Physical functioning in pediatric survivors of childhood posterior fossa brain tumors

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Background. Survival rates for children diagnosed with posterior fossa brain tumors (PFBTs) have improved significantly over the past several decades, and long-term functioning assessments have become priorities. These evaluations have occurred frequently in adults but only rarely in children. This study describes a cross-sectional assessment of physical functioning in pediatric survivors of PFBTs using the Bruininks-Osteretsky Test of Motor Performance, Second Edition (BOT-2).

Methods. Primary analyses compared BOT-2 scores to normative data using 1-sample t tests for each gross motor subscale (Bilateral Coordination, Balance, Running Speed/Agility, Strength) and motor-area composite (Body Coordination and Strength and Agility). Second, the cohort was stratified by diagnostic or treatment variables. Group differences and groups vs norms were evaluated using independent 2-sample and 1-sample t tests, respectively. Primary analyses compared BOT-2 scores with normative data using 1-sample t tests for each gross motor subscale (Bilateral Coordination, Balance, Running Speed/Agility, Strength) and motor-area composite (Body Coordination and Strength and Agility). Second, the cohort was stratified by diagnostic or treatment variables. Group differences and groups vs norms were evaluated using independent 2-sample and 1-sample t tests, respectively.

Results. Mean age of 30 participants was 11.4 years (range, 4.9y–18.2y), and mean time from diagnosis was 6.1 years (range, 1.1y–16.7y). Cerebellar astrocytoma (43.3%) and medulloblastoma (40%) were the most common diagnoses. As a group, significantly decreased functioning, compared with norms, was observed in Balance ($P < .001$) and Running Speed/Agility ($P = .005$). Specifically in Balance, 21 (70%) participants performed below or well-below average. Participants with a non-astrocytoma performed significantly lower than norms in all areas, independent of age at diagnosis. Survivors with tumors infiltrating the vermis demonstrated significantly lower Body Coordination than norms ($P < .001$).

Conclusions. Pediatric survivors of PFBTs demonstrated decreased physical functioning, most notably in Balance. These data underscore the need for further research and implementation of physical activity programs aimed specifically at approaches to minimize physical limitations.

Keywords: cancer survivor, disability, long-term outcome, pediatric, physical function, posterior fossa brain tumor.
Recognizing this deficit, the Bloom Scale and the Karnofsky Performance Scale became 2 of the most commonly used early standardized tools for evaluating functional outcomes in pediatric brain tumor patients. The Bloom Scale rated ‘global functioning’ using parent-report and clinical observation to describe “disability” and was commonly used throughout the 1980’s and early 1990’s. In these papers, the majority of childhood brain tumor survivors were described as “mildly disabled or not at all.” However, the Bloom Scale is purely observational, relatively crude, and poorly defined and therefore may not precisely evaluate changes after treatment, leading to an underestimation of severity and range of deficits. The Karnofsky Performance Scale (KPS) has also been used to evaluate physical performance in long-term survivors of pediatric medulloblastoma. The KPS, which was developed for patients actively undergoing chemotherapy, is not particularly applicable to children as it concentrates on activities of daily living and the ability to work. As a result, the Bloom Scale and KPS are no longer used as frequently in this population. Similar to the KPS, the Lansky or Play Performance Scale assesses global functioning but was specifically designed for the pediatric oncology population. Although it too has been used in assessing the overall functioning of pediatric PFBT survivors, it was developed as a parent-report measure, and the solitary score provides very limited information regarding the specific details of physical functioning.

In a shift towards evaluation of more encompassing aspects of functioning, some authors have evaluated neurodevelopmental outcomes, health status, or quality of life in survivors of pediatric CNS tumors. However, these measures were designed to evaluate multiple domains of functioning (ie, physical, social, emotional, etc.) and do not provide a comprehensive representation of physical function as an independent concept. Additionally, the health status and quality of life measures used were all questionnaires and did not involve any observational aspect of assessing physical function.

Despite the broad range of methods used in describing the physical functioning of survivors of pediatric brain tumors, the literature does document some neurologic sequelae and physical functioning late effects in this population. However, the vast majority of existing studies have evaluated adults previously diagnosed with brain tumors as children. Therefore, there is a paucity of information regarding the physical functioning of children (ie, aged 18 years or younger) who have completed treatment for a brain tumor. Understanding the physical functioning of young survivors of PFTBs can provide valuable information into the limitations faced by a group of individuals who are still undergoing developmental changes. Additionally, evaluating this pediatric population may offer insight into opportunities for education and rehabilitation.

Therefore, the goal of this paper was to describe the physical functioning of pediatric survivors of PFBTs using a standardized, observational measure of physical performance validated for children and adolescents.

**Materials and Methods**

**Study Design and Participants**

This study was a component of a larger cross-sectional study, and all eligible participants were identified using a prospectively maintained database of all children with brain tumors treated at SickKids in Toronto, Canada. Eligible participants completed informed consent in accordance with the ethics review board.

Inclusion criteria were: (i) aged 4 to 18.9 years old at the time of assessment; (ii) diagnosis of a PFTB of any type that required surgery at least 1 year earlier (with no maximum time since diagnosis as long as the participant was still under 19 years of age); (iii) no chemotherapy or radiation therapy received within the last 6 months; and (iv) no evidence of progressive disease.

**Procedures**

A standard introduction letter describing the project was mailed to eligible families, and a follow-up telephone call was made by the research project coordinator to ask families if they were interested in participating. Those who agreed scheduled an appointment to complete the assessment by a registered physiotherapist.

**Measure of Physical Functioning**

The Bruininks-Osteretsky Test of Motor Performance, Second Edition (BOT-2) was used for the evaluation of physical performance. The BOT-2 is a 53-item tool (with 8 subscales) of motor functioning for children aged 4 to 21 years. Its measurement properties have been well established and include clinical groups such as children with developmental coordination disorder and high-functioning autism. Additionally, the BOT-2 has been used in studies in other pediatric neurological populations as well as pediatric oncological conditions. This measure is norm referenced (age and gender matched) and allows scores to be compared with those of children without any known physical performance limitations.

The 4 gross motor subscales were used in assessing physical functioning in this study: Bilateral Coordination (7 items), Balance (9 items), Running Speed/Agility (5 items), and Strength (5 items). These 4 subscales took approximately 30 to 40 minutes to administer, depending on the abilities of each participant. Subscale scores were combined into 2 motor-area composite scores: Body Coordination (includes Bilateral Coordination and Balance) and Strength and Agility (includes Running Speed/Agility and Strength). Both subscale and motor-area composite z scores were also categorized into 5 clinical groups: well-above average (z score ≥ 2), above average (z score 1 – 2), average (z score 1 to -1), below average (z score -1 to -2), and well-below average (z score ≤ -2).

**Data Collected**

Once consent was provided and the physical assessment was complete, the following data were collected from the medical chart: body mass index (BMI), age at diagnosis, tumor type, postoperative mutism, use of chemotherapy, use of radiation therapy, and need for permanent hydrocephalus treatment. Additionally, tumor location was identified by 1 of the co-investigators (A.K.), who was blinded to diagnosis and functional outcome using preoperative MRI if available. (If unavailable, a postoperative scan was used, and clinical judgment was used to label tumor location.) Tumor location was defined as visualization on MRI of actual tumor infiltration of the structure (not just displacement from mass effect). Location was then divided into 2 cerebellar regions: “vermis” and “not vermis.” Any brainstem involvement was also noted.

**Analysis**

Data were analyzed using SPSS v.21 (IBM). Descriptive statistics were calculated for the sample. Z scores were calculated for each of the 4 gross motor subscales using age- and gender-specific scores in the BOT-2 manual. Z scores were also calculated for the 2 motor-area composites in the gross motor domain.
The primary analysis was a comparison of PFBT survivors with the published normative data using 1-sample t tests. Comparisons were performed for the z scores on the 2 motor-area composites as well as z scores for all 4 gross motor subscales. The significance level was set at $P = .05$. Additionally, z score descriptions and distributions were further examined to offer insight into the clinical meaning of the observed scores.

Second, to examine the impact of tumor type, age at diagnosis, tumor location, need for permanent hydrocephalus treatment, chemotherapy, radiotherapy, and postoperative mutism, patients were grouped according to these characteristics, and a second set of comparisons was conducted. Specifically, differences between these groups, as well as each group, vs normative data were evaluated using independent 2-sample and 1-sample $t$ tests, respectively. For between-group analyses, the significance level was $P < .05$, but for comparisons to normative data, the significance level was reduced to $P = .01$ due to multiple comparisons. Additionally, physical function relative to time since diagnosis was examined for the entire cohort, as well as for the cerebellar astrocytoma (CA) and medulloblastoma (MB)/ependymoma (EP)/brainstem astrocytoma (BSA) groups, using Pearson correlations and trends over time were examined.

The final analysis involved model building to determine if there was a combination of patient-related or treatment-related variables that predicted physical functioning. Independent variables, with $P < .10$ on univariate analyses for each motor-area composite, were then entered into a stepwise linear regression model.

**Results**

Ninety patients were deemed eligible for the larger study, of which 62 consented. Of that 62, 30 (48%) agreed to the physical assessment portion of the study between December 2006 and December 2009. Of those who consented to the larger study, there were no significant differences between those participating in the physical assessment ($n = 30$) and those who did not ($n = 32$) based on current age (participants mean, 11.4 years; nonparticipants mean, 11.8 years; $P = .65$), age at diagnosis (participants mean, 5.2 years; nonparticipants mean, 5.1 years; $P = .88$), or tumor type (43% participants with cerebellar astrocytoma; 47% nonparticipants with cerebellar astrocytoma). There was also no difference in age at diagnosis based on tumor type (cerebellar astrocytoma, 5.25 years; non-astrocytoma, 5.20 years; $P = .96$).

Fifty percent of participants were male, and ages at time of assessment ranged from 4.9 to 18.2 years with a mean age of 11.4 years (SD = 4.1). The mean time since diagnosis was 6.1 years (range, 1.1 y–16.7 y; SD = 4.2), and the 2 most common diagnoses were cerebellar astrocytoma (43.3%) and medulloblastoma (40%). The sample characteristics are provided in Table 1.

BMI was also collected for 26 of the 30 participants. Height data at the time of the study was not available retrospectively for 4 participants, so BMI could not be calculated. Overall, 73% of the 26 participants had a BMI in the normal or underweight ranges, and the distribution was greater for the females than males (see Table 2).

Motor-area composite z score distributions (ie, Body Coordination and Strength and Agility), as well as each of the 4 subscales, were evaluated using Q-Q plots and found to be normally distributed.

Statistically significant differences were observed between the cohort and normative data for both Body Coordination ($P < .001$, 95% CI, -1.44 to -0.76) and Strength and Agility ($P = .041$, 95% CI -0.96 to -0.023). To further analyze which motor areas demonstrated the greatest limitations, the 4 subscales within these motor-area composites were examined (Fig. 1). Statistically significant differences between participant z scores and the normative sample were observed on Bilateral Coordination ($P = .021$, 95% CI, -0.78 to -0.07), Balance ($P < .001$; 95% CI, -1.66 to -0.97), and

### Table 1. Sample Characteristics of Respondents ($n = 30$)

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>No. (%)</th>
</tr>
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<tbody>
<tr>
<td>Medulloblastoma</td>
<td>12 (40)</td>
</tr>
<tr>
<td>Cerebellar astrocytoma</td>
<td>13 (43.3)</td>
</tr>
<tr>
<td>Ependymoma</td>
<td>2 (6.7)</td>
</tr>
<tr>
<td>Brainstem glioma</td>
<td>3 (10)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Age distribution (years) at time of assessment:</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>4.9–7.9</td>
<td>9 (30)</td>
</tr>
<tr>
<td>8.0–12.9</td>
<td>7 (23.3)</td>
</tr>
<tr>
<td>13.0–18.2</td>
<td>14 (46.6)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Age distribution (years) at time of diagnosis:</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>0–3.9</td>
<td>11 (36.6)</td>
</tr>
<tr>
<td>4.0–11.9</td>
<td>19 (63.3)</td>
</tr>
</tbody>
</table>

| Deepest Tumor Location:                        |  
| Deep Cerebellar Hemisphere                     | 8 (26.7) |
| Vermis                                          | 22 (73.3) |

| Brainstem Involvement:                         |  
| Yes                                             | 9 (30)  |
| No                                              | 21 (70) |

| Received chemotherapy:                         |  
| Yes                                             | 13 (43.3) |
| No                                              | 17 (56.7) |

| Radiotherapy:                                   |  
| Craniospinal                                    | 9 (30)  |
| Local PF                                        | 2 (6.7) |
| None                                            | 19 (63.3) |

| Postoperative mutism:                          |  
| Yes                                             | 3 (10)  |
| No                                              | 23 (90) |

| Permanent hydrocephalus treatment:             |  
| VP shunt                                        | 7 (23.3) |
| ETV                                             | 3 (10)  |
| None                                            | 20 (66.7) |

### Table 2. Body mass index (BMI) of all respondents ($n = 26$)

<table>
<thead>
<tr>
<th>Male no. (%)</th>
<th>Female no. (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Severely wasted (BMI &lt;0.1st percentile)</td>
<td>2 (7)</td>
</tr>
<tr>
<td>Wasted (BMI &lt; 3rd percentile)</td>
<td>2 (8)</td>
</tr>
<tr>
<td>At risk for wasting (BMI: 3rd–15th percentile)</td>
<td>3 (12)</td>
</tr>
<tr>
<td><strong>Within normal limits (BMI: 15th–85th percentile)</strong></td>
<td>11 (42)</td>
</tr>
<tr>
<td>Overweight (BMI: &gt; 85th percentile)</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Obese (BMI: &gt; 97th percentile)</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Severely obese (BMI: &gt; 99.9th percentile)</td>
<td>1 (4)</td>
</tr>
</tbody>
</table>
Running Speed/Agility ($P = .005$; 95% CI, -1.04 to -0.20) using $P = .05$ as the level of significance. Z scores were further examined to offer insight into the clinical meaning of these scores. As seen in Fig. 2, 47% of the participants had below average functioning in Balance, and an additional 23% were well-below average, for a total of 70% demonstrating clear balance deficits. A large proportion of this cohort also demonstrated significant deficits in the Running Speed/Agility and Strength domains, with 40% and 30% in the combined below and well-below average categories, respectively. Bilateral Coordination had the smallest proportion, with only 23% of participants showing clear deficits compared with normative data.

When BOT-2 data were examined by tumor type (ie, CA vs MB/EP/BSA), there was only a significant difference between groups in the Strength and Agility motor-area composite ($P = .044$; 95% CI, -1.81 to -0.026). However, as seen in Fig. 3, when these 2 groups were independently compared with normative data, the CA subgroup was not statistically significantly different from normative data (although Body Coordination was quite low) using $P < .05$, but the MB/EP/BSA group was statistically lower than norms in both motor-area composites (Body Coordination $P < .001$; 95% CI, -1.68 to -0.96; Strength and Agility $P = .005$; 95% CI, -1.46 to -0.17).

Figure 4 illustrates z score distributions for participants diagnosed at aged 3 years or younger vs those aged 4 years and older. There were no significant differences between groups in either motor-area composite. However, when compared with norms, Body Coordination was statistically lower for both the younger ($P = .01$; 95% CI, -1.88 to -0.32) and older groups ($P < .001$; 95% CI, -1.46 to -0.74).

Preoperative MRI was used to identify tumor location in 22 (73.3%) participants. Unfortunately, only postoperative scans were available for the other 8 PFBT survivors. As shown in Fig. 5, Body Coordination was significantly lower than norms if the tumor infiltrated the vermis ($P < .001$; 95% CI, -1.56 to -0.95), and Strength and Agility was also quite low in this group but not below the $P < .01$ threshold. There were no significant differences between the 2 tumor location groups in either motor-area composite. $\chi^2$ analysis revealed an association between tumor type (CA vs MB/EP/BSA) and tumor location (infiltration of vermis vs no...
Additionally, the relationships between time since diagnosis and physical functioning were considered. Across the whole sample, there was no relationship observed between time since diagnosis and either Body Coordination or Strength and Agility. Some qualitative trends were noted, so an additional analysis examined relationships when the group was stratified by tumor type. Fig. 6 illustrates the distribution of z scores for each subscale against time since diagnosis for each tumor group. No significant relationships for these 4 BOT-2 subscales were observed, but there appeared to be a pattern of greater improvement in physical functioning over time for the CA group compared with the MB/EP/BSA group.

A final analysis was performed to evaluate the impact of other potentially relevant medical variables. Participants were grouped (yes/no) on the following variables: need for permanent hydrocephalus treatment, chemotherapy, radiotherapy and postoperative mutism. Body Coordination and Strength and Agility were analyzed using independent t tests for each group. When Body Coordination was examined for each variable, the only significant difference observed was between participants with (n = 3) and without (n = 27) postoperative mutism (P = .033; 95% CI, 0.10–2.20). Alternately, in the Strength and Agility analyses, participants who had received chemotherapy (P = .018; 95% CI, 0.19–1.93) or radiotherapy (P = .008, 95% CI, 0.35–2.18) demonstrated significantly lower scores than those who had not received these adjuvant therapies. Additionally, the 3 participants who suffered postoperative mutism also demonstrated significantly lower scores in Strength and Agility (P = .003, 95% CI, 0.81–3.51).

As these 3 variables, as well as tumor type, were found to be significant (ie, P < .05) for Strength and Agility in independent sample t tests, a series of stepwise regressions was conducted to determine what combination of variables best predicted this component of physical functioning. Hence, tumor type, receipt of chemotherapy, radiotherapy, and postoperative mutism were included. The analysis revealed that only postoperative mutism (β = −0.56, P < .001) and chemotherapy (β = −0.47, P = .002) were included in the stepwise model and accounted for a significant amount of the variance in Strength and Agility (R² = 0.49, F = 13.12, P = .001). Tumor type and radiotherapy did not significantly contribute to the model.

Discussion

To the best of our knowledge, this is the first study specifically looking at the physical functioning of pediatric brain tumor survivors. These results indicate that children and adolescents previously diagnosed with a PFBT demonstrated significantly lower gross motor physical functioning compared with normative data on the BOT-2. These findings are consistent with the existing (primarily adult) literature that has documented sensorimotor deficits in more than 50% of PFBT survivors and as high as 100% in 1 report. Even after treatment for a cerebellar astrocytoma, Aarsen et al. (2004) demonstrated long-standing deficits such as motor neglect. Balance and Running Speed/Agility appeared to explain the largest portions of variance within the Body Coordination and Strength and Agility motor-area composites, respectively. This information is not surprising, as balance deficits are known to be 1 of the primary physical limitations faced by survivors of PFBTs. As evidenced by z scores, 70% of the cohort was functioning either...
below or well-below average in the area of Balance. This large percentage clearly demonstrates that this young population struggled with activities on the BOT-2 that required a moderate amount of balance, such as standing on 1 foot or walking forward on a line. Although these specific activities may not be regularly performed in isolation on a daily basis, they are essential for successful performance of activities such as putting on boots (from a standing position), kicking a ball, or maneuvering in small spaces. These results are consistent with the findings by Hudson et al. (2003), who reported that 31.7% of adult survivors of all childhood CNS tumors (not just PFBTs) reported physical performance limitations and participation restrictions in activities such as bathing, dressing, shopping, and going to school. Although balance rehabilitation may be more difficult than improving strength or speed, these data suggest that PFBT survivors should be supported to engage in ongoing activities to challenge and improve their balance abilities.

Balance requires a complex interaction and integration of a variety of body systems that need to be intact for full functioning. It has long been known that lesions to the cerebellum cause ataxia, or a loss of coordination and balance, but the literature has only more recently described the functional impact of lesions to different parts of the cerebellum. It has been shown that damage to the vermis and deep cerebellar nuclei are more highly associated with decreased control of balance than tumor size or age at diagnosis and that children and adolescents may achieve more complete recovery if the cerebellar nuclei are not injured. Schoch et al. (2006) analyzed clinical ataxia scores as well as posturography (ie, a balance assessment at the impairment level of the ICF) and found that surgical damage to the cerebellar nuclei had a greater impact on balance function than either chemotherapy or radiation. Although we did not have the same detailed level of imaging, our finding that tumors infiltrating the vermis caused greater disability, is supported.

When compared with normative data, only the non-astrocytoma group had scores significantly lower in both Body Coordination and Strength and Agility. However, when the 2 tumor groups were compared, the only significant difference observed...
was in Strength and Agility, not Body Coordination. These results are similar to earlier findings by Ronning et al. (2005) who found decreased neurological functioning in children with medulloblastoma compared with those with astrocytoma, but the results were not statistically different. Alternately, Roncadin et al. (2008) found that children previously diagnosed with medulloblastoma demonstrated significantly lower scores on self-reported functional independence and health-related quality of life than those diagnosed with cerebellar astrocytoma, but no observational measures of physical functioning were used. The additional analyses in this study were performed to gain further insight into group differences. The decreased performance in Strength and Agility in the MB/EP/BSA group was hypothesized to be a compound effect of additional chemotherapy and/or radiation. In the regression analysis, it was found that receipt of chemotherapy did in fact contribute to the model of prediction of functioning in this motor area composite. This finding suggests that chemotherapy-related toxicity, such as peripheral neuropathy, may lead to sustained deficits even years after completion of treatment; however, neuropathy was not specifically addressed in this study. Although an association between tumor type and location was observed, all participants had tumors infiltrating either the deep cerebellar hemisphere and/or vermis. Therefore, the impact of tumor on deep cerebellar structures in all participants could provide some rationale for why balance and coordination deficits were present in most participants and no difference in Body Coordination between tumor type groups was observed. Further research with longitudinal assessments, a larger sample and/or the inclusion of additional domains and/or variables may offer greater insight into differences between tumor types and physical functional outcomes.

When age at diagnosis was examined, our findings demonstrated no significant difference between the PFBT survivors who had been diagnosed prior to the age of 4 years vs those who were older at the time of diagnosis. These findings are similar to those of Helseth et al. (1999) who found that power of locomotion was not correlated to age at diagnosis. Similarly, Konczak et al. (2005) found no difference in postural motor control in children who underwent surgery prior to 4 years of age vs those diagnosed in adolescence. However, other published findings examining pediatric brain tumor survivors are contradictory. Roncadin et al. (2008) found that in survivors of cerebellar astrocytoma, younger age at diagnosis was associated with decreased functional independence on a self-report questionnaire. Similarly, there are reports demonstrating that diagnosis of a pediatric brain tumor at a younger age appears to be associated with greater disability or decreased strength and overall fitness; whereas others have found that older children were at increased risk of motor impairment. However, in each of these cases of contrasting reports, methodological issues for extrapolation to this particular pediatric population were of concern. Two studies used the Bloom Scale, 1,19,21 and 1 used no standardized measure, 1 and 1 used a child self-report questionnaire. 3,33 Finally, Ness et al. (2005) used questionnaires administered only to adult survivors, so caution should be taken when generalizing results to children who are PFBT survivors.

Overall, this group of young PFBT survivors demonstrated lower physical performance in all areas of gross motor functioning, but the degree to which these deficits are due to the tumor and subsequent treatment vs possible disuse over time is unknown. Many parents lean towards overprotection after a child’s cancer diagnosis. Parents may sometimes limit their children’s participation in activities due to fear of injury, primarily related to the degree of ataxia and motor impairment suffered, as they may perceive the child as fragile or vulnerable. Additionally, it may be the child or adolescent who self-limits physical activity. Individuals may be aware of their physical and/or cognitive deficits and may feel stigmatized or lack the confidence to participate in social physical activities. Wrotniak et al. (2006) found that children with lower perceptions of their physical abilities reported lower levels of physical activity than peers. There is evidence to show that this decreased activity may translate long term. Results from the Childhood Cancer Survivor Study showed that adult survivors of pediatric brain tumors were 2.5 times more likely than siblings to report physical performance limitations and demonstrated overall physical performance slightly lower than published expected norms for individuals in their 7th decade of life when the median age of the cohort was 22 years. Overall, young PFBT survivors face a range of physical limitations due to their tumor and treatment, and these functional deficits may be compounded by a more sedentary lifestyle due to a multitude of internal or external factors.

There are several limitations to this study. First, it is possible that individuals who completed the physical assessment component of the study were systematically different, ie, higher or lower functioning, than individuals who did not consent to any part of the study or those who only completed the nonobservational portion. If so, it is possible that these results are not entirely generalizable to the larger pediatric PFBT population. Therefore, standardized observational physical functioning assessments should be considered for all PFBT survivors at follow-up medical visits, and these data could be analyzed for greater generalizability. Second, aside from the observational portion of the assessment, all other data were collected retrospectively, and a degree of inaccuracy was possible. For example, only 3 PFBT survivors were found to have convincing evidence of cerebellar mutism, which was likely an underestimation of the true incidence. Further, only postoperative MRI scans were available for identifying tumor location in 8 participants, so a degree of inaccuracy was possible. Again, collecting these data in a prospective manner would eliminate these possible inaccuracies. Third, although the BOT-2 has been used in other pediatric neurological and oncological populations, its measurement properties specifically within the PFBT population have not been examined. Wang and Su (2009) reported low sensitivity (33%) and high specificity (84%) for the Balance section of the BOT-2. It has been suggested that the BOT-2 may better identify children without balance impairments than those with deficits. In a population where balance is the primary physical limitation, it may be valuable to explicitly examine the reliability and validity of the BOT-2 in this unique population. Another limitation is that impairments such as ataxia or hemiparesis or participation-level items such as school or sport participation were not evaluated. Although the absence of these domains does not impact the value of this new information, future studies could include these items and relationships between domains explored. Finally, due to the small sample size and therefore limited statistical power, caution should be taken when interpreting these results. A prospective examination of all pediatric PFBT survivors at multiple time points after treatment completion would allow for a greater sample size and broader generalizability.
Conclusions

These data are the first to report long-term physical functioning of children and adolescents who are PFBT survivors using a standardized, observational measurement tool. Overall, decreased functioning in gross motor skills was observed, but balance in particular was extremely low. Unfortunately, despite these deficits, ongoing rehabilitation programs and long-term resources for these survivors to engage in physical activity are broadly unavailable. Future research should focus on the implementation and evaluation of physical activity programs for PFBT survivors. These studies should examine the impact of exercise on physical performance limitations and any possible approaches to minimize these limitations. Finally, there are many unanswered questions about the possibility of ongoing recovery, whether therapy should be maximized at different points (eg, when chemotherapy-induced neuropathy is at its worst or after it has improved) and the extent to which physical activity promotes better function. Further research must evaluate whether encouragement from PFBT clinicians to patients and families to engage in physical activity programs will result in better outcomes.

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References


