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Parent's Perceptions about their Child's Illness in Pediatric Cancer: Links with Caregiving Burden and Quality of Life

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Parent's Perceptions about their Child's Illness in Pediatric Cancer: Links with Caregiving Burden and Quality of Life

Resumo

O presente estudo teve por objetivo contribuir para a compreensão da qualidade de vida em pais de crianças com cancro. Especificamente, esta investigação examinou o papel das percepções dos pais acerca da doença em termos de gravidade e interferência na vida da criança na sua própria qualidade de vida (QdV) de forma direta e indireta através da sobrecarga percebida. Pretendeu-se ainda examinar se as referidas associações entre as variáveis em estudo diferiam de acordo com a fase de tratamento da criança.

A amostra do presente estudo transversal foi constituída por 277 pais de crianças ou adolescentes com diagnóstico de cancro, divididos em dois grupos clínicos: 126 pais de crianças em tratamento e 151 pais de crianças fora de tratamento. Os participantes preencheram questionários de auto-resposta que avaliaram as suas percepções da gravidade e interferência da doença na vida da criança, a sua sobrecarga e sua QdV. A informação relativa ao diagnóstico, condição e intensidade de tratamento foi reportada pelo oncologista pediátrico que acompanhava a criança.

Os resultados mostraram que os pais de crianças em tratamento reportavam percepções mais negativas acerca da doença (gravidade e interferência na vida da criança), níveis mais elevados de sobrecarga e pior QdV, quando comparados com os pais de crianças fora de tratamento. Para ambos os grupos, apenas a percepção de interferência da doença na vida da criança se mostrou negativamente associada à QdV. Por sua vez, a associação entre a percepção de gravidade e QdV não se mostrou significativa. Adicionalmente, as percepções dos pais acerca da doença (gravidade e interferência) mostraram estar negativamente associadas à QdV por meio da sobrecarga. Por fim, a associação entre a sobrecarga e a QdV foi moderada pela condição de tratamento, com associações mais fortes encontradas para os pais de crianças em tratamento.

Os resultados deste estudo reforçam a relevância da investigação e intervenção com famílias no contexto do cancro pediátrico, no sentido de identificar as necessidades específicas dos pais em diferentes fases de tratamento, promover a sua adaptação nesta situação de adversidade e, consequentemente, promover também o bem-estar das próprias crianças.

Palavras-chave: Percepções acerca da doença, Sobrecarga, Qualidade de Vida, Cancro Pediátrico.

Abstract

This study examined the direct and indirect links, via the caregiving burden, between parents' perceptions about the severity of their child's illness and its interference in his or her life and the parents' quality of life (QoL). The participants were 277 parents of children with malignant cancer, divided into two clinical groups according to treatment status: 126 parents of children on-treatment and 151 parents of children off-treatment. Self-reported questionnaires assessed parents' perceptions of illness severity and interference in the child's life, caregiving burden and QoL. Pediatric oncologists provided information about diagnosis, treatment status and intensity. Parents of children on-treatment reported more negative perceptions about the illness (severity and interference), higher levels of caregiving burden and worse QoL, when compared to parents of children off-treatment. Furthermore, parents' perceptions about their child's illness (severity and interference) were negatively linked to parents' QoL through caregiving burden. Additionally, the link between caregiving burden and parents' QoL was moderated by child treatment status, with stronger associations found for parents of children on-treatment. This study's findings are discussed in terms of their relevant implications for intervention with families in the pediatric cancer context.

Keywords: Perceptions of illness, Caregiving burden, Quality of Life, Pediatric cancer.

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Lista de Abreviaturas

QdV - Qualidade de Vida

QoL - Quality of Life

Introdução

O cancro pediátrico é a segunda causa de morte em países desenvolvidos (Kaatsch, 2010). Além de considerada uma condição de saúde de risco (Anderzén-Carlsson, Kihlgren, Svantesson, & Sørli, 2007), esta doença interfere significativamente na vida diária da criança e da sua família (McGrath, Paton, & Hoff, 2005; Patterson, Holm, & Gurney, 2004). Sabe-se que os pais têm um papel fundamental na manutenção da qualidade do funcionamento familiar. E sabe-se igualmente que perturbações no funcionamento familiar podem influenciar a própria evolução da doença (Kazak, Rourke, & Navsaria, 2009). Assim e, dado do impacto da doença no bem-estar parental, torna-se relevante compreender as suas próprias vivências em torno da doença e tratamentos. O presente estudo enquadra-se no âmbito da Psicologia Pediátrica, uma disciplina que, cruzando os conhecimentos da Medicina Pediátrica e da Psicologia da Criança, procura compreender e auxiliar as crianças, adolescentes, e também as suas famílias (Roberts & Steele, 2009).

O diagnóstico de cancro pediátrico envolve uma série de desafios para a criança, mas também para os outros membros da família, devido, entre outros aspetos, aos efeitos secundários adversos dos tratamentos e à necessidade de hospitalizações frequentes (Long & Marsland, 2011). Exige-se uma reorganização nos papéis e responsabilidades, no sentido de se incluir na dinâmica familiar um tratamento complexo e as necessidades da criança doente (Alderfer & Kazak, 2006). Os pais têm um papel cada vez mais ativo na implementação dos procedimentos médicos (Klassen et al., 2011). Para além das tarefas relativas à doença e tratamento, os pais têm ainda de conciliar as tarefas domésticas, a ocupação profissional, as próprias necessidades e as dos outros membros da família (Patterson et al., 2004; Wolfe-Christensen et al., 2010; Young, Dixon-Woods, Findlay, & Heney, 2002).

Os défices na qualidade de vida dos pais de crianças com cancro (Klassen et al., 2008) poderão decorrer desta ‘carreira inesperada’ enquanto cuidadores de uma criança com uma condição crónica, e que pode ser percebida como sobrecarga (Raina et al., 2004). Um dos fatores que parece explicar o desgaste destes pais refere-se à forma como percecionam a doença, quer no que toca a limitações impostas à vida da criança (Litzelman, Catrine, Gagnon, & Witt, 2011), quer em termos de gravidade da condição clínica (Turner-Henson, Holaday, & Swan, 1992).

A perceção que os pais têm da doença bem como as suas tarefas enquanto cuidadores podem variar consoante a fase da doença. Durante os tratamentos, os procedimentos médicos aversivos debilitam o estado físico da criança e interferem nas suas atividades diárias (Anderzén-Carlsson et al., 2007; McCaffrey, 2006; Patterson et al., 2004). Adicionalmente, aumentam as tarefas dos pais necessárias à boa prossecução do tratamento, bem como a necessidade de fornecer apoio emocional à criança (Young et al., 2002).

Assim, é legítimo considerar que a pior QdV reportada pelos pais de crianças em tratamento (Litzelman et al., 2011), poderá ser explicada, em parte, pela maior sobrecarga destes pais.

O objetivo central deste estudo é compreender de que forma a percepção dos pais acerca da doença, em termos de gravidade e interferência na vida da criança, influencia a sua qualidade de vida, examinando o papel mediador da sobrecarga percebida. Avaliou-se ainda o papel moderador da condição de tratamento da criança (i.e., em tratamento *vs.* fora de tratamento) na relação entre as variáveis referidas. Acreditamos que compreender a forma como os pais percebem a doença da criança se torna fundamental à compreensão do seu comportamento e adaptação perante esta experiência adversa. Pretende-se adicionalmente, através dos resultados deste estudo empírico, gerar informação fundamental à sinalização dos pais em risco, que necessitam de ações de intervenção focalizadas, que minimizem o impacto da doença e tratamento na sua vida e bem-estar.

Seguidamente apresenta-se a tese no formato de artigo científico, tal como será submetido a periódico internacional com arbitragem científica.

Parent's Perceptions about their Child's Illness in Pediatric Cancer: Links with Caregiving Burden and Quality of Life

The incidence rate of childhood cancer is about 100-150 cases per million children and adolescents under 15 years of age (Michaud, Suris, & Viner, 2007). Receiving the diagnosis of a chronic illness, such as childhood cancer, poses significant challenges to the child or adolescent, and also to the family (Kazak, 1989; Kazak, Rourke, & Navsaria, 2009) as the caregiving system (Rolland, 2012).

Even though much of the literature has focused on the child's psychological adjustment, there has been increasing consensus regarding the importance of understanding the needs of the entire family throughout the experience of childhood cancer (Alderfer & Kazak, 2006). Parents of children with cancer are more at risk of psychological distress than parents of healthy children (see Vrijmoet-Wiersma et al., 2008 for a review). Evidence has also suggested that parents of children with cancer report worse quality of life (QoL), compared to parents of healthy children (Witt et al., 2010) and population norms (Eiser, Eiser, & Stride, 2005; Klassen et al., 2008). Consistent with a family systems perspective, this increased risk can affect the parents as individuals and also produce a ripple effect on the family, also affecting the children. According to the socio-ecological framework applied to the context of chronic conditions (Kazak, 1989; Kazak et al., 2009) the child's adaptation and development cannot be understood without taking into account contextual sources of influence, such as parental adjustment. Research has systematically found significant associations between parental distress and poor child adjustment (Robinson, Gerhardt, Vannatta, & Noll, 2007; Wolfe-Christensen et al., 2010). These findings suggest the need for family-based interventions in pediatric oncology, in order to promote parental adaptation to a child's illness, and consequently protect the ill child from adverse effects of the cancer experience. The present research focusing on parents' perceptions about the child's illness, the caregiving burden and QoL seeks to understand parents' experiences and inform future family-based interventions in the context of this condition.

Parents' Subjective Perceptions about Childhood Cancer

When parents are confronted with a pediatric chronic condition diagnosis, their perception about the illness and its treatment are important predictors of parental adaptation (Kazak et al., 2009). In a study of parents of children undergoing or having previously undergone cancer treatment at a Malaysian hospital, those who perceived their child's current medical condition in a more negative way reported poorer adjustment (Othman, Mohamad, Hussin, & Blunden, 2011).

Despite the advances in treatment and improvements in prognosis and survival rates (Michaud et al., 2007), childhood cancer is still considered a life-threatening condition (Anderzén-Carlsson, Kihlgren, Svantesson, & Sørli, 2007; Yeh, 2002; Young, Dixon-Woods, Findlay, & Heney, 2002). In fact, childhood cancer is viewed as a serious medical condition, which

would generally be expected to only occur in late adulthood (Grootenhuis & Last, 1997a), and for that reason it is considered to be a non-normative event (Rolland, 2012). Following cancer diagnosis, parents must face the uncertainty and unpredictability of the course of the illness and its treatment (Grootenhuis & Last, 1997b; Hung, Wu, & Yeh, 2004). Concerns about their child's health and chances of survival are frequently reported by parents of children/adolescents with this condition (Grootenhuis & Last, 1997b; Patterson, Holm, & Gurney, 2004). A study by Boman, Viksten, Kogner and Samuelsson (2004) found that unlike pediatric diabetes, where levels of parental uncertainty decreased over time, pediatric cancer was associated with parents' constant fear of losing the child. Even if diagnosis and treatment are followed by a remission period, pediatric oncologists cannot guarantee a full recovery (McCaffrey, 2006). Parents are often concerned about the possibility that treatment will be ineffective, that the cancer will recur, or that children will suffer later effects of aggressive medical protocols (e.g., cardiac toxicity, sterilization, limb loss) (Boman, Lindahl, & Björk, 2003; Fletcher, 2010; Grootenhuis & Last, 1997b; McCaffrey, 2006; Patterson et al., 2004).

Another area of concern for parents, alongside the severity of the condition, is the interference in the child's life (Patterson et al., 2004). Childhood cancer is perceived as a life-changing condition, involving significant disruption to the functional status and daily routine of the ill child. The invasive and intensive treatment of childhood cancer exposes the child to different stressors. These include aversive medical procedures (e.g., needles or blood draw, lumbar punctures, bone marrow tests, chemotherapy) and unpleasant side-effects (e.g., pain, mouth ulcers, weakness, fatigue, nausea, vomiting, toxic reactions, infections, hair loss, and weight loss or gain) (Anderzén-Carlsson et al., 2007; McCaffrey, 2006; Yeh, 2002). A complex regimen of long-term hospitalizations and frequent visits to the outpatient services result in disruption to school attendance and isolation from peers (McCaffrey, 2006; McGrath, Paton, & Huff, 2005, 2005; Patterson et al., 2004; Yeh, 2002). Childhood cancer patients can be limited in their abilities to deal with daily living routines (Litzelman, Catrine, Gangnon, & Witt, 2011), due to a range of physical, social and cognitive impairment caused by the disease and its treatment. Thus they may depend significantly on family caregiver support.

Caregiving Burden

Given the improvements in treatment for pediatric cancer, many children receive a large part of their treatment and supportive care at home. Consequently, parents are increasingly taking a more active role in the care of their ill child (Klassen et al., 2007, 2011). Caring for a child with cancer requires additional nursing and the performance of technical tasks (Young et al., 2002). These include administering medication, monitoring for toxicity and other side-effects, communicating with the health care team, and planning frequent clinic visits and hospitalizations (von Essen, Sjöden, & Mattsson, 2004; Wolfe-Christensen et al., 2010). Parents must also deal with

new and complex information related to illness and medical treatment (Othman et al., 2011; Wolfe-Christensen et al., 2010) and make important decisions throughout the course of the treatment (Mercer & Ritchie, 1997). Furthermore, children who suffer pain and discomfort require additional emotional support (Wolfe-Christensen et al., 2010; Young et al., 2002).

Parents of chronically ill children are exposed to considerable burden as a result of maintaining multiple and time-consuming roles (Turner-Henson, Holaday, & Swan, 1992). In addition to illness and treatment-related demands, parents must continue to perform household tasks, balance occupational activities and meet the physical and emotional needs of other family members (McCaffrey, 2006; McGrath, et al., 2005; Patterson et al., 2004; Wolfe-Christensen et al., 2010; Young et al., 2002). Frequently, parents use sick leave or vacation time, or they quit their jobs, which can result in income variations and possibly financial difficulties (Fletcher, 2010; McGrath et al., 2005; Patterson et al., 2004). Additionally, excessive responsibilities may reduce the time that parents have to take care of themselves and their own health (Fletcher, 2010; Klassen et al., 2008), and they may give up leisure activities they had before the onset of the childhood cancer (McCaffrey, 2006). As such, when parents face the diagnosis, a new and sudden role of caregiving is added to already existing roles. Consequently, they may experience physical, personal, emotional and financial hardships (Fletcher, 2010) and caregiving burden (Wolfe-Christensen et al., 2010).

Implications of Caring for a Child with Cancer on Parents' Quality of Life

The “unexpected career” of a caregiver of a child with a health condition poses different and challenging demands (Raina et al., 2004, p.14) that can compromise the physical and psychological health of the caregiver (Klassen et al., 2007; Raina et al., 2004). Much of the research on pediatric cancer has focused on the negative effects of this condition on parents' adjustment, such as anxiety (Dahlquist, Czyzewski, & Jones, 1996; Grootenhuis & Last, 1997b; Kazak & Barakat, 1997), depression (Dahlquist et al., 1996; Grootenhuis & Last, 1997b) and post-traumatic stress symptoms (Kazak & Barakat, 1997). More recently, however, researchers have also focused on the impact of childhood cancer on parents' positive outcomes such as QoL. The results so far suggest clinically important deficits in QoL reported by parents of children with cancer compared to parents of healthy children (Witt et al., 2010) and to population norms (Eiser et al., 2005; Klassen et al., 2008).

One of the factors explaining the poorer QoL reported by parents of children with cancer is caregiving burden (Klassen et al., 2011). More specifically, Litzelman and colleagues (2011) found that limitation of children's activity, as reported by parents, was associated with lower parental QoL in the mental domain, via the mediating effect of the caregiving burden and stress. Additionally, in a study with parents of chronically ill children, Turner-Henson and colleagues (1992) found that

when parents perceive the illness as more severe or serious they spend a greater amount of time in caregiving activities. Together these results suggest that when parents perceive their child's illness to be more severe and/or as posing limitation on the child's activity, they may feel overwhelmed with extra caregiving demands. The full availability required to manage symptoms and lifestyle limitations can become burdensome (Raina et al., 2004), and this therefore explains the lower rates of health-related quality of life (Klassen et al., 2011).

Impact of the Child's Treatment Status: On or Off-Treatment Phase

Existing research has focused on parental adjustment in different phases of the illness: after diagnosis, during the treatment phase, and after the end of treatment. The majority of the studies relied on samples of parents with children undergoing cancer treatment at the time of the assessment (see Klassen et al., 2007 for a review). Other studies compared the adaptation of parents of children on active treatment and of parents of children who had completed treatment at the time of the assessment. These studies, using two groups of parents according to their child's treatment status (i.e., on-treatment vs. off-treatment), consistently suggested that parents of children on-treatment were at more risk of higher levels of anxiety (Boman et al., 2003; Othman et al., 2011) and depression (Boman et al., 2003; von Essen et al., 2004; Dockerty, Williams, McGee, & Skegg, 2000) and poorer QoL (Litzelman et al. 2011; von Essen et al., 2004).

The present study sought to understand the impact that having a child undergoing or having undergone medically invasive procedures (e.g., chemotherapy, radiation, surgery and/or transplant) had on parental perceptions about the illness, caregiving burden and QoL. We expected the child's treatment status to influence not only parents' perceptions about the illness, but also the nature and degree of caregiving demands. Parents of children undergoing cancer treatment procedures deal with greater stressors, compared to those whose children have finished treatment. Parents may witness their child experiencing pain and unpleasant side-effects and lifestyle restrictions due to aggressive medical procedures (Anderzén-Carlsson et al., 2007; McCaffrey, 2006; Patterson et al., 2004). Additionally, they may also struggle with the uncertainty about the effectiveness of treatment (McCaffrey, 2006). When a child is receiving treatment, medical, nursing and emotional care demands increase considerably. Understandably, parents wish to be at the hospital, accompanying their ill child. Parental attention is greatly focused on providing comfort to a child who is suffering (Anderzén-Carlsson et al., 2007; Young et al., 2002). Furthermore, during the active treatment phase, parents struggle with different strategies to gain their child's co-operation with aversive medical procedures (e.g., taking medicine, physical exams) (Young et al., 2002). At the same time, parents must maintain their other roles in the family and in the wider social arena, such as in the sphere of employment. In this context, it becomes particularly difficult to balance job demands with multiple family needs, including

financial needs that can arise or increase due to taking prolonged leave or where one or both of the parents give up their job altogether. The combination of these strains can lead to exhaustion (Patterson et al., 2004), increasing the levels of caregiving burden, which, in turn, may negatively impact QoL (Klassen et al., 2011).

The Current Study

The present research sought to examine the links between parents' perceptions about their child's illness and the former's QoL. Four specific objectives were defined. The first was to assess whether parents' perceptions about the illness (severity and the degree of interference in the child's life), caregiving burden and QoL were different according to the child's treatment status, on-treatment and off-treatment. The second was to identify the associations between parents' perceptions about the illness (severity and interference in the child's life) and their QoL. The third objective was to identify a potential process by which parents' perceptions of illness (severity and interference in the child's life) would be linked to parental QoL, by testing the mediating role of the caregiving burden. The fourth and final objective was to examine if the aforementioned mediation processes differed according to the child's treatment status (children on-treatment vs. children off-treatment).

Four hypotheses were devised:

Hypothesis 1: Parents with children on-treatment would report higher perceptions of illness (severity and interference in the child's life), higher caregiving burden and lower QoL, compared to parents of children off-treatment;

Hypothesis 2: Parents' perceptions about the illness (severity and degree of interference in the child's life) would be negatively associated with parents' QoL;

Hypothesis 3: Caregiving burden would be associated with more negative perceptions about illness (severity and interference in the child's life) and with poorer QoL;

Hypothesis 4: Caregiving burden would mediate the links between parents' perceptions of illness severity and interference in the child's life and QoL.

A key aspect of our study was to examine if the mediation models would be moderated by the child's treatment status. Given the scarce literature comparing children and parents' adaptation in different treatment stages of pediatric cancer, we established no specific hypothesis due to the exploratory nature of these analyses.

Method

Participants

Participants were 277 parents of children¹ diagnosed with cancer.

¹ For simplicity, we used the term children when referring to this study's sample composed of both children and adolescents.

Parents, mostly mothers (82.7%, $n = 229$) and married (81.2%, $n = 225$), were between 25 and 68 years old ($M = 42.35$; $SD = 6.48$). Most of the parents came from a low socio-economic background (55.6%, $n = 154$). Children were diagnosed with leukemias (38.2%, $n = 106$), lymphomas (25.2%, $n = 70$), bone sarcomas (13.7%, $n = 38$), brain cancers (5.8%, $n = 16$), soft tissue sarcomas (3.2%, $n = 9$), or other types of cancer (10.6%, $n = 29$). For 9 of the cases, diagnosis details were missing.

The sample was divided into two clinical groups according to treatment status, one composed of 126 parents of children on-treatment and the other of 151 parents of children off-treatment. Parents were included in the on-treatment group if their children were undergoing treatment (active or maintenance) at the time they filled in the assessment protocol. If children had completed treatment for a cancer diagnosis parents were included in the off-treatment group. Parents of children undergoing palliative treatment were excluded. Descriptive results for both groups are depicted in Table 1.

In order to examine if there were differences between clinical groups with regard to clinical and socio-demographic characteristics, independent t -tests and chi-square analysis were conducted. On-treatment group participants were parents of older children compared to the participants in the off-treatment group. A statically significant difference was found regarding time elapsed since diagnosis, with parents of children belonging to the off-treatment group reporting a longer period of time since diagnosis. Clinical groups also differed according to intensity of treatment. Most children belonging to the on-treatment group were receiving treatment assessed as very intensive, whereas the majority of those in the off-treatment group had received, in the past, moderately intensive treatments. The samples did not differ in any of the other variables, as depicted in Table 1.

Measures

Parents' Perception of the Severity of the Child's Illness. For the purpose of this study, a one-item measure assessing parents' subjective perception of the severity of their child's illness was created. Participants answered the question "On a scale of 1 to 10, to what extent do you think that your child's health problem is severe?" on a 10-point Likert scale from 1 (*Not severe*) to 10 (*Very severe*).

Parents' Perception of the Illness' Interference in the Child's Life. This one-item measure created for the purpose of the present research assessed the parents' subjective perception of the impact of the cancer on the child. Participants were asked "On a scale of 1 to 10, to what extent do you think that your child's health problem interferes in his/her life?" and answered on a 10-point Likert scale from 1 (*Not disabling*) to 10 (*Very disabling*).

Caregiver Burden Scale - Revised. The objective burden subscale of this self-report questionnaire was used in the present study (Montgomery & Kosloski, 2006; Portuguese version: Carona, Faria-Morais, Nazaré, & Canavarró, 2008). This subscale assessed the negative psychological state resulting from the impact of the caregiving responsibilities on the caregiver's

life (e.g., “Have your caregiving responsibilities decreased time you have for yourself?”). Participants answered 6 items on a 5-point Likert scale ranging from 1 (*Not at all*) to 5 (*A great deal*).

EUROHIS-QOL. This questionnaire is a measure derived from the WHOQOL-100 and the WHOQOL-BREF instruments (Schmidt, Mühlan, & Power, 2006; Portuguese version: Pereira, Melo, Gameiro, & Canavarro, 2011). The EUROHIS-QOL is a one-dimensional measure comprising 8 items in the following domains of QoL: physical, psychological, social and environmental. Participants answered items on a 5-point Likert scale ranging from 1 (*Very poor/Very dissatisfied/Not at all*) to 5 (*Very good/Very satisfied/Completely*). An overall QoL score was obtained from the mean of the items’ scores.

Intensity of Treatment Rating Scale (ITR -3.0). This scale provides a classification of the intensity of pediatric cancer treatment based on medical chart review (Kazak et al., 2011; Portuguese version: Santos, Crespo, Canavarro, & Pinto, *in preparation*). The pediatric oncologists were asked to classify treatment into one of four levels of intensity, from level 1 (*Least intensive treatment*) to 4 (*Most intensive treatment*), based on diagnosis, stage, risk for the patient, phase of illness (new diagnosis or relapse), and treatment data criteria. Rater reliability for a Portuguese sample in a prior study was excellent ($kappa=.97$; $p>.001$) (Santos, Crespo, Canavarro, & Pinto, *in preparation*).

Clinical and Socio-demographic characteristics. Parents provided other relevant clinical information (e.g. time since diagnosis) and socio-demographic data (e.g. gender, age, marital status, socio-economic status-SES). SES was determined according to a classification system based on parents’ jobs and educational level (Simões, 1994).

Procedures

The sample was collected at the pediatric oncology services of three Portuguese state hospitals: the Portuguese Institute of Oncology (IPO-Porto), Pediatric Unit-Coimbra Hospital and University Centre, and São João Hospital. Between June 2012 and March 2013 all parents, through consecutive sampling, were invited to participate in the study. Formal authorizations for sample collection were obtained from the Ethics Committees of these institutions.

Parents of children with malignant cancer were included whenever the following inclusion criteria were met: (1) the parent was the child’s primary caregiver; (2) the child was aged between 8 and 19 years old; (3) the child was at least 3 months post-diagnosis; and (4) the child had had an off-treatment status for less than 5 years.

Participants were approached either in inpatient or outpatient settings of the aforementioned institutions. The study’s aims were explained to all participants, and informed consent forms were obtained, according to procedures of the local institutional ethical boards. The assessment protocol was administered under the supervision of a trained research assistant, while subjects were waiting for their medical appointments and procedures.

Parents with difficulties in filling in the protocol were personally aided by the research assistant. The child's pediatric oncologist provided diagnosis groups and assessed the treatment intensity and treatment status.

Results

Descriptive Analyses and Zero-order Correlations

The statistical analyses were conducted with Statistical Package for Social Sciences, version 20 (SPSS, Inc., Chicago, IL). Descriptive statistics and correlations for study variables for both groups are presented in Table 2. We performed a MANOVA to assess if the two clinical groups differed regarding parents' perceptions of illness severity and interference on the child's life, caregiving burden and QoL. A statistically significant multivariate effect was found, Wilks' Lambda = .79, $F(4, 272) = 18.53$, $p = .04$, partial $\eta^2 = .21$. The univariate analyses showed that groups differed in all the study's main variables. Parents of children on-treatment reported their child's illness as more severe ($M = 7.50$, $SD = 2.19$), $F(1, 275) = 32.90$, $p < .001$, partial $\eta^2 = .11$, and interfering more in the child's life ($M = 7.26$, $SD = 2.38$), $F(1, 275) = 66.87$, $p < .001$, partial $\eta^2 = .20$, than parents of children off-treatment ($M = 5.79$, $SD = 2.69$ and $M = 4.62$, $SD = 2.90$, respectively). Caregiving burden was higher for parents of children on-treatment ($M = 3.24$, $SD = .93$) compared to parents of children off-treatment ($M = 2.71$, $SD = .98$), $F(1, 275) = 20.67$, $p < .001$, partial $\eta^2 = .07$. Finally, parents of children on-treatment reported lower QoL ($M = 3.42$, $SD = .53$) than their counterparts whose children were off-treatment ($M = 3.64$, $SD = .53$), $F(1, 275) = 11.39$, $p = .001$, partial $\eta^2 = .04$.

Correlational analyses indicated that parents' perception of illness interference in the child's life was associated with QoL in the expected negative direction. However, we found no significant association between parents' perception of the severity of their child's illness and QoL. Parents' reports of caregiving burden were associated with lower QoL for both groups. Parents' perceptions of illness severity and interference in the child's life were both positively associated with caregiving burden in both groups.

Moderated Mediation Analyses

Our proposed mediation models hypothesized that the links between parents' perceptions of the severity of their child's illness and its interference in the child's life were linked to higher levels of caregiving burden, which in turn, would be linked to lower QoL; in addition, we sought to examine if these links were different for the two clinical groups. In order to test these two models, we conducted a multiple regression and a bootstrapping procedure (Preacher & Hayes, 2008; Hayes, 2013), according to which indirect effects are assessed based on a point estimate and bootstrapped 95% confidence interval; an indirect effect is considered significant if its confidence interval does not include 0. We used the SPSS macro PROCESS and tested Hayes' (2013) proposed model number 59 (see

Figure 1 for an application of this model to the present data). For the model using perception of illness severity as the independent variable, results showed significant indirect effects on the outcome QoL via the mediator caregiving burden for both groups. There was only one significant interaction in this model, caregiving burden x clinical group ($B = -.19$; $t = -3.03$, $p < .01$), indicating that the association between caregiving burden and QoL was different for parents with children on-treatment and parents with children off-treatment. Similar results were found for the model where perception of the illness interference on the child's life was used as the independent variable, with the same significant interaction ($B = -.21$; $t = -3.11$, $p < .01$).

In order to examine how the association between caregiving burden and QoL varied according to clinical group, we performed regression analyses. Results revealed that the interaction between clinical group and caregiving burden was significantly related to QoL ($B = -.18$; $t = -2.95$, $p < .05$). The significant interaction effects were plotted using Mod-Graph (Jose, 2008). Post hoc simple slopes analyses showed that the QoL of parents of children undergoing treatment, $t = -6.86$, $p < .001$, was especially affected by caregiving burden, compared to that of parents of children off-treatment, $t = -2.77$, $p < .05$ (see Figure 2).

Given that the two groups only differed in one path of the proposed mediation models (caregiving burden's association with QoL) and that this path differed in terms of strength and not direction of association, we opted to present results with the total sample for the two mediation models. We used SPSS macro INDIRECT provided by Preacher & Hayes (2008) to assess the significance of direct and indirect effects. For the present analyses we performed a bootstrap resampling procedure with 10,000 samples [95% bias-corrected and accelerated confidence intervals (BCa 95% CI)].

We tested two simple mediation models. For model 1, parents' perception of the severity of the child's illness was the independent variable, whereas for model 2 parents' perception of the illness' interference in the child's life was the independent variable. For both models, the outcome was QoL and caregiving burden was the mediator.

Results indicated that indirect effects were significant for both models. For model 1, caregiving burden (*point estimate* = $-.02$; CI = $-.04/-0.01$) mediated the marginal link between parents' perception of the severity of the child's illness and QoL (see Figure 3); the R^2 for QoL was $.16$. For model 2, caregiving burden (*point estimate* = $-.03$; CI = $-.04/-0.01$) mediated the link between parents' perception of the illness' interference in the child's life and QoL; the R^2 for QoL in this model was $.18$ (see Figure 4).

Discussion

In the present study, we posed and found general support for four hypotheses. First, parents of children undergoing treatment reported more negative perceptions about the child's illness (severity and illness

interference), higher levels of caregiving burden and poorer QoL than parents of children who were already off-treatment. Secondly, we found negative associations between parents' perception about the interference of their child's illness and QoL. Contrary to that which was expected, we did not find significant associations between parents' perception about the severity of their child's illness and QoL. Thirdly, confirming our third and fourth hypotheses, when parents perceived their child's illness as more severe and as interfering more with the child's life, they also experienced higher levels of caregiving burden and, in turn, they reported poorer QoL. Finally, for parents with children undergoing treatment, the link between caregiving burden and QoL was stronger than for parents of children off-treatment.

Parents of Children On and Off-Treatment

Mean differences between groups

Consistent with our first hypothesis, parents of children undergoing treatment reported their child's illness as more severe and as having greater interference in their child's life, compared to their counterparts whose children were off-treatment. Moreover, the former group of parents also reported higher levels of caregiving burden and lower QoL than the latter. The cycles of treatment required in pediatric cancer are extremely unpleasant and painful for the child (Anderzén-Carlsson et al., 2007; McCaffrey, 2006; Yeh, 2002). Understandably, due to the visibly aggressive side-effects of treatment (e.g., loss or gain of weight, weakness, skin reactions and sickness related to therapy or infections) parents may perceive their child's health status as severely compromised. During the specific period of treatment there are also great limitations to the child's lifestyle, such as being absent from school, being in isolation to avoid infections and generally being unable to do sports or outdoor activities. Moreover, caregivers face additional demands, such as closely monitoring side-effects, attempting to gain the child's cooperation with exams and medical procedures and providing emotional support to a child whose well-being is compromised (Young et al., 2002), alongside the typical and day-to-day demands of caring for a chronically ill child. Finally, studies reported that having to watch their child suffering during aversive treatment was consistently reported as a source of distress by parents (Othman et al., 2011; Patterson et al., 2004; Young et al., 2002), which will likely have a negative impact on parents' overall QoL.

The moderating role of the child's treatment status on the link between caregiving burden and QoL

Results showed that in the tested mediation models, the only path which was different according to the child's treatment was the path between caregiving burden and parents' QoL, with stronger associations being found for the group of parents with children currently undergoing treatment.

The finding that the link between caregiving burden and parents' QoL

is especially strong during an active treatment phase supports the idea that during this period parents are confronted with great disruptions in their daily life, compared to the period after cancer treatment has been completed. Previous studies suggest that parents of children with cancer on-treatment may experience excessive burden due to nursing and technical and emotional care demands, added to the usual demands of parenting in general (Wolfe-Christensen et al., 2010; Young et al., 2002).

As the children's dependence on others increases, parents must attend to their needs, while also monitoring adherence to treatment protocols (Young et al., 2002). In addition, other important responsibilities must be maintained (e.g., giving support to other children; housekeeping; employment duties) (Patterson et al., 2004; Young et al., 2002). Although parents try to keep their other roles, the needs of the ill child usually become the priority during this period. Furthermore, during an active treatment phase, due to heightened caregiving tasks, parents may neglect their own health (e.g., worse eating habits, lower sleep quality, less physical exercise), which can negatively impact their QoL (Klassen et al., 2008).

Parents' Perception of the Child's Illness and QoL: Direct and Indirect Effects via Caregiving Burden

Direct effects

Our second hypothesis, stating that parents' perceptions about the severity of the illness and its interference in the child's life would be negatively associated with parents' QoL, received partial support. We only found evidence for a negative association between parents' perception of the interference of the illness in the child's life and their QoL appraisals. These findings are in accordance with previous research suggesting that a child's worse health status, rated by parents in terms of lifestyle limitations or restrictions, was associated with lower QoL of parents (Klassen et al., 2008; Klassen et al., 2011; Litzelman et al., 2011). A study by Patterson and colleagues (2004) also found that the child's emotional reaction to losing out on normal life and activities (e.g., missing school and time with friends) was an important concern reported by parents of children with cancer.

However, our results suggest that although childhood cancer is considered a serious and life-threatening condition, being associated with a constant fear of losing the child (Boman et al., 2004), the perceived severity of the illness did not have a significant direct association with parents' QoL. One possible explanation for this unexpected finding is the role of coping strategies endorsed by parents, such as being positive and hopeful, which were not measured in the present study. Although there is limited research regarding the role of hope among parents of children suffering from cancer, this appears to be an important resource in the adaptation of these parents (Kylmä & Juvakka, 2007). Parents may perceive the illness as a severe condition, but maintain a positive attitude, i.e., focus on the possibility that the treatment will be successful and that the child will survive. Dockerty and

colleagues (2000) found that the majority of parents of children with cancer believed that their child would get better. An important coping behavior often reported by parents of children with cancer consists of actively seeking informational support, often given by health care providers (Patterson et al., 2004). If the medical care team shares adequate information about the illness and its treatment, parents may be aware of the severity of the condition and simultaneously hopeful and confident of a favorable therapeutic outcome.

Indirect effects via caregiving burden

We found that parents' perceptions of illness (severity and interference in the child's life) were indirectly linked to parents' QoL via caregiving burden, confirming our fourth hypothesis. Litzelman and colleagues (2011) found that caregiving burden and stress mediated the relationship between child impairment or functional limitations, rated by parents of children with cancer and brain tumors, and parental QoL. Our research strengthens these results and adds to them by showing that in addition to parents' perception of illness interference in their child's life, their perception about the illness severity is also indirectly linked to QoL via caregiving burden.

Although there is little research on the links between illness perceptions and the burden of the caregiver, according to the caregiving process and caregiver burden model, certain characteristics of the child (e.g., level of dependence in daily activities) increase the demands upon the caregiver, and consequently add to the burden they experience (Raina et al., 2004). It is possible that when parents believe that the illness has a greater impact on their child's life, they also perceive a higher degree of the child's dependence on them. Furthermore, if the child's illness is perceived as a health condition involving severe medical risks, parents may struggle with a more complex and demanding regime of treatment to which children may adhere, placing extra supportive care on them. The focus of attention is monitoring the child's health and well-being, minimizing the disruptions in daily routines, and providing emotional and instrumental support (Patterson et al., 2004; Young et al., 2002). For parents, simultaneously dealing with different emotional and practical concerns may lead to role strain (Young et al., 2002), which, in turn, may negatively influence their QoL (Klassen et al., 2011).

At the same time, parents must balance other family roles (e.g., giving support to other children and their spouse) and occupational roles (Patterson et al., 2004). These roles might also be impaired due to caregiving demands. Studies on the context of caregivers of children with cerebral palsy and with asthma support this argument, showing that higher levels of caregiving burden were linked to compromised QoL through the deterioration of social support (Carona, Crespo, & Canavarro, 2013) and family relationships (Crespo, Silva, Carona, Canavarro, & Dattilio, 2011). In addition to the negative impact on these interpersonal supportive sources, caregiving burden is also likely to interfere with intrapersonal factors. Parents may see their abilities to function in other roles as compromised (Young et al., 2002), an

awareness which may be associated with worse self-perception. Previously research has shown that excessive caregiving demands are associated with a lower sense of mastery and self-esteem, which can, in turn, negatively impact parents' QoL (Klassen et al., 2011).

Limitations and Strengths

One of the limitations of the present research is that we used a cross-sectional study design which did not allow for the examination of the direction of causality among the study variables. Although the relationships we tested in the two mediation models were hypothesized according to past literature, it is possible, for instance, that caregiving burden is associated with more negative perceptions about the child's illness, which in turn, are linked to worse QoL. Another potential methodological concern was the use of two single-item scales to assess parents' perception of the illness. Future research should use multiple-item measures to capture the complexity of parents' perceptions about childhood cancer experience. A further limitation arises from the differences between the two groups of parents in time elapsed since diagnosis, which was longer for parents of children off-treatment. Time since diagnosis is considered an important factor in understanding the family adaptation to chronic illness (Rolland, 1987, 2012). Therefore, results should be interpreted with caution, because the role of the clinical situation may interact with time since diagnosis. Additionally, children in the on-treatment group were receiving treatment considered as more intensive than children who had completed treatment. Given the important role of parents in implementing medical procedures and supportive care, this difference in treatment intensity may also contribute to explain the higher levels of caregiving burden reported by parents of children on-treatment. Finally, in our study parents of children in palliative treatment were excluded, so results cannot be generalized to parents in this situation. Future research should address the caregiving experience and adaptation outcomes of parents of children with malignancies known to be incurable. However, this study also has important strengths. According to Kazak (2009), few studies include subjective perceptions of parents about their child's illness, which can be significant aspects for understanding families' adaptation. This study provides insight into caregiving burden as a potential mechanism by which parents' perceptions of their child's health problem can adversely influence their QoL. A better understanding of the relations between illness perceptions and caregiving burden may help to identify those parents at higher risk of poorer QoL and provide insights for interventions with families, as described below.

Implications and conclusions

The present study's findings convey relevant implications for health professionals in pediatric oncology. First, results showed that parents' perceptions of the illness were associated with the burden they experience. Therefore, the health care team can be particularly attuned to those parents who hold more negative perceptions about their child's condition; providing

information about the illness and its treatment seems to be an important aspect for parents' adaptation. In addition, the links between caregiving burden and parents' QoL reinforce the need for health professionals to identify those who would benefit from additional support, namely informational, emotional and instrumental support, which have been pointed out as crucial aspects for coping with a challenging situation (Vrijmoet-Wiersma et al., 2008). Interventions with parents would be particularly important during the treatment active phase, when the child's health is perceived as more threatened and their dependence on caregivers may be at the highest. Such support may reduce caregiver burden and have a positive impact on parents' QoL. Given that parental and child adaptation are closely intertwined (Sameroff, 2009), identifying parents whose QoL may be compromised is paramount to promoting not only the parents' adaptation to the adverse context of childhood cancer, but also that of the whole family.

References

- Alderfer, M. A., & Kazak, A. E. (2006). Family issues when a child is on treatment for cancer. In R. Brown (Ed.), *Pediatric hematology/oncology: A biopsychosocial approach* (pp. 53-74). New York: Oxford University Press.
- Anderzén-Carlsson, A., Kihlgren M., Svantesson, M., & Sørli, V. (2007). Children's fear as experienced by the parents of children with cancer. *Journal of Pediatric Nursing*, 22(3), 233-244. doi:10.1016/j.pedn.2007.03.003
- Boman, K. K., Lindahl, A., & Björk, O. (2003). Disease-related distress in parents of children with cancer at various stages after the time of diagnosis. *Acta Oncologica*, 42(2), 137-146. doi:10.1080/02841860310004995
- Boman, K. K., Viksten, J., Kogner, P., & Samuelsson, U. (2004). Serious illness in childhood: The different threats of cancer and diabetes from a parent perspective. *The Journal of Pediatrics*, 145(3), 373-379. doi:10.1016/j.jpeds.2004.05.043
- Carona, C., Crespo, C., & Canavarro, M. C. (2013). Similarities amid the difference: Caregiving burden and adaptation outcomes in dyads of parents and their children with and without cerebral palsy. *Research in Developmental Disabilities*, 34(3), 882-893. doi:10.1016/j.ridd.2012.12.004
- Carona, C., Faria-Morais, T., Nazaré, B., & Canavarro, M. C. (2008). *Escalas de desgaste do cuidador- Revistas [Caregiver Burden Scales - Revised]*. Unpublished manuscript.
- Crespo, C., Carona, C., Silva, N., Canavarro, M. C., & Dattilio, F. (2011). Understanding the quality of life for parents and their children who have asthma: Family resources and challenges. *Contemporary Family Therapy*, 33(2), 179-196. doi:10.1007/s10591-011-9155-5
- Dahlquist, L. M., Czyzewski, D. I., & Jones, C. L. (1996). Parents of

- children with cancer: A longitudinal study of emotional distress, coping style, and marital adjustment two and twenty months after diagnosis. *Journal of Pediatric Psychology*, 21(4), 541-554. doi:10.1093/jpepsy/21.4.541
- Dockerty, J. D., Williams, S. M., McGee, R., & Skegg, D. C. G. (2000). Impact of childhood cancer on the mental health of parents. *Medical and Pediatric Oncology*, 35(5), 475-483. doi:10.1002/1096-911X(20001101)35:5<475::AID-MPO6>3.0.CO;2-U
- Eiser, C., Eiser, J. R., & Stride, C. B. (2005). Quality of life in children newly diagnosed with cancer and their mothers. *Health and Quality of Life Outcomes*, 3(29), 1-5. doi:10.1186/1477-7525-3-29
- Fletcher, P.C. (2010). My child has cancer: The costs of mothers' experiences of having a child with pediatric cancer. *Issues in Comprehensive Pediatric Nursing*, 33(3), 164-184. doi:10.3109/01460862.2010.498698
- Grootenhuis, M. A., & Last, B. F. (1997a). Adjustment and coping by parents of children with cancer: A review of the literature. *Supportive Care in Cancer*, 5(6), 466-484. doi:10.1007/s005200050116
- Grootenhuis, M. A., & Last, B. F. (1997b). Parents' emotional reactions related to different prospects for the survival of their children with cancer. *Journal of Psychosocial Oncology*, 15(1), 43-62. doi:10.1300/J077v15n01_04
- Hayes, A. F. (2013). *Introduction to Mediation, Moderation, and Conditional Process Analysis*. New York: Guilford Press.
- Hung, J. W., Wu, Y.-H., & Yeh, C.-H. (2004). Comparing stress levels of parents of children with cancer and parents of children with physical disabilities. *Psycho-Oncology*, 13(12) 898-903. doi:10.1002/pon.868
- Jose, P. (2008). *ModGraph-I: A programme to compute cell means for the graphical display of moderational analyses: The internet version, Version 2.0*. Retrieved from <http://www.victoria.ac.nz/psyc/paul-jose-files/modgraph/>
- Kazak, A. (1989). Families of chronically ill children: A systems and social ecological model of adaptation and challenge. *Journal of Consulting and Clinical Psychology*, 57(1), 25-30. doi:10.1037/0022-006X.57.1.25
- Kazak, A. E., & Barakat, L. P. (1997). Brief report: Parenting stress and quality of life during treatment for childhood leukemia predicts child and parent adjustment after treatment ends. *Journal of Pediatric Psychology*, 22(5), 749-758. doi:10.1093/jpepsy/22.5.749
- Kazak, A. E., Hocking, M. C., Ittenbach, R. F., Meadows, A. T., Hobbie, B., DeRosa, B. W., ... Reilly, A. (2011). A revision of the intensity of treatment rating scale: Classifying the intensity of pediatric cancer treatment, *Pediatric Blood Cancer*, 59(1), 96-99.

- doi:10.1002/psc.23320
- Kazak, A. E., Rourke, M. T., & Navsaria, N. (2009). Families and other systems in pediatric psychology. In M. Roberts & R. Steele (Eds.), *Handbook of pediatric psychology* (pp. 656 - 671). New York: Guilford.
- Klassen, A. F., Klaassen, R., Dix, D., Pritchard, S., Yanofsky, R., O'Donnell, M., ... Sung, L. (2008). Impact of caring for a child with cancer on parents' health-related quality of life. *Journal of Clinical Oncology*, 26(36), 5884-5889. doi:10.1200/JCO.2007.15.2835
- Klassen, A. F., Raina, P., McIntosh, C., Sung, L., Klaassen, R. J., O'Donnell, M., ... Dix, D. (2011). Parents of children with cancer: Which factors explain differences in health-related quality of life. *International Journal of Cancer*, 129(5), 1190-1198. doi:10.1002/ijc.25737
- Klassen, A., Raina, P., Reineking, S., Dix, D., Pritchard, S., & O'Donnell, M. (2007). Developing a literature base to understand the caregiving experience of parents of children with cancer: A systematic review of factors related to parental health and wellbeing. *Supportive Care in Cancer*, 15(7), 807-818.
- Kylmä, J. & Juvakka, T. (2007). Hope in parents of adolescents with cancer: Factors endangering and engendering parental hope. *European Journal of Oncology Nursing*, 11(3), 262-271. doi:10.1016/j.ejon.2006.06.007
- Litzelman, K., Catrine, K., Gangnon, R., & Witt, W. P. (2011). Quality of life among parents of children with cancer or brain tumors: The impact of child characteristics and parental psychosocial factors. *Quality of Life Research*, 20(8), 1261-1269. doi:10.1007/s11136-011-9854-2
- McCaffrey, C. N. (2006). Major stressors and their effects on the well-being of children with cancer. *Journal of Pediatric Nursing*, 21(1), 59-66. doi:10.1016/j.pedn.2005.07.003
- McGrath, P., Paton, M. A., & Huff, N. (2005). Beginning treatment for pediatric acute myeloid leukemia: The family connection. *Issues in Comprehensive Pediatric Nursing*, 28(2), 97-114. doi:10.1080/01460860590950881
- Mercer, M., & Ritchie, J. A. (1997). Tag-team parenting of children with cancer. *Journal of Pediatric Nursing*, 12(6), 331-341. doi:10.1016/S0882-5963(97)80059-0
- Michaud, P.-A., Suris, J. C., & Viner, R. (2007). *The adolescent with a chronic condition: Epidemiology, developmental issues and health care provision*. Geneva: World Health Organization.
- Montgomery, R. J. V., & Kosloski, K. (2006). *The league of experienced family caregivers: Measure development*. Milwaukee, WI: University of Wisconsin-Milwaukee.
- Othman, A., Mohamad, N., Hussin, Z. A., & Blunden, S. (2011). Factors related to parental well being in children with cancer. *International*

- Proceedings of Economics Development & Research*, 5(1), 255-260.
- Patterson, J. M., Holm, K. E., & Gurney, J. G. (2004). The impact of childhood cancer on the family: A qualitative analysis of strains, resources, and coping behaviors. *Psycho-oncology*, 13(6), 390-407. doi:10.1002/pon.761
- Pereira, M., Melo, C., Gameiro, S., & Canavarro, M. C. (2011). Estudos psicométricos da versão em português europeu do índice de qualidade e vida EUROHIS-QOL-8 [Psychometric studies of the European Portuguese version of the quality of life index EUROHIS-QOL-8]. *Laboratório de Psicologia*, 9, 109-123.
- Preacher, K.J., & Hayes, A.F. (2008). Asymptotic and resampling strategies for assessing and comparing indirect effects in multiple mediator models. *Behavior Research Methods*, 40(3), 879–891. doi:10.3758/BRM.40.3.879
- Raina, P., O'Donnell, M., Schweltnus, H., Rosenbaum, P., King, G., Brehaut, J., ... Wood, E. (2004). Caregiving process and caregiving burden: Conceptual models to guide research and practice. *BioMed Central Pediatrics*, 4(1), 1-13. doi:10.1186/1471-2431-4-1
- Robinson, K. E., Gerhardt, C. A., Vannatta, K., & Noll, R. B. (2007). Parent and family factors associated with child adjustment to pediatric cancer. *Journal of Pediatric Psychology*, 32(4), 400-410. doi:10.1093/jpepsy/jsl038
- Rolland, J. S. (1987). Family illness paradigms: Evolution and significance. *Family Systems Medicine*, 5(4), 467-486. doi:10.1093/jpepsy/jsl038
- Rolland, J. S. (2012). Mastering family challenges in serious illness and disability. In F. Walsh (Ed.), *Normal Family Processes* (pp. 452-482). New York: The Guilford Press.
- Sameroff, A. J. (2009). *The transactional model of development: How children and contexts shape each other*. Washington, DC: American Psychological Association.
- Santos, S., Crespo, C., Canavarro, M. C., & Pinto, A. Versão portuguesa da escala de classificação da intensidade do tratamento 3.0 no cancro pediátrico [Portuguese version of intensity of treatment rating scale 3.0 in pediatric cancer]. Manuscript in preparation.
- Schmidt, S., Mühlhan, H., & Power, M. (2006). The EUROHIS-QOL 8-item index: Psychometric results of a cross-cultural field study. *European Journal of Public Health*, 16(4), 420–428. doi:10.1093/eurpub/cki155
- Simões, M. R. (1994). *Investigações no âmbito da aferição nacional do teste das Matrizes Progressivas de Raven [Raven's Progressive Matrices: Aferition studies]*. Unpublished Doctoral Dissertation, Faculdade de Psicologia e de Ciências da Educação, Universidade de Coimbra, Coimbra, Portugal
- Turner-Henson, A., Holaday, B., & Swan, J. H. (1992). When parenting becomes caregiving: Caring for the chronically ill child. *Family &*

- Community Health*, 15(2), 19-30. doi:10.1097/00003727-199207000-00006
- von Essen, L., Sjöden, P. O., & Mattsson, E. (2004). Swedish mothers and fathers of a child diagnosed with cancer. *Acta Oncologica*, 43(5), 474-479. doi:10.1080/02841860410029348
- Vrijmoet-Wiersma, C. M. J., van Klink, J. M. M., Kolk, A. M., Koopman, H. M., Ball, L. M., & Egeler, R. M. (2008). Assessment of parental psychological stress in pediatric cancer: A review. *Journal of Pediatric Psychology*, 33(7), 694–706. doi:10.1093/jpepsy/jsn007
- Witt, W. P., Litzelman, K., Wisk, L. E., Spear, J. A., Catrine, K., Levin, N., & Gottlieb, C. A. (2010). Stress-mediated quality of life outcomes in parents of childhood cancer and brain tumor survivors: A case-control study. *Quality of Life Research*, 19(7), 995-1005. doi:10.1007/s11136-010-9666-9
- Wolfe-Christensen, C., Mullins, L. L., Fedele, D. A., Rambo, P. L., Eddington, A., & Carpentier, M. Y. (2010). The relation of caregiver demand to adjustment outcomes in children with cancer: The moderating role of parenting stress. *Children's Health Care*, 39(2), 108-124. doi:10.1080/02739611003679881
- Yeh, C.-H. (2002). Life experience of Taiwanese adolescents with cancer. *Scandinavian Journal of Caring Science*, 16(3), 232-239. doi:10.1046/j.1471-6712.2002.00066.x
- Young, B., Dixon-Woods, M., Findlay, M., & Heney, D. (2002). Parenting in a crisis: Conceptualising mothers of children with cancer. *Social Science & Medicine*, 55(10), 1835-1847. doi:10.1016/S0277-9536(01)00318-5

Conclusão

Os resultados do presente estudo mostraram que, como esperado, os pais de crianças em tratamento, comparativamente aos pais de crianças fora de tratamento, percecionavam a doença como mais grave e interferente na vida da criança e experienciavam maior sobrecarga e pior QdV. Para ambos os grupos, os resultados mostraram que pais que percecionavam a doença como mais interferente na vida da criança reportavam pior QdV. No caso da perceção da gravidade da doença, a sua associação com a QdV não se mostrou significativa. Os resultados sugerem ainda que quando os pais percecionavam a doença como mais grave e interferente, reportavam maior sobrecarga e, conseqüentemente pior QdV. Por fim, a associação entre a sobrecarga e a QdV mostrou ser mais forte para os pais de crianças em tratamento.

Estes resultados podem auxiliar no planeamento e implementação de estratégias de intervenção empiricamente sustentadas e direcionadas para os pais de crianças com cancro. Em primeiro lugar, as significações que os pais atribuem à doença parecem promover níveis de adaptação diferentes. Por essa razão, os profissionais de saúde têm um importante papel na identificação daqueles pais que beneficiariam de informação adequada e realista acerca da doença e dos procedimentos médicos, no sentido de amortecer o efeito adverso de perceções negativas acerca do estado de saúde e interferência da doença na vida da criança. Em segundo lugar, os pais, enquanto principais cuidadores, poderiam beneficiar de intervenções que reduzissem a sobrecarga decorrente dos cuidados com a criança. Assim, a prestação de apoio instrumental e emocional poderia ser útil na minimização da influência negativa da ‘carreira inesperada’ destes pais. Por fim, este tipo de intervenções poderia ser particularmente útil no período em que a criança está em tratamento, pois a condição de saúde mais debilitada e a intensificação dos cuidados médicos com a criança aumentam as responsabilidades dos pais, interferindo negativamente na sua QdV.

Entendemos que a equipa de profissionais de saúde se confronta com o desafio de estar particularmente atenta aos pais em maior risco, garantindo e adequando as ações de intervenção às necessidades específicas de cada pai e família em diferentes fases da doença. A inclusão dos pais no processo da doença e do tratamento promove não só o bem-estar do indivíduo mas, simultaneamente, a adaptação da criança e de todo o sistema familiar à experiência adversa do cancro pediátrico. Efetivamente, intervir com os pais da criança doente, bem como com outros membros da família, é um dos importantes focos da Psicologia Pediátrica (Kazak et al., 2009).

Se é verdade que grande parte da investigação e programas de intervenção no âmbito da oncologia pediátrica se tem focado na criança doente, também é verdade que temos assistido a um crescente interesse em compreender a vivência parental da doença oncológica. O diagnóstico de uma doença crónica num membro da família impõe desafios importantes ao indivíduo doente, mas também aos restantes membros da família (Rolland, 1987, 2012), principalmente quando a doença exige cuidados especiais, como é o caso do cancro pediátrico. Kazak (1989), aplicando a perspetiva socio-ecológica ao contexto das condições crónicas pediátricas, defende que a adaptação da criança à doença é determinada pelos seus contextos sociais, nomeadamente os pais, os irmãos, a escola. De facto, os estudos têm salientado a existência de uma relação entre o bem-estar parental e a

adaptação da criança com cancro (Robinson et al., 2007; Wolfe-Christensen et al., 2010). Por essa razão, compreender os fatores promotores de uma melhor adaptação parental torna-se fundamental quer para os próprios pais enquanto indivíduos, quer também, de uma forma indireta, para as crianças com doença oncológica.

Referências adicionais

- Kaatsch, P. (2010). Epidemiology of childhood cancer. *Cancer Treatment Reviews*, 36(4), 277-285. doi:10.1016/j.ctrv.2010.02.003
- Long, K. A., & Marsland, A.L. (2011). Family Adjustment to Childhood Cancer: A Systematic Review. *Clinical Child and Family Psychology Review*, 14(1), 57-88. doi:10.1007/s10567-010-0082-z
- Roberts, M. C., & Steele, R. C. (Eds.). (2009). *Handbook of pediatric psychology* (4th ed.). New York: Guilford.

Tabelas

Table 1

Socio-demographic and clinical characteristics of the sample's two clinical groups: on-treatment and off-treatment

	On-Treatment Group (<i>n</i> = 126)	Off-Treatment Group (<i>n</i> = 151)	Differences between groups
Age (<i>M/SD</i>)	41.73 (6.25)	42.86 (6.64)	$t = 1.45; p >.05$
Gender (<i>n</i>)			
Male	25	23	
Female	101	128	$\chi^2_{(1)} = .72; p >.05$
Marital status (<i>n</i>)			
Single	5	4	
Married	94	131	
Cohabiting	9	5	
Divorced	2	6	
Separated	13	-	
Widowed	3	5	$\chi^2_{(5)} = 10.24; p >.05$
SES (<i>n</i>)			
Low	68	86	
Medium	42	53	
High	16	12	$\chi^2_{(2)} = 1.71; p >.05$
Child's Age (<i>M/SD</i>)	12.69 (3.55)	13.54 (3.47)	$t = 2.00; p = .05$
Child's Gender (<i>n</i>)			
Male	72	73	
Female	54	78	$\chi^2_{(1)} = 1.79; p >.05$
Time since diagnosis in months (<i>M/SD</i>)	15.75 (19.58)	40.23 (26.05)	$t = 8.69; p < .001$
Intensity of Treatment (<i>n</i>)			
1 "least intensive"	3	4	
2 "moderately intensive"	34	65	
3 "very intensive"	65	55	
4 "most intensive"	22	21	$\chi^2_{(3)} = 10.30; p <.05$
Missing	2	3	

Table 2

Descriptive statistics and matrix of inter-correlations among study variables for on-treatment group (figures in bold font) and off-treatment group (figures in non-bold font)

Variable	Parents					Children	
	1	2	3	4	5	6	7
<i>Parents</i>							
1. Perception of Child's Illness Severity							
2. Perception of Interference in the Child's Life	.45*** /.50***						
3. Caregiving Burden	.19* /.20*	.39*** /.35***					
4. QoL	-.04 /.06	-.26** /.25**	-.54*** /.23**				
5. Age	-.11 /.03	.03 /.06	.02 /.01	-.02 /.12			
<i>Children</i>							
6. Gender	.05 /.11	.11 /.18*	.09 /.11	-.09 /.08	-.01 /.01		
7. Age	.01 /.02	.13 /.05	.10 /.05	-.09 /.14	.46*** /.44***	.17 /.08	
Mean	7.50 /5.79	7.26 /4.62	3.24 /2.71	3.42 /3.64	41.73 /42.86		12.69 /13.54
SD	2.19 /2.69	2.38 /2.90	.93 /.98	.53 /.53	5.25 /6.64		3.55 /3.47
<i>Cronbach's alpha</i>			.87 /.88	.83 /.84			

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

Figuras

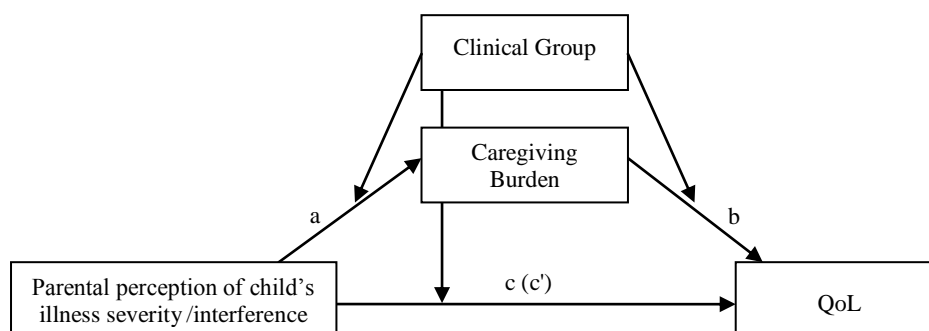


Figure 1. The proposed conceptual model depicting clinical group according treatment status as moderator of the link between parents' perception of child's illness severity/interference and QoL mediated by caregiving burden.

Notes. Path a: The effect of the independent variable (IV) on the proposed mediator (M); Path b: The effect of M on the dependent variable (DV) partialling out the effect of IV; Path c: The total effect of IV on DV; Path c': The direct effect of IV on DV after controlling for M.

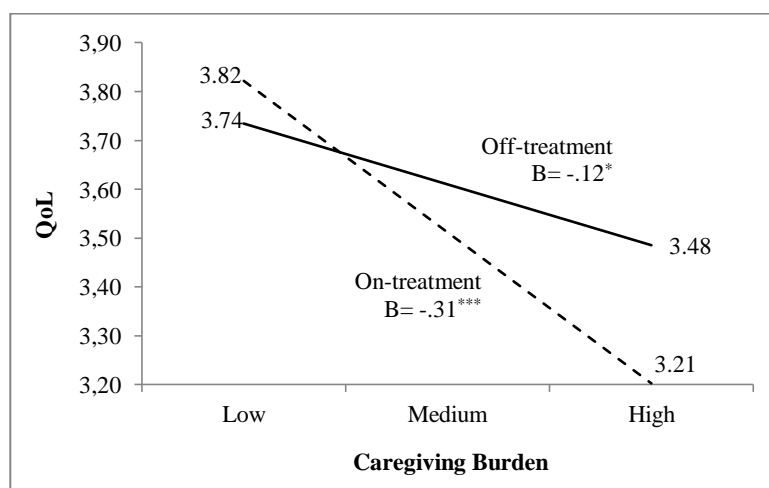


Figure 2. The moderating effect of clinical group (in-treatment and off-treatment) on the link between caregiving burden and QoL.

Note. The values represent maximum and minimum of QoL for each clinical group.

* $p < .05$. *** $p < .001$ for simple slope (B).

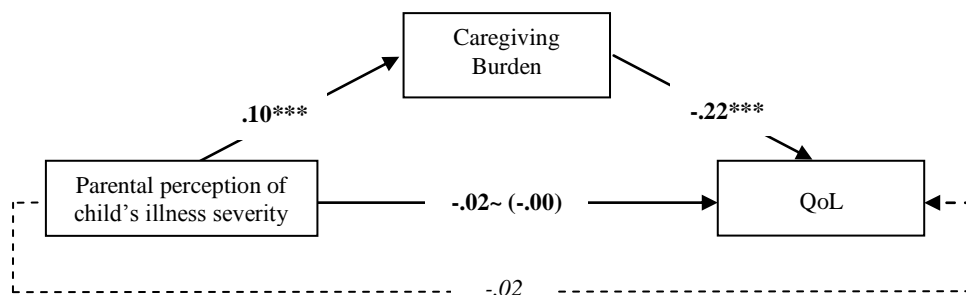


Figure 3. Model 1, depicting mediating effect of caregiving burden on the link between parents' perception of the child's illness severity and QoL.

Notes. The value inside parentheses represents the direct effect of the independent variable on the dependent variable after controlling for the mediator. Non italic bold figures represent unstandardized coefficients for direct paths; italic non-bold figure represents unstandardized coefficient for the indirect path.

$\sim p = .06$. *** $p < .001$; the indirect effect was significant at 95% CI [-.04/-.01].

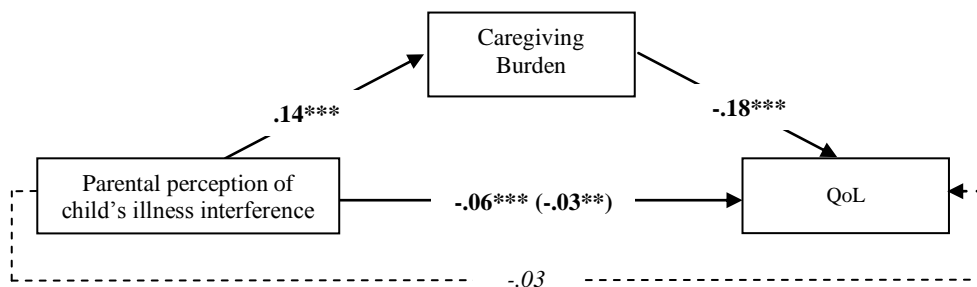


Figure 4. Model 2, depicting the mediating effects of caregiving burden on the link between parents' perception of the illness interference on the child's life and QoL.

Notes. The value inside parentheses represent the direct effect of independent variable on dependent variable after controlling for the mediator. Non italic bold figures represent unstandardized coefficients for direct paths; italic non-bold figure represents unstandardized coefficient for the indirect path.

** $p < .01$. *** $p < .001$; the indirect effect was significant at 95% CI [-.04/-.01].