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The mediating role of visuospatial planning skills on adaptive function among young adult survivors of childhood brain tumors

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Keywords: Rey-Osterrieth, Executive Function, Brain Tumor, Adaptive Function, Adulthood, Long-term outcomes

Abstract

The Boston Qualitative Scoring System (BQSS) was used as a method to examine executive skills on the Rey-Osterrieth complex figure (ROCF). Young adult survivors of childhood brain tumor (N = 31) and a demographically-matched comparison group (N = 33) completed the ROCF copy version and Grooved Pegboard, and informants were administered the Scales of Independent Behavior-Revised (SIB-R) and Behavior Rating Inventory of Executive Function (BRIEF). Survivors had significantly lower BQSS planning and SIB-R community living skills and greater perseveration. Mediation analyses found that BQSS planning skills mediate the relationship between group and community living skills. Convergent findings of the BRIEF Planning, and discriminant findings with the BQSS Fragmentation, BRIEF Emotional Control, and Grooved Pegboard support the planning construct as the specific mediator in this model. Together, these findings highlight the role of planning skills in adaptive functions of young adult survivors of childhood brain tumor.

The mediating role of visuospatial planning skills on adaptive function among young adult survivors of childhood brain tumor

Executive function difficulty is among the commonly reported neuropsychological sequelae of pediatric brain tumor and treatment in short-term (Howarth, et al., 2013; Kahalley et al., 2013; Palmer et al., 2010) as well as long-term, adult survivors (Brinkman et al., 2012; Ellenberg et al., 2009). In fact, in survivors, executive function is more likely to be affected than emotional and psychological well-being (Howarth et al., 2013; Ness et al., 2008; Zeltzer et al., 2008). Domains of executive functioning such as attention, working memory, and problem solving are commonly noted difficulties for survivors of childhood brain tumor (Dennis et al., 1991; Reddick et al., 2003; Spiegler, Bouffet, Greenbert, Rutka, & Mabbott 2004).

Understanding executive dysfunction in this population is important as it has been shown to be related to everyday adaptive function abilities in other populations. For example, previous research has utilized both performance- and informant report-based measures of executive functioning to identify important predictors of adaptive behaviors in neurologically impaired populations such as traumatic brain injury (TBI; Brookshire, Harvey, Song, & Zhang, 2004; Gioia & Isquith, 2004; Mangeot, Armstrong, Colvin, Yeates, & Taylor 2002), autism spectrum disorders (Liss et al., 2001), Alzheimers disease (Farias, Harrell, Neumann, & Houtz, 2003) and epilepsy (Culhane-Shelburne, Chapieski, Hiscock, & Glaze, 2002).

Planning skill is under the umbrella of executive function and is defined as identifying and organizing the steps needed to complete a task or achieve a goal (Lezak, 2004). Planning is thought to be important in everyday activities. Indeed, in childhood epilepsy, planning skill was found to predict adaptive functioning (Culhane-Shelburne et al., 2002). In survivors of childhood

brain tumor, planning skill has been described by self-and informant report measures. Short-term survivors, 1 year from diagnosis of medulloblastoma were reported to be in the average range on planning and organization as measured by the Behavior Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, & Kenworthy, 2000; Palmer et al., 2010). Long-term cancer survivors on average five years post-diagnosis, however, had elevations on planning at a rate of 25% (Kahalley et al., 2013). Further, of all BRIEF subscales, survivors with planning elevations were most highly referred for clinical follow up suggesting that planning difficulties may reflect untreated or inadequately treated behavioral concerns (Kahalley et al., 2013). Thus, planning may be especially important to examine in long-term survivors (5 years or greater since diagnosis). Yet the previously mentioned study sample included a combined group of brain tumor survivors and survivors of non-CNS cancer (acute lymphoblastic leukemia). When comparing long-term brain tumor survivors to non-CNS cancer survivors on planning, Howarth et al. (2013) reported that in adolescent brain tumor survivors, on average 6 years post-diagnosis, planning and organization skills were significantly lower than both healthy siblings and non-CNS solid tumor survivors. This suggests that planning may be at risk in long-term survivors of CNS tumors. In contrast, Howarth et al. (2013) found that the adolescent brain tumor survivors showed no significant differences on emotional control subscale of the BRIEF compared with healthy siblings and non-CNS solid tumor survivors. This is important to note as it suggests that survivors do not show a global impairment across executive skills.

Taken together, previous research has provided valuable information on reported planning skills in childhood survivors of a brain tumor. While report measures can have ecological validity, information from an unstructured paper and pencil measure could provide complimentary information about planning. To the authors' knowledge, performance-based

measures of planning have yet to be studied in young adult survivors of childhood brain tumor. To better understand planning skills in this population, the current study aimed to examine planning utilizing a performance-based process approach with the Boston Qualitative Scoring System (BQSS; Stern et al., 1994; 1999). The process method examines executive skills by observing a patient's approach to an unstructured task (Kaplan, 1990; Libon, Swenson, Ashendorf, Bauer, & Bowers, 2013). The BQSS (Stern et al., 1994; 1999) is a comprehensive scoring system that provides quantifiable data by evaluating qualitative aspects of the patient's approach to the construction of the Rey-Osterrieth Complex Figure (ROCF). BQSS is a reliable and valid measure of executive functions in typical populations (Stern et al., 1994; 1999) and research has found expected group differences in clinical populations such as schizophrenia (Mahurin, Eckert, Velligan, Hazelton, & Miller, 1997), Attention Deficit Hyperactivity Disorder (ADHD; Schreiber, Javorsky, Robinson, & Stern, 1999), and TBI (Javorsky, Rosenbaum, & Stern, 1999).

Poorer planning using the BQSS was reported in individuals diagnosed with ADHD compared with a typically developing group (Schreiber et al., 1999). Lower overall organization and planning skills also were found in individuals diagnosed with obsessive compulsive disorder compared with a typically developing group (Shin et al., 2004). Further, planning on the BQSS significantly enhanced the prediction of memory performance on the ROCF above depression status alone in depressed elderly patients and a typically developing group (Elderkin-Thompson et al., 2004). Thus, the first aim of this study was to utilize the BQSS to examine planning difficulties in young adult survivors of childhood brain tumor and compare with a typically developing group.

Furthermore, the relationship between planning skills and adaptive functioning could be

important to understanding the effect of executive functions on daily living skills. Previous research has yielded significant findings between executive functioning and adaptive functioning using performance-based tests and informant-report measures in pediatric neurological populations. In childhood TBI, higher rated general executive problems were related to lower adaptive behavior ratings (Mangeot et al., 2002). Moreover, specific executive function constructs, shifting attention and planning skills, were the strongest predictors of general adaptive functioning in children with epilepsy (Culhane-Shelburne et al., 2002). Survivors of childhood brain tumor also experience difficulties of adaptive functions (Carpentieri et al., 2003; Papazoglou, King, Morris, & Krawiecki, 2009; Stargatt et al., 2006). However, little is known with regard to how executive skills, in particular planning skills, contribute to these deficits in adaptive behaviors. Community living skills, in particular, are an important set of advanced adaptive function skills especially relevant to young adult survivors of childhood brain tumor as they transition to living independently. The second aim of this study was to better understand the role of planning skills in community living skills in brain tumor survivors and a comparison group.

The current study aimed to first, examine adaptive function, and executive performance using the BQSS (including planning skill) in young adult survivors of childhood brain tumor compared with a typically developing group and second, investigate a mediation model such that the effect of group (brain tumor vs. comparison) on adaptive functioning is predicted through the indirect effect of BQSS planning skills. It was hypothesized that first, young adult survivors of childhood brain tumor would exhibit both lower executive and adaptive functioning compared with typically developing adults, and second, planning on the BQSS would mediate the relationship between group and adaptive functioning (community living skills).

Secondary analyses were conducted to provide support for the specificity of planning skill as the mediating variable in the hypothesized conceptual model of group predicting community living skills. Given the challenges in parsing out the visuospatial and general cognitive aspect from the BQSS planning measure, we attempted to find support for planning specificity in three ways. First, to add support to the construct of planning in this conceptual model, an informant report of planning on the BRIEF was tested in the mediation model, which we expected to be significant (concurrent validity). Second, to separate planning as a specific skill in this model rather than representing global executive skills, we tested a discriminant mediator that we do not believe to be disrupted in survivors, informant report of emotional control on the BRIEF. In addition, we tested a BQSS executive variable that we did not expect to be different between groups, fragmentation. Third, to separate motor skill from planning skill, motor skill was tested in place of the mediator in the model and also was not expected to be significant.

2. Methods

2.1 Participants

Thirty-one young adult survivors of childhood brain tumor and thirty-three demographically matched healthy participants between the ages of 18 and 30 years participated in the current study. The study was approved by the local Institutional Review Board and all participants provided written informed consent. Young adult survivors of childhood brain tumor were recruited from Brain Tumor Foundation of Georgia newsletter and opt-in letters mailed to survivors, and identified by: first, a previous longitudinal study, in which they participated as children (Carlson-Green, Morris, & Krawiecki, 1995) and second, a local children's hospital.

Given that the current study required survivors to contact the researchers after receiving a letter, the sample may not be representative of all young adult survivors of childhood brain tumor. All survivors passed a test of visual acuity (Rosenbaum Pocket Vision Screener). Basic motor skill was assessed with the MMSE “write a sentence” and “copy two pentagons” items (Folstein, Folstein, & McHugh, 1975). If participants were not able to adequately complete these tasks, they were not included in the current study.

Survivors were an average age of 8.84 years of age at the time of diagnosis ($SD= 4.82$, age range: 1-19 years) and 13.72 years since time of diagnosis ($SD=5.21$, range: 4-24 years). Treatment history included neurosurgery (96.8%), chemotherapy (41.9%), radiation treatment (58.1%), and other neurological complications included: hydrocephalus (77.4%) and hormone deficiency (61.3%). Young adult survivors had diverse tumor pathologies, including astrocytic (38.7%), embryonal (41.9%), tumors of the sellar region (craniopharyngioma; 12.9%), pineal (3.2%), and oligodendroglial (3.2%). Tumor locations were as follows posterior fossa (71.0%), brainstem (6.5%), pituitary (3.2%), ventricles (9.7%), temporal lobe (6.5%), and occipital lobe (3.2%).

The comparison group comprised of students in the psychology research participant pool at local universities, friends of survivors, and individuals from the community. Participants in this group passed the visual acuity and basic motor screen described earlier. Comparison participants were screened with the Structured Clinical Interview for DSM-IV Axis I Diagnosis to rule out current psychopathology (SCID; First, Spitzer, Gibbon & Williams, 1995); only those who did not meet criteria for any current Axis I disorders were included. Groups were not significantly different on any demographic variable (Table 1). Participants were on average 22 years at testing and identified as 66% Caucasian, 20% African American, 6% Hispanic, 5%

Asian, 3% mixed race, and 5% not reported. SES was on average 2.36 ($SD=1.14$). Groups were not significantly different on whether they were financially independent or dependent ($\chi^2(1, N=62)=0.98, p=.32$).

2.2 Measures

Executive function: The *Boston Qualitative Scoring System* (BQSS; Stern, et al., 1999) was utilized to obtain a qualitative assessment of visuospatial and executive functions from the copy trial of the Rey-Osterrieth Complex Figure (ROCF). The ROCF is a paper and pencil neuropsychological measure of visuospatial constructional ability (copy trial) with subsequent memory trials of figure construction (Meyers & Meyers, 1995). The executive measure for the copy trial was analyzed in the current study within the context of standardized ROCF instructions. As participants constructed the figure, the examiner would follow the participant and draw along piece by piece, numbering the order of the components. The ROCF raw score for the copy trial was utilized for a measure of ROCF copy performance. Measures of executive function were derived from the figure drawing based on qualitative assessment of how drawing was approached. There are eleven qualitative executive function scores that can be calculated by this method. Planning was chosen because it was our main construct of interest. Perseveration and fragmentation were used because they have also been shown to correlate with other executive function measures (Stern et al., 1994; 1999). Neatness was included originally to be used as an indirect measure of motor skill. We calculated qualitative scores for executive subscales of interest according to criteria presented in the BQSS Manual (Stern et al., 1999). Planning skills were defined by the BQSS manual as “the order in which the elements are drawn, placement on the page, placement within the figure, and overall integrity of the production” (range 0-4; 0=poor planning, 4=good planning). Neatness, our proxy for motor skill, was scored

on “how neatly a figure is drawn as evidenced by the number of wavy lines, gaps and overshoots, cross-outs, and rounded corners” (range 0-4, 0=extremely messy, 4=very neat). Perseveration was measured based on “the extent of recognizably inappropriate repetitions” (range 0-4; 0=extreme perseveration, 4=no perseveration) and fragmentation was determined on “the integration of the information (i.e., whether or not the individual elements are drawn as whole units)” (range 0-4; based on number of times fragmentations occurred) (Stern et al., 1999). This qualitative evaluation results in a quantitative score on an ordinal scale. The BQSS z scores for all subscales were calculated based on a sample of 30 healthy adults (Hartman and Porter, 1998). The perseveration variable was highly skewed and was noted to have a more bimodal distribution reflecting either impairment at greater than 1.5 SD below the mean, or not impaired with a positive z value. Thus, perseveration was converted to a dichotomous variable for analyses.

Due to the qualitative nature of scoring the ROCF using the BQSS, two research assistants were trained on scoring. All 64 ROCF copy trials were scored by each research assistant. Overall reliability of the BQSS scores using intra-class correlation was large at ($\alpha=0.96$). Any scoring discrepancies were discussed and agreed upon by referring to the BQSS Manual.

Adaptive functioning: The *Scales of Independent Behavior-Revised* (SIB-R; Bruininks, Woodcock, Weatherman, & Hill, 1996) was administered as a structured interview to informants of participants that consists of four domains with age-based norms. The four domains include Motor Skills, Social Interaction and Communication skills, Personal Living Skills, and Community Living Skills. This study utilized the Community Living Skills (time and punctuality, money and value, work skills, and home/community) for primary analyses. Criteria

for informant included that this person was knowledgeable of the participants' daily living skills for at least the past year. Informants for our participants were generally parents, spouses or roommates.

Informant report of executive function in daily life: Behavior Rating Inventory of Executive Function -Informant form of the Behavior Rating Inventory of Executive Function-Adult Version (BRIEF; Roth, Isquith, & Gioia, 2005; ages 18-90 years old) was administered to parents or significant others to obtain a general rating of executive function behaviors in home and social environments. A subset of the current sample (n=50) received this measure. The BRIEF informant measures nine different aspects of executive functioning on empirically derived clinical scales, including the Inhibit, Shift, Emotional Control, Self-Monitor, Initiate, Working Memory, Plan/Organize, Task Monitor, and Organization of Materials scales. The BRIEF has demonstrated good reliability, with high test-retest reliability (.82 for parents) internal consistency (alphas - .80 to .98). Convergent and divergent validity have been established with other measures of emotional and behavioral functioning, and the BRIEF has also demonstrated utility in differentiating clinical and non-clinical children and adolescents with Attention Deficit Hyperactivity Disorder. For purposes of this study, we used the Plan/Organize subscale T score as a second measure of planning skill to add conceptual support to the mediation model in secondary analyses. The Emotional Control subscale T score was identified as an executive skill that was not expected to be disrupted in survivors and was used as a control or discriminant executive measure in the secondary analyses.

Motor skill: The Grooved Pegboard (Ruff & Parker, 1993) dominant hand z-score was used to measure motor skill. This task requires the participant to fit key-hole-shaped pegs into holes in a pegboard using only one hand at a time, going as quickly as possible. The raw score is

the amount of time in seconds that it takes the participant to place pegs into all the holes on the board. Z- scores computed from normative data (Bornstein, 1985) were used in the current study. The dominant hand z-score was tested in secondary analyses as the mediator in the relationship between group and community living skills as a control task.

Socioeconomic status (SES): Hollingshead Four-Factor Index of Social Status (Hollingshead, 1975). SES was calculated for descriptive purposes. A qualitative score from 1 to 5 (1=high, 5=low) is derived using this scale as a measure of SES by assessing current occupation and education of participants, participants' parents, or caregivers (based on the financial dependence of the participant). For the purposes of assessing group differences, SES was dichotomized into low and high SES (low=3,4,5= 34%, high=1,2= 57%, SES not reported=9%).

Intelligence: The Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999) is a commonly used abbreviated test of intelligence. It includes four core subtests derived from the unabbreviated Wechsler Adult Intelligence Scale III: Vocabulary, Similarities, Matrix Reasoning, and Block Design. These subtests measure: ability to verbally define words, verbal and perceptual reasoning, and perceptual-motor problem solving. The Full Scale Intelligence Quotient standard scores were used as a measure of general cognitive outcome for descriptive purposes. This measure was not used as covariate or mediator in the analyses due to previously described limitations in developmental populations (Dennis et al., 2009).

2.4 Statistical Approach

All variables were examined for normality, outliers, and that they met assumptions for all statistical tests used. A confound was defined as a demographic variable that both is significantly different between groups and is correlated with the dependent variable (community living skills).

Variables that met these criteria would be added as a covariate in subsequent analyses. To test for potential confounds, independent samples t-tests and chi-square tests were employed to test group differences on demographic variables (age, sex, ethnicity, and SES) and bivariate correlations were employed to test the relationship between demographic variables and community living skills.

For aim 1, to test group differences, non-parametric Mann-Whitney U tests were performed with BQSS variables, which are ordinal in nature and ROCF copy raw score. Independent samples t-tests were used to test group differences on community living. Further, group differences in the proportion of participants who performed at a significant level of impairment on each variable were tested with Fisher exact tests. Impairment was defined as a z-score of -1.5 or lower.

For aim 2, hierarchical regression analyses were utilized to test the hypothesized mediation model. The SPSS “indirect” script for SPSS (<http://www.afhayes.com/spss-sas-and-mplus-macros-and-code.html>) was used to test the model with group membership (survivors vs. comparison) as the independent variable, community living skills as the dependent variable, and BQSS planning skills as the hypothesized mediator. With regard to the statistical significance of the model, prior research has introduced concerns regarding the Baron and Kenny (1986) model for mediation for lacking power, and the Sobel test for not being able to detect effect size in small samples (≤ 100) (Preacher & Hayes, 2004). Thus, bootstrapping (10,000 samples) was employed to confirm the mediation model. The bootstrapping approach (Preacher, Rucker & Hayes, 2007) is nonparametric and estimates effect sizes accurately with small samples and skewed distributions by resampling with replacement. It provides a 95% confidence interval; a significant effect will result in a confidence interval that does not include zero. While the

bootstrapping approach has many advantages, one limitation is that it can increase the likelihood of a type 1 error. Therefore, to add support to the bootstrapping results, we also conducted a Test of Joint Significance which is based on the statistical significance of the a and b paths using regression. If *both* paths are significant, the indirect effect is considered statistically significant. While this test is not a direct comparison of the effects of *c* and *c'*, it is less vulnerable to type 1 error. This test also shows comparable results with other alternatives (Hayes & Scharkow, 2013).

For secondary specificity analyses, separate regression models were employed using the bootstrapping approach described above. In all models, group was the independent variable and community living skills (SIB-R) was the dependent variable. The following mediators were tested in four separate models: report of planning/organization (BRIEF), emotional control (BRIEF), BQSS fragmentation, and motor skill (Grooved Pegboard).

3. Results

As shown in Table 1, independent samples t-tests and chi-square tests demonstrated that groups were not significantly different on any demographic variable. Further, the following demographic variables were not significantly correlated with the community living skills: age at testing ($r=-0.04, p=0.75$), socioeconomic status ($r=0.17, p=0.20$), ethnicity ($r=0.14, p=0.30$), and sex ($r=0.06, p=0.63$). Given the small correlations of demographic variables with the dependent variable, these variables did not meet criteria for a potential confound variable. This also indicates that demographic variables would not be good extraneous variables in analyses because they do not remove significant variance from the dependent variable. Thus, these variables were not added as covariates in subsequent analyses.

3.1 Aim 1: Group Differences on Executive and Adaptive Functioning

On executive scales of the BQSS, survivors scored significantly lower on planning and perseveration compared with the comparison group (see Table 2). The groups did not show significant differences on neatness and fragmentation. Similarly, survivors evidenced a higher percent impairment on planning (32%) and perseveration (29% survivors impaired); however no differences in impairment on the executive function scales of neatness and fragmentation were detected. With regard to community living, survivors demonstrated significantly lower functional community living skills and significantly greater impairment (16% survivors impaired). Although means for both groups suggest that on average, functioning across BQSS executive skills and community living adaptive functioning were within normal limits, examining proportion of impairment within each group revealed a greater proportion of survivors with difficulties in these abilities.

3.2 Aim 2: Mediation Model

Aim 2 tested a model in which the group and community living skills would be associated through a mediator, planning skills. In other words, group differences in community living skills were expected to be influenced by planning skill on the BQSS. Group had a significant effect on community living skills (path c: $B=-12.94$, $SE=4.80$, $p=0.01$) such that brain tumor history related to lower community living (see Figure 1). Planning skill was significant when regressed on group (path a: $B=-6.56$, $SE=2.71$, $p=0.02$), such that brain tumor history related to lower planning skill. The effect of planning on community living was also statistically significant (path b: $B=0.45$, $SE=0.22$, $p=0.05$) such that better planning was related to higher community living skills. The significance of both a and b paths suggest a statistically significant indirect effect based on the Test of Joint Significance. The statistical significance of the entire model of the indirect effect of group on community living through planning skills, was tested

with bootstrapping with 10,000 samples and was significant ($B=-2.88$, $SE=1.97$, $CI: -8.64, -0.28$) such that brain tumor history was related to lower community living skills through lower planning skill. This mediation model, accounts for 13.40% (adjusted R^2) of the variance in community living skills.

Model Specificity

Using the same bootstrapping approach, we examined the specificity of the above model by testing four independent models. These were informant report of planning (concurrent validity), BQSS fragmentation and informant report of emotional control (discriminant validity), and motor skill (discriminant validity); each substituted for the mediating variable of BQSS planning. As predicted, informant report of planning skills significantly mediated the relationship between group and community living skills ($CI: -12.46, -.90$). In contrast, emotional control as the mediating variable did not lend to a statistically significant model ($CI: -6.12, 4.37$) as predicted and nor did BQSS fragmentation ($CI: -3.41, .64$). The mediation model of group differences predicting community living skills through motor skill also was not statistically significant ($CI: -5.45, 5.14$).

4. Discussion

The current study aimed to examine young adult survivors' planning skills during the ROCF and community living skills compared with a healthy comparison group. Results suggest that young adult survivors have greater difficulty in community living skills (16% impaired) relative to the healthy comparison group. It is important to note that while survivors were significantly lower on community living, the mean score was average with a range of 61-135 which suggests a wide range of functioning that is not unexpected in this heterogeneous sample of young adult survivors of brain tumor. These findings are consistent with previous literature on

adaptive functioning in survivors of childhood brain tumor (Papazoglou et al., 2009; Stargatt et al., 2006). With regard to executive functions, survivors were not globally impaired on all BQSS executive skills measured, but instead showed areas of specific difficulty. Survivors performed significantly lower and had higher percent impairment on planning skill, and made more perseverative errors than the healthy comparison group. In contrast, there were no significant group differences on neatness and fragmentation between the groups. Reduced planning performance in long-term survivors is consistent with other long-term studies using informant of planning (Howarth et al., 2013; Kahalley et al., 2013). We have extended planning skill findings to longer-term survivors (13 years post-diagnosis), with a process-based performance measure of planning during the ROCF.

The second aim of this study found that the primary mediation model was significant; brain tumor history was related to poorer planning performance, which was associated with lower community living skills. The current study was cross-sectional in nature and longitudinal studies would be needed to test a causal link between planning skill and adaptive function. Recent research has shown that the bootstrapping method employed, while increasing power for small samples, can increase the rate of type 1 errors (Hayes & Scharkow, 2013; Koopman, Howe, Hollenbeck, & Sin, 2015). With that in mind, the Test of Joint Significance was used to increase confidence in the bootstrapping result, and this test was also statistically significant.

Anderson (2002) discussed two fundamental problems with neuropsychological assessments of executive function that are relevant to the current discussion on planning skills. First, assessments designed to measure executive functions tap into processes of both executive and non-executive functions. As a visuospatial drawing task, the ROCF requires motor and visuospatial functioning to complete the task. Therefore, these components also are involved in

the BQSS planning measure. To increase confidence that the construct of planning was critical to the current model in this study and that it was not due to motor impairments, motor skill of the Grooved Pegboard was tested as a control or discriminant task. The lack of a significant mediational effect with motor skill provides preliminary evidence that the significance of planning in the model, measured by the ROCF is not due to fine motor skill on the ROCF. This is supported by ROCF planning studies that separate motor functioning from executive functioning in survivors of brain tumors. Maddrey et al. (2005) found motor impairments to be independent of visuoconstructive ability as measured by the ROCF copy trial in young adult survivors of childhood brain tumor. Other studies have shown that poor performance on the ROCF copy is due to visuospatial skills and other executive functions (Schmahmann and Sherman, 1998; Vaquero, Gomez, Quintero, Gonzalez-Rosa, & Marquez, 2008).

The BQSS planning is inherently linked to visuospatial skills given the ROCF task design and is much more difficult to separate without also removing the effect of planning. Thus, in order to increase confidence in the planning construct, we tested another planning measure in the model, informant-reported planning skills on the BRIEF, and this model was also statistically significant. This provided convergent evidence on the specificity of planning skill in this relationship. The significance of the mediation with two complimentary planning measures, performance and informant report-based planning, highlights an important role of planning in daily functioning that requires further investigation. One may speculate how visuospatial planning skills are important for community living skills. For example, tasks such as navigating to new areas in the community or understanding public transportation. It is possible that fine motor visuospatial planning shares some of the common skills of real world planning but it may also be distinct from a person's ability to skillfully negotiate the steps required in planning

transportation. These distinctions in planning are important to explore and require additional empirical research to better understand the ecological validity of performance measures such as the planning on the BQSS.

To increase confidence that the model was specific to the construct of planning rather than another executive function, we selected an informant report of emotional control on the BRIEF and BQSS fragmentation as additional discriminant measures that were not expected to be disrupted in survivors. Both discriminant models were not statistically significant and provided further support for a specific executive difficulty of planning within this model.

A second problem with executive function measures is ecological validity and the variability of the measure predicting real life behavior (Anderson, 2002). Research has shown that cognitive tests are at best moderately predictive of daily functional behaviors (Payn, Hyman, Shores, & North, 2011). Performance measures are limited because one-on-one testing provides more structure than the real world. The ROCF does structure the participant with paper, pencil, a desk, and administrator presence. However, it does not structure a participant's approach to planning the task. Previous research in childhood brain tumors primarily has assessed planning skills with report-based measures (Howarth et al., 2013; Kahalley et al., 2013; Palmer et al., 2010) and while these scales provide valuable information about planning skills they are nonetheless subjective in nature. Informant report relies on insight of the reporter and can be influenced by his/her biases. Thus, questionnaires are at risk of over-inflation or underrepresentation of reporting. Palmer et al. (2010) examined executive functioning in pediatric patients with medulloblastoma using neurocognitive assessments and parent-report measures and emphasized the importance of utilizing both questionnaires and assessments to have a better understanding of executive functioning. Employing a performance-based measure

of planning concurrently in the current study complements the assessment of planning specifically. The current findings extend prior research on executive function difficulties in survivors on report-based measures (Howarth et al., 2013; Kahalley et al., 2013; Palmer et al., 2010) to a process measure of planning on the ROCF. Executive difficulties remain apparent in this group of long-term survivors on average 13 years from diagnosis, albeit with a wide range of outcomes.

The current findings should be considered in the context of the study limitations. The survivors in this study had a range of tumor types, location, and treatments. However, different tumor locations and treatments have been associated with executive dysfunction. The current sample of survivors was relatively small which limited the ability to explore more complex analyses comparing mediating variables and important secondary relationships with cancer-related variables. Yet in exploratory analyses, radiation treatment was significantly associated with planning, and age at diagnosis was related to both planning and community living skills. In addition, the sample size may limit the representativeness to the population. Therefore, future research may consider examining the relationships between these variables with a larger sample. That said, the long-term nature of the current sample, on average 13 years since diagnosis, is unique as this patient group is often not engaged in regular contact with healthcare providers, and thus, more difficult to recruit.

Little research exists on the relationship between planning skills and adaptive behaviors in survivors of childhood brain tumor. The current study demonstrates that planning skills may be important in adaptive functioning, such as community living skills. With the current study's preliminary model, we are inviting future researchers to continue to assess how executive skills and specifically planning may contribute to adaptive outcomes. This line of research may help to

identify areas on which to focus intervention that may in turn optimize adaptive functioning skills in survivors. Understanding these relationships are particularly important as it allows clinicians to isolate one of the underlying cognitive processes that may impact a young adult's independent living skills.

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Figure 1: The relationship between group and community living skill was significantly mediated by the indirect effect of planning skill.

Note: Model accounts for 13% of the variance in community living skill.

Table 1
Similar Demographic Characteristics Between Groups

	Survivors	Controls	Group difference	
	Percent		χ^2	p
Sex	58% F	61% F	.04	.84
Ethnicity	74% Caucasian	55% Caucasian	3.22	.07
SES	26% High	55% High	1.73	.19
	Mean (<i>SD</i>)		t	p
Age	22.47 (2.57)	21.48 (3.09)	-1.40	.17

Note. Independent samples t-test were used to evaluate age differences between groups; Pearson Chi-square was used for categorical variables of sex, ethnicity, and socioeconomic status (SES). SES was dichotomized into high SES (1,2) and low SES (3,4,5). Three participants chose not to report their ethnic identity.

Table 2
Group Means and Statistical Differences on Executive, Adaptive, Visuospatial, and Motor Measures

	Survivors	Controls	Group differences		Survivors	Controls	
	Mean (SD)		<i>t</i>	<i>p</i>	Percent Impairment (<i>n</i>)		<i>p</i>
Intelligence (FSIQ)*	98.17 (17.50)	112.25 (8.90)	3.90	<0.001	14% (4)	0% (0)	0.05
Visuospatial Construction (ROCF copy raw)**	30.73 (4.55)	33.34 (2.55)	2.81	.007	-	-	-
Community living skills** (SIB-R)	104.52 (20.69)	117.45 (17.67)	2.70	0.009	16% (5)	0% (0)	0.02
Boston Qualitative Scoring System (BQSS)	Mean (SD)		<i>U</i>	<i>p</i>	Percent Impairment (<i>n</i>)		<i>p</i>
Planning*	42.77 (12.41)	49.33 (9.13)	361.50	0.03	32% (10)	6% (2)	0.01
Neatness	47.68 (13.54)	53.76 (10.91)	390.50	0.06	7% (2)	0% (0)	0.23
Perseveration*	44.13 (15.21)	50.73 (9.61)	396.50	0.02	29% (9)	6% (2)	0.02
Fragmentation	46.39 (8.12)	48.18 (7.59)	447.50	0.36	10% (3)	6% (2)	0.66
	Mean (SD)		<i>t</i>	<i>p</i>	Percent Impairment (<i>n</i>)		<i>p</i>
Plan/organize (BRIEF)	50.05 (11.53)	44.30 (7.34)	-2.16	.04	10% (2)	0% (0)	.16
Emotional Control (BRIEF)	46.05 (8.94)	45.10 (7.05)	-0.42	.68	5% (1)	0% (0)	.40
Motor skill (GP) z-score	-1.28 (1.40)	-0.21 (.64)	3.96	<.001	22.6% (7)	3% (1)	.03

Notes: FSIQ= Full Scale IQ; ROCF=Rey-Osterrieth Complex Figure; SIB-R=Scales of Independent Behavior, Revised; BRIEF=Behavior Rating Inventory of Executive Function; GP=Grooved Pegboard test. BQSS Executive Function variables were converted from z scores to T scores for ease of interpretation. All analyses were run using z scores. Impairment was defined as a z score of 1.50 standard deviations below mean or greater. Independent samples t-test and Mann-Whitney U tests were used to assess group differences on BQSS measures. Fisher exact test was used to assess group differences in number of impaired participants. *Significant group differences between survivors and comparison group at $p \leq .05$. ** Significant group differences between survivors and comparison group at $p \leq .01$.

