **marital conflict** -For 2/3 of the parents, the spouses was the most important source of emotional and practical **support**. However, in the case of 13 parents, the spouses were so disturbed by the diagnosis that they could not provide much effective help

							-23 couples agreed regarding the impact of cancer on the **quality of their marital relationship**
Patterson et al., 2004	USA	Cross-sectional	N = 26 (26 mothers; 19 fathers)	$N_{married}$ = 23; N_{single} = 3	1 – 9 years (M = 4) post-treatment	Qualitative focus groups	Although the partner is an important source of **support**, participants also acknowledged **conflict**
Penn et al., 2009	UK	Longitudinal	N = 35 (35 'parents')	?	1, 6 and 12 months post-diagnosis	FSS	Perceived marital **support** was consistent across time from diagnosis until 1 year post-diagnosis
Shapiro & Shumaler, 1987	California	Cross-sectional	N = 26 (26 mothers, 26 fathers)	?	6 months – 2 years post-diagnosis	Qualitative interviews & self-developed questionnaires	Male participants reported more **marital dissatisfaction** than did their partner did
Tremolada et al., 2012	Italy	Longitudinal	N = 94 (94 mothers)	?	1, 6, 12 months post-diagnosis	EFI-Cancer interview	**Couple connectedness** increased significantly from diagnosis to one year post-diagnosis
Wijnberg-Williams, Van de Hiel et al., 2015	The Netherlands	Longitudinal	N = 98 (85 mothers, 79 fathers at T1; 58 mothers, 57 fathers at T2)	$N_{married}$ = 158 individuals $N_{widowed}$ = 1; $N_{divorced}$ = 5	0-14 days post-diagnosis, 5 years post-diagnosis	MMQ CSI	-No significant difference at diagnosis regarding the amount of avoidance, incongruent and destructive **communication** between

			female participants and male participants. Female partners did experience less mutual understanding and sharing in their relationship than the male partner -Significant increase in **marital dissatisfaction** from diagnosis to 5 years later in female participants. Change in male participants was not significant -No differences in **marital dissatisfaction** between parents of whom the children had survived (86 parents), relapsed (8 parents) or were deceased (21 parents) -When compared with population based controls, no significant differences in **marital dissatisfaction** between parents of children with cancer and the comparison group of men and women at diagnosis and five years later

							-No significant differences in **marital dissatisfaction** between female participants and male participants at diagnosis and 5 years later
Wills, 1999	China	Longitudinal	N =9 (9 mothers, 8 fathers)	$N_{married}$ = 9	0-4 weeks, 4 months post diagnosis	Qualitative interviews	-Some participants reported an increase in **conflict** frequency; while others reported no changes or even having fewer arguments, as they channeled all their energy into caring for the sick child and had no time to argue -All but two female participants stated that their spouses were the main source of **support** -Male participants stated that they wanted to receive more **support** from their wives
Wills, 2009	China	Longitudinal	N = 8 (8 fathers)	$N_{married}$ = 8	1 and 4 months post diagnosis	Qualitative interviews	-All male participants unanimously identified their wives as most supportive person. However, some male participants also stated that

							they would like to have received more **support** from their wives
Wittrock et al., 1994	USA	Cross-sectional	N = 17 (17 mothers, 17 fathers) Control group = 32 (32 mothers, 32 fathers)	$N_{married}$ = 17	On-treatment and off-treatment	DAS	-No differences between parents of children with cancer and parents with healthy children regarding **general marital adjustment**
Yeh, 2002	Taiwan	Cross-sectional	N = 164 (164 mothers, 164 fathers)	$N_{married}$ = 157; $N_{divorced}$ = 7	0.08 – 9.42 years post-diagnosis (*M* = 1.88 year)	MSS	-Female and male participants whose children had been diagnosed within the past 2 months reported greater **marital dissatisfaction** than parents whose children were in off-treatment groups -No differences in **marital dissatisfaction** between new diagnosed group and relapsed group -Female participants reported less **marital satisfaction** than the male participants did

Notes. [1]Only the participants completing the couple functioning measures are listed. [2]Time since diagnosis or time since the end of treatment, with means or medians, is listed if this information was available in the manuscript; if not available, other descriptors of the treatment status of the sample (i.e., on-treatment, off-treatment) are provided if available. [3]Only the measures assessing couple functioning are listed; abbreviations are defined below. [4]Only findings relevant to this systematic review are reported here; the relevant domains of couple functioning are provided in bold.

MAT – Locke Wallance Marital Adjustment Questionnaire; DAS – Dyadic Adjustment Scale; MMQ – Maudsley Marital Questionnaire; MMPI – Minnesota Multiphasic Personality Inventory; ENRICH – ENRICH; PERCEIVED PARTNER CRITISISM AND AVOIDANCE – Scale adapted from the Cancer Support Inventory; FSS – Family Support Scale; CSI – Communication Skills Inventory; CSI – Couples Satisfaction Index; MSS – Marital Satisfaction Scale

Supplementary Table 8 – Chapter 10

APIM-analyses

		SDC	CDC	NDC
PIP_F				
Actor Effect	M → M	-0.467 (0.940)	-2.305 (1.105) *	0.732 (0.977)
	F → F	-0.302 (0.693)	-0.582 (0.882)	2.175 (0.966) *
	D	-0.165 (1.174)	-1.723 (1.471)	-1.443 (1.412)
Partner Effect	F → M	1.081 (0.653)	1.208 (0.865)	-0.598 (0.943)
	M → F	-1.641 (0.998)	-1.858 (1.127)	-0.153 (1.001)
	D	2.722 (1.198) *	3.066 (1.477) *	-0.444 (1.413)
Time Effect	M	-0.904 (0.326) **	-0.910 (0.346) **	-0.952 (0.337) **
	F	-1.981 (0.346) ***	-2.074 (0.353) ***	-1.982 (0.345) ***
PIP_D				
Actor Effect	M → M	0.376 (0.828)	-2.583 (0.886) **	1.145 (0.832)
	F → F	0.259 (0.649)	0.821 (0.851)	0.337 (0.975)
	D	0.117 (1.064)	-3.403 (1.320) **	0.808 (1.341)
Partner Effect	F → M	-0.183 (0.575)	1.156 (0.694)	0.182 (0.803)
	M → F	-3.074 (0.934) **	-3.142 (1.088) **	1.745 (1.011)
	D	2.890 (1.108) **	4.297 (1.377) **	-1.563 (1.350)
Time Effect	M	-0.054 (0.287)	-0.105 (0.278)	-0.187 (0.287)
	F	-0.957 (0.324) **	-0.883 (0.341)**	-0.930 (0.349) **
DASS DEPRESSION				
Actor Effect	M → M	0.047 (0.242)	-0.392 (0.265)	0.504 (0.237) *
	F → F	0.121 (0.189)	0.118 (0.259)	0.368 (0.284)
	D	-0.074 (0.308)	-0.511 (0.383)	0.135 (0.377)
Partner Effect	F → M	-0.104 (0.168)	-0.214 (0.207)	0.065 (0.229)
	M → F	-0.818 (0.272) **	-0.467 (0.331)	0.118 (0.294)
	D	0.714 (0.321) *	0.253 (0.403)	-0.053 (0.380)

Time Effect	M	0.018 (0.084)	-0.043 (0.083)	-0.030 (0.082)
	F	-0.268 (0.094) **	-0.234 (0.104) *	-0.252 (0.102) *
DASS ANXIETY				
Actor Effect	M → M	-0.018 (0.123)	-0.062 (0.139)	0.182 (0.127)
	F → F	-0.010 (0.129)	-0.055 (0.188)	0.021 (0.208)
	D	-0.008 (0.179)	-0.007 (0.244)	0.161 (0.250)
Partner Effect	F → M	-0.161 (0.085)	-0.204 (0.109)	-0.077 (0.123)
	M → F	-0.777 (0.185) ***	-0.227 (0.240)	0.101 (0.216)
	D	0.617 (0.205) **	0.023 (0.272)	-0.177 (0.254)
Time Effect	M	0.053 (0.043)	0.027 (0.044)	0.051 (0.044)
	F	-0.214 (0.064) **	-0.185 (0.075) *	-0.168 (0.074)*
DASS STRESS				
Actor Effect	M → M	0.331 (0.203)	-0.066 (0.236)	0.225 (0.211)
	F → F	0.070 (0.162)	-0.088 (0.230)	0.094 (0.255)
	D	0.260 (0.262)	0.021 (0.344)	0.131 (0.339)
er Effect	F → M	-0.075 (0.141)	-0.149 (0.184)	-0.016 (0.203)
	M → F	-0.844 (0.234) ***	-0.152 (0.294)	0.021 (0.264)
	D	0.0769 (0.275) **	0.003 (0.361)	-0.038 (0.341)
Time Effect	M	0.017 (0.070)	-0.030 (0.074)	-0.022 (0.073)
	F	-0.243 (0.081) **	-0.211 (0.092) *	-0.194 (0.091) *
MMQ RELATION				
Actor Effect	M → M	-1.183 (0.604)*	-2.231 (0.601) ***	2.504 (0.484) ***
	F → F	-1.907 (0.369) ***	-2.659 (0.428) ***	2.620 (0.466) ***
	D	0.724 (0.716)	0.428 (0.817)	-0.117 (0.706)
Partner Effect	F → M	-0.875 (0.420) *	-1.046 (0.470) *	1.647 (0.467) ***
	M → F	-0.097 (0.531)	-0.294 (0.547)	1.370 (0.483) **
	D	-0.778 (0.686)	-0.752 (0.802)	0.277 (0.706)
Time Effect	M	0.476 (0.209) *	0.274 (0.188)	0.269 (0.167)
	F	0.421 (0.184) *	0.108 (0.171)	0.225 (0.167)
MMQ SEXUAL				
Actor Effect	M → M	-0.694 (0.491)	-1.600 (0.533) **	1.288 (0.473) **
	F → F	-0.509 (0.299)	-0.567 (0.368)	0.827 (0.414) *

	D	-0.185 (0.583)	-1.033 (0.735)	0.461 (0.680)
Partner Effect	F → M	-0.441 (0.341)	-0.168 (0.417)	0.614 (0.456)
	M → F	-0.222 (0.431)	-0.831 (0.470)	0.463 (0.429)
	D	-0.219 (0.558)	0.663 (0.718)	0.151 (0.678)
Time Effect	M	0.271 (0.170)	0.182 (0.167)	0.186 (0.163)
	F	0.033 (0.149)	-0.073 (0.147)	-0.023 (0.148)

Note. The table presents estimated regression coefficients (with standard error in brackets) for the actor and partner effect for males (M) and females (F), and the difference in actor (resp. partner) effects between males and females (D); and the estimated regression effects for the effect of the time since diagnosis in men and women. A separate APIM was fitted for each combination of dyadic coping subscale and outcome

DATA STORAGE FACT SHEETS

Data Storage Fact Sheet – Chapter 2

% Name/identifier study: Family adjustment when facing pediatric cancer: The role of parental psychological flexibility, dyadic coping and network support
% Author: Marieke Van Schoors
% Date: December 2019

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., De Paepe, A., Lemiere, J., Morez, A., Norga, K., Lambrecht, K., Goubert, L., & Verhofstadt, L. L. (2019). Family adjustment when facing pediatric cancer: The role of parental psychological flexibility, dyadic coping and network support. *Frontiers Psychology*, *10*:2740. doi: 10.3389/fpsyg.2019.02740

* Which datasets in that publication does this sheet apply to?: Quantitative datasets (SPSS/R files)

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [] other (specify)

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [x] file(s) describing the transition from raw data to reported results. Specify: For each step, a separate SPSS file is stored
- [x] file(s) containing processed data. Specify: SPSS/R outcome files
- [ ] file(s) containing analyses. Specify:
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 3

% Name/identifier study: Parents' perspectives of changes within the family functioning after a pediatric cancer diagnosis: A multi family member interview analysis
% Author: Marieke Van Schoors
% Date: January 2018

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., De Mol, J., Morren, H., Verhofstadt, L. L., Goubert, L. & Van Parys, H. (2018). Parents' perspectives of changes within the family functioning after a pediatric cancer diagnosis: A multi family member interview analysis. *Qualitative Health Research, 28*, 1229-1241. doi: 10.1177/1049732317753587

* Which datasets in that publication does this sheet apply to?: Qualitative datasets

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [ ] other (specify):

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [] file(s) describing the transition from raw data to reported results. Specify

- [x] file(s) containing processed data. Specify: All interviews were transcribed and saved as word document. All interviews were

blinded by the use of pseudonyms.
- [x] file(s) containing analyses. Specify: All analyses were done in MAXQDA. For every interview, a separate file with analyses was saved.
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 4

% Name/identifier study: Siblings' experiences of everyday life in a family where one child is diagnosed with blood cancer: A qualitative study
% Author: Marieke Van Schoors
% Date: January 2018

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., De Mol, J., Laeremans, N., Verhofstadt, L. L., Goubert, L. & Van Parys, H. (2019). Siblings' experiences of everyday life in a family where one child is diagnosed with blood cancer: A qualitative study. *Journal of Pediatric Oncology Nursing, 36*, 131-142. doi: 10.1177/1043454218818067

* Which datasets in that publication does this sheet apply to?: Qualitative datasets

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [ ] other (specify):

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group

- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [] file(s) describing the transition from raw data to reported results. Specify

- [x] file(s) containing processed data. Specify: All interviews were transcribed and saved as word document. All interviews were blinded by the use of pseudonyms.
- [x] file(s) containing analyses. Specify: All analyses were done in MAXQDA. For every interview, a separate file with analyses was saved.
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 5

% Name/identifier study: Associations between family functioning and child adjustment after pediatric cancer diagnosis: A meta-analysis
% Author: Marieke Van Schoors
% Date: September 2016

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., Caes, L., Knoble, N., Goubert, L., Verhofstadt, L. L. & Alderfer, M. (2017). Associations between family functioning and child adjustment after pediatric cancer diagnosis: A meta-analysis. *Journal of Pediatric Psychology*, *42*, 6-18. doi: 10.1093/jpepsy/jsw07

* Which datasets in that publication does this sheet apply to?: Quantitative datasets (SPSS/R files)

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [] YES / [x] NO

If NO, please justify: the analysis was done by the second author

* On which platform are the raw data stored?

- [] researcher PC
- [] research group file server
- [x] other (specify): PC of the second author

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [] main researcher
- [] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [x] other (specify): second author

3b. Other files

---

* Which other files have been stored?

- [] file(s) describing the transition from raw data to reported results. Specify: …
- [ ] file(s) containing processed data. Specify:
- [x] file(s) containing analyses. Specify: Pearson's *r* correlations of the included studies
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP

- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 6

% Name/identifier study: Family members dealing with childhood cancer: A study on the role of family functioning and cancer appraisal.
% Author: Marieke Van Schoors
% Date: June 2019

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., De Paepe, A., Norga, K., Cosijns, V., Morren, H., Vercruysse ,T., Goubert, L. & Verhofstadt, L. L. (2019). Family members

dealing with childhood cancer: A study on the role of family functioning and cancer appraisal. *Frontiers*. doi: 10.3389/fpsyg.2019.01405

* Which datasets in that publication does this sheet apply to?: Quantitative datasets (SPSS/R files)

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [] other (specify)

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [x] file(s) describing the transition from raw data to reported results. Specify: For each step, a separate SPSS file is stored
- [x] file(s) containing processed data. Specify: SPSS/R outcome files
- [ ] file(s) containing analyses. Specify:
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 7

% Name/identifier study: Siblings dealing with pediatric cancer: A family- and context-oriented approach.
% Author: Marieke Van Schoors
% Date: October 2020

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of

Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., Sels, L. Goubert, L., & Verhofstadt, L. L. (2020). Siblings dealing with pediatric cancer: A family- and context-oriented approach. *Manuscript accepted for publication in Journal of Pediatric Oncology Nursing.*

* Which datasets in that publication does this sheet apply to?: Quantitative datasets (SPSS/R files)

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [] other (specify)

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [x] file(s) describing the transition from raw data to reported results. Specify: For each step, a separate SPSS file is stored
- [x] file(s) containing processed data. Specify: SPSS/R outcome files
- [ ] file(s) containing analyses. Specify:
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...

- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 8

% Name/identifier study: The family practice of support giving after a pediatric cancer diagnosis: A multi-family member interview analysis
% Author: Marieke Van Schoors
% Date: 25/1/2018

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., De Mol, J., Verhofstadt, L. L., Goubert, L. & Van Parys, H. (2019). The family practice of support giving after a pediatric cancer diagnosis: A multi-family member interview analysis. *European Journal of Oncology Nursing, 44:* 101712. doi: 10.1016/j.ejon.2019.101712

* Which datasets in that publication does this sheet apply to?: Qualitative datasets

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [ ] other (specify):

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [ ] file(s) describing the transition from raw data to reported results. Specify
- [x] file(s) containing processed data. Specify: All interviews were transcribed and saved as word document. All interviews were blinded by the use of pseudonyms.
- [x] file(s) containing analyses. Specify: All analyses were done in MAXQDA. For every interview, a separate file with analyses was saved.
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO

Data Storage Fact Sheet – Chapter 10

% Name/identifier study: Couples dealing with pediatric blood cancer: A study on the role of dyadic coping
% Author: Marieke Van Schoors
% Date: 25/1/2018

1. Contact details

==========

1a. Main researcher

- name: Marieke Van Schoors
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: marieke.vanschoors@hotmail.com

1b. Responsible Staff Member (ZAP)

- name: Lesley Verhofstadt (promotor PhD project)
- address: Henri Dunantlaan 2 - 9000 Gent - Belgium
- e-mail: lesley.verhofstadt@ugent.be

If a response is not received when using the above contact details, please send an email to data.pp@ugent.be or contact Data Management, Faculty of Psychology and Educational Sciences, Henri Dunantlaan 2, 9000 Ghent, Belgium.

2. Information about the datasets to which this sheet applies

==========

* Reference of the publication in which the datasets are reported:

Van Schoors, M., Loeys, T., Goubert, L., Berghmans, G., Ooms, B., Lemiere, J., Norga, K., & Verhofstadt, L. (2019). Couples dealing with pediatric blood cancer: A study on the role of dyadic coping. *Frontiers*. doi: 10.3389/fpsyg.2019.00402

* Which datasets in that publication does this sheet apply to?: Quantitative datasets (SPSS/R files)

3. Information about the files that have been stored

==========

3a. Raw data

---

* Have the raw data been stored by the main researcher? [x] YES / [ ] NO

If NO, please justify:

* On which platform are the raw data stored?

- [x] researcher PC
- [x] research group file server
- [] other (specify)

* Who has direct access to the raw data (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group
- [ ] all members of UGent
- [ ] other (specify): ...

3b. Other files

---

* Which other files have been stored?

- [x] file(s) describing the transition from raw data to reported results. Specify: For each step, a separate SPSS file is stored
- [x] file(s) containing processed data. Specify: SPSS/R outcome files
- [ ] file(s) containing analyses. Specify:
- [x] files(s) containing information about informed consent: the IC's were storaged on paper.
- [ ] a file specifying legal and ethical provisions
- [ ] file(s) that describe the content of the stored files and how this content should be interpreted. Specify: ...
- [ ] other files. Specify: ...

* On which platform are these other files stored?

- [x] individual PC
- [x] research group file server
- [ ] other: ...

* Who has direct access to these other files (i.e., without intervention of another person)?

- [x] main researcher
- [x] responsible ZAP
- [ ] all members of the research group

- [ ] all members of UGent
- [ ] other (specify): ...

4. Reproduction

==========

* Have the results been reproduced independently?: [ ] YES / [x] NO