



Published in final edited form as:

*Pediatr Blood Cancer*. 2018 June ; 65(6): e26976. doi:10.1002/pbc.26976.

## A Prospective Study of Family Predictors of Health-Related Quality of Life in Pediatric Brain Tumor Survivors

**Lauren F. Quast,**

The Children's Hospital of Philadelphia

**Peter C. Phillips,**

The Children's Hospital of Philadelphia and the Perelman School of Medicine at the University of Pennsylvania

**Yimei Li,**

The Children's Hospital of Philadelphia and the Perelman School of Medicine at the University of Pennsylvania

**Anne E. Kazak,**

Nemours Children's Health System and Sidney Kimmel School of Medicine at Thomas Jefferson University

**Lamia P. Barakat, and**

The Children's Hospital of Philadelphia and the Perelman School of Medicine at the University of Pennsylvania

**Matthew C. Hocking**

The Children's Hospital of Philadelphia and the Perelman School of Medicine at the University of Pennsylvania

### Abstract

**Background**—The objective of this study was to examine prospectively the associations between family functioning at the end of tumor-directed treatment and the health-related quality of life (HRQL) of pediatric brain tumor survivors (PBTS) approximately nine months later.

**Procedure**—Thirty-five PBTS (age 6 to 16) and their mothers completed measures of family functioning and survivor HRQL within five months of completing tumor-directed therapy (baseline) and again approximately nine months later (follow-up).

**Results**—Survivor-rated general family functioning at baseline significantly predicted mother proxy- and self-reported survivor HRQL at follow-up when controlling for survivor HRQL at baseline and relevant demographic and treatment-related variables.

**Conclusions**—Family functioning is a key factor contributing to survivor HRQL and should be screened throughout the course of tumor-directed treatment. Psychosocial interventions directed

---

Correspondence concerning this article should be addressed to Matthew C. Hocking, Division of Oncology, The Children's Hospital of Philadelphia, Philadelphia, PA 19104., hockingm@email.chop.edu; Phone: 267-426-5561. Fax: 215-590-4183.

Conflict of Interest: The authors have no conflicts of interest to disclose.

toward improving general family functioning may improve survivor well-being following the completion of treatment.

### Keywords

brain tumor; family functioning; quality of life

As the pediatric brain tumor survivor (PBTS) population grows, increased attention is being directed towards health-related quality of life (HRQL). Five-year survival rates for children with brain or central nervous system tumors have increased to 73.8% for malignant brain tumors and 96.6% for non-malignant tumors over the last 30 years due to treatment advances.<sup>1</sup> However, survivors often experience challenges after treatment, including medical, neurocognitive, psychosocial and behavioral impairments, that negatively impact autonomy.<sup>2</sup> HRQL comprises the broader impact of an illness on aspects of physical, emotional, social and school functioning<sup>3</sup> and is a key marker of survivorship quality.

Peterson and Drotar's theoretical model of childhood cancer survivorship<sup>4</sup> integrates medical variables and constructs from systems theory<sup>5</sup> to explain survivor HRQL. The model illustrates connections between premorbid family functioning, disease/treatment variables, family adaptation and functioning following illness, and various survivor outcomes, including HRQL. The model suggests bidirectional associations between family functioning and survivor outcomes over time. For example, survivor late effects may disrupt family life<sup>6</sup> by altering routines and roles within the family. Conversely, family adaptation to survivor sequelae may facilitate child adjustment and promote better HRQL.

PBTS generally have poorer HRQL across multiple domains compared to other childhood cancer survivors or healthy controls,<sup>7</sup> due to tumor- and treatment-related late effects.<sup>2, 8,9</sup> Within PBTS, HRQL is associated with a range of survivor, disease, treatment and sociodemographic variables. Specifically, executive function difficulties,<sup>8,10</sup> low IQ,<sup>11</sup> infratentorial tumor site,<sup>12</sup> radiation therapy, and/or chemotherapy<sup>13,14</sup> are correlated with poorer HRQL for PBTS. However, findings are mixed regarding the influence of contextual factors, including socio-economic status (SES) and annual household income, on survivor HRQL.<sup>8,12,15</sup>

Among youth with chronic illness, family functioning has important implications for well-being beyond medical factors.<sup>16</sup> Within childhood cancer, family functioning predicts psychosocial HRQL over and above treatment variables<sup>17</sup> and a recent meta-analysis underscores associations between better survivor HRQL and positive indices of family functioning.<sup>18</sup> Cross-sectional studies of PBTS also demonstrate positive associations between family functioning and survivor HRQL.<sup>19, 20</sup>

Only one study has examined the prospective influence of family functioning on PBTS HRQL indicating a significant gap in knowledge.<sup>12</sup> This study enrolled 35 youth, measured family variables and HRQL at 1, 6 and 12 months after diagnosis and did not find significant associations between family functioning and later survivor HRQL. This study was limited by relying on parent-reported family functioning and focusing on the first year post-diagnosis. Additional prospective studies documenting the associations between family functioning and

survivor outcomes are essential to elucidating whether potentially modifiable family factors are viable targets for interventions seeking to enhance survivor HRQL.

The completion of tumor-directed therapy marks a vulnerable time when families assume primary responsibility for care from medical team. Given the improved survival rate of PBTS and challenges from late effects, it is important to identify early predictors of HRQL in this group. This study examined the prospective influence of family functioning within five months of the end of tumor-directed treatment (baseline) on survivor HRQL nine months later (follow-up). We hypothesized that family functioning at baseline would be positively associated with survivor HRQL at follow-up when controlling for survivor HRQL at baseline and relevant medical and demographic variables.

## Method

### Participants

Participants included 35 PBTS aged 6–16 years and their mothers. Inclusion criteria specified that PBTS (1) received a form of tumor-directed treatment, including any combination of resection, chemotherapy, and radiation therapy, (2) completed all tumor-directed treatment within the previous five months, and (3) were expected to live at least six months. Exclusion criteria included a history of cognitive or developmental delay prior to brain tumor diagnosis and non-English speaking. The study team determined eligibility through review of medical records and communications with the primary neuro-oncologist. No participants were excluded for cognitive difficulties attributed to tumor-directed treatments. Although participation was open to any primary caregiver, only survivors' mothers participated.

Fifty of the 76 (65.8%) families approached agreed to participate. Reasons for not participating included “too much going on” ( $n = 3$ ), scheduling conflicts ( $n = 9$ ), lack of interest ( $n = 8$ ) and passive refusal ( $n = 9$ ). Survivor race, age, and tumor type did not differ between consenting and non-consenting families. Thirty-five of the 50 survivor/mother dyads who completed baseline assessments completed follow-up assessments approximately nine months after baseline. Reasons for not completing follow-up included relapse ( $n = 5$ ), death ( $n = 2$ ), withdrawal ( $n = 2$ ), follow-up at outside medical center ( $n = 1$ ), and passive refusal ( $n = 5$ ). Disease-related and demographic variables did not differ between those who did and did not complete follow-up visits. At baseline, survivors (51.4% female) were 11.00 years of age ( $SD = 2.71$ ), 1.44 years ( $SD = 1.88$ ) from diagnosis and 2.76 months ( $SD = 1.32$ ) from the conclusion of tumor-directed therapy. Mothers were 42.23 years old ( $SD = 5.71$ ) and primarily in a partnered relationship (77.1%). See Table 1 for complete sample characteristics.

### Procedure

The current data are part of a larger study that recruited from a pediatric neuro-oncology program in the northeast United States. All procedures had institutional review board approval and participants provided consent and assent prior to completing study procedures. Baseline visits occurred within five months after completing tumor-directed treatment ( $M =$

2.64 months,  $SD = .11$  months, range 3 weeks – 4.9 months). Follow-up visits occurred approximately 8.79 months ( $SD = 3.83$  months, range 5.33 – 21.13 months) after baseline, however five families (14.2%) had follow-up visits more than 12 months after baseline. Time from baseline to follow-up was not significantly associated with survivor HRQL. Mothers completed measures of demographics, family functioning, family management, and survivor HRQL. PBTS completed measures of family functioning and HRQL. Obtaining ratings of HRQL and family functioning from survivors and mothers addresses the limitations of single-source designs and allows for examination of effects of each source separately.<sup>21</sup> Information on tumor type, treatment, and time since diagnosis was obtained from medical records. Study visits occurred in conjunction with outpatient medical appointments and typically lasted 60 minutes. Participants received a gift card as a thank you.

## Measures

**Pediatric Quality of Life Inventory 4.0 (PedsQL)<sup>22</sup>**—The PedsQL is a widely-used 23-item measure of a child’s physical, emotional, social, and school functioning. The age-appropriate version was used for each survivor (young child: 6–7 years; child: 8–12 years; teen: 13–16 years). Respondents noted how much of a problem each item has been in the past month on a 5-point Likert scale ranging from *never a problem* to *almost always a problem*. Items were reverse scored and linearly transformed on a 0–100 scale, with higher scores representing better HRQL. Survivors and mothers completed the PedsQL at both study visits and only total HRQL scores were used in analyses. The PedsQL has excellent psychometric properties, distinguishes between healthy youth and youth with chronic health conditions, and has been associated with notable indicators of health.<sup>3</sup> In the current sample, the Cronbach alphas for survivor-reported HRQL at baseline were .87 for survivors younger than 12 and .82 for survivors 12 years or older. At follow-up, alphas were .92 for younger survivors and .91 for older survivors. Alphas were .85 and .89 for parent proxy-report at baseline for younger and older survivors, respectively, and .94 and .91 at follow-up.

**McMaster Family Assessment Device (FAD)<sup>23</sup>**—The 12-item General Functioning Scale from the FAD assessed general family functioning. Participants rated how well statements (e.g., “In times of crisis, we can turn to each other for support”) described their family on a 4-point Likert scale ranging from *strongly agree* to *strongly disagree*. The FAD generates scores from 1 to 4 with higher scores indicating greater levels of family dysfunction and scores above 2.0 representing poor family functioning. Mothers and survivors age 8 and higher completed the FAD at baseline. The FAD is considered a “well-established” measure of family functioning and has been used frequently with families of children with a chronic health condition.<sup>24</sup> Alphas were .89 for survivors and .81 for mothers.

**Family Management Measure (FaMM)<sup>25</sup>**—The FaMM is a 53-item questionnaire that measures parents’ perceptions of how their family manages their child’s condition and incorporates care into everyday life. The Family Life Difficulty (FLD) subscale assessed parents’ perceptions of the extent to which their family has difficulty integrating demands related to their child’s chronic illness into family life. The FLD subscale includes 14

statements, such as “it seems as if our child’s condition controls our family life”, to be rated on a 5-point scale ranging from *strongly disagree* to *strongly agree*. Higher values signify greater difficulty in managing the child’s condition. The FaMM has been shown to be a valid and reliable measure of family life difficulty in families of children with chronic illness.<sup>25</sup> Mothers completed the FaMM at baseline and the Cronbach alpha for the FLD subscale was .81.

### Statistical Power and Data Analyses

Power analyses revealed that 29–55 participants were needed to detect effect sizes ranging from .25–.50 for multiple regression analyses with 3–5 predictors.<sup>26</sup> Descriptive analyses were conducted to describe demographic characteristics, tumor-related variables, family functioning, and HRQL. The distribution of scores for primary variables was checked for violations of normality. Preliminary analyses used paired sample t-tests to evaluate differences in mother- and survivor-rated HRQL and one-sample t-tests to compare survivor HRQL to normative values. Associations between demographic and medical variables and HRQL at follow-up were examined using correlations, t-tests and analyses of variance to identify potential covariates. Pearson bivariate correlations examined associations between survivor HRQL and family variables. Baseline family functioning variables that were significantly correlated to follow-up survivor HRQL were entered into a linear multiple regression to test their respective contributions to survivor HRQL at follow-up, while controlling for survivor HRQL at baseline and other identified covariates. Effect sizes were calculated using Cohen’s *d*,  $R^2$  and Cohen’s  $f^2$ . All data were analyzed using SPSS version 24 with alpha level set to  $p = .05$ .

## Results

### Descriptive and Preliminary Analyses

Table 2 presents means and standard deviations for all variables. At baseline, mothers rated survivor HRQL as significantly worse than survivors rated their own HRQL,  $p < .05$ , Cohen’s  $d = 0.42$ . Both survivor- and mother-rated survivor HRQL at baseline were significantly lower than that of the normative sample<sup>8</sup>,  $p < .01$ . Notably, 45.7–48.5% of survivors were below the cut-point for at-risk status (69.7 for self-report and 65.4 for parent-proxy report<sup>3</sup>). At follow-up, HRQL, as rated by both survivors and mothers, remained significantly worse than that of the normative sample,  $p < .01$ , with 34–40% of survivors below the cut-point for at-risk status. There was no difference between self-rated and mother-rated survivor HRQL at follow-up. Change in survivor-rated HRQL from baseline to follow-up ranged from –17.43 to 18.48 with an average absolute change in HRQL of 9.09 ( $SD = 5.95$ ). Change in mother-rated survivor HRQL ranged from –35.87 to 48.91 with an average absolute change of 10.65 ( $SD = 10.25$ ). Survivors reported worse family functioning (FAD) than mothers,  $p < .01$ . Half of survivors and 11.8% of mothers reported scores of 2.0 or higher on the FAD, indicating poor family function.

Many demographic and disease variables were not related to survivor HRQL at follow-up, including survivor sex, age, age at diagnosis, and time since diagnosis. Additionally, tumor location (infratentorial v. supratentorial) and whether or not treatment included radiation

therapy or surgical resection were not related to survivor HRQL at follow-up. However, survivors who received chemotherapy ( $n = 11$ ;  $M = 58.50$ ,  $SD = 18.76$ ) had significantly worse parent-reported survivor HRQL at follow-up compared to those who did not receive chemotherapy ( $n = 23$ ;  $M = 78.40$ ,  $SD = 18.30$ ),  $p < .01$ . WHO tumor grade was unrelated to survivor- and mother-reported survivor HRQL at follow-up. Survivor self-reported HRQL at follow-up was higher for those with mothers who identified being in a partnered relationship ( $n = 27$ ;  $M = 75.60$ ,  $SD = 15.54$ ) compared to those not identifying a partner ( $n = 8$ ;  $M = 60.91$ ,  $SD = 12.23$ ),  $p < .05$ . Maternal educational level also was related significantly to mother-rated survivor HRQL at follow-up,  $p < .01$ , with mothers having at least a college degree ( $n = 17$ ;  $M = 81.14$ ,  $SD = 15.75$ ) reporting higher survivor HRQL than mothers with a high school degree or less ( $n = 10$ ;  $M = 56.30$ ,  $SD = 22.44$ ). Family income based on the three levels reported in Table 1 was unrelated to survivor- or mother-reported survivor HRQL at follow-up.

### Correlational Analyses

Pearson bivariate correlations (Table 3) revealed significant associations between worse survivor-rated family functioning at baseline and lower survivor- and mother-rated survivor HRQL at follow-up. Additionally, greater mother-rated FLD was associated with worse mother-rated survivor HRQL at follow-up.

### Linear Regression Analyses

**Survivor-Rated HRQL**—A hierarchical multiple regression (Table 4) tested the predictive strength of baseline family functioning on survivor-rated HRQL at follow-up. Baseline survivor-rated HRQL and mother's partner status were entered in the first step, followed by baseline survivor-rated FAD in the second step. The overall model explained 62% of the variance in survivor-rated HRQL at follow-up,  $F(3, 24) = 15.45$ ,  $p < .001$ , and had a large effect (Cohen's  $f^2 = 1.94$ ). Adding baseline survivor-rated family functioning significantly improved the model  $R^2 = .11$ ,  $p < .01$  with worse family functioning predicting worse survivor-rated HRQL at follow-up,  $\beta = -.43$ ,  $p < .01$ .

**Mother-Rated HRQL**—A second hierarchical multiple regression (Table 5) tested the strength of baseline family variables as predictors of mother-rated survivor HRQL at follow-up. Mother-rated survivor HRQL at baseline, maternal education level and whether or not the survivor received chemotherapy were entered in the first step, followed by survivor-rated family functioning (FAD) and FLD (FaMM) at baseline in the second step. The overall model explained 66% of the variance in mother-rated survivor HRQL at follow-up,  $F(5, 19) = 10.19$ ,  $p < .01$ , and had a large effect (Cohen's  $f^2 = 2.67$ ). While adding family functioning variables into the model did not significantly improve the amount of variance explained in mother-rated survivor HRQL at follow-up,  $R^2 = .07$ ,  $p > .05$ , survivor-rated family functioning at baseline emerged as a significant predictor,  $\beta = -.31$ ,  $p < .05$ . Maternal education,  $\beta = .35$ ,  $p < .05$ , and chemotherapy treatment,  $\beta = -.46$ ,  $p < .01$ , also significantly predicted mother-rated survivor HRQL at follow-up.



## Discussion

Prior research has focused largely on medical and survivor-related (e.g., executive function) predictors of HRQL in PBTS and generally ignored family functioning variables. The current study highlights the prospective influence of family functioning on survivor HRQL after tumor-directed treatment and offers support for considering social-contextual factors with medical factors as predictors of survivor HRQL.<sup>4</sup> Consistent with our hypotheses, better survivor-rated family functioning at the conclusion of tumor-directed treatment was associated with higher survivor HRQL approximately nine months later as rated by both the survivor and mother, even when controlling for baseline HRQL. These findings are similar to prior studies illustrating the influence of positive family functioning on youth adjustment in pediatric burn,<sup>27</sup> asthma,<sup>28</sup> and traumatic brain injury (TBI)<sup>29</sup> populations.

The pattern of findings highlights the importance of measuring family factors from multiple perspectives and using multiple approaches. Survivor and mother ratings of family functioning were significantly different from one another with four times more survivors than mothers rating family functioning in the poor range. This warrants further study and could be due to choice of measure, differential effects of response bias or simply different perspectives. Additionally, survivor and mother ratings of family functioning demonstrated discrepant associations with survivor HRQL. Only survivor-rated family functioning, but not mother-rated family functioning or integration of illness demands into family routines (FLD), was a significant predictor of survivor HRQL in regression models. Notably, the associations between survivor-rated family functioning and survivor HRQL were found across raters of survivor HRQL. Mother-rated FLD was more strongly correlated with their perceptions of survivor HRQL than their perceptions of general family functioning. This suggests that the components of family functioning most relevant to survivor HRQL vary by rater and underscores the importance of differential reporting by family members.<sup>30</sup> Future studies should assess family functioning from multiple perspectives and include various components of family functioning, including communication, cohesion and problem-solving.<sup>21</sup> Observation-based approaches<sup>24</sup> offer another way to measure this dynamic construct that reduces reporter bias and enhances validity. For example, ratings of families' interactions have been shown to moderate functional impairments in pediatric TBI.<sup>31</sup>

The prospective associations between family functioning and survivor HRQL are notable given that they occurred when controlling for baseline HRQL and pertinent medical and demographic factors related to HRQL at follow-up. Future research is needed to replicate these findings and determine the influence of family variables over and above variables not examined in this study, including survivor neurocognitive functioning (e.g., executive functioning). Such research could establish family functioning as an important factor to assess during off-therapy appointments and a potential target for interventions aiming to enhance survivor HRQL.

Both child and mother reports suggest that the HRQL for PBTS was worse compared to community norms,<sup>9</sup> with 34–48% of survivors in the at-risk range.<sup>3</sup> The current sample reported similar HRQL scores to those reported in other studies of pediatric brain tumor populations.<sup>12</sup> Meeske and colleagues<sup>15</sup> found that for PBTS, psychosocial functioning

scores were highest for those who completed treatment within the past twelve months, but lowest for children who had been off tumor-directed treatment for over 12 months, likely due to the emergence of late effects. This pattern differs from survivors of other childhood cancers, who generally show increases in HRQL farther out from treatment.<sup>15</sup> Future longitudinal research that follows children for a longer period beyond the completion of treatment could examine when declines in HRQL typically develop. This information may help determine the most appropriate time to implement intervention services or added support.

The longitudinal nature of the HRQL data offers important insights into the trajectory of PBTS HRQL in the year following the conclusion of tumor-directed treatment. Within-subjects comparisons of survivor HRQL at baseline and follow-up indicated significant improvements in mother-rated survivor HRQL across the nine month period with medium-sized effects, while self-reported survivor HRQL did not change significantly. Relatedly, mothers rated survivor HRQL as lower than survivors self-rated at baseline but this difference disappeared at follow-up. Such discrepancies between mother and survivor perceptions of HRQL are consistent with prior research with samples of PBTS in the initial months to years following diagnosis and the conclusion of treatment.<sup>8,13,32</sup>

Study findings offer implications for clinical care for PBTS. First, notable modifiable and non-modifiable risk factors for poor survivor HRQL emerged that can be evaluated with routine screeners. Several non-modifiable risk factors, including single-parent status, lower caregiver educational attainment and treatment with chemotherapy, were associated with lower survivor HRQL at follow-up. This suggests targeting supportive resources to families meeting these characteristics to mitigate their role in HRQL. Second, evaluating family functioning routinely throughout the trajectory of tumor-directed treatment may identify families who would benefit from additional support. Lastly, providing evidence-based interventions that enhance family functioning (e.g, problem-solving skills therapy) to families with identified risk factors may promote better survivor HRQL over time. Such approaches have had success in pediatric TBI.<sup>33</sup> For example, better family adaptation to illness demands earlier in the disease course, including defined roles in managing survivor health and low levels of conflict, might moderate survivor HRQL over time due to being more responsive to survivors' needs.<sup>4</sup>

The present study has a number of strengths including a longitudinal design and obtaining multiple perspectives on survivor HRQL and family functioning. However, the findings should be considered in the context of study limitations. First, there was a large time range from baseline to follow-up, though this was unrelated to survivor HRQL at either time point. Second, the participation rate was low, resulting in a relatively small sample that was primarily Caucasian from relatively higher income families with well-educated mothers who tended to be in a partnered relationship. While the sample characteristics represent the population of our cancer center,<sup>34</sup> greater diversity in terms of demographics and family structure would enhance generalizability, particularly since demographics have been associated with PBTS HRQL in prior research<sup>8</sup> and the current study. Additionally, the small sample limited our ability to evaluate the impact of tumor-directed treatment combinations on HRQL. Finally, only 70% of those enrolled completed both visits. Over half of the



participants lost to follow-up (n=8 or 53.3% of those missing) were due to non-controllable factors, including relapse and death. Multi-site studies may facilitate larger and more representative samples.

Overall, findings highlight a significant subset of PBTS experience poor HRQL following tumor-directed therapy and put forward family functioning as a key predictor of HRQL in this population worthy of increased clinical and research attention. Evidence-based screeners and interventions may improve family functioning and HRQL in this vulnerable population.<sup>35</sup> The transition to survivorship may be a period of particular focus, given potential challenges related to returning to “normal” routines and patterns of family interactions.

## Acknowledgments

This research was supported by the National Cancer Institute at the National Institutes of Health (Grant number: 1R03CA162970 to M.C.H.). Yimei Li was partly supported by the Abramson Cancer Center Biostatistics Core funded by the NIH/NCI Cancer Center Support Grant P30CA016520.

## Abbreviation

<b>HRQL</b>	Health-related quality of life
<b>PBTS</b>	Pediatric Brain Tumor Survivor
<b>PedsQL</b>	Pediatric Quality of Life Inventory 4.0
<b>FAD</b>	Family Assessment Device
<b>FaMM</b>	Family Management Measure
<b>FLD</b>	Family Life Difficulty
<b>TBI</b>	Traumatic Brain Injury

## References

1. [Accessed December 15, 2016] Central Brain Tumor Registry of the United States. 2016 CBTRUS Fact Sheet. Available: <http://www.cbtrus.org/factsheet/factsheet.html>
2. Turner CD, Rey-Casserly C, Liptak CC, et al. Late effects of therapy for pediatric brain tumor survivors. *Journal of Child Neurology*. 2009; 24:1455–1463. [PubMed: 19841433]
3. Varni JW, Burwinkle TM, Seid M, et al. The PedsQL 4.0 as a pediatric population health measure: feasibility, reliability, and validity. *Ambulatory Pediatrics*. 2003; 3:329–341. [PubMed: 14616041]
4. Peterson CC, Drotar D. Family impact of neurodevelopmental late effects in survivors of pediatric cancer: review of research, clinical evidence, and future directions. *Clinical Child Psychology and Psychiatry*. 2006; 11:349–366. [PubMed: 17080773]
5. Bronfenbrenner, M. *The ecology of human development*. Cambridge, MA: Harvard University Press; 1979.
6. Brannan AM, Heflinger CA. Distinguishing caregiver strain from psychological distress: Modeling the relationship between child, family, and caregiver variables. *Journal of Child and Family Studies*. 2001:405–418.
7. Macartney G, Harrison MB, VanDenKerkhof E, et al. Quality of life and symptoms in pediatric brain tumor survivors: a systematic review. *Journal of Pediatric Oncology Nursing*. 2014; 31:65–77. [PubMed: 24608699]

8. Barrera M, Atenafu EG, Schulte F, et al. Determinants of quality of life outcomes for survivors of pediatric brain tumors. *Pediatric Blood & Cancer*. 2017
9. Varni JW, Limbers C, Burwinkle TM. Literature review: health-related quality of life measurement in pediatric oncology: hearing the voices of the children. *Journal of Pediatric Psychology*. 2007; 32:1151–1163. [PubMed: 17347186]
10. Netson KL, Ashford JM, Skinner T, et al. Executive dysfunction is associated with poorer health-related quality of life in pediatric brain tumor survivors. *Journal of Neurooncology*. 2016; 128:313–321.
11. Reimers TS, Mortensen EL, Nysom K, et al. Health-related quality of life in long-term survivors of childhood brain tumors. *Pediatric Blood & Cancer*. 2009; 53:1086–1091. [PubMed: 19499581]
12. Penn A, Lowis SP, Stevens MC, et al. Family, demographic and illness-related determinants of HRQL in children with brain tumours in the first year after diagnosis. *Pediatric Blood & Cancer*. 2009; 53:1092–1099. [PubMed: 19743518]
13. Kuhlthau KA, Pulsifer MB, Yeap BY, et al. Prospective study of health-related quality of life for children with brain tumors treated with proton radiotherapy. *Journal of Clinical Oncology*. 2012; 30:2079–2086. [PubMed: 22565004]
14. Yock TI, Bhat S, Szymonifka J, et al. Quality of life outcomes in proton and photon treated pediatric brain tumor survivors. *Radiotherapy & Oncology*. 2014; 113:89–94. [PubMed: 25304720]
15. Meeske K, Katz ER, Palmer SN, et al. Parent proxy-reported health-related quality of life and fatigue in pediatric patients diagnosed with brain tumors and acute lymphoblastic leukemia. *Cancer*. 2004; 101:2116–2125. [PubMed: 15389475]
16. Kazak, AE., Rourke, MT., Navsaria, N. Families and other systems in pediatric psychology. In: Roberts, M., Steele, R., editors. *Handbook of pediatric psychology*. Vol. 4. 2009. p. 656–671.
17. Barakat LP, Marmer PL, Schwartz LA. Quality of life of adolescents with cancer: family risks and resources. *Health and Quality of Life Outcomes*. 2010; 8:63. [PubMed: 20584303]
18. Van Schoors M, Caes L, Knoble NB, et al. Associations between family functioning and child adjustment after pediatric cancer diagnosis: A meta-analysis. *Journal of Pediatric Psychology*. 2017; 42:6–18. [PubMed: 28173163]
19. Barakat LP, Li Y, Hobbie WL, et al. Health-related quality of life of adolescent and young adult survivors of childhood brain tumors. *Psychooncology*. 2015; 24:804–811. [PubMed: 25111013]
20. Kulkarni AV, Piscione J, Shams I, et al. Long-term quality of life in children treated for posterior fossa brain tumors. *Journal of Neurosurgery: Pediatrics*. 2013; 12:235–240.
21. Holmbeck GN, Li ST, Schurman JV, et al. Collecting and managing multisource and multimethod data in studies of pediatric populations. *Journal of Pediatric Psychology*. 2002; 27:5–18. [PubMed: 11726675]
22. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Medical Care*. 2001; 39:800–812. [PubMed: 11468499]
23. Epstein NB, Baldwin LM, Bishop DS. The McMaster family assessment device. *Journal of Marital and Family Therapy*. 1983; 9:171–180.
24. Alderfer MA, Fiese BH, Gold JJ, et al. Evidence-based assessment in pediatric psychology: family measures. *Journal of Pediatric Psychology*. 2008; 33:1046–1061. discussion 1062–1044. [PubMed: 17905801]
25. Knafl K, Deatrick JA, Gallo A, et al. Assessment of the psychometric properties of the Family Management Measure. *Journal of Pediatric Psychology*. 2011; 36:494–505. [PubMed: 19451173]
26. Soper, D. [Accessed December 15, 2016] A-priori sample size calculator. Available: <http://www.danielsoper.com/statcalc/calc01.aspx>
27. Landolt MA, Grubenmann S, Meuli M. Family impact greatest: predictors of quality of life and psychological adjustment in pediatric burn survivors. *Journal of Trauma and Acute Care Surgery*. 2002; 53:1146–1151.
28. Sawyer MG, Spurrier N, Whaites L, et al. The relationship between asthma severity, family functioning and the health-related quality of life of children with asthma. *Quality of Life Research*. 2000; 9:1105–1115. [PubMed: 11401043]

29. Wade SL, Michaud L, Brown TM. Putting the pieces together: preliminary efficacy of a family problem-solving intervention for children with traumatic brain injury. *Journal of Head Trauma Rehabilitation*. 2006; 21:57–67. [PubMed: 16456392]
30. De Los Reyes A. Introduction to the special section: More than measurement error: Discovering meaning behind informant discrepancies in clinical assessments of children and adolescents. *Journal of Clinical Child and Adolescent Psychology*. 2011; 40:1–9.
31. Wade SL, Zhang N, Yeates KO, et al. Social Environmental Moderators of Long-term Functional Outcomes of Early Childhood Brain Injury. *JAMA Pediatrics*. 2016; 170:343–349. [PubMed: 26902662]
32. Penn A, Shortman RI, Lowis SP, et al. Child-related determinants of health-related quality of life in children with brain tumours 1 year after diagnosis. *Pediatric Blood & Cancer*. 2010; 55:1377–1385. [PubMed: 20981692]
33. Narad ME, Minich N, Taylor HG, et al. Effects of a Web-Based Intervention on Family Functioning Following Pediatric Traumatic Brain Injury. *Journal of Developmental and Behavioral Pediatrics*. 2015; 36:700–707. [PubMed: 26461100]
34. Bureau USC. State & County QuickFacts. Available: <http://quickfacts.census.gov/qfd/states/42000.html>
35. Kazak AE. Evidence-based interventions for survivors of childhood cancer and their families. *Journal of Pediatric Psychology*. 2005; 30:29–39. [PubMed: 15610982]

**Table 1**

## Sample Characteristics of Participants

Variables	<i>n</i> (%) or <i>M</i> ± <i>SD</i>
Survivor age in years	11.0 ± 2.7
Survivor Gender	
Male	17 (48.6)
Female	18 (51.4)
Survivor Race	
Caucasian	30 (85.7)
African-American	4 (11.4)
Other	1 (2.9)
Tumor-related characteristics	
Tumor types	
Astrocytoma	10 (28.6)
Low grade glioma	8 (22.9)
Ependymoma	4 (11.4)
Ganglioglioma	4 (11.4)
Other *	9 (25.7)
WHO grade	
I	17 (48.6)
II	9 (25.7)
III	4 (11.4)
IV	3 (8.6)
N/A (germinoma)	2 (5.7)
Tumor location	
Infratentorial	13 (37.1)
Supratentorial	22 (62.9)
Treatment	
Surgical resection only	13 (37.1)
Chemotherapy only	3 (8.6)
Radiation therapy only	1 (2.9)
Surgery and chemotherapy	3 (8.6)
Surgery and radiation therapy	10 (28.6)
Surgery, chemotherapy, radiation therapy	5 (14.3)
Years since diagnosis	1.4 ± 1.9
Months since completion of treatment	2.6 ± 1.3
Mother age in years	42.2 ± 5.7
Mother Education <sup>a</sup>	
High school degree or less	10 (30.3)
Some college/vocational school	6 (18.2)
Graduated from 4 year college or higher	17 (51.5)

Variables	<i>n</i> (%) or <i>M</i> ± <i>SD</i>
Total Household Income <sup><i>b</i></sup>	
< \$34,000	7 (22.6)
\$34,000 – \$100,000	11 (35.5)
>\$100,000	13 (41.9)
Relationship status	
In partnered relationship	27 (77.1)
Not in partnered relationship	8 (22.9)

Note.

<sup>*a*</sup>N = 33,

<sup>*b*</sup>N = 31

\* Other tumor types include: medulloblastoma (n=3), germinoma (n=2), craniopharyngioma (n=1), germ cell tumor (n=1), meningioma (n=1), and neurocytoma (n=1).

Author Manuscript

Author Manuscript

Author Manuscript

Author Manuscript

**Table 2**

## Mean Values for Primary Variables

Variable	Baseline <i>M (SD)</i>	Follow-Up <i>M (SD)</i>	Normative Values (if applicable)
Survivor-rated survivor HRQL	69.36 (11.89) <sup>a</sup>	72.25 (15.96)	83.84 <sup>b</sup>
Mother-rated survivor HRQL	62.80 (17.55) <sup>a</sup>	71.96 (20.48)	82.70 <sup>c</sup>
Survivor-rated Family Functioning (FAD)	1.88 (.54) <sup>d</sup>		
Mother-rated Family Functioning (FAD)	1.55 (.39) <sup>d</sup>		
Mother-rated Family Life Difficulty (FaMM)	28.42 (7.91)		

<sup>a</sup>Mother-rated survivor HRQL was significantly lower than survivor-rated HRQL at baseline,  $p < .05$ , Cohen's  $d = 0.42$ .

<sup>b</sup>Survivor-rated survivor HRQL at baseline and follow-up were significantly lower than normative values,  $p < .01$ .

<sup>c</sup>Mother-rated survivor HRQL at baseline and follow-up were significantly lower than normative values,  $p < .01$ .

<sup>d</sup>Survivor-rated family functioning was significantly worse than mother-rated family functioning,  $p < .01$ .



**Table 3**

Bivariate Correlations Among Key Study Variables

	N	2	3	4	5	6	7	8
1 Follow-up Survivor-rated HRQL	35	.65**	.73**	.26	-.74**	-.14	-.30	-.06
2 Follow-up Mother-rated Survivor HRQL	34	---	.47**	.47**	-.54**	-.24	-.38*	-.03
3 Baseline Survivor-rated HRQL	33	---	---	.41**	-.47**	.09	.07	.06
4 Baseline Mother-rated Survivor HRQL	35	---	---	---	-.13	-.08	-.23	-.16
5 Baseline Survivor-rated Family Functioning (FAD)	29	---	---	---	---	.15	.23	-.06
6 Baseline Mother-rated Family Functioning (FAD)	35	---	---	---	---	---	.41**	.32
7 Baseline Mother-rated FLD (FaMM)	34	---	---	---	---	---	---	.36*
8 Time from Baseline to Follow-Up	35	---	---	---	---	---	---	---

Note. Higher scores on the FAD indicate worse family functioning.

\*  $p < .05$ ;

\*\*  $p < .01$ .

**Table 4**

Hierarchical Regression Analysis Predicting Survivor-Rated HRQL at Follow-up From Family Variables

	$R^2$	Survivor-Rated HRQL at Follow-Up $\beta$
Step 1	.51 **	
Survivor-Rated Survivor HRQL at Baseline		.68 **
Mother Partner Status		.18
Step 2	.11 *	
Survivor-Rated Survivor HRQL at Baseline		.43 **
Mother Partner Status		.14
Survivor-Rated Family Functioning (FAD)		-.43 **
Total Adjusted $R^2$	.62 ** <sup>a</sup>	

Note. Higher scores on the FAD indicate worse family functioning.

\*  $p < .05$ ;

\*\*  $p < .01$ .

<sup>a</sup>  $F(3, 24) = 15.45, p < .001$ .

**Table 5**

Hierarchical Regression Analysis Predicting Mother-Rated Survivor HRQL at Follow-Up From Family Variables

	$R^2$	Mother-rated Survivor HRQL at Follow-Up $\beta$
Step 1	.61 <sup>**</sup>	
Mother-Rated Survivor HRQL at Baseline		.37 <sup>*</sup>
Maternal Education Level		.43 <sup>**</sup>
Treatment Including Chemotherapy		-.37 <sup>*</sup>
Step 2	.05	
Mother-Rated Survivor HRQL at Baseline		.35 <sup>*</sup>
Maternal Education Level		.35 <sup>*</sup>
Treatment Including Chemotherapy		-.46 <sup>**</sup>
Survivor-Rated Family Functioning (FAD)		-.31 <sup>*</sup>
Mother-Rated FLD (FaMM)		.23
Total Adjusted $R^2$	.66 <sup>**a</sup>	

Note. Higher scores on the FAD indicate worse family functioning.

\*  $p$  .05;

\*\*  $p$  .01.

<sup>a</sup>  $F(5, 19) = 10.19, p < .01$ .