

Motor Function in School-Aged Children With Positional Plagiocephaly or Brachycephaly

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Objective: To determine whether children with a history of positional plagiocephaly/brachycephaly (PPB) show persistent deficits in motor development.

Methods: In a longitudinal cohort study, we completed follow-up assessments with 187 school-aged children with PPB and 149 participants without PPB who were originally enrolled in infancy. Primary outcomes were the Bruininks-Oseretsky Test of Motor Proficiency-Second Edition (BOT-2) composite scores.

Results: Children with PPB scored lower than controls on the BOT-2. Stratified analyses indicated that differences were restricted to children who had moderate-severe PPB. No consistent differences were observed in children who had mild PPB.

Conclusion: Children who had moderate-severe PPB in infancy show persistent differences in motor function. We suggest close developmental monitoring and early intervention to address motor deficits. (Pediatr Phys Ther 2020;32:107–112)

Key words: brachycephaly, motor skills, plagiocephaly

INTRODUCTION

Positional skull deformation has been associated with infants' motor development and is known to co-occur with conditions that affect muscle tone and mobility.^{1–5} For example, up to 97% of infants with positional plagiocephaly have also been found to have torticollis.³ Similarly, skull deformation is common among preterm infants, who lack the motor development to maintain midline head position when lying in the supine position.^{6–8} Infants with positional plagiocephaly/brachycephaly (PPB) are also more likely than controls to have delays in motor development and abnormal muscle tone.⁴

Much less is known about the *persistence* of motor deficits in children with PPB, and results from existing studies are

mixed. One study found a high rate of motor asymmetry among children who had positional plagiocephaly and torticollis in infancy,⁹ though this resolved for most children by age 2 years. In another study, children with and without PPB were evaluated for posture, flexibility, and balance at ages 3 to 5 years.¹⁰ Those with PPB had worse craniocervical posture, flexibility, and balance than controls.

In previous research, developmental assessments were completed of 235 children with PPB and 237 controls in infancy (age 7 months, on average) and at ages 18 and 36 months.^{11–13} Robust differences were observed in motor development in infancy, with children with PPB scoring lower than controls in both gross motor and fine motor development. Although children with PPB continued to receive lower motor scores than controls, these differences were reduced in magnitude and were no longer statistically significant by age 36 months.¹³

The goal of this study was to determine whether persistent differences in motor development are observed in school-aged children with and without a history of PPB. In addition to examining group differences, we explored differences as a function of PPB severity and in relation to clinically relevant modifiers. This included stratification based on history of torticollis, which in some studies has been associated with worse developmental outcomes,¹⁴ and history of orthotic helmet treatment. In separate analyses, we examined the effects of interventions targeting motor development (ie, physical therapy or occupational therapy).

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METHODS

The study used a prospective cohort design to assess motor development in children with a history of PPB in infancy (cases) and children without PPB (controls). In phase 1, we recruited infants with PPB at the time of their diagnosis in the craniofacial center at a large pediatric hospital.¹¹ We recruited the first 8 control infants through area pediatricians. All remaining controls were recruited through a participant registry, composed of families who agreed at the time of their child's birth to be contacted for research. To confirm the presence or absence of PPB, infants received 3-dimensional (3D) imaging of their head shape. Images were de-identified, randomly sorted, and rated by 2 craniofacial pediatricians who were unaware of infants' PPB diagnosis. Ratings of deformation were made on a 4-point scale (0 = none, 1 = mild, 2 = moderate, and 3 = severe). Interrater agreement was excellent for the presence/absence of deformation ($\kappa = 0.80$) and good for severity ratings (weighted $\kappa = 0.72$). We used the average of the 2 ratings to categorize infants as having PPB (diagnosed PPB and severity rating ≥ 0.5), controls (no diagnosed PPB and severity rating = 0), and affected controls (no diagnosed PPB and severity rating ≥ 0.5).

We reapproached children with PPB and controls when they were 7 years and older for phase 2. Families received \$200 compensation for participating and a written summary of their child's evaluation results. The study was approved by the Institutional Review Board.

Children With PPB

Phase 1 inclusion criteria were a clinical diagnosis of PPB, confirmed on 3D imaging; age 4 to 11 months at diagnosis; English reported as the primary language spoken in the home; and the child's biologic mother was available for participation. Exclusions were prematurity (<34 weeks' gestational age); diagnosed neurodevelopmental condition (eg, Down syndrome); and sensory impairment that would interfere with infants' participation in assessment (eg, deafness or blindness). We enrolled 235 infants with PPB, representing 52% of those approached.⁶ We did not approach 2 children who had a clinical diagnosis of PPB but did not have discernible deformation on 3D imaging, and 7 children diagnosed with neurodevelopmental conditions (eg, 22q11 deletion syndrome, mitochondrial disorder, and Chiari malformation) after enrollment in phase 1. One child in the case group died prior to phase 2. This left a total of 225 potentially eligible case participants. Ten families could not be located, 9 declined participation, and 19 were unresponsive to outreach efforts. A total of 187 families consented to participate between August 2014 and March 2018, representing 83% of those eligible.

Controls

In addition to the exclusions listed for cases, controls were excluded if they had any previously diagnosed craniofacial condition or any evidence of deformation on the 3D imaging completed in phase 1 ($n = 70$). Two children were diagnosed with neurodevelopmental conditions after phase 1 enrollment (eg, Chiari malformation and neurofibromatosis) and 1 child

died prior to phase 2, leaving 164 controls eligible for follow-up. Three families could not be located, 1 family declined to participate, and 11 families were unresponsive. One hundred forty-nine control families consented to participate between December 2014 and February 2018, representing 91% of those eligible.

Study Measures

Participants completed an individually administered neuropsychological assessment battery, including measures of cognition and academic achievement (reported in Collett et al¹⁵) and motor function assessed using the Bruininks-Oseretsky Test of Motor Proficiency-Second Edition (BOT-2).¹⁶ Composite scores from the BOT-2 were derived for fine manual control, manual coordination, body coordination, strength and agility, and the total motor composite. T scores (average = 50, standard deviation [SD] = 10) were generated for composite indices using separate norms for males and females, and scaled scores (average = 10, SD = 3) were generated for subtests. The BOT-2 was administered by trained psychometrists and video recorded for coding of interexaminer reliability. Approximately 30% of the tests administered were reviewed for reliability by 1 of the investigators. Average item-level agreement ranged from 94% to 99%.

We used clinician ratings of 3D infants' head shape images, described earlier, to quantify PPB severity among cases. Children with PPB were categorized as having had "mild PPB" (average severity rating = 0.5-1.0) or "moderate-severe PPB" (average severity rating ≥ 1.5) as infants.

Caregivers completed a semistructured interview, which confirmed and updated family demographic information; categorized the child's participation in occupational therapy or physical therapy; and documented any newly diagnosed medical conditions.

Analyses

We calculated descriptive statistics to summarize participant demographic and clinical characteristics, separately for cases and controls. We used separate linear regression analyses with robust standard errors to compare children with PPB and controls on primary motor outcomes. Analyses were adjusted for the following a priori covariates known by previous research or clinical impression to be related to both PPB and motor function or to motor function alone (ie, as either confounding or precision variables): socioeconomic status (SES) based on the total Hollingshead¹⁷ score (continuous), age at assessment, sex, and race/ethnicity (white, non-Hispanic vs non-white or Hispanic). We also completed descriptive analyses examining case-control differences on BOT-2 subscales to identify any areas of strength or weakness. As this is the first study to evaluate motor skills in school-aged children with and without a history of PPB, we did not adjust *P* values for multiple comparisons to avoid "missing" potentially important associations that warrant further research. Rather than considering *P* values to be dichotomous tests of significance, we evaluated the magnitude and consistency of effect sizes (ES) across outcomes as well as the precision of effect estimates.

To determine the magnitude of group differences, we calculated standardized mean difference ES using the adjusted group difference divided by the root mean square error. The interpretation of ES is somewhat subjective and dependent on the research design, outcomes assessed, and other factors.¹⁸ An ES of less than 0.25 was considered “small,” 0.25 to 0.50 “moderate,” and greater than 0.50 “large.” To evaluate for potential attrition bias, we repeated these analyses using inverse probability weighting.¹⁹ For these analyses, we determined propensity scores for phase 2 participation based on phase 1 characteristics (SES, sex, race/ethnicity, PPB severity, and Bayley-3 scores at age 36 months). These scores estimate the probability of being lost to follow-up based on covariates that are measured in all participants, both those retained and those lost to follow-up. We then used these scores to generate propensity weights for linear regression analyses. In essence, these analyses give additional “weight” to the observations of participants who were retained in the study but were similar to participants who were lost to follow-up.

To evaluate case-control differences by PPB severity, we repeated these analyses for children with PPB who had (a) “mild PPB” in infancy versus controls and (b) had “moderate/severe PPB” versus controls. We also examined case-control differences separately for children with and without a history of torticollis, and for children with and without a history of orthotic helmet treatment. Finally, we evaluated the effects of developmental intervention (occupational therapy or physical therapy) on phase 2 motor scores using censored normal regression.²⁰ This approach assumes that the scores of children who received intervention are “left censored” and, while their true scores in the absence of intervention are unknown, they are presumed to be at least as low as the scores observed.

RESULTS

Three hundred thirty-six children ($n = 187$ PPB cases, $n = 149$ controls) were reenrolled for phase 2. Nonparticipating families were lower in SES, more likely to have moderate-severe PPB in infancy (90% vs 72% among participants), and more likely to have had developmental delays as infants/toddlers (53% vs 38% among participants). Sixteen participants ($n = 8$ PPB cases, $n = 8$ controls) were unable to complete an in-person assessment (eg, due to family relocation out of the country), and therefore completed only a caregiver interview. Detailed phase 2 participation data are summarized in Collett et al.¹⁵ Motor assessments were considered invalid for 1 control and 1 child with PPB due to child refusal, and motor testing could not be completed (eg, due to time constraints) for 3 children with PPB and 1 control. Test data were therefore available for 1 or more outcomes for 177 children with PPB and 139 controls.

Most participants in both groups were male (66% children with PPB, 57% controls), white and non-Hispanic (70% children with PPB, 60% controls), and from middle to upper SES families (ie, Hollingshead categories I-II; 80% children with PPB, 78% controls) (Table 1). Mean age at participation was 9.0 years ($SD = 0.8$) for children with PPB and 8.8 years ($SD = 0.6$) for controls. Among children with PPB, 52 (28%) had “mild” PPB as infants and 135 (72%) had “moderate-severe”

TABLE 1
Characteristics of Children With and Without Positional
Plagiocephaly/Brachycephaly at School Age

Characteristic	Controls	Cases
	n (%) or Mean (SD)	n (%) or Mean (SD)
Total	149 (100.0)	187 (100.0)
Gender		
Female	64 (43.0)	64 (34.2)
Male	85 (57.0)	123 (65.8)
Age, y	8.8 (0.6)	9.0 (0.8)
Race/ethnicity		
White, non-Hispanic, or Latino	89 (59.7)	130 (69.5)
Asian or Pacific Islander	4 (2.7)	9 (4.8)
Black	5 (3.4)	0 (0.0)
Hispanic or Latino	20 (13.4)	22 (11.8)
Mixed race or ethnicity	31 (20.8)	26 (13.9)
Socioeconomic status		
I (highest)	39 (26.2)	69 (36.9)
II	77 (51.7)	80 (42.8)
III	24 (16.1)	27 (14.4)
IV	8 (5.4)	10 (5.3)
V (lowest)	1 (0.7)	1 (0.5)
Child delivered ≤ 38 wk		
No	139 (93.3)	146 (78.1)
Yes	10 (6.7)	41 (21.9)
Interventions received		
Occupational therapy	12 (8.1)	34 (18.2)
Physical therapy	8 (5.4)	93 (49.7)
Any intervention	15 (10.1)	106 (56.7)
Torticollis		
No	146 (98.0)	103 (55.1)
Yes	3 (2.0)	84 (44.9)
Helmet treatment		
No		122 (65.2)
Yes		64 (34.2)
Severity of plagiocephaly		
Mild		52 (27.8)
Moderate		101 (54.0)
Severe		34 (18.2)

PPB. Eighty-four children with PPB (45%) and 3 controls (2%) had a history of torticollis. Children with PPB were more likely than controls to have received occupational therapy or physical therapy (57% PPB cases, 10% controls). Sixty-four (34%) children with PPB received orthotic helmet treatment for their skull deformation.

BOT-2 scores for children with and without PPB were slightly lower than expected relative to test norms, with average T scores on the composite scales ranging from 41.2 ($SD = 6.8$) to 45.8 ($SD = 7.2$) for children with PPB and 42.6 ($SD = 7.3$) to 46.8 ($SD = 7.9$) for controls (Table 2). Children with PPB scored lower than controls on the BOT-2 strength and agility ($ES = -0.31$, $P = .01$) and total motor composites ($ES = -0.25$, $P = .03$) (Table 2). Children with PPB also scored lower on all other BOT-2 composite scales, although these differences were smaller in magnitude and imprecise ($ES = -0.17$ to -0.16 , P values = .14 to .17). Among the BOT-2 subscales, case-control differences were most prominent for running speed and agility ($ES = -0.24$), strength ($ES = -0.27$), and fine motor integration ($ES = -0.26$) (Table 2).

TABLE 2

Comparison of Mean Motor Scores in Children With and Without Positional Plagiocephaly/Brachycephaly as Infants

BOT-2 Scale ^a	Cases vs Controls									
	Controls		Cases		Unadjusted			Adjusted ^b		
	n	Mean (SD)	n	Mean (SD)	Mean Difference (95% CI)	ES	P Value	Mean Difference (95% CI)	ES	P Value
Total motor composite	139	42.6 (7.3)	175	41.2 (6.8)	− 1.38 (−2.96 to 0.19)	−0.20	.09	− 1.70 (−3.21 to −0.18)	−0.25	.03
Strength and agility	139	43.1 (7.5)	176	41.4 (7.8)	− 1.75 (−3.45 to −0.05)	−0.23	.04	− 2.30 (−4.01 to −0.59)	−0.31	.01
Running speed and agility	139	11.9 (3.3)	177	11.3 (3.8)	− 0.60 (−1.39 to 0.19)	−0.17	.14	− 0.87 (−1.67 to −0.07)	−0.24	.03
Strength	139	12.3 (4.4)	176	11.4 (4.0)	− 0.92 (−1.85 to 0.02)	−0.22	.06	− 1.12 (−2.07 to −0.16)	−0.27	.02
Fine manual control	139	46.8 (7.9)	176	45.8 (7.2)	− 1.01 (−2.70 to 0.68)	−0.13	.24	− 1.19 (−2.83 to 0.45)	−0.17	.16
Fine motor precision	140	11.7 (3.7)	177	11.6 (3.7)	− 0.09 (−0.92 to 0.73)	−0.03	.82	− 0.16 (−1.00 to 0.68)	−0.04	.70
Fine motor integration	140	15.2 (3.8)	177	14.3 (3.7)	− 0.88 (−1.72 to −0.04)	−0.23	.04	− 0.93 (−1.76 to −0.11)	−0.26	.03
Manual coordination	139	43.5 (7.8)	176	42.5 (7.8)	− 1.00 (−2.73 to 0.73)	−0.13	.26	− 1.25 (−2.93 to 0.42)	−0.17	.14
Manual dexterity	139	12.0 (3.7)	177	12.0 (3.8)	− 0.03 (−0.86 to 0.81)	−0.01	.95	− 0.18 (−1.00 to 0.64)	−0.05	.67
Upper limb coordination	139	12.5 (4.3)	176	11.8 (4.1)	− 0.70 (−1.63 to 0.22)	−0.17	.14	− 0.87 (−1.79 to 0.05)	−0.22	.06
Body coordination	139	44.8 (8.1)	177	43.5 (7.8)	− 1.28 (−3.05 to 0.48)	−0.16	.16	− 1.28 (−3.09 to 0.53)	−0.16	.17
Bilateral coordination	139	13.6 (4.1)	177	13.3 (4.0)	− 0.30 (−1.19 to 0.59)	−0.07	.51	− 0.39 (−1.26 to 0.48)	−0.10	.38
Balance	139	12.4 (4.3)	177	11.7 (4.2)	− 0.69 (−1.65 to 0.26)	−0.16	.16	− 0.66 (−1.66 to 0.34)	−0.15	.20

Abbreviations: BOT-2, Bruininks-Oseretsky Test of Motor Proficiency-Second Edition; CI, confidence interval; ES, standardized effect size.

^aAverage norm-referenced score for BOT-2 composite scales is a T score = 50 ± 10, and for subtests is a scaled score = 10 ± 3.^bAdjusted for sex, socioeconomic status (continuous), race (white/nonwhite), and age (continuous, months).

In analyses stratified by the severity of PPB in infancy, meaningful group differences were observed only among children who had moderate-severe PPB as infants (Figure). Children with moderate-severe PPB scored lower than controls on the strength and agility, fine manual control, body coordination, and total motor composites (ES = −0.35 to −0.26, *P* values = .01 to .05).

The case-control difference for manual coordination was smaller in magnitude and was not statistically significant (ES = −0.21, *P* = .22). Review of BOT-2 subscales suggests that differences are most prominent in running speed and agility (ES = −0.30), strength (ES = −0.29), fine motor integration (ES = −0.35), and upper limb coordination (ES = −0.28). Among children

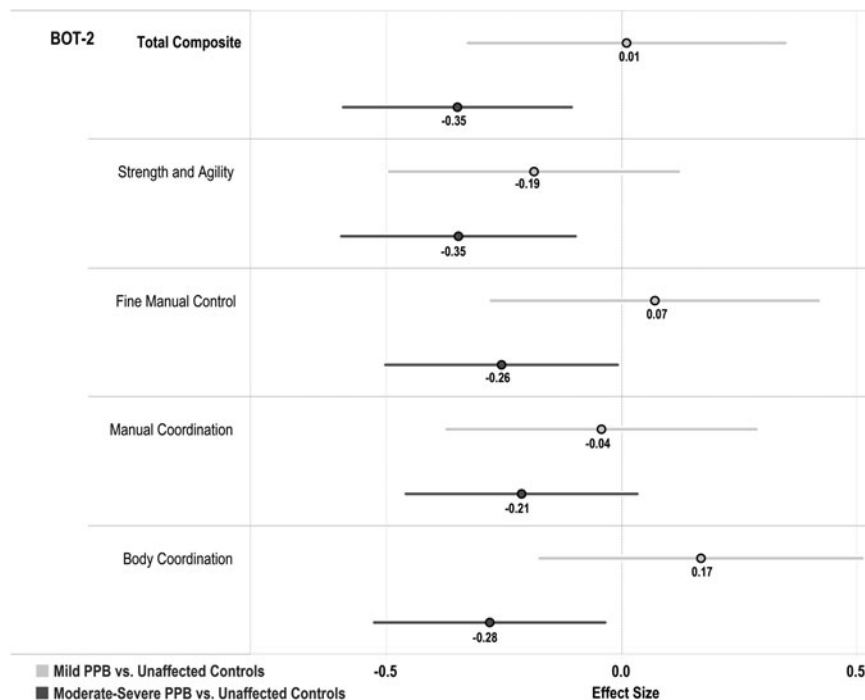


Fig. Case-control differences in BOT-2 motor scores by PPB severity, mild PPB versus controls, and moderate-severe PPB versus controls. BOT-2 indicates Bruininks-Oseretsky Test of Motor Proficiency-Second Edition; PPB, positional plagiocephaly/brachycephaly.

who had mild PPB, effect sizes were small and there were no statistically significant differences observed ($ES = -0.23$ to 0.17 , P values = .14 to .96).

Case-control differences were similar for children with and without a history of torticollis (Table 3). For children without torticollis, effect sizes ranged from -0.30 to -0.15 ($P = .03$ to $.27$) and for children with torticollis effect sizes were -0.31 to -0.08 ($P = .04$ to $.45$). Receipt of orthotic helmet treatment did not appreciably affect motor outcomes. Observed differences were larger when accounting for differential attrition using inverse probability weighting, with ES for overall group differences ranging from -0.35 to -0.19 . Similarly, effect sizes were increased when adjusting for the anticipated effects of motor interventions using censored normal regression ($ES = -0.70$ to -0.56). In other words, when adjusting for the effects of developmental intervention and the effects of differential attrition (eg, higher rates of attrition among children who had developmental delays as infants or toddlers), we expect the true differences between children with and without PPB to be larger than those observed.

DISCUSSION

Children with a history of moderate to severe PPB scored lower on measures of motor development than unaffected peers, while no consistent case-control differences were observed for children with mild PPB. These results are comparable to our findings in this cohort on other outcome measures.¹⁵ Although these findings warrant replication, they suggest that the clinical management of PPB should differ depending on its severity in infancy. Among infants with moderate-severe PPB, close developmental monitoring and intervention appear warranted. As motor skills are the earliest and most prominent areas of delay in this population, these would be the most likely “target” for intervention. In addition to offsetting motor skill deficits, addressing early motor development may have benefits for other areas of development. Specifically, there is evidence that infant and toddler motor skills are associated with the development of language, cognition, and academic achievement.²¹

Further, although effects are modest, there is emerging evidence that physical therapy and related interventions delivered in infancy can help to reduce skull deformation associated with PPB.^{22,23}

We did not find any differences based on history of orthotic helmet treatment, suggesting that treatments to modify skull shape do not necessarily influence motor development. Similarly, we did not find differences in school-age motor outcomes as a function of torticollis in infancy. Although there is considerable overlap between torticollis and PPB, other associated motor vulnerabilities (eg, hypotonia) may confer greater risk for poor developmental outcomes. This would make intuitive sense for some of the motor tasks for which we observed the largest case-control differences, such as strength and agility. Aside from the Bayley-3 evaluations in early childhood, we do not have specific measures of muscle tone or other areas of motor function (eg, range of motion) to evaluate this possibility in our sample. Findings in the literature on torticollis (independent of PPB) are mixed, with some studies showing that children with torticollis are at risk for poor long-term outcomes (eg, higher than expected rates of parent-reported neurodevelopmental coordination disorders and attention-deficit hyperactivity disorder¹⁵) and others showing typical development on standardized measures relative to controls.²⁴ Prospective studies assessing muscle tone and range of motion in infants with PPB are needed to clarify these associations and may help to identify specific motor deficits to address with intervention.

A high proportion of the participants with PPB in our sample received community-based interventions targeting motor development (ie, occupational therapy or physical therapy), which lends support to the notion that motor skills are an area of vulnerability. However, the rate of intervention was likely inflated by children's participation in phase 1 of this research. That is, we provided parents with their child's test results and encouraged them to share this information with their pediatrician whenever developmental concerns were indicated, in order to inquire about the need for further assessment and intervention. If anything, the high rate of developmental intervention among children with PPB in our sample might have

TABLE 3

Comparison of Mean BOT-2 Scores for Children With and Without Positional Plagiocephaly/Brachycephaly by Torticollis

		Torticollis						
		No n = 103			Yes n = 84			P Value Group Difference
BOT-2 Scale ^b	Controls ^a n = 146	Mean Difference ^c (95% CI)	ES	P Value	Mean Difference ^c (95% CI)	ES	P Value	
Total motor composite	Reference	−1.75 (−3.49 to −0.01)	−0.26	.05	−1.57 (−3.48 to 0.35)	−0.23	.11	.10
Strength and agility	Reference	−2.25 (−4.24 to −0.26)	−0.30	.03	−2.33 (−4.53 to −0.14)	−0.31	.04	.04
Fine manual control	Reference	−1.74 (−3.50 to 0.02)	−0.25	.05	−0.55 (−2.69 to 1.58)	−0.08	.61	.14
Manual coordination	Reference	−1.14 (−3.10 to 0.82)	−0.15	.26	−1.36 (−3.47 to 0.75)	−0.18	.21	.35
Body coordination	Reference	−1.18 (−3.26 to 0.90)	−0.15	.27	−1.16 (−3.40 to 1.09)	−0.15	.31	.45

Abbreviations: BOT-2, Bruininks-Oseretsky Test of Motor Proficiency-Second Edition; CI, confidence interval; ES, standardized effect size.

^aExcludes 3 controls with torticollis.

^bAverage norm-referenced score for the BOT-2 composite scales is a T score = 50 ± 10 .

^cAdjusted for sex, socioeconomic status (continuous), race (white/non-white), and age (continuous, months).

led to an underestimate of case-control differences in the population, as indicated by our analyses using censored normal regression. Unfortunately, because interventions were provided in multiple community settings, we were unable to more specifically examine effects related to the type, timing, or intensity of treatment.

Importantly, our findings do not imply that PPB causes developmental delay. We believe that the opposite is more likely (ie, children with motor deficits are more likely to develop PPB in infancy, particularly in the context of frequent supine positioning). These early motor deficits might also predict an elevated risk for later developmental concerns, including those less obviously related to motor functions (cognition, language). Future, prospective studies tracking development prior to the onset of PPB are needed to confirm this impression. If this hypothesis were accurate, PPB might serve as a marker of developmental risk, prompting providers to more closely monitor development and intervene early.

Scores on the BOT-2 were lower than expected relative to test norms for children with and without PPB. This is somewhat surprising, particularly for children in the control group, given that the sample was low risk (ie, children with known neurodevelopmental conditions were excluded). Although interrater reliability was excellent and the BOT-2 was administered according to standardized instructions, study examiners may have been more stringent than examiners who participated in the norming of the measure. If this were true, test scores for both groups would presumably have been affected and case-control differences would have been unaltered. Fatigue might have also been a factor, as the BOT-2 was the final assessment in an extensive battery (~4.5 hours total). The order of administration was consistent for children with and without PPB, and we therefore do not believe that fatigue would have influenced the group differences observed. Regardless, this highlights the importance of including a control group for comparison rather than relying only on test norms for comparison, which in this instance may have overestimated deficits associated with PPB.

Although retention in the study was excellent overall, there was differential attrition among children who had delays in early childhood, more severe PPB, and lived in lower SES households. Analyses adjusting for attrition suggest that, if anything, observed case-control differences may be an underestimate of true group differences.

Clinically, we recommend close developmental monitoring for children with moderate-severe PPB and interventions to address their motor deficits when present (eg, occupational or physical therapy). As the motor development of children with mild PPB did not differ from controls in this study, we would advise reassurance and anticipatory guidance for the parents of these children. Our finding that orthotic helmet treatment was unrelated to developmental outcomes suggests that developmental interventions for PPB should be primarily based on underlying skill deficits rather than skull deformation. Indeed, for all children with PPB, recommendations for parents to promote motor development may have the dual benefits of reducing deformation and improving motor and other developmental outcomes.

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