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ORIGINAL PAPER

Infant/toddler motor skills as predictors of cognition and language in children with and without positional skull deformation

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Abstract

Purpose To estimate associations between early motor abilities (at two age points, 7 and 18 months on average) and cognitive/language outcomes at age 3. To determine whether these associations are similar for children with and without positional plagiocephaly and/or brachycephaly (PPB).

Methods The Bayley Scales of Infant/Toddler Development 3 were given at all age points to 235 children with PPB and 167 without PPB. Linear regressions assessed longitudinal associations between fine and gross motor scales and cognition/language. Item analyses examined the contributions of specific motor skills.

Results Associations between 7-month motor skills and cognition/language were modest overall (effect sizes [ES] = − 0.08 to 0.10, $p = .13$ to .95). At 18 months, both fine and gross motor skills were associated with outcomes for children *with* PPB (ES = 0.21 to 0.41, $p < .001$ to .01), but among those *without* PPB, only fine motor skills were associated with outcomes (ES = 0.21 to 0.27, $p < .001$ to .001).

Conclusions Toddlers' motor skills were associated with cognition and language at 3 years, particularly among children with PPB. Interventions targeting early motor development in infants and toddlers with PPB may have downstream benefits for other outcomes.

Keywords Plagiocephaly · Brachycephaly · Motor skills · Infant · Development

Introduction

Infant positional skull deformation, including plagiocephaly (i.e., asymmetric head shape) and brachycephaly (i.e., lower than normal ratio of skull length:width), is common in countries that have programs to prevent sudden infant death syndrome, typically by encouraging parents to place their infants in a supine sleep position [1, 2]. Prevalence data are limited, but the best estimates suggest that 20–30% of infants show some degree of positional plagiocephaly and/or brachycephaly (PPB) [3, 4], which may persist into the preschool years [5]. As a result, increasing numbers of children have been referred to craniofacial centers and neurosurgery clinics for evaluation of skull deformation, with a concomitant increase in treatments such as orthotic helmets and bands [1, 2].

Although initially considered a purely cosmetic condition, PPB has been associated with other medical problems and with an increased risk of developmental delay [6]. In a recent review paper, 13 of 19 studies reviewed reported developmental delays among infants with PPB. In children < 2 years of age, motor skills were the most commonly affected domain. This is not surprising given the known link between skull deformation and limitations in head and neck movement (e.g., torticollis) and associations between skull deformation and environmental restrictions on movement (e.g., consistent sleep positioning, limited 'tummy time') [6].

In a series of longitudinal assessments, we observed motor skill deficits among infants and toddlers with PPB relative to

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unaffected children [7–9]. By age 36 months, group differences in motor skills were reduced, while deficits in cognition and language were more pronounced [7]. We have not assumed that these deficits are *caused* by skull malformation. Rather, we have hypothesized that PPB may be a marker of developmental risk [7]. For example, infants with subtle neuromotor deficits may be less able to reposition themselves and therefore at greater risk for developing skull deformation. These neuromotor deficits may have a cascading effect on other areas of development. Bornstein et al. [10] showed that infant motor-exploratory behavior (e.g., independent sitting, crawling) predicts academic achievement in adolescence, via cognition in middle childhood. Similarly, others have shown that infant motor skills are associated with language development and reading acquisition [11]. In children with PPB, these findings may imply that early interventions targeting motor development would have downstream benefits for other areas of development.

The goal of this study was to investigate predictive relations between infant/toddler motor abilities and cognition and language in preschoolers with a history of PPB. We examined associations between motor skills assessed at approximately ages 7 and 18 months and cognitive and language outcomes assessed at 3 years of age. We addressed the following questions: (1) Do motor skills predict cognition and language among children with PPB? (2) Are predictive associations similar for children with and without PPB? (3) Among children with PPB, do specific domains of early motor skill (e.g., fine vs. gross motor; head control vs. sitting) predict differently across different outcome domains (e.g., language vs. cognition, expressive vs receptive language)?

Methods

Participants

Families were enrolled after obtaining informed consent using procedures approved by the Institutional Review Board at Seattle Children's Hospital (SCH).

Infants with PPB Infants with PPB were recruited from the Seattle Children's Craniofacial Center following a clinical evaluation of skull deformation [9]. Eligibility criteria included: (1) a diagnosis of PPB by a craniofacial specialist; (2) infant age was between 4 and 11 months, and (3) families were able to complete a study visit within 4 weeks of the child's diagnosis. Exclusions were: (1) history of prematurity (< 35-week gestation), (2) a diagnosed neurodevelopmental condition, brain injury, or significant hearing or vision impairment; (3) presence of a major malformation or $\geq$ 3 minor extracranial anomalies; [12] (4) a non-English speaking mother; (5) history of adoption or out-of-home placement; and (6) family plans to move out of state before project completion. Between June 2006 and February 2009, we recruited 235 infants with PPB, representing 52% of all eligible case subjects. Participating children were similar to non-participants with regard to demographic characteristics and severity of cranial deformation [9].

Infants without PPB The first eight infants without PPB were recruited through area pediatric practices. All remaining infants without PPB were recruited from a registry of families who agreed shortly after their child's birth to be contacted for research participation at a later date. Parents were contacted by phone when their child was 4 to 11 months old, and those who expressed interest in the project were screened to determine eligibility. In addition to the exclusions listed for children with PPB, children were excluded from this group if their parent reported a known or suspected history of PPB or other craniofacial anomalies. Among the infants meeting this criterion, we selected those who were most similar to infants in the PPB group in terms of infants' age, gender, ethnicity, and family socioeconomic status (SES) [13]. Two hundred thirty-seven infants without known DP were recruited between March 2007 and February 2009, representing 90% of those eligible for participation. Twenty-seven families declined participation.

Confirmation of PPB Digital three-dimensional (3D) photographs were taken of every infant in order to quantify and characterize skull shape. To confirm the presence or absence of PPB, these images were de-identified, randomly sorted, and rated by two physician dysmorphologists who were unaware of infants' enrollment status. The overall severity of cranial deformation was rated on a 4-point ordinal scale (0 = none, 1 = mild, 2 = moderate, 3 = severe). Inter-rater agreement for case status (i.e., presence or absence of PPB using the overall severity of cranial deformation scale) was excellent ($\kappa = 0.80$), and exact agreement for each of the four severity categories was adequate (weighted $\kappa = 0.72$). Two children with a diagnosis of PPB did not exhibit discernible skull deformation on 3D imaging at time 1 and were eliminated from analyses, as were five children with medical conditions that may affect neurodevelopment (e.g., Chiari malformation, chromosomal anomaly). Of the remaining 228 children with diagnosed PPB, 207 (91%) were assessed at time 3 and were included in the analyses. Among the 237 infants without previously diagnosed skull deformation at time 1, clinician ratings of 3D imaging identified 70 infants with some degree of PPB. Because the developmental course for these infants has been found to differ from that of unaffected infants, we excluded these participants from subsequent analyses. The resulting control sample at time 1 included 167 unaffected children, of which 158 (95%) participated at time 3.

Measures

Infants were first assessed at an average age of 7 months (time 1). Children were re-assessed at average ages of 18 and 36 months (time 2 and time 3, respectively). At each time point, infants or children were given the Bayley Scales of Infant and Toddler Development, 3rd Edition (Bayley-3) [14]. The Bayley-3 yields composite scores for cognitive, language, and motor development. Sub-scale scores are derived for expressive and receptive language and for fine and gross motor development. Raw scores are converted to norm-referenced standard scores (average = 100, standard deviation [SD] = 15) for composite scales and scaled scores (average = 10, SD = 3) for language and motor sub-scales. Gestational age was calculated using maternal report of due date and birth date. We corrected Bayley-3 scores for prematurity for children born between 35 and 37 weeks gestation, and for those born at 37 weeks gestation but weighing < 6 pounds. The Bayley-3 was administered by trained psychometrists who were unaware of children's case status, though on occasion, this may have been compromised by children's appearance or information volunteered by parents. Assessments were videotaped, and approximately 10% were reviewed for reliability by one of the authors (BC). Scoring agreement on individual items (kappa) was 0.84 to 0.90.

After testing, psychometrists indicated whether they considered the evaluation "valid" or "invalid" due to child behavior (e.g., persistent non-compliance) or testing circumstances (e.g., child illness). One or more Bayley-3 scores were dropped for one child with PPB and six children without PPB due to examiner ratings of invalidity.

Medical and intervention history Interviews were completed with mothers at time 1 to document demographic characteristics, maternal and child prenatal history, and child medical and developmental history. At time 2 and time 3, abbreviated interviews were conducted to obtain information about newly diagnosed medical conditions, treatments the child received for PPB, and participation in developmental interventions.

Statistical analysis

The distributions of demographic characteristics and motor and other neurodevelopmental scores at each time point were calculated for children with and without PPB. We examined associations between the Bayley-3 fine and gross motor subscales, assessed at time 1 and time 2, and Bayley-3 cognitive, language, and motor outcomes at time 3.

Separately for children with and without PPB, we used linear regression to estimate the univariate associations between motor skills assessed at times 1 and 2 with language (expressive and receptive), cognitive, and motor (fine and gross) scores at time 3. Raw scores were converted to a z-score for each of these outcomes to maximize the sensitivity of the analyses and the interpretability of the regression coefficients, and 95% confidence intervals were calculated using robust standard errors. We also examined the univariate relations between individual fine and gross motor test items (e.g., "sitting with support") and time 3 outcomes. These items are considered 'passed' if a child demonstrates the skill during testing. Very low or high item passing rates would not generate meaningful statistics—that is, an item would not have predictive value if it were passed or failed by the majority of infants tested. We therefore restricted all analyses of individual motor items to those with a pass or failure rate of at least 5%. In lieu of inferential statistics, given the number of comparisons, these associations were plotted to identify those

Table 1 Demographic characteristics by diagnosis group

Characteristic	Unaffected controls		Children with PPB	
	N	%	*N*	%
Total	158	100.0	207	100.0
Age at time 1 (months), mean (SD)	6.9	1.7	7.2	1.6
Age at time 2 (months), mean (SD)	18.3	0.7	18.5	0.7
Age at time 3 (months), mean (SD)	35.9	1.1	36.5	1.2
Gender				
Female	69	43.7	72	34.8
Male	89	56.3	135	65.2
Ethnicity				
White (non-Hispanic)	100	63.3	143	69.1
Asian/Pacific Islander	4	2.5	11	5.3
Black	5	3.2	0	0.0
Hispanic or Latino	19	12.0	23	11.1
Native American	30	19.0	30	14.5
Socioeconomic status				
I (highest)	36	22.8	74	35.7
II	68	43.0	81	39.1
III	32	20.3	31	15.0
IV	18	11.4	15	7.2
V (lowest)	4	2.5	6	2.9
Intervention services				
Speech	7	4.4	31	15.0
Physical therapy	1	0.6	90	43.5
Occupational therapy	5	3.2	22	10.6
Hearing	1	0.6	4	1.9
Developmental	2	1.3	9	4.3
0–3 services	9	5.7	28	13.5
Any intervention	15	9.5	109	52.7

Missingness: age at time 2 (1 control, 1 child with PPB), speech services (3 controls, 10 children with PPB), physical therapy (3 controls, 4 children with PPB), occupational therapy (3 controls, 4 children with PPB), hearing therapy (3 controls, 3 children with PPB), developmental services (2 controls, 6 children with PPB), 0–3 services (3 controls, 4 children with PPB)

items with particularly robust or consistent associations with time 3 outcomes.

Results

Demographic characteristics and information about intervention services prior to time 3 are summarized in Table 1. Average age at the first assessment was 7.0 months (SD = 1.6) and ages 18.4 (SD = 0.7) and 36.2 (SD = 1.2) months at time 2 and time 3, respectively. Relative to children without PPB, children with PPB were more likely to be male, white, and of higher socioeconomic status (SES). Fifty-three percent of children with PPB received developmental intervention prior to their time 3 visit, the most common being physical therapy (44%).

Infant motor skills (time 1) and cognition and language at 36 months

There was little evidence for associations of Bayley-3 fine, gross, or composite motor scores at time 1 with any outcome at time 3 in children with PPB. Effect sizes (ES) ranged from − 0.05 to 0.10 (p values = 0.13 to 0.95) (Table 2). Analyses of individual items are provided in Fig. 1, and effect sizes (ES) and 95% confidence intervals for all items are provided in Supplementary Table 1. One grasping item, the thumb-fingertip grasp of a food pellet, was positively associated with time 3 expressive and receptive language and cognitive outcomes (ES = 0.37 to 0.40), but there was little consistent evidence for other grasping items (Fig. 1). There were several consistent but modest associations between gross motor items (e.g., making stepping movements, walking with support, and walking sideways with support) and time 3 outcomes (see Fig. 1 and Supplementary Table 1).

In unaffected infants, associations between the Bayley-3 motor sub-scales and composite score at time 1 and outcomes at time 3 were negligible (Table 2). Similarly, none of the individual fine or gross motor items at time 1 were consistently associated with time 3 outcomes (see Fig. 1 and Supplementary Table 2).

Toddler motor skills (time 2) and cognition and language at 36 months

Bayley-3 motor scores at time 2 for children with PPB were positively associated with all outcomes at time 3 (ES = 0.21 to 0.45, p values < 0.001 to 0.01) (Table 2). Time 2 fine motor items related to crayon/pencil grasp and drawing were positively associated with multiple time 3 outcomes, with effect sizes ranging from 0.12 to 0.78 (p values < 0.001 to 0.52; see Fig. 2 and Supplementary Table 1). Among gross motor items, balancing on one foot with support, walking down stairs, and running with coordination were all positively associated with time 3 outcomes.

Table 2 Association between summary motor scores and time 3 language and cognitive outcomes in cases and controls

Case group	Time point and domain	Outcome											
		Expressive language				Receptive language				Cognitive			
		Estimate	95% CI		p value	Estimate	95% CI		p value	Estimate	95% CI		p value
Children with PPB	*Time 1*												
	Summary: fine	− 0.05	− 0.18	0.08	0.47	− 0.05	− 0.20	0.09	0.49	0.00	− 0.11	0.12	0.95
	Summary: gross	0.09	− 0.05	0.23	0.19	0.09	− 0.08	0.25	0.30	0.10	− 0.03	0.23	0.13
	Total motor	0.04	− 0.11	0.18	0.60	0.03	− 0.13	0.19	0.70	0.07	− 0.06	0.19	0.30
	Time 2												
	Summary: fine	0.36	0.13	0.59	0.002	0.42	0.17	0.66	0.001	0.41	0.17	0.65	0.001
	Summary: gross	0.21	0.05	0.36	0.01	0.29	0.12	0.47	0.001	0.30	0.16	0.44	< 0.001
	Total motor	0.36	0.15	0.56	0.001	0.45	0.23	0.68	< 0.001	0.45	0.26	0.65	< 0.001
Unaffected controls	*Time 1*												
	Summary: fine	− 0.04	− 0.17	0.10	0.61	0.01	− 0.12	0.15	0.83	0.03	− 0.07	0.14	0.52
	Summary: gross	− 0.08	− 0.20	0.05	0.25	− 0.04	− 0.18	0.09	0.53	− 0.02	− 0.12	0.09	0.74
	Total motor	− 0.06	− 0.19	0.07	0.35	− 0.02	− 0.16	0.12	0.75	0.00	− 0.10	0.11	0.95
	Time 2												
	Summary: fine	0.21	0.09	0.33	0.001	0.26	0.12	0.39	< 0.001	0.27	0.18	0.37	< 0.001
	Summary: gross	0.10	− 0.07	0.27	0.25	0.12	− 0.03	0.27	0.12	0.03	− 0.10	0.16	0.67
	Total motor	0.21	0.04	0.38	0.02	0.26	0.09	0.42	0.002	0.20	0.07	0.34	0.003

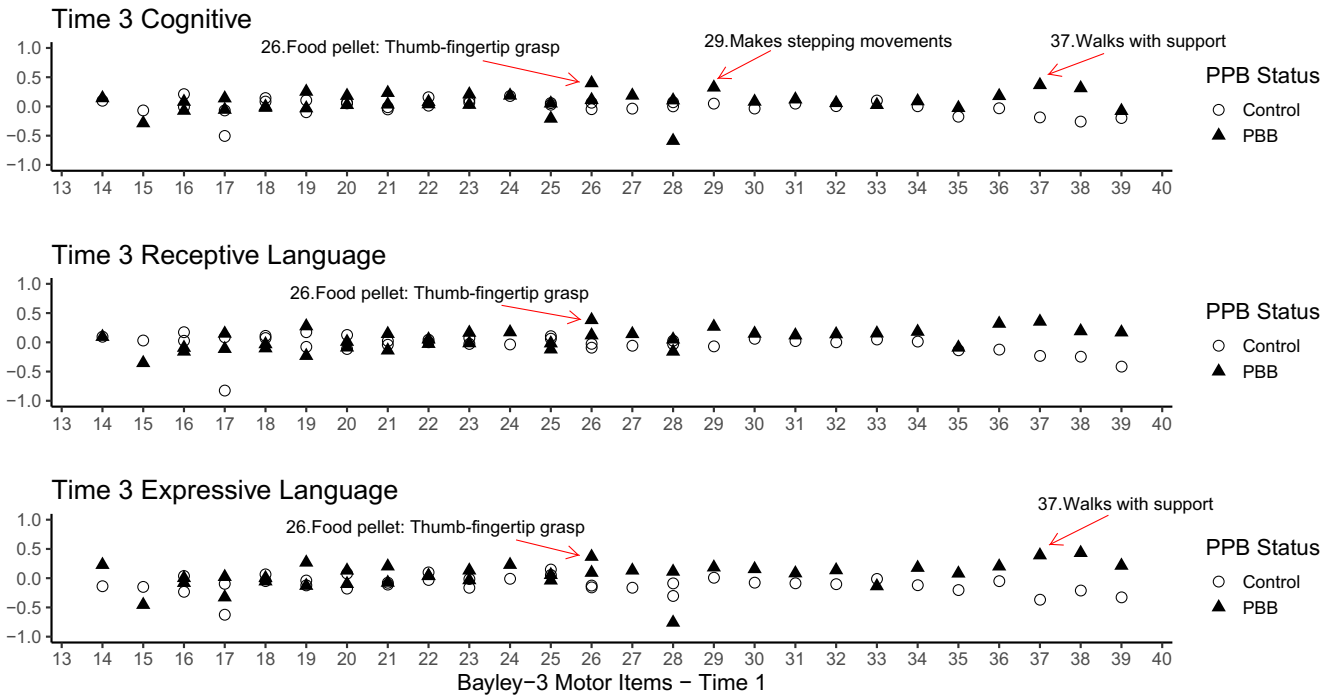

Fig. 1 Motor skills at time 1 in relation to cognition, receptive language, and expressive language at age 36 months. Values represent the standardized effect size change in the Bayley-3 scores at age 36 months for each motor item passed at time 1

Bayley-3 fine motor and total motor scores for unaffected controls were associated with all time 3 outcomes (ES = 0.20 to 0.27, p values < 0.001 to 0.02), while associations between these outcomes and Bayley-3 gross motor scores were negligible. Among individual items, those related to crayon/pencil grasp and imitating drawing were modestly positively associated with time 3 cognitive and language scores (see Fig. 2 and Supplementary Table 2). No other consistent associations between time 2 motor skills and time 3 outcomes were observed.

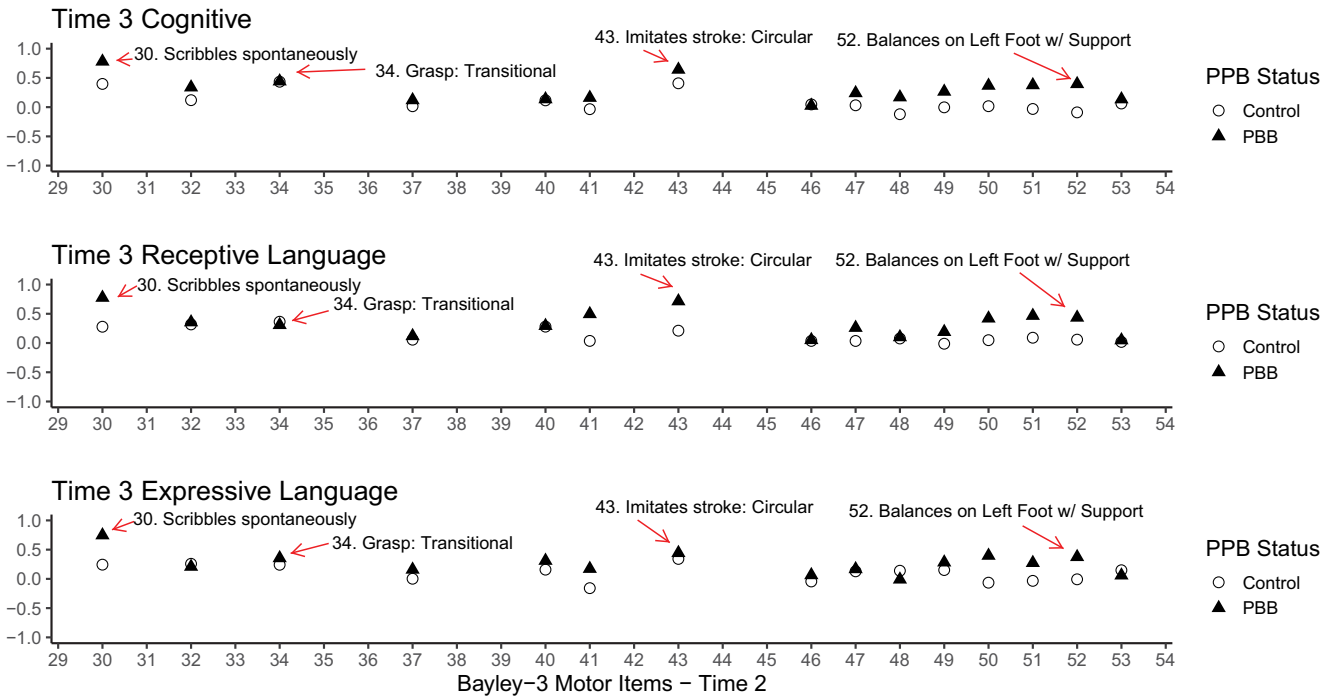

Fig. 2 Motor skills at time 2 in relation to cognition, receptive language, and expressive language at age 36 months. Values represent the standardized effect size change in Bayley-3 scores at age 36 months for each motor item passed at time 2

Discussion

Associations between infant motor skills, assessed at an average age of 7 months, and cognitive/language outcomes at age 36 months were modest overall. Among children with a history of PPB, motor skills in infancy (e.g., walking with support, grasping a food pellet) were moderately but inconsistently associated with cognition and language. Infant motor skills in controls were largely unrelated to outcomes at 36 months. Associations were stronger between motor skills assessed at 18 months and time 3 outcomes. Among toddlers with and without PPB, fine motor skills such as grasping a crayon or pencil and imitating line drawings were significantly associated with cognition and language at 36 months. Toddlers' gross motor skills (e.g., balancing on one foot) were associated with time 3 outcomes for PPB cases but not controls.

These findings are partially consistent with previous literature, showing that infant and toddler motor skills are predictive of later language, academic achievement, and cognitive outcomes. Bornstein et al. [10] and others [15–17] have proposed that motor skills in early childhood serve as a 'catalyst' for other aspects of development, through active engagement of the environment and caregivers. For example, as a child becomes more mobile, caregivers may be more likely to use language contingent on the child's activity (e.g., commenting on a toy or other object that the child approaches) [18, 19]. Similarly, fine motor activity may promote symbolic play and caregiver-child conversation (e.g., using blocks to build a house). Although these types of motor achievements would not be considered necessary and sufficient for development, deficits may place children at a relative disadvantage.

As observed in previous research with clinical and non-clinical samples [15, 16], associations between motor skills and outcomes at age 36 months were stronger among children with PPB versus unaffected controls. We have previously shown that children with PPB score lower than controls on developmental measures through age 36 months [7], and associations among developmental domains might be more apparent in children with developmental delays [20]. Alternatively, this may reflect the greater variability in development in children with PPB versus controls, in both their early motor skills and cognitive and language outcomes at age 36 months. This variability may provide the range necessary to detect an association. In our sample, cognition and language were more consistently associated with fine versus gross motor skills. These skills, developed in the first 2 years, may also correlate more closely than gross motor skills with the types of cognitive problem-solving tasks featured on the Bayley-3 exam that require visual-motor abilities (e.g., puzzles, pegboard, etc.).

Study limitations include the use of a relatively broad assessment of motor development. Previous studies have often utilized serial assessment of very specific motor milestones (e.g., walking). Although our study more closely resembles the information gathered in early intervention settings, it may lack the precision of a more detailed motor assessment. Finally, many of our participants, especially those with a history of PPB, received developmental interventions between infancy and age 36 months. If anything, such interventions might make it more difficult to detect an association. Specifically, children with early motor delays are more likely to receive interventions like physical therapy or occupational therapy, which may benefit other outcomes.

As we have emphasized in previous papers [7–9], the observed associations between PPB and early development do not necessarily imply that skull deformation *causes* delays. Although this cannot be definitively ruled out based on our research, we have hypothesized the opposite. That is, we propose that early delays in motor development make children more likely to develop skull deformation in the context of routine supine positioning and limited opportunities for 'tummy time' and motor exploration. PPB might therefore be considered a 'marker' of developmental risk that could prompt further assessment and intervention. Findings from this study suggest that motor skills assessed at age 18 months were consistently and significantly associated with language and cognitive outcomes among children with PPB when they reached preschool age. A holistic developmental approach to physical therapy and occupational therapy for children with PPB intervention is warranted [21–23], as such interventions might lay the developmental foundation for the later acquisition of cognitive and language skills. Early motor deficits are among the first developmental issues observed in this population and targeted intervention or prevention efforts may help to improve these and other developmental outcomes. For example, coaching caregivers to provide a variety of fine and gross motor activities during the first 1 to 2 years may help to develop other problem solving and language skills. We are continuing to follow this cohort into early school age and will therefore have an opportunity to examine early motor development and developmental trajectories in relation to more precise cognitive and language outcomes.

Compliance with ethical standards

Conflict of interest None of the authors have any conflicts of interest to disclose.

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