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The Role of Contextual Threat in Predicting Self-Reported Distress among Siblings of Children with Cancer

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Abstract Each year, 14,000 children are diagnosed with cancer in the United States. Prolonged, intensive treatment regimens disrupt the entire family system, but effects on siblings are poorly understood. In this preliminary investigation, we employed a risk and resistance framework to study adjustment among 30 siblings (aged 10–17) of children undergoing cancer treatment. We examined whether or not objective stress associated with the cancer experience (contextual threat) predicted sibling distress and explored demographic and disease-related predictors of sibling adjustment. Contextual threat was positively associated with sibling-reported distress, independent of sibling age, gender, birth order relative to the child with cancer, and cancer treatment intensity. From among the demographic and disease-related factors, only younger birth order relative to the child with cancer was independently associated with sibling distress. These results suggest that a subset of siblings may be at increased risk for elevated distress in the face of their brother's or sister's illness.

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Over the past 30 years, significant biomedical advances in the treatment of childhood cancers have led to 5-year relative survival rates exceeding 80 % (Howlader et al., 2012). However, these improved survival rates are achieved through prolonged, intensive treatment protocols requiring families to alter their roles, responsibilities, and day-to-day patterns of functioning (Long & Marsland, 2011). In addition to emotional strain, primary caregivers spend extended periods of time in the outpatient clinic or hospital and assume responsibility for home-based treatments. These treatment demands can result in chronic disruption of the family system and can influence the psychological health of all family members, including siblings (Alderfer & Kazak, 2006).

The psychosocial impact of a childhood cancer diagnosis on patients and parents is well-researched (e.g., Patenaude & Kupst, 2005; Vrijmoet-Wiersma et al., 2008). However, the impact on siblings is not as well understood (Alderfer & Noll, 2006). Siblings may be particularly vulnerable during the diagnosis and treatment periods, as attention is shifted to the child with cancer and caregivers are less available to siblings both physically and emotionally. Indeed, a recent review of sibling functioning in the face of childhood cancer found that siblings endorse higher levels of negative emotion, lower levels of positive emotion, and reduced quality of life when compared to their peers (Alderfer et al., 2010).

Although the majority of siblings endorse normative levels of depression and anxiety, an elevated percentage of siblings falls into the clinical ranges on these scales (Alderfer et al., 2010). This pattern of results suggests that a subgroup of

siblings is at increased risk for elevated distress. A meta-analysis examining siblings of children with various chronic illnesses suggests that siblings' internalizing or externalizing symptoms are not moderated by demographic factors such as sibling gender, age, or birth order (Vermaes, van Susante, & van Bakel, 2012). Higher sibling distress may be associated with higher treatment intrusiveness and mortality risks (Vermaes et al., 2012), especially when assessed at time points more proximal to the cancer diagnosis (Alderfer et al., 2010). However, frequent mixed findings suggest that illness-related variables do not solely account for elevations in sibling distress. Rather, demographic or illness factors may interact with other aspects of the sibling experience to influence sibling adjustment, but the mechanisms through which this may occur remain unclear.

There are many frameworks for understanding how stressors associated with a major life event, such as childhood cancer, may influence individual functioning. One general model of adjustment that was developed to explain variations in distress in the context of pediatric chronic illness is the risk and resistance model (Wallander, Varni, Babani, Banis, & Wilcox, 1989; Wallander & Varni, 1998). This framework considers factors likely to contribute to patients' psychosocial adjustment to pediatric chronic illnesses, including the role of disease and disability parameters, functional dependence, psychosocial stressors, intrapersonal factors (e.g., competence, temperament), social-ecological factors (e.g., family environment, social support), and stress processing factors (e.g., appraisals and coping strategies). Due to the complexity of the model, the authors advocate focusing on components of the larger model or submodels (Wallander & Varni, 1998). Accordingly, aspects of this model have been used to explain the psychosocial adjustment of siblings of children with various medical illnesses, including cancer (Maurice-Stam, Grootenhuis, Brons, Caron, & Last, 2007; Robinson, Gerhardt, Vannatta, & Noll, 2007). The work of Taylor, Fuggle, and Charman (2001) focused on maternal variables including mothers' distress, social support, and awareness of siblings' attitudes about the illness. The current work takes a broader approach by incorporating aspects of the disease, psychosocial stress, and social-ecological domains in predicting distress among siblings of children with cancer (Fig. 1).

Implicit in the risk and resistance model is the idea that the illness itself is only one factor that may influence a child's adjustment. In addition to aspects of the cancer or its treatment (e.g., treatment intensity), a sibling's experience is likely to be influenced by developmental and contextual factors such as instrumental and emotional social support, family structure and dynamics, financial security, and the presence of other stressors that may be unrelated to cancer. Thus, the present research focuses on the role of *contextual threat* in predicting sibling distress. Contextual threat is defined as an aggregate measure of objective stress surrounding the cancer experience (Brown & Harris, 1978). Rather than focusing on discreet predictors, contextual threat encompasses a set of circumstances that are considered as a whole given the likelihood that they act synergistically to influence functioning. Siblings experiencing more concurrent stressors in addition to the cancer, along with fewer resources to help them cope with these stressors, have higher levels of contextual threat and may endorse greater distress. The contextual threat approach has been applied in studies explaining child or adolescent onset of mental health problems (e.g., Dunn, Abbott, Croudace, Wilkinson, Jones, Herbert, & Goodyer, 2011; Rudolph & Hammen, 1999) and in studies linking greater life stress and contextual threat to increased asthma symptomatology and poorer biological parameters (e.g., Chen, Hanson, Paterson, Griffin, Walker, & Miller, 2006; Wolf, Nicholls, & Chen, 2008). To our knowledge, the contextual threat framework has not been applied in studies of sibling functioning.

The overarching goal of this study was to identify factors that may place siblings at greater risk for elevated distress following their brother's or sister's cancer diagnosis. Our primary aim was to examine the hypothesis that higher levels of objective stress (contextual threat) surrounding the cancer experience would predict greater sibling-reported distress. In addition to contextual threat, we carried out exploratory analyses to investigate whether sibling distress was correlated with demographic or illness-related variables including treatment intensity, time since diagnosis, and sibling age, gender, and birth order relative to the child with cancer.

Methods

Participants

Thirty siblings of children with cancer from 22 families were enrolled between October, 2007 and June, 2010 from two different children's hospitals. Inclusion criteria were (a) having a brother or sister currently on treatment for cancer, diagnosed at least six months earlier; (b) fluency in English; and (c) being between the ages of 10 and 17. Exclusion criteria included death of the child with cancer and history of intellectual disability and/or life-threatening disease in the sibling. At site A, 25 eligible siblings were identified during the recruitment period. Of these, eight refused due to the sibling being too busy, the parents being overwhelmed, or the sibling not being interested in the study, resulting in a sample size of 17 (68 % of those eligible).

At site B, eligible siblings were identified from among families participating in an ongoing study of sibling adjustment to childhood cancer (MRSG 05-213 awarded to MAA). The purpose of the larger project was to test a social contextual model including family and peer

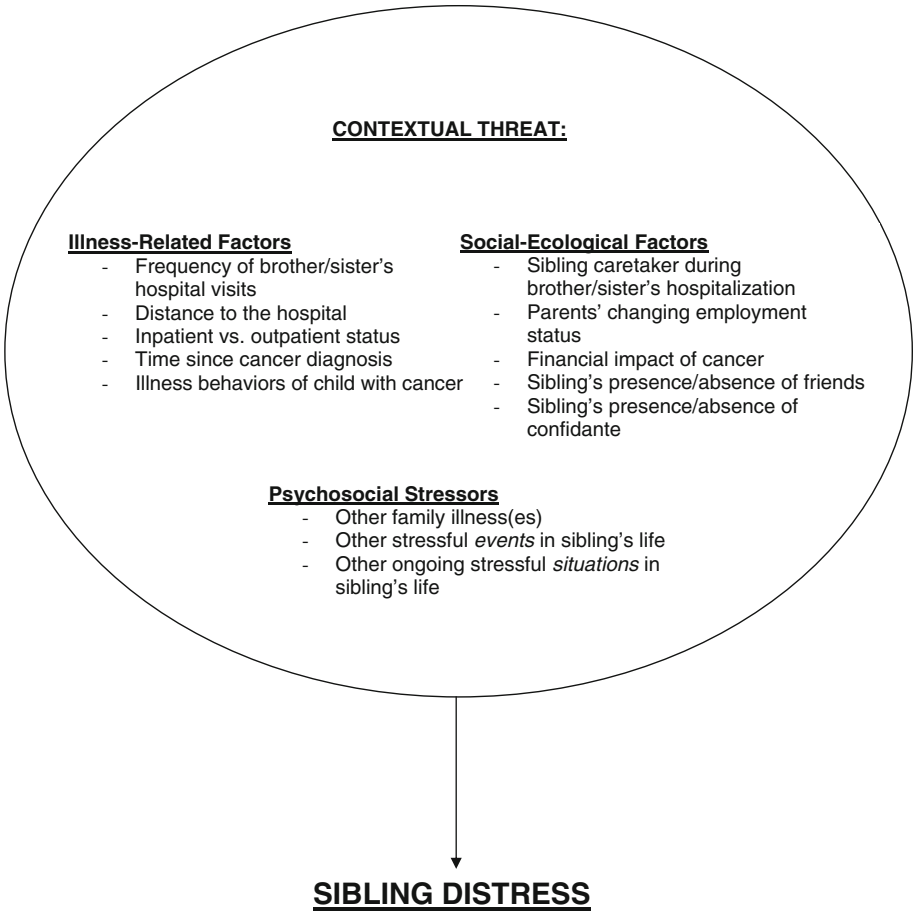

Fig. 1 Aspects of Wallander and Varni's (1998) risk and resistance model integrated with the contextual threat framework

influences on sibling adjustment. The present pilot work differs from the parent project due to our holistic approach to quantifying contextual threat. The enrollment rate for the parent project was 81 %. Siblings not taking part in the parent study (e.g., in families with more than one sibling) and eligible for this sub-study were approached for participation; all consented ($n = 13$). The study was approved by the Institutional Review Boards at both sites.

Procedure

At site A, a registry was used to identify families of children who had been diagnosed at least 6 months earlier and who were currently receiving active cancer treatment. A letter was sent to these families inviting siblings to participate. To follow up, families were reminded about the study by a member of the medical treatment team during clinic visits, who also confirmed that the sibling met inclusion criteria; this information was used to calculate the response rate for Site A (see "Participants" section). At Site B, information regarding family constellation was collected during the first phase of the parent project. Letters inviting participation were sent to families with at least one sibling who was not participating in the parent project, and this was followed up with a telephone call to families 2–3 weeks after the letter was sent. At both sites, brief telephone screenings were conducted to ensure eligibility.

Data collection occurred in the participants' homes ($n = 24$), in the hospital library or a conference room ($n = 3$), or in the university study offices ($n = 3$). Siblings participated in 30–60 min semi-structured interviews during which they provided information on their experiences of having a brother or sister with cancer (qualitative results to be reported separately). After siblings completed these semi-structured interviews, they spent approximately 15-min providing open-ended, verbal responses to questions probing the contextual details of the cancer experience. Finally, they completed a battery of written psychosocial questionnaires.

Subsequently, information about diagnosis and treatment intensity was extracted from the medical charts of the children with cancer through a standardized procedure (Werba et al., 2007) by a masters-level research assistant with 5 years of experience working with pediatric oncology populations (Site A) or a pediatric psychologist specializing in childhood cancer (Site B). Sibling participants were compensated for participation with $25 store gift cards and entered into a lottery to receive a larger incentive prize.

Contextual Threat

Contextual details about the cancer experience were gathered by asking participants a series of targeted questions in a format modeled after semi-structured interviews that were developed to quantify the level of contextual threat associated with stressful life events (e.g., Rudolph & Hammen, 1999; Williamson et al., 2003). Questions were derived from a review of the empirical literature and from clinical experience with families of children with cancer. Some probes targeted information specific to cancer-related events (e.g., "How often does your brother/sister need to stay overnight in the hospital?"), while other questions examined events or coping resources that were not directly related to the child's cancer diagnosis (e.g., "Is there anyone else in your family who is currently ill?" or "Do you feel that there is someone in your life with whom you can discuss your feelings, if you ever want to?").

Our method was based on the approach of Williamson et al. (2003), who have shown that their Stressful Life Events Schedule (SLES) has adequate inter-rater reliability (kappas = .67–.75), one week test–retest reliability (intraclass coefficient (ICC) = .93), and parent–child agreement (ICC = .81). They have demonstrated concurrent validity between the SLES and the Life Events Checklist (Johnson & McCutcheon, 1980; ICC = .83) and the Life Events and Difficulty Schedule (Monck & Dobbs, 1985; kappa = .77). According to this methodology, a team consisting of the interviewer (K.L.) and two raters with experience in pediatric psychology (L.E. & A.M.) convened for a consensus meeting. The interviewer presented written descriptions of the details surrounding each sibling's cancer experience to the other two team members, who were blinded to participants' self-reported distress scores. The following information was included in the summary paragraphs and therefore was considered when assigning an overall contextual threat rating: (a) frequency of hospital visits, (b) distance to the hospital, (c) inpatient versus outpatient status, (d) recency of diagnosis, (e) typical sibling caretaker during hospital visits (e.g., parent, extended family, non-relative, none), (f) frequency of the ill child being sick when at home, (g) change in employment status of parent(s) after diagnosis, (h) presence of illness in other family members, (i) financial impact of the cancer, (j) presence/absence of friends, (k) presence/absence of someone to talk to, (l) other stressful events occurring since the ill child was diagnosed, and (m) other ongoing stressful situations. The team of raters discussed the objective amount of stress surrounding the cancer experience for each participant, and contextual threat ratings were derived for each sibling by consensus using the following scale: (1) little/no contextual threat, (2) some contextual threat, (3) moderate contextual threat, or (4) high contextual threat. Standardized rating procedures included comparing and contrasting contextual details across siblings. Raters were aware of sibling pairs from the same family.

Self-Report Instruments

After being interviewed, siblings completed a short battery of questionnaires measuring distress. The Perceived Stress Scale (PSS; Cohen, Kamarck, & Marmelstein, 1983) is a 10-item self-report instrument that measures the extent to which participants appraise their life circumstances to be uncontrollable or unpredictable. Test–retest reliability is high (r = .85; Cohen et al., 1983), and internal consistency is good (alpha in the present sample = .77). The wording of eight of the 10 PSS questions was modified slightly to make the language more understandable for children and adolescents (e.g., the phrase "control irritations" was replaced with "handle things that bother you;" the phrase "difficulties were piling up so high that you could not overcome them" was replaced with "had so many problems that you could not handle them").

Siblings also completed the Child Depression Inventory (CDI; Kovacs, 1981), a 27-item self-report questionnaire that assesses the frequency and severity of depressive symptoms. It has relatively high levels of test–retest reliability and predictive validity (Ialongo, Edelsohn, & Kellam, 2001; Mattison, Handford, Kales, & Goodman, 1990), along with adequate construct (Worchel, Rae, Olson, & Crowley, 1992) and discriminant validity (Carey, Faulisch, Greshman, Ruggiero, & Enyart, 1987). In the present sample, internal consistency was high (alpha = .89). This measure has been validated in children and adolescents, ages 7–17.

The Revised Child Manifest Anxiety Scale (RCMAS, Reynolds & Richmond, 1985) is a 37-item self-report measure of child and adolescent anxiety. Internal consistency is good, with Cronbach's alpha calculated to be .72 in the present sample. Test–retest reliability is adequate, with 1-week, 5-week, and 9-month Pearson correlations of .88, .77, and .68, respectively (Wisniewski, Mulick, Genshaft, & Coury, 1987; Reynolds, 1981). Concurrent validity is strong when compared to the State Trait Anxiety Inventory for Children (r = .88; Muris, Merckelbach, Ollendick, King, & Bogie, 2002). This measure has been validated for children and adolescents, ages 6–19.

Finally, siblings completed the Children's Posttraumatic Stress Disorder Reaction Index (PTSD-RI; Pynoos, Frederick, Nader, & Arroyo, 1987), which is a 20-item self-report measure that corresponds to the diagnostic criteria for PTSD. Although not designed as a diagnostic tool, comparison of PTSD-RI scores with clinical diagnoses of PTSD in a large community sample resulted in the following categories of posttraumatic stress reaction severity:

none (score <12), mild (score 12–24), moderate (score 25–39), and severe (score >39; Pynoos et al., 1993). The scale has been shown to have good reliability and validity (Pynoos et al., 1993), with internal consistency in the present sample estimated at alpha = .81.

Treatment Intensity

The Intensity of Treatment Rating Scale 2.0 (ITR-2; Werba et al., 2007) is a 7-question instrument developed specifically for pediatric cancer diagnoses, classifying treatment intensity (1 = least intense, 4 = most intense) on the basis of treatment duration, side effects, and recovery time. This measure has high interrater reliability (r = .87) and content validity (r = .95; Werba et al., 2007).

Data Analysis

Preliminary analyses revealed that the depression and perceived stress variables were positively skewed. Square root transformations resulted in acceptable distributions of these variables. Anxiety, posttraumatic stress, contextual threat, and treatment intensity variables did not require transformation. Given the statistical (Table 1) and conceptual overlap among measures of depression, anxiety, perceived stress, and posttraumatic stress, z-scores from these measures were averaged to form a composite distress score that was used in all analyses. Descriptive statistics of the original scales including range, mean, and standard deviation were calculated to characterize the sample.

Initial Pearson product-moment correlation analyses and independent samples t-tests were performed to determine bivariate associations between sibling distress and sibling age, gender, birth order relative to the child with cancer, treatment intensity, and time since diagnosis. Demographic factors associated with sibling distress were entered as control variables in subsequent regression models. Multiple regression was used to examine our primary hypothesis that higher levels of objective stress (contextual threat) surrounding the cancer experience would predict greater sibling-reported distress. Here, control variables were entered into the first step and contextual threat into the second step of a regression model predicting sibling distress. Finally, exploratory analyses employed multiple regression to investigate whether each demographic and disease factor predicted sibling distress while controlling for the other demographic and disease factors and contextual threat.

The sample size of 30 was determined based upon theoretical saturation and verification of the qualitative portion of this study (data presented separately). Using an alpha level of .05, a sample size of 30, and five predictors, our power to detect change in R^2 equivalent to a medium-sized effect (f^2 = .15) when adding the predictor of interest to a regression equation predicting sibling distress was .53.

Results

Sample

Thirty siblings ages 10–17 completed the study. Siblings' mean age was 13.90 (SD = 2.28), with 63 % in early adolescence (≤14 years) and the remainder in later adolescence (≥15 years). The sample included 19 male and 11 female siblings, and time since diagnosis ranged from seven to 33 months (M = 14.47, SD = 7.13). Twenty-three sibling participants (77 %) were older than the child with cancer. All 30 siblings were Caucasian. Cancer diagnoses included leukemia (n = 12), lymphoma (n = 1), brain tumor (n = 3), rhabdomyosarcoma (n = 3), neuroblastoma (n = 1), osteosarcoma (n = 1), and thyroid cancer (n = 1). All children with cancer were receiving active treatment at the time of data collection, either for a first diagnosis (n = 20, 91 %) or relapse (n = 2, 9 %). To determine whether there were differences between participants recruited at Site A versus Site B, a series of t-tests (age, time since diagnosis, treatment intensity, contextual threat, and distress) and Chi square tests (gender, presence of additional siblings, and birth order relative to the child with cancer) were carried out and confirmed that the two groups were equivalent (p's > .05).

To put the following results into context, we provide a summary of the percentage of our sample who endorsed elevated rates of symptomatology on clinical measures. With regard to posttraumatic stress, 13 % of participants (n = 4) showed no posttraumatic stress reaction (score <12), 57 % (n = 17) showed mild reactions (score 12–24), and 30 % (n = 9) showed moderate to severe reactions (score ≥25; Table 2). With regard to depression symptoms, 7 % (n = 2) of siblings fell into the clinical range on the CDI (T score ≥70). Three percent of siblings (n = 1) fell into the borderline range of anxiety symptoms on the RCMAS (T score between 60 and 70), while another 7 % fell into the clinical range on this measure (n = 2, T score ≥70). Based on distributions in the general population, it is expected that 16 % of children would have T scores ≥60 and 2 % would have T scores ≥70 on clinical measures.

Bivariate Analyses

Consistent with our primary hypothesis, initial Pearson correlations showed significant positive associations between contextual threat and sibling distress (Table 1). Next, we examined other variables that might influence

Table 1 Correlations among demographic and disease factors, contextual threat, and sibling distress

	Age	Time since diagnosis	Treatment intensity	Contextual threat	Depression	Anxiety	Perceived stress	Posttraumatic stress
Contextual threat	.21	−.17	.39*	1.00				
Depression	.39*	−.06	.37*	.50**	1.00			
Anxiety	.31†	−.09	.44*	.38*	.76**	1.00		
Perceived stress	.35†	−.20	.13	.47**	.65**	.66**	1.00	
Posttraumatic stress	.38*	.00	.29	.31†	.70**	.72**	.64**	1.00
Distress composite	.42*	−.11	.39*	.48*	.89**	.90**	.86**	.88*

** $p < .01$; * $p < .05$; † $p < .10$

Table 2 Descriptive statistics: measures of sibling distress, treatment intensity, and contextual threat

	Minimum	Maximum	Mean	*SD*
Depression T scores[a] (CDI)	40	84	45.70	9.69
Anxiety T scores (RCMAS)	25	76	45.40	12.53
Perceived stress raw scores[b] (PSS)	4	31	11.97	5.53
Posttraumatic stress raw scores[c] (PTSD-RI)	7	53	22.03	9.91
Treatment intensity raw scores[d] (ITR-2)	1	4	2.43	.63
Contextual threat raw scores[e]	1	4	2.03	1.07

Higher scores indicate greater distress, more intense treatment regimens in the child with cancer, and greater levels of contextual threat

[a] In the normative population, T scores have a mean of 50 and a standard deviation of 10; [b] possible range for the PSS is 0–40; [c] possible range for the PTSD-RI is 0–80; [d] possible range for the ITR-2 is 1–4; and [e] possible range for contextual threat is 1–4

siblings' distress, including sibling age, time since diagnosis, treatment intensity, sibling gender, and birth order relative to the child with cancer. Results showed positive correlations of sibling distress with older sibling age and greater treatment intensity. Time since diagnosis was not significantly correlated with sibling distress.

Categorical variables (relative birth order and gender) were assessed using independent samples t-tests. Siblings who were younger than the child with cancer scored higher on the composite measure of distress than siblings who were older than the child with cancer (Younger: $M(SD) = 1.11(1.23)$, Older: $M(SD) = -.26(.55)$, $t(28) = -2.65$, $p = .04$, *Cohen's* $d = 1.44$). There were no significant gender differences in distress (Female: $M(SD) = .29(.89)$; Male: $M(SD) = -.14(.88)$; $t(28) = 1.30$, $p = .20$, *Cohen's* $d = .49$).

Contextual Threat as a Predictor of Sibling Distress

To examine whether the association between contextual threat and sibling distress is independent of sibling characteristics and disease factors, we conducted a linear regression analysis in which covariates (treatment intensity, sibling age, and relative birth order) were entered into Step 1 and contextual threat was entered into Step 2 of a model predicting sibling distress. Results of these analyses are presented in Table 3 and show a positive association of contextual threat with sibling distress independent of treatment intensity, sibling age, and relative birth order. The same pattern of results was obtained when also controlling for treatment intensity and gender.

Associations between Illness and Demographic Factors and Sibling Distress

Exploratory regression analyses were conducted to further examine the finding that siblings younger than the child with cancer experienced greater distress than those older than the child with cancer. In these analyses, covariates were entered into Step 1 and sibling relative birth order was entered into Step 2 of models predicting sibling distress. After controlling for age, treatment intensity, and contextual threat, regression analyses revealed that siblings who were younger than the child with cancer endorsed significantly greater distress than siblings older than the child with cancer (Table 3). The same approach was used to examine the independent effects of sibling age and treatment intensity. Findings showed a trend for older siblings to report greater distress than younger siblings, but this effect did not reach significance. There was no independent association between treatment intensity and sibling distress once demographic factors and contextual threat were entered into the model.

It should be noted that these results are considered preliminary due to the small sample size and dependencies in the data (30 sibling participants from 22 families). To address data dependencies, analyses were repeated using a subset of participants randomly selected to represent one sibling from each family ($n = 22$). The same pattern of regression results was obtained using this subsample, except that the finding for the independent effect of sibling age on distress no longer reached a marginal significance level.

Discussion

The current study is grounded in the risk and resistance model (Wallander & Varni, 1998) and considers aspects of the illness (e.g., treatment, time since diagnosis), social ecology (e.g., social support), and psychosocial stress (e.g., presence of other stressors unrelated to cancer) domains in order to calculate a holistic rating of the degree of objective stress (contextual threat) associated with having a brother or sister with cancer. Our findings support a positive association between contextual threat and sibling-reported distress. This relationship holds independent of demographic and illness-related variables. Exploratory analyses suggested that relative birth order also independently contributed to sibling distress such that siblings younger than the child with cancer endorsed greater distress than those older than the child with cancer. With regard to sibling age, there was a marginal effect in the full sample for older siblings to endorse greater distress. However, this finding was no longer evident when analyses were repeated with a subsample ($n = 22$) of siblings representing one sibling from each family. There were no independent effects of sibling gender, treatment intensity, or time since diagnosis on sibling distress.

Examination of contextual threat is novel in the pediatric sibling literature and was carried out in response to evidence that a subgroup of siblings endorse elevated distress in the face of their brother's or sister's cancer diagnosis (Alderfer et al., 2010). Based on semi-structured stress interviews that have been employed in examinations of child and adolescent depression and anxiety (Rudolph & Hammen, 1999; Williamson et al., 2003), our goal was to quantify the degree of objective stress associated with the cancer experience by considering contextual factors and assigning threat values based on holistic descriptions of the stressor. This approach is consistent with the premise that aspects of stressful situations have synergistic rather than additive effects on sibling stress (Sameroff, 2000). In other words, the whole is greater than the sum of its parts. Overall, our findings are consistent with reports suggesting that greater contextual threat contributes to the onset and/or course of child and adolescent depression, bipolar disorder, and eating disorders (Kim, Miklowitz, Biuckians, & Mullen, 2007; Rojo, Conesa, Bermudez, & Livianos, 2006; Rudolph et al., 2000). By applying this framework to the

Table 3 Results of separate regression models examining the independent effects of contextual threat, treatment intensity, relative birth order, and sibling age on sibling distress ($N = 30$)

Predictor	Distress		
	Beta	p value	Adj R^2
Regression examining contextual threat			
Step 1			.45
Sibling age	.35	.02	
Relative birth order	.53	.005	
Treatment intensity	−.08	.65	
Step 2			.56
Sibling age	.26	.06	
Relative birth order	.60	.001	
Treatment intensity	−.11	.51	
Contextual threat	.38	.01	
Regression examining treatment intensity			
Step 1			.57
Sibling age	.28	.04	
Relative birth order	.54	.000	
Contextual threat	.34	.01	
Step 2			.56
Sibling age	.26	.06	
Relative birth order	.60	.001	
Contextual threat	.38	.01	
Treatment intensity	−.11	.51	
Regression examining relative birth order			
Step 1			.33
Sibling age	.36	.03	
Treatment intensity	.28	.11	
Contextual threat	.30	.10	
Step 2			.56
Sibling age	.26	.06	
Treatment intensity	−.11	.51	
Contextual threat	.38	.01	
Relative birth order	.60	.001	
Regression examining sibling age			
Step 1			.50
Relative birth order	.66	.000	
Treatment intensity	−.18	.31	
Contextual threat	.45	.004	
Step 2			.56
Relative birth order	.60	.001	
Treatment Intensity	−.11	.51	
Contextual threat	.38	.01	
Sibling age	.26	.06	

population of siblings of children with cancer, we begin to address one of the most fundamental limitations of the sibling literature. While the majority of existing work focuses on identifying mean differences in psychopathology symptoms between siblings and controls or normative data, our approach is in line with the developmental psychopathology framework and begins to shed light on why some siblings endorse ongoing distress while others show resilience. In doing so, we lay the groundwork for future work identifying at-risk siblings who may benefit from intervention.

Although higher levels of contextual threat predicted greater sibling distress in our sample, it is important to note that the majority of participants did not endorse clinically significant levels of depression or anxiety. These findings are consistent with the conclusions of a recent review paper reporting that the majority of siblings do not show clinically significant levels of depression or anxiety and that qualitative themes of stress and negative emotionality generally are not reflected in responses to standardized symptom questionnaires (Alderfer et al., 2010). However, fifty-seven percent of siblings in the current study endorsed mild posttraumatic stress reactions and 30 % endorsed moderate to severe reactions. These rates of posttraumatic stress are consistent with previous work (Alderfer, Labay, & Kazak, 2003; Packman, Gong, VanZutphen, Shaffer, & Crittenden, 2004) and suggest that elevations in posttraumatic stress may be relatively common in siblings of children with cancer. Future research may benefit from including measures of sibling functioning that reflect variations in normal functioning rather than psychopathology per se, such as perceived and posttraumatic stress, quality of life, mood, academic functioning, and somatic complaints.

The few existing studies of sibling functioning that have considered the role of demographic factors have yielded inconsistent findings with regard to sibling age and gender (Alderfer et al., 2010). Our results are consistent with findings of a recent meta-analysis which reported that sibling adjustment to a brother's or sister's chronic health condition does not vary by sibling age or gender (Vermaes et al., 2012). However, our findings differ with regard to the significant role of birth order relative to the child with cancer, which has been examined less frequently in the sibling literature and has yielded inconsistent findings both within and across empirical investigations. For example, one study showed that siblings younger than the child with cancer endorsed higher levels of loneliness, but this study did not show differences in self-reported state anxiety as a function of sibling birth order (Hamama, Ronen, & Feigin, 2000). The current findings that siblings younger than the child with cancer reported more distress may reflect a difference in how the cancer experience is perceived. For example, siblings younger than the child with cancer may conceptualize their older brother or sister as a stable or protective figure whose illness leads to a fundamental loss of security. Alternately, siblings who are older than the child with cancer may assume increased responsibility at home, which may serve as an active coping mechanism and give them a defined role in helping the family handle the challenges of childhood cancer, or patterns of parenting may differ according to birth order. Regardless of the explanation, our data provides initial evidence that siblings who are younger than the child with cancer may be more vulnerable to distress and may benefit from supportive interventions.

With regard to disease factors, the current study found that time since diagnosis was not associated with sibling distress and that treatment intensity was no longer a significant predictor of sibling distress after demographic variables and contextual threat were entered into the model. The extant literature examining time since diagnosis has reported mixed results (Alderfer et al., 2010), with some studies showing that relationships between time since diagnosis and sibling functioning were nonsignificant (e.g., Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003). Consistent with the results of the recent review of sibling adjustment to chronic health conditions (Vermaes et al., 2012), the current findings showed significant bivariate correlations between treatment intensity and sibling distress. However, these effects were no longer significant after the effects of contextual threat were taken into account. One possible explanation for this pattern of findings is that aspects of the illness and its treatment are considered as part of the contextual threat rating. Although treatment intensity is not probed specifically, it is likely that responses to questions about inpatient versus outpatient treatment and the illness behaviors of the child with cancer may reflect treatment intensity. Alternately, the relatively minor role of disease factors on siblings' functioning may suggest that the context in which the cancer is experienced, rather than the cancer diagnosis per se, is more important for siblings' psychosocial functioning.

Methodological strengths of the current study include recruitment of a more homogeneous sample than much of the extant literature, with the inclusion of only adolescent siblings of children who are at least 6-months post diagnosis and on active treatment. Furthermore, 90 % of data were collected outside the hospital setting, thereby assessing sibling distress in a setting more reflective of day-to-day functioning. Despite these strengths, the current study included a small sample size and employed a cross-sectional design, which limits our ability to identify patterns that develop over time. Further, the lack of a control group limits the extent to which we can determine whether our findings represent a departure from normative

developmental processes, and the considerable age range of the sample (10–17) may obscure developmental variations in functioning. The overrepresentation of Caucasian families may underestimate the strength of these relationships in non-Caucasian families, who have been shown to have higher levels of cumulative risk (Koinis-Mitchell et al., 2008) and where different patterns of family roles and relationships may influence the meaning ascribed to having an ill brother or sister and may result in higher levels of sibling caretaking. Finally, results should be interpreted with caution since the contextual threat details were provided by the siblings themselves before completing the distress measures. It is possible that siblings who are more attuned to the stressfulness of the cancer experience may offer more threatening details than those who experience less subjective stress. On the other hand, the positive association may represent siblings' accurate perceptions of the objective levels of threat surrounding the cancer diagnosis and their resulting distress. Future research would benefit from gathering information regarding contextual threat from a caretaker, teacher, or healthcare professional in addition to the sibling.

Despite these limitations, the current findings represent a first step toward identifying factors that may contribute to competence versus maladjustment in siblings of children with cancer. Clinically, links between greater contextual threat and heightened distress lay the groundwork for developing criteria to identify siblings who may be at increased risk for ongoing adjustment difficulties and/or who may benefit from a higher level of psychosocial care.

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