Emotional distress in parents of long-term survivors of childhood acute lymphoblastic leukemia

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Abstract

Objective: The current study investigated the occurrence of emotional distress in parents of long-term survivors of childhood acute lymphoblastic leukemia (ALL) and identified factors associated with parent emotional distress symptoms.

Methods: Parents of 127 long-term survivors of childhood ALL treated on a chemotherapy-only protocol at St. Jude Children's Research Hospital participated in the study. Parents completed standard ratings of emotional distress, caregiver strain, and child physical, emotional, and psychosocial functioning. Multivariable hierarchical linear regression analyses were used to examine associations between symptoms of caregiver strain, survivor functioning, and parent emotional distress. Covariates included parent education, survivor age, survivor sex, and time since childhood cancer diagnosis.

Results: On average, few parents reported significant symptoms of emotional distress. Clinically significant levels of anxiety and depression were reported by 7.1% and 3.1% of parents, respectively. Only 3.9% of parents endorsed significant symptoms of posttraumatic stress. Perceived caregiver strain was significantly associated with symptoms of parent anxiety, depression, and posttraumatic stress. Parent-report of child emotional functioning was significantly associated with symptoms of parent anxiety.

Conclusions: Most parents of long-term survivors of ALL exhibit low levels of emotional distress in the context of rates observed in the general population. Perceived caregiver strain was significantly associated with parent emotional distress. Further research is required to examine specific sources of caregiver strain, as well as other risk and protective factors associated with parent emotional distress symptoms.

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Introduction

Significant progress has been made in the treatment of childhood cancer [1]. Long-term survival rates for acute lymphoblastic leukemia (ALL), the most common form of childhood cancer, have improved from less than 10% in the 1960s to over 90% today [2,3]. Thus, the majority of children diagnosed with ALL are living into adulthood to join a growing population of long-term childhood cancer survivors [4]. However, modern treatment is not without cost. Survivors are at risk for treatment-related late-effects including organ dysfunction [5,6], neurocognitive impairment [7,8], emotional distress [9], and reduced quality of life [10,11]. These morbidities suggest that the impact of childhood ALL continues well past its cure.

While recent literature has emphasized long-term follow-up of survivors, relatively little attention has been paid to the long-term impact of survivorship on parent functioning. Despite high cure rates, the diagnosis and treatment of childhood ALL remain a potential stressor for parents and families [12]. Research has consistently reported elevated levels of parental emotional distress at diagnosis with declining levels throughout treatment [13,14]; however, parent emotional distress through the course of long-term survivorship is less well understood. Studies of posttraumatic stress symptoms (PTSS) in parents of childhood cancer survivors have garnered attention, but yield varied results [15]. For example, while some reports suggest that up to 43.7% of parents of survivors of childhood cancer experience moderate to severe PTSS [16–19], others indicate that they have significantly lower levels of PTSS than parents of healthy controls [20]. The prevalence of symptoms of depression and anxiety in parents of childhood cancer survivors is difficult to estimate as past study samples have differed with respect to disease type, location, and treatment modality. Moreover,

methodological differences related to assessment of symptoms are apparent. This underscores the need for additional research with homogeneous samples to better characterize prevalence rates of distress symptoms, clarify discrepancies reported in previous studies, and identify factors that place parents at increased risk for emotional distress symptoms.

Long-term survivorship may involve new stressors associated with treatment-related late effects that have the potential to adversely affect child and parent functioning. Peterson and Drotar [21] have speculated that late effects of childhood cancer treatment may be associated with reduced child quality of life and increased caregiver strain (i.e. difficulties associated with caring for a child), each of which may independently contribute to ongoing parent emotional distress. Caregiver strain may be especially salient in survivors of childhood cancer who are at risk of multiple organ dysfunction (i.e. endocrinopathies, cardiopulmonary complications), neurocognitive impairment (i.e. memory problems, behavioral dysregulation), and emotional distress (i.e. anxiety), each of which may independently contribute to parent emotional distress. Studies in non-cancer pediatric populations have demonstrated associations between child functioning, caregiver strain, and parent stress [22,23]. Specifically, decreased child functioning has been associated with increased caregiver strain and increased parent emotional distress. To our knowledge, these relations have yet to be examined in parents of childhood ALL survivors. Nevertheless, theoretical models posit that among children with physical illness, maternal mental health may be directly influenced by child disease parameters (i.e. brain involvement), strain associated with providing child care, and social–ecological factors including child adaptation and demographic factors [24,25]

The current study sought to examine the prevalence of emotional distress in parents of long-term survivors of childhood ALL, and to investigate potential associations among symptoms of child functioning, caregiver strain, and parent emotional distress symptoms. We hypothesized that the majority of parents of long-term survivors would not report significant emotional distress. We further hypothesized that parent perception of child functioning and caregiver strain would be associated with parent emotional distress symptoms, including symptoms of anxiety, depression, and posttraumatic stress.

Method

Participants

Long-term survivors of childhood ALL treated at St. Jude Children's Research Hospital (SJCRH) and their parents were approached to take part in the study. Eligible survivors were treated on a chemotherapy only protocol without the use of prophylactic radiation (Total XV) [3], were a minimum of five years post diagnosis and at least eight years of age at the time of evaluation. Per the Total XV protocol, survivors were treated on either a low risk or standard/high risk protocol. Patients were treated on the standard/high risk arm if they had evidence of CNS involvement at diagnosis. Treatment regimens were similar across protocols and included triple intrathecal chemotherapy (i.e. methotrexate, hydrocortisone, and cytarabine) and high dose intravenous methotrexate. However, standard/high risk patients received higher amounts of high dose intravenous methotrexate and underwent a reintensification treatment phase following consolidation, during which they received more triple intrathecal doses. Survivors were excluded if they had a history of head injury, neurological condition unrelated to ALL treatment, or genetic disorder associated with neurocognitive impairment. A total of 192 parents of children <18 years of age were recruited to complete measures of parent emotional distress, caregiver strain, and child functioning. Among these, 7 families were lost to follow-up, 2 were pending evaluation at the time of data analysis, 2 survivors completed only a portion of the study, and 54 declined. One hundred twenty seven parents (66.1%) participated in the study. Only one parent (i.e. mother or father) completed measures for each child.

Procedure

Study approval was obtained from the SJCRH Institutional Review Board. All potentially eligible patients were contacted by telephone or during a clinic visit to determine interest in study participation. Informed parent consent and child assent were obtained prior to study participation. Survivors and their parents participated in the study during their annual follow-up visit to SJCRH. This visit is an established aspect of survivorship care provided to patients treated at SJCRH who are at least five years post-diagnosis. Parents were recruited to complete measures of parent emotional distress, caregiver strain, and child functioning. A licensed psychologist was present to assist with questionnaire completion, to answer questions, and to provide follow-up recommendations when needed. Data was collected from February 2010 to December 2012.

Measures

Primary outcomes

Parent anxiety and depressive symptoms: Parent emotional distress was assessed using the Brief Symptom Inventory 18 (BSI-18) [26]. Parents responded to 12 questions measuring anxiety and depressive symptomology over the past 7 days. Each dimension contains 6 items rated on a five-point Likert scale (0 *Not at all* to 4 *Extremely*) with higher scores indicating greater levels of emotional

distress. Responses were summed for each dimension to obtain raw scores [26]. Raw scores were converted into sex specific T-scores using national normative data derived from adults in the general population. T-scores >63 were considered to represent clinically significant emotional distress. Using this threshold 10% of the general population would be expected to report clinically significant emotional distress symptoms. The BSI-18 has shown adequate internal consistency (α=0.74–0.91) and test–retest reliability (range: 0.68–0.90) [26,27].

Parent posttraumatic stress symptoms: Symptoms of parent posttraumatic stress were evaluated using The Impact of Event Scale-Revised (IES-R) [28]. The IES-R is a 22-item scale measuring current subjective response to a specific trauma [28]. The scale is comprised of three subscales corresponding to the three criteria for posttraumatic stress disorder identified in the Diagnostic and Statistical Manual of Mental Disorders-IV-Text Revision (DSM-IV): avoidance, hyperarousal, and intrusion [29]. Responses for each scale are measured on a five-point Likert type scale (0 *Not at all* to 4 *Extremely*). The sum of items in each subscale, as well as a total score of all items, was calculated with higher scores indicating higher levels of stress. Overall levels of PTSS are calculated following DSM-IV criteria; 1 symptomatic score in intrusion, 3 symptomatic scores in avoidance, and 2 scores in hyperarousal represent clinically significant levels of PTSS [29]. The IES-R has shown high internal consistency (α=0.96; intrusion: 0.87–0.94; avoidance: 0.84–0.87; hyperarousal: 0.79–0.91) [28,30,31].

Independent variables and covariates

Caregiver strain: The Caregiver Strain Questionnaire (CGSQ) is a 21-item questionnaire designed to measure parent strain associated with caring for a child [32]. Sources of strain assessed include: demands on time, disruption of relationships, negative health effects, financial burden, sacrifice, disruption of social life, worry and guilt, fatigue, and embarrassment [32]. Parents were asked to rate on a five-point Likert scale (0 *Not at all* to 4 *Very Much*) how much of a problem each item has been over the past 6 months. Responses reflect 3 dimensions used to calculate an overall measure of caregiver strain: externalized subjective caregiver strain, internalized subjective caregiver strain, and objective caregiver strain [32]. Internal consistency across subscales is adequate (alpha range 0.74–0.93) [32].

Child functioning: Parents reported on their child's functioning using the Pediatric Quality of Life Inventory (PedsQL) 4.0 Generic Core Scales parent report, a measure designed to assess health related quality of life (HRQOL) in children and adolescents [33]. Two versions of the parent report PedsQL were used in this study, the 8–12-

year-old proxy version and the 13–18-year-old proxy version. These versions measure identical features of HRQOL. The questionnaire contains 23 items reflecting four dimensions of child functioning: physical, emotional, social, and school functioning. Parents were asked to rate how frequently each item has been a problem for their child over the past month. Items were rated using a five-point Likert scale (0 *Never* to 4 *Almost Always*) and were reverse scored and linearly transformed from 0 to 100, with higher scores reflecting better HRQOL [33]. The PedsQL parent report has shown high internal consistency across subscales (α=0.73–0.93) when used in cancer populations [34].

The PedsQL Cancer Module (PedsQL-Cancer) parent report is a 27-item questionnaire used to measure HRQOL in childhood cancer patients and survivors [34]. The questionnaire is comprised of 8 scales: pain and hurt, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance, and communication. Nausea, procedural anxiety, treatment anxiety, and communication were not evaluated in the current study due to limited applicability in a long-term survivorship population. PedsQL-Cancer scoring methods are consistent with the PedsQL [34]. High internal consistency has been demonstrated across subscales (α=0.81–0.93) [34].

Select PedsQL and PedsQL-Cancer parent report subscales were combined to create 4 child functioning composites. Constructs covering similar domains of functioning were paired to reduce subsequent modeling. Four composites were evaluated: physical functioning (physical functioning and pain and hurt), emotional functioning (emotional functioning and worry), social functioning (social functioning and perceived physical appearance), and cognitive functioning (school functioning and cognitive problems). Physical functioning (Cronbach's α=0.87) and social functioning (Cronbach's α=0.83) displayed good reliability. Reliability for emotional functioning (Cronbach's α=0.90) and cognitive functioning (Cronbach's α=0.92) was excellent.

Covariates: Parent and survivor demographic characteristics were included as covariates. Specifically, parent sex, child sex, mother education (e.g. 12 years=high school, 14 years=some college, 16 years=bachelor's degree), child age, age at diagnosis, time since diagnosis, and treatment risk status (i.e. low vs. standard/high) were abstracted from the child's medical record and through parent report.

Statistical analysis

Questionnaires were scored according to published methods, and values were calculated for each exposure and outcome variable. If less than half of the items comprising a variable were missing, item responses were

imputed using the mean of endorsed items for that variable. Descriptive statistics for demographic and questionnaire data were calculated. Prevalence of parent emotional distress was assessed by calculating the frequency of parent posttraumatic stress, anxiety, and depressive symptoms defined by scores exceeding pre-specified levels (e.g. T-score >63). To investigate the association between parent report of child functioning, caregiver strain, and parent emotional distress symptoms, we performed a series of hierarchical linear regression analyses. Entry order for the hierarchical regression models was theory driven [35] and based on causal priority. Covariates known to influence parent emotional distress outcomes were adjusted for in each model and entered in the first step (i.e. child age, child sex, time since diagnosis, mother education). The second step consisted of one of the four child functioning composites (physical, emotional, social, and cognitive). Caregiver strain was always entered in the final step of each model. This method was repeated for each parent emotional distress outcome (anxiety, depression, and posttraumatic stress). Separate models were examined for each child quality of life domain because of concerns with multicollinearity as well as for the purpose of examining the unique contribution of specific dimensions of child functioning to parent emotional distress. Results were not adjusted for multiple comparisons [35,36].

Results

The majority of participating caregivers were mothers (79%). Mother education ranged from 8 to 20 years (mean = 13.9, SD = 2.5). Parent and survivor characteristics are presented in Table 1. Parents of survivors reported, on average, few clinically significant symptoms of emotional distress (depression: mean = 44.8, SD = 7.4; anxiety: mean = 45.6, SD = 9.8). Elevated symptoms of anxiety and depression (T-score ≥ 63) were reported by 7.1% and 3.1% of parents, respectively according to established cut-off

Table 1. Participant characteristics

	N (%)	Mean (SD)
Parent sex		
Female	100 (78.70)	
Male	27 (21.30)	
Child sex		
Female	67 (52.80)	
Male	60 (47.20)	
Treatment risk status		
Low	83 (65.87)	
Standard/high	43 (34.13)	
Child age (years)		12.30 (2.83)
Age at diagnosis (years)		4.65 (2.50)
Time since diagnosis (years)		7.64 (1.53)
Mother education (years)		13.85 (2.47)
Father education (years)		13.87 (3.10)

scores. Scores on the IES-R reflected minimal levels of posttraumatic stress symptomology (intrusion: mean = 5.8, SD = 6.3; avoidance: mean = 4.1, SD = 5.5; hyperarousal: mean = 2.5, SD = 4.2). Though a subset of parents reported symptoms of intrusion (28.3%), avoidance (6.3%), hyperarousal (10.2%), only 3.9% of parents reported globally elevated symptoms of PTSS. Parent-reported caregiver strain and child functioning are presented in Table 2. Strong positive correlations were observed among parent emotional distress symptoms (see Supplemental Table 1). In contrast, parent emotional distress symptoms were negatively correlated with parent report of child functioning (see Supplemental Table 2).

Anxiety symptoms

Multivariable hierarchical linear regression models were conducted to examine if caregiver strain was independently associated with parent anxiety after accounting for

Table 2. Parent emotional distress, caregiver strain, and child functioning

	Mean (SD)	Range
Parent emotional distress		
Anxiety[a]	45.6 (9.8)	38–81
Depression[a]	44.8 (7.4)	40–81
Intrusion[b]	5.8 (6.3)	0.0–29.0
Avoidance[b]	4.1 (5.5)	0.0–24.0
Hyperarousal[b]	2.5 (4.2)	0.0–20.0
Posttraumatic stress[b]	12.3 (15.0)	0.0–70.0
Parent strain[c]		
Objective	1.32 (.61)	1.0–3.9
Internalized subjective	1.99 (.99)	1.0–4.8
Externalized subjective	1.67 (.54)	1.0–3.3
Total strain	1.66 (.58)	1.0–4.0
Child functioning[d]		
Physical	86.86 (14.88)	37.5–100
Emotional	79.49 (20.02)	25–100
Social	85.24 (18.58)	0.0–100
School	75.08 (21.29)	15–100
Child functioning cancer[d]		
Pain	84.65 (19.99)	0.0–100
Worry	89.24 (20.87)	0.0–100
Physical appearance	78.87 (24.84)	0.0–100
Cognitive problems	68.69 (23.67)	0.0–100
Child functioning composites[d,e]		
Physical	86.42 (14.58)	0.0–100
Emotional	83.14 (18.40)	0.0–100
Social	82.83 (17.20)	0.0–100
Cognitive	71.88 (21.42)	0.0–100

[a]BSI-18: T-scores M = 50, SD = 10
[b]IES-R: item raw scores 0–4, higher scores = higher levels of stress.
[c]CGSQ: item raw scores 0–4, higher scores = higher caregiver strain.
[d]PedsQL and PedsQL-Cancer: Item raw scores 0–4 are reverse scored and linearly transformed from 0 to 100, higher scores = better health-related quality of life (HRQOL).
[e]Child Functioning Composites: PedsQL and PedsQL-Cancer subscales were combined to create 4 child functioning composites. Item raw scores 0–4 are reverse scored and linearly transformed from 0 to 100, higher scores = better HRQOL.

demographics and survivor functioning. Separate models were examined for each child functioning composite (i.e., physical, emotional, social, and cognitive). Demographics entered in Step 1 were differentially associated with anxiety across models. Specifically, mother education was significantly associated with anxiety in the physical and emotional functioning models but not in the social or cognitive models. The addition of survivor functioning composites in Step 2 significantly improved model fit (physical: $\Delta R^2 = 0.08$, $p = 0.002$; emotional: $\Delta R^2 = 0.14$, $p < 0.001$; social: $\Delta R^2 = 0.06$, $p = 0.008$; cognitive: $\Delta R^2 = 0.03$ $p = 0.05$), as did the addition of caregiver strain in Step 3 (physical:, $\Delta R^2 = 0.11$, $p < 0.001$; emotional: $\Delta R^2 = 0.07$, $p < 0.002$; social $\Delta R^2 = 0.12$, $p < 0.001$; cognitive: $\Delta R^2 = 0.15$, $p < 0.001$). In the final models including survivor physical functioning, social functioning, and cognitive functioning, only caregiver strain (all p's < 0.001) accounted for a significant proportion of variance in parental anxiety symptoms. However, in the final model for emotional functioning, both survivor emotional functioning ($p = 0.05$) and caregiver strain ($p = 0.002$) contributed significantly to parental anxiety symptoms. Results are provided in Supplemental Table 3.

Depressive symptoms

Multivariable hierarchical linear regression analyses were performed to examine the association between caregiver strain and parent depressive symptoms. Across analyses, demographics entered in Step 1 were not significantly associated with depression ($R^2 = 0.02$). However, in models including physical functioning, emotional functioning, and social functioning, the addition of child functioning in Step 2 significantly improved model fit (physical: $\Delta R^2 = 0.05$, $p = 0.01$; emotional: $\Delta R^2 = 0.14$, $p < 0.001$; social: $\Delta R^2 = 0.06$, $p = 0.01$). Child functioning was not statistically significant in the cognitive functioning model. The addition of caregiver strain in Step 3 improved model fit for all dimensions of child functioning (physical: $\Delta R^2 = 0.13$, $p = <0.001$; emotional: $\Delta R^2 = 0.07$, $p = 0.002$; social: $\Delta R^2 = 0.12$, $p < 0.001$; cognitive: $\Delta R^2 = 0.15$, $p < 0.001$). In the final models including survivor physical functioning, social functioning, and cognitive functioning, only caregiver strain (all p's < 0.001) accounted for a significant proportion of variance in parent depressive symptoms. However, in the final model for emotional functioning, both survivor emotional functioning ($p = 0.05$) and caregiver strain ($p = 0.002$) contributed significantly to parent depressive symptoms. Results are provided in Supplemental Table 4.

Posttraumatic stress symptoms

Multivariate hierarchical linear regression analysis was used to examine if caregiver strain was independently

Table 3. Multivariable hierarchical linear regression models for posttraumatic stress symptoms

Exposure	β	R^2	ΔR^2
3a. Physical functioning			
Step 1		0.03	
Mother education	0.04		
Child age	0.02		
Child sex	−0.03		
Time since diagnosis	−0.08		
Step 2		0.12	
Child physical functioning	0.004		0.09[†]
Step 3		0.47	
Caregiver strain	0.7[†]		0.35[†]
3b. Emotional functioning			
Step 1		0.03	
Mother education	0.04		
Child age	0.02		
Child sex	−0.03		
Time since diagnosis	−0.08		
Step 2		0.18	
Child emotional functioning	−0.02		0.15[†]
Step 3		0.47	
Caregiver strain	0.68[†]		0.30[†]
3c. Social functioning			
Step 1		0.03	
Mother education	0.06		
Child age	0.02		
Child sex	−0.03		
Time since diagnosis	−0.08		
Step 2		0.19	
Child social functioning	−0.08		0.16[†]
Step 3		0.48	
Caregiver strain	0.66[†]		0.29[†]
3d. Cognitive functioning			
Step 1		0.03	
Mother education	0.03		
Child age	0.02		
Child sex	−0.03		
Time since diagnosis	−0.08		
Step 2		0.11	
Child cognitive functioning	0.05		0.08[†]
Step 3		0.48	
Caregiver strain	0.72[†]		0.37[†]

[†]$P < 0.001$

associated with parent PTSS after accounting for demographics (Step 1) and child functioning (Step 2). Separate models were examined for each child functioning composite (i.e., physical, emotional, social, and cognitive). Across models, demographics entered in Step 1 were not significantly associated with parent PTSS ($R^2 = 0.03$). The addition of child functioning composites significantly improved model fit (physical: $\Delta R^2 = 0.09$, $p = 0.001$; emotional: $\Delta R^2 = 0.15$, $p < 0.001$; social: $\Delta R^2 = 0.16$, $p < 0.001$; cognitive: $\Delta R^2 = 0.08$, $p = 0.001$); however, in the final model, only caregiver strain (all p's <0.001) accounted for a significant proportion of variance in explaining parent PTSS. Model results are presented in Table 3.

Discussion

Results of this study suggest that, on average, parents of long-term ALL survivors experience few clinically significant symptoms of emotional distress. While symptoms of emotional distress were within normative expectations, a small subset of parents experienced elevated symptoms of anxiety, depression, and PTSS. Caregiver strain and parent perception of survivor emotional functioning contributed significantly to parent emotional distress symptoms. These results underscore the importance of continued research to further understand factors that place subgroups of parents of long-term survivors at-risk for emotional distress symptoms.

Though the prevalence of parent emotional distress in survivors of childhood cancer has been examined in recent literature, results have been inconsistent [15]. For example, Kazak *et al.* [19] reported that 43.7% of mothers and 35.3% of fathers experienced moderate to severe PTSS. However, other reports have found no significant elevation in PTSS symptoms in parents of long-term childhood cancer survivors compared to parents of healthy controls [20,37]. It is possible that discrepancies observed across previous studies may result from differences in patient populations studied, measurement tools, and time from treatment (i.e. duration of survivorship). Importantly, research to date has largely focused on parents of heterogeneously-treated survivors across multiple childhood cancer diagnoses with little emphasis placed on understanding parent emotional distress in specific populations of long-term survivors. As such, results from the current study expand upon existing research, by contributing data from parents of a large sample of long-term survivors of childhood ALL treated on a chemotherapy-only protocol which minimizes risk of many serious late effects associated with the use of irradiation. Overall, most parents in our study did not report elevated symptoms of anxiety, depression, or PTSS, supporting findings of more contemporarily treated cohorts [20,37].

Understanding sources of variability in parental emotional distress outcomes remains important. Our results highlight associations between child functioning, caregiver strain, and parent emotional distress. Perceived caregiver strain consistently contributed to parent emotional distress in our sample. Child emotional functioning was the only survivor characteristic to contribute to parent emotional distress after accounting for caregiver strain. Of note, survivor emotional functioning composite scores showed the greatest variability in our sample. This increased variability may, in part, explain why significant associations were only observed with emotional functioning in the final models. Alternatively, parent emotional distress may be most closely related to child emotional functioning, as symptoms of parent emotional distress may directly influence perceptions and reporting of child emotional functioning.

Overall, these results support findings from other pediatric populations that suggest caregiver strain may be a mediator between child functioning and parent emotional distress [22,23]. Parents who experience elevated strain may be experiencing unique sources of stress related to their child's functioning following cancer treatment (e.g. financial burden, disruption of family and social life, and worry). This perceived strain may, in turn, contribute to increased symptoms of emotional distress. As such, parents of long-term survivors may benefit from interventions designed to manage caregiver strain. Programs designed to educate and provide resources for parents coping with and caring for long-term survivors as well as opportunities for social and emotional support may help reduce perceptions of strain. Further research is needed to identify specific sources of strain that may contribute to increased parent emotional distress and to develop interventions to reduce parent strain and distress. Specifically, research efforts should examine the contribution of additional factors such as child behavior and physical late effects to parent strain and emotional distress symptoms.

Results of the current study should be considered in the context of several limitations. First, survivor emotional, social, cognitive, and physical functioning were obtained through parent proxy report rather than self-report. Past research has indicated that child and parent report of functioning may differ from self-report and that these may further differ within healthy compared to chronically ill samples [38]. For example, a review of parent–child concordance on measures of child quality of life found that parents of children with health problems reported lower quality of life than reported by the child while parents of children of healthy children reported higher quality of life than reported by the child. Thus, future studies should incorporate both child and parent report. The use of only one parent, primarily mothers, is another limitation of this study. Because of the small number of fathers participating in our study we were unable to statistically examine differences in reporting by parent. Past research has indicated that mothers and fathers differentially report child functioning as well as cope differently in response to stressors. Moreover reliance solely on parent report may result in reporting bias. For example, parents experiencing emotional distress may have negative cognitions and attributions that influence their perceptions not only of caregiver strain but also of child functioning. The cross-sectional nature of this study precludes discussing temporal patterns between parent emotional distress, strain, and child functioning. Thus, our results do not support statements of casual associations. Future research may benefit from longitudinal research designs to better understand temporal and causal relations between parent and child functioning. Notably, the current study included

parents of long-term survivors of childhood ALL who were treated on a chemotherapy-only protocol at SJCRH. While examining this population allowed for the exploration of associations between parent emotional distress, strain, and child functioning in a homogenously-treated sample, the study may lack generalizability to other childhood cancers treated on different therapeutic trials. Specifically, parents of long-term survivors of ALL treated on chemotherapy-only protocols may be at reduced risk of emotional distress than parents of long-term survivors of ALL treated using more intensive CNS directed therapies (i.e. cranial irradiation). Historically, prophylactic radiation therapy was a critical component of successful treatment for ALL, though was associated with a number of adverse late effects. Thus, parents of children treated on more intensive treatment protocols may experience higher levels of emotional distress compared to parents of children treated on contemporary chemotherapy only protocols. It is also possible that parents of children treated at SJCRH received more or less supportive care than parents of children treated at other medical institutions. Therefore, the prevalence of emotional distress symptoms among parents of children treated at SJCRH may not be representative of parents of patients treated at other institutions. Likewise, parents who participated may have more or less emotional distress compared to parents who did not participate. Finally, this study was limited by a lack of existing normative data; we were unable to compare reported levels of parent emotional distress, caregiver strain, and child quality of life in our sample to the general population. Future studies should include healthy children and their parents as controls.

In summary, our results suggest that, on average, parents of long-term survivors of childhood ALL treated with contemporary therapy experience few clinically significant symptoms of anxiety, depression, and PTSS. Our results support an association between symptoms of caregiver strain and parent emotional distress. Continued research to further understand factors that place parents of long-term survivors at-risk for emotional distress symptoms, as well as protective factors that may mitigate or prevent emotional distress, is warranted.

Acknowledgements

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