Development at Age 36 Months in Children With Deformational Plagiocephaly

AUTHORS: Brent R. Collett, PhD,[a,b] Kristen E. Gray, MS,[c] Jacqueline R. Starr, PhD,[c,d,e] Carrie L. Heike, MD,[f,g] Michael L. Cunningham, MD, PhD,[f,g] and Matthew L. Speltz, PhD[a,b]

Departments of [a]Psychiatry and Behavioral Sciences, [c]Epidemiology, and [f]Pediatrics, University of Washington, Seattle, Washington; Department of [b]Psychiatry and Behavioral Medicine, and [g]Seattle Children's Craniofacial Center, Seattle Children's Hospital, Seattle, Washington; [d]Department of Clinical and Translational Research, The Forsyth Institute, Cambridge, Massachusetts; and [e]Department of Oral Health Policy and Epidemiology, Harvard School of Dental Medicine, Boston, Massachusetts

KEY WORDS

plagiocephaly, developmental assessment, preschool age, Back to Sleep

ABBREVIATIONS

3D—3 dimensional
BSID-III—Bayley Scales of Infant and Toddler Development, Third Edition
CI—confidence interval
DP—deformational plagiocephaly
RR—relative risk
SES—socioeconomic status

Dr Collett assisted in study conceptualization and design, participated in recruitment and data collection, supervised infant/toddler examiners, led data analyses, drafted the initial manuscript, and approved the final manuscript as submitted; Ms Gray assisted with conceptualization and study design, particularly data analyses and interpretation; assisted with manuscript preparation; provided a critical review of the manuscript; and approved the final manuscript as submitted; Dr Starr assisted in conceptualization and study design, particularly data analyses and interpretation; provided a critical review of the manuscript; and approved the final manuscript as submitted; Dr Heike assisted with conceptualization and study design, particularly collection of 3-dimensional cranial imaging data; provided a critical review of the manuscript; and approved the final manuscript as submitted; Dr Cunningham assisted with study conceptualization and design, provided medical oversight during participant enrollment (eg, to determine eligibility when questions arose for children with other medical conditions), provided a critical review of the manuscript, and approved the final manuscript as submitted; and Dr Speltz conceptualized and designed the study, obtained National Institutes of Health funding, supervised data collection, assisted with data analyses and interpretation, provided a critical review of the manuscript, and approved the final manuscript as submitted.

(Continued on last page)

WHAT'S KNOWN ON THIS SUBJECT: Infants and toddlers with deformational plagiocephaly (DP) score lower on developmental measures than children without DP and lower than expected relative to test norms.

WHAT THIS STUDY ADDS: This study is the first to examine developmental outcomes in preschool-aged children with DP relative to demographically similar children without DP using a standardized, clinician administered assessment.

abstract

OBJECTIVES: Infants and toddlers with deformational plagiocephaly (DP) have been shown to score lower on developmental measures than unaffected children. To determine whether these differences persist, we examined development in 36-month-old children with and without a history of DP.

METHODS: Participants included 224 children with DP and 231 children without diagnosed DP, all of who had been followed in a longitudinal study since infancy. To confirm the presence or absence of DP, pediatricians blinded to children's case status rated 3-dimensional cranial images taken when children were 7 months old on average. The Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III) was administered as a measure of child development.

RESULTS: Children with DP scored lower on all scales of the BSID-III than children without DP. Differences were largest in cognition, language, and parent-reported adaptive behavior (adjusted differences = −2.9 to −4.4 standard score points) and smallest in motor development (adjusted difference = −2.7). Children in the control group who did not have previously diagnosed DP but who were later rated by pediatricians to have at least mild cranial deformation also scored lower on the BSID-III than unaffected controls.

CONCLUSIONS: Preschool-aged children with a history of DP continue to receive lower developmental scores than unaffected controls. These findings do not imply that DP causes developmental problems, but DP may nonetheless serve as a marker of developmental risk. We encourage clinicians to screen children with DP for developmental concerns to facilitate early identification and intervention. *Pediatrics* 2013;131:e109–e115

Deformational plagiocephaly (DP) refers to flattening of the infant skull secondary to external forces. The prevalence of DP in the United States has increased from 5% in the 1990s to 20% to 30% at present,[1–3] an increase largely attributed to the successful "Back to Sleep" campaign.[4] Many clinicians consider DP to be a minor cosmetic condition, although DP has been associated with heightened risk for developmental delays in infants and toddlers.[5–10] Data on the persistence of DP-associated delays are less consistent.[11–13] Existing studies are limited by the use of parent observations rather than clinician-administered measures, and most have relied on retrospective evaluations of development and comparisons to test norms.

To determine whether DP is associated with development from diagnosis through age 3 years, we initiated a longitudinal study of 235 children diagnosed with DP and 237 demographically similar controls. Participants were previously assessed at an average age of 7 and 18 months (Time 1 and Time 2, respectively) using the Bayley Scales of Infant and Toddler Development–Third Edition (BSID-III).[14] At both assessments, children with DP received lower BSID-III scores than controls.[5,10] In this study, we sought to examine whether (1) these group differences persisted at age 36 months (Time 3), (2) findings were altered by participation in developmental interventions, and (3) outcomes among cases were affected by demographic and clinical variables.

METHODS

Participants

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

Infants With DP

The parents of infants with DP were approached for participation at the time of their child's diagnosis at the Seattle Children's Hospital Craniofacial Center (see Speltz et al[10]). Patients were eligible if they had been diagnosed with DP by a craniofacial specialist, were aged 4 to 11 months, and families were able to complete a study visit within 4 weeks of the child's diagnosis. Exclusions were (1) history of prematurity (<35 weeks' gestation); (2) a diagnosed neurodevelopmental condition, brain injury, or significant hearing or vision impairment; (3) presence of a major malformation or ≥3 minor extracranial anomalies[15]; (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. We recruited 235 infants with DP between June 2006 and February 2009, representing 52% of eligible patients. Participants were similar to nonparticipants with regard to demographic characteristics and DP severity.[10]

Infants Without DP

In addition to the exclusions listed for infants with DP, infants without DP were excluded if they had been diagnosed with DP or any other craniofacial anomaly. The first 8 infants in this group were identified through pediatric practices. Remaining infants were identified from a pool of families who agreed to be contacted for research participation when their child was born. 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. We selected controls who were most similar to infants in the DP cohort in terms of infants' age, gender, ethnicity, and family socioeconomic status (SES).[16] 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.

Measures

Severity of Cranial Deformation

Three-dimensional (3D) cranial images were obtained during participants' Time 1 study visit using the 3dMD Cranial System (see Speltz et al[10]). Images were deidentified, randomly sorted, and rated for severity by 2 craniofacial pediatricians (MC and CH) who were unaware of DP status. A 4-point ordinal scale (none, mild, moderate, severe) was used to rate cranial deformation. The mean of the 2 raters' scores was used to represent the overall severity of each participant's cranial deformation. Interrater agreement (weighted κ) was 0.72 for severity category and (κ) 0.80 for the presence or absence of any deformation.

Bayley Scales of Infant and Toddler Development, Third Edition (BSID-III)

The BSID-III[14] yields composite scores for cognitive, language, and motor development and for parent reports of the child's adaptive behavior. Subscale 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, SD = 15) for composite scales and scaled scores (average = 10, SD = 3) for language and motor subscales. Gestational age was calculated using maternal report of due date and birth date. We corrected BSID-III 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 BSID-III was administered by trained psychometrists, who were blinded to children's case status, although on occasion this may have been compromised by some children's appearance or information shared by parents. Assessments were videotaped, and ~10% were reviewed for reliability by one of the authors (BC). Scoring agreement on individual

items (κ) was 0.84 to 0.90. Mothers were asked to complete the BSID-III adaptive development scale and return the form by mail.

Medical and Intervention History

Interviews were completed with mothers at Time 1 to document demographic characteristics and medical history, including history of suspected or confirmed torticollis. At Time 3, an abbreviated interview was conducted for cases and controls to obtain information about newly diagnosed medical conditions and participation in developmental interventions. Because the duration of services varied, participants were only classified as receiving an intervention if they had ≥4 treatment sessions or ≥2 months of monitoring in a "Birth-to-Three" early intervention program. These cutoffs were used to differentiate children receiving only assessments from those participating in ongoing intervention. For cases, the Time 3 interview also documented the use of orthotic treatments for DP.

Assessment Procedures

We scheduled participants' Time 3 visit within 12 weeks of the child's 3-year birthday and set an upper age limit of 42 months. Parents unable to participate in a full assessment completed the interview by phone and were mailed the BSID-III adaptive behavior scale. After testing, psychometrists indicated whether they considered the evaluation "valid" or "invalid" due to child behavior (eg, noncompliance) or testing circumstances (eg, child illness). One or more BSID-III scores were dropped for 1 child with DP and 6 children without DP due to examiner ratings of invalidity.

Data Analyses

We used pediatricians' ratings of Time 1 3D surface images to categorize participants into 3 groups: DP Cases (infants with a diagnosis of DP, confirmed with clinician ratings), Unaffected Controls (infants with no previous diagnosis of DP and no evidence of DP with clinician ratings), and DP Controls (infants without previously diagnosed DP but detectable dysmorphology based on our pediatricians' ratings of 3D images).

Linear regression analyses with robust standard errors were used to compare Time 3 standard scores for DP cases and unaffected controls on the BSID-III cognitive, language, motor, and adaptive behavior composites. We also examined differences in scaled scores on BSID-III language and motor subscales. In categorical analyses, we used Poisson regression models with robust standard errors to estimate the relative risk (RR) of children with and without DP receiving a standard score <85 on the BSID-III composite scales, a conventional threshold for risk of developmental delay. We adjusted all analyses for children's age (in months), gender, race/ethnicity (white and non-Hispanic vs non-White or Hispanic), and SES.

In secondary analyses, we used linear regression to compare average BSID-III scores of unaffected controls and DP controls. Among cases, we examined developmental outcomes as a function of initial severity, coded as "mild" or "moderate to severe." In separate regression analyses, we also examined developmental outcomes among cases as a function of children's gender; history of suspected or confirmed torticollis; and history of orthotic treatment. In addition to adjusting for demographic covariates, we adjusted these analyses for Time 1 DP severity (continuous).

The foregoing analyses do not account for the fact that some children received developmental interventions before being assessed. This could bias case-control differences toward the null because these interventions were far more common among DP cases (see Table 1) and may have improved their developmental outcomes on average. We therefore repeated the primary analyses using censored normal regression.[17] This approach assumes that we cannot know how participants receiving intervention would have scored without treatment, but their estimated scores would likely have been at least as low as their observed scores (ie, their scores are "left censored"). This method therefore provides an estimate of bias by accounting for the anticipated increase in BSID-III scores after intervention. Analyses were completed by using the Stata SE 12.0 software package.[18]

RESULTS

Two hundred fifteen children with and 224 children without diagnosed DP completed the BSID-III at Time 3. A parent interview or BSID-III adaptive behavior scale was completed for an additional 9 children with DP and 7 controls, resulting in partial or complete data for 224 children with DP and 231 children without DP (95% and 97% of the original cohorts, respectively). Compared with participants, families lost to follow-up (11 children with DP, 6 children without DP) had lower SES (71% Hollingshead categories III–IV compared with 28% among participants), and a greater proportion were non-White or Hispanic (59% compared with 35% among participants). Participants and nonparticipants were similar in their average DP severity at Time 1.

Children with and without DP were predominantly male, of white race, and of middle to upper SES (Table 1). The mean age at Time 3 for children in both groups was ~36 months. Seventy-nine children with DP (35.3%) received orthotic treatment. Eighty-two children with DP (36.6%) and 15 children without DP (6.5%) participated in ≥4 intervention sessions or ≥2 months of Birth-to-Three monitoring.

TABLE 1 Demographic and Clinical Characteristics for Children With DP (Cases) and Without Deformational Plagiocephaly (Controls)

Characteristic	Cases (n = 224)		Controls (n = 231)	
	n	%	n	%
Gender				
Male	145	64.7	139	60.2
Female	79	35.3	92	39.8
Age in months at Time 3 (mean, SD)[a]	36.5(1.2)		36.0(1.1)	
Race/ethnicity				
Caucasian	153	68.3	144	62.3
Asian/Pacific Islander	12	5.4	12	5.2
Black/African American	0	–	6	2.6
Hispanic/Latino	26	11.6	28	12.1
Mixed race/Other	33	14.7	41	17.7
Familial SES (mean, SD)	47.1 (12.4)		46.9 (11.6)	
I (high)	80	35.7	60	26.0
II	89	39.7	100	43.3
III	34	15.2	46	19.9
IV	15	6.7	20	8.7
V (low)	6	2.7	5	2.2
History of torticollis[b]				
Suspected	9	4.0	3	1.3
Confirmed	89	39.7	2	0.9
None	126	56.3	226	97.8
Orthotic helmet or band therapy[b]				
Yes	79	35.3	0	–
No	145	64.7	231	100.0
Developmental interventions[b]				
Physical or occupational therapy	104	46.4	11	4.8
Speech/language therapy	34	15.2	14	6.1
Birth-to-Three monitoring	31	13.8	16	6.9
Other	38	17.0	18	7.8
Any Developmental Interventions[c]	122	54.5	38	16.5
≥4 intervention sessions or ≥2 mo Birth-to-Three early intervention monitoring	82	36.6	15	6.5
DP severity in infancy				
None	2	0.9	163	70.6
Mild	101	45.1	66	28.6
Moderate-severe	121	54.0	2	0.9

[a] Range in age = 34.0 to 41.7 mo in cases, 33.3 to 40.9 mo in controls.
[b] Variables assessed at Time 3, including children with any history of the condition or treatment.
[c] Refers to children who received ≥1 of the listed interventions by Time 3.

Fifty-four percent of cases had "moderate" or "severe" DP based on pediatricians' reviews of subjects' 3D images at Time 1 (Table 1). Among controls, 163 (70.6%) had no discernible skull dysmorphology, 66 (28.6%) had "mild" dysmorphology, and 2 (0.9%) had "moderate" to "severe" dysmorphology. We excluded from further analyses 2 children with diagnosed DP who did not have discernible skull dysmorphology. We also excluded from analysis 7 children with DP and 2 children without DP (both of who had evidence of dysmorphology on 3D imaging) who, after study enrollment, were diagnosed with other medical conditions that could affect neurodevelopment.[5]

This left 215 DP cases, 163 unaffected controls and 66 DP controls for analysis.

Developmental Outcomes for DP Cases Versus Unaffected Controls

Cases scored lower on average than unaffected controls on all BSID-III composite scales (Table 2). The largest differences were observed in language (adjusted difference = –4.4, 95% confidence interval [CI] = –6.8 to –2.0) and cognitive development (adjusted difference = –2.9 95% CI = –4.6 to –1.1). The smallest difference was in motor development (adjusted difference = –2.7, 95% CI = –5.1 to –0.3). DP cases also scored lower than unaffected controls on the receptive language, expressive language, and fine motor subscales.

DP cases were more likely than unaffected controls to receive scores <85 in language (RR = 7.9, 95% CI = 1.8–35.1), motor (RR = 4.3, 95% CI = 1.0–17.9), and cognitive development (RR was not calculated because none of the controls scored in the delayed range; Table 3). Children with DP were only slightly more likely than controls to score in the delayed range on the adaptive behavior (parent report) composite (RR = 1.7, 95% CI = 0.7–4.4).

Secondary Analyses

DP controls scored lower than unaffected controls on all BSID-III composite scales, with adjusted group differences of –2.9 to –3.5 standard score points. DP controls also scored lower on the receptive language (adjusted difference = –0.56, 95% CI = –1.1 to 0.0) and fine motor subscale (adjusted difference = –0.76, 95% CI = –1.5 to 0.0).

Among DP cases, outcomes were similar for children with "mild" and "moderate-severe" DP and for those with and without a history of orthotic treatment. Female DP cases scored lower than male DP cases in cognitive development (adjusted difference = –2.7, 95% CI = –5.1 to –0.3), although their scores were similar in all other areas. Average scores were similar for children with and without a history of torticollis except on the composite motor and gross motor scales, on which cases with torticollis scored on average 4.2 and 0.6 points higher, respectively (95% CI = 1.0–7.4 and 0.1–1.1, respectively).

In censored normal regression analyses, the magnitude of all differences between DP cases and unaffected controls were

TABLE 2 Average Standardized[a] BSID-III Scores and Adjusted[b] Group Comparisons for Children With DP (Cases) and Without DP (Controls)[c]

BSID-III Domain	Cases, n = 215 mean (SD)	Controls, n = 163 mean (SD)	Case vs unaffected control differences			
				95% CI		
			Adj. diff.	Lower	Upper	P value
Cognitive[d]	97.3 (8.3)	99.4 (8.2)	−2.9	−4.6	−1.1	.001
Language[d]	105.0 (11.9)	108.7 (10.8)	−4.4	−6.8	−2.0	<.0005
Receptive language[d]	10.9 (2.1)	11.3 (1.9)	−0.6	−1.1	−0.2	.005
Expressive language[d]	10.8 (2.2)	11.6 (2.1)	−0.9	−1.3	−0.4	<.0005
Motor[d]	100.4 (11.1)	102.5 (10.5)	−2.7	−5.1	−0.3	.030
Fine motor[d]	10.6 (2.4)	11.1 (2.3)	−0.6	−1.1	−0.1	.022
Gross motor[d]	9.5 (1.8)	9.7 (1.9)	−0.3	−0.7	0.1	.185
Adaptive behavior[e]	101.1 (13.9)	104.8 (13.0)	−4.0	−7.0	−0.9	.011

[a] Standard scores for the cognitive, language, motor, and adaptive behavior composite scales have a normative mean of 100 and SD of 15. Scaled scores for the receptive language, expressive language, fine motor, and gross motor subscales have a normative mean of 10 and SD of 3.

[b] Adjusted for child age (in mo), gender, SES (Hollingshead total, measured continuously), and ethnicity (white, non-Hispanic vs nonwhite or Hispanic).

[c] Data were dropped for 2 children in the DP group who did not have evidence of DP based on clinician ratings of 3D head photographs and 68 children in the non-DP group who had mild or greater posterior skull flattening or asymmetry. Data were also dropped for 7 children with DP and 2 children without DP who were diagnosed with medical conditions with developmental implications after enrollment in the study.

[d] Scores rated as "invalid" by the examiner were dropped from analyses. At Time 3, cognitive scores were rated invalid for 1 child without DP; language scores were rated invalid for 3 children without DP; and motor scores were rated invalid for 1 child with DP and 5 children without DP.

[e] Adaptive behavior data were missing for 31 children with DP and 18 children without DP at Time 3. Another 3 children with DP and 3 children without DP were not seen for a clinic assessment, and only parent-reported adaptive behavior data were available.

TABLE 3 Percentage of Children With DP (Cases) and Without DP (Controls)[a] Who Received Standard Scores <85 on the BSID-III and Adjusted RR[b]

BSID-III domain	Percent Delayed (Standard Score <85)		Adjusted RR		
				95% Confidence Interval	
	Cases (n = 215)	Controls (n = 163)	RR	Lower	Upper
Cognitive[c,d]	3.1	0.0	—	—	—
Language[c]	5.3	1.2	7.9	1.8	35.1
Motor[c]	5.8	1.8	4.3	1.0	17.9
Adaptive[e]	6.2	4.8	1.7	0.7	4.4

[a] Data were dropped for 2 children in the DP group who did not have evidence of DP based on clinician ratings of 3D head photographs, and 68 children in the non-DP group who had mild or greater posterior skull flattening or asymmetry. Data were also dropped for 7 children with DP and 2 children without DP who were diagnosed with medical conditions with developmental implications after enrollment in the study.

[b] Adjusted for child age (in mo), gender, SES (Hollingshead total, measured continuously), and ethnicity (white, non-Hispanic vs nonwhite or Hispanic).

[c] Scores rated as "invalid" by the examiner were dropped from analyses. At Time 3, cognitive scores were rated invalid for 1 child without DP; language scores were rated invalid for 3 children without DP; and motor scores were rated invalid for 1 child with DP and 5 children without DP.

[d] RR was not calculated for cognitive scores given that no children in the unaffected control group scored <85.

[e] Adaptive behavior data were missing for 31 children with DP and 18 children without DP at Time 3. Another 3 children with DP and 3 children without DP were not seen for a clinic assessment, and only parent reported adaptive behavior data were available.

greater after accounting for the estimated effects of developmental interventions. Group differences favoring controls increased by 2.0 to 3.3 standard score points on the BSID-III composites and 0.6 to 0.7 scaled score points on BSID-III subscales.

DISCUSSION

Children with a history of DP continued to score lower than unaffected controls on the BSID-III at age 36 months. Importantly, this observation is based on a sample from which we excluded cases and controls with conditions that elevate the risk of developmental delays (eg, prematurity, sensory impairments). We observed few differences among DP cases as a function of clinical and demographic characteristics. Consistent with the study by Miller and Clarren,[12] a large proportion of children with DP received developmental services in the community. Assuming even a modest treatment effect, use of interventions likely reduced the magnitude of observed group differences, as is suggested by our findings using censored normal regression analyses. Finally, "DP controls" (ie, infants enrolled as healthy controls without previously diagnosed DP, but later found to have skull dysmorphology on 3D imaging) consistently scored lower on the BSID-III than unaffected controls. This suggests that the association between DP and development is not due merely to bias in patients referred to a specialty clinic.

These observations are consistent with comparisons at ages 7 and 18 months in this cohort,[5,10] and they provide the first evidence of a persistent association between DP and development in preschoolers using clinician-administered assessments. Although the strength of the association between DP and development is relatively modest, its clinical relevance is suggested by the consistency of this association over time and the persistence of group differences despite the high proportion of children with DP who received developmental intervention. Compared with those observed during infancy, group differences in gross motor development were diminished, whereas differences persisted or increased in language and cognition. This specificity may reflect the nature of the developmental interventions received because physical and occupational therapy were the most common interventions (46.4% of cases, compared with 4.8% of controls) and may have ameliorated motor deficits.

We know of only 1 other study that tracked the development of children with DP to preschool age,[11] which relied on parent-reported developmental outcomes and test norms for comparison. In that study, 11% of children with DP aged 3 to 5 years scored in the "delayed" range relative to test norms on the Ages and Stages Questionnaire.[19] One risk when relying on test norms for comparison is that research participants may differ from the normative sample in characteristics that are associated with development (eg, SES), potentially leading to an underestimation of the risk of scoring below a clinical cutoff relative to demographically similar peers. In our sample, few children scored in the "at-risk" range compared with the proportion indicated by test norms (see Table 3), likely reflecting the middle to high SES of the families in our sample and the exclusion of children with known neurodevelopmental liabilities. Nonetheless, children with DP in this study were ~2 to 8 times more likely to receive "at-risk" scores than unaffected controls, and we would expect the absolute percentage of delayed children with DP to be higher in samples with broader representation of SES.

Although we cannot rule out DP as the cause of developmental delays, a more parsimonious explanation for the observed case-control differences is the reverse: infants with early developmental risk are more apt to develop DP. There is some evidence for this possibility in population-based studies,[2] and the high prevalence of previously undiagnosed medical conditions that emerged in our sample of children with DP suggests an underlying developmental liability that preceded skull deformation. It may be that DP is the result of the interaction between positioning practices and neurodevelopmental vulnerability. Although the majority of children are now placed in supine position for sleep, only a minority develop DP. Those with developmental concerns, who may be less able to reposition themselves, may be more likely to develop skull deformation. This idea is consistent with data reported by Fowler et al[6] who found that infants with DP were more likely than controls to have abnormal muscle tone on neurologic exams. DP might therefore serve as a physical "marker" of developmental risk, evident before delays are fully manifest and testable, which could be used to identify children who need additional evaluation and intervention.

That the severity of DP in infancy was unrelated to developmental outcomes at Time 3 argues further against the notion that DP causes later developmental delays. Using the model proposed here, it may be that early neurodevelopmental concerns place an infant at risk for some degree of DP, with severity of deformation mediated by parents' positioning practices. Or parents may be better motivated to participate in repositioning and other exercises when their child's skull deformity is severe, with beneficial effects on later development.

Strengths of this study included objective measures of development, 3D imaging of children with and without diagnosed DP to identify skull dysmorphology, the inclusion of demographically similar controls for comparison, and an excellent retention rate. Limitations include recruitment of participants through a specialty craniofacial clinic (DP cases) and via a participant pool (controls), rather than population-based sampling. Children in both samples might differ from the general population in ways that would be difficult to measure but that might nonetheless affect developmental outcomes. Furthermore, the participation rate among children with DP was lower than desired, in part because of the requirement that families be seen for the study ≤4 weeks after the child's diagnosis. However, we did not find evidence of participation bias as a function of demographic (eg, SES) or clinical (eg, DP severity) variables.[10] The rate of developmental intervention might have been elevated by feedback provided to participants as part of this study. We provided all families with feedback regarding their child's BSID-III scores, and when children scored in the "at-risk" range on the BSID-III we recommended follow-up with the child's primary care provider. This likely prompted some families who otherwise would not have received assessment to seek out developmental intervention in the community. However, this should have had a similar effect on families in both groups, and therefore does not account for the disparity between DP cases and controls. Moreover, censored regression analyses suggested that accounting for this source of bias would only magnify the observed differences.

CONCLUSIONS

Developmental differences between children with and without DP persist through age 36 months. Additional study is needed to determine whether these differences continue in school-aged children and to confirm these findings in prospectively studied population-based samples. In the meantime, we encourage clinicians to provide developmental screening and monitoring for infants with DP and to offer early intervention services when warranted.

REFERENCES

1. Argenta LC, David LR, Wilson JA, Bell WO. An increase in infant cranial deformity with supine sleeping position. *J Craniofac Surg*. 1996;7(1):5–11
2. McKinney CM, Cunningham ML, Holt VL, Leroux B, Starr JR. Characteristics of 2733 cases diagnosed with deformational plagiocephaly and changes in risk factors over time. *Cleft Palate Craniofac J*. 2008;45(2):208–216
3. Kane AA, Mitchell LE, Craven KP, Marsh JL. Observations on a recent increase in plagiocephaly without synostosis. *Pediatrics*. 1996;97(6 pt 1):877–885
4. Turk AE, McCarthy JG, Thorne CH, Wisoff JH. The "back to sleep campaign" and deformational plagiocephaly: is there cause for concern? *J Craniofac Surg*. 1996;7(1):12–18
5. Collett BR, Starr JR, Kartin D, et al. Development in toddlers with and without deformational plagiocephaly. *Arch Pediatr Adolesc Med*. 2011;165(7):653–658
6. Fowler EA, Becker DB, Pilgram TK, Noetzel M, Epstein J, Kane AA. Neurologic findings in infants with deformational plagiocephaly. *J Child Neurol*. 2008;23(7):742–747
7. Hutchison BL, Stewart AW, Mitchell EA. Characteristics, head shape measurements and developmental delay in 287 consecutive infants attending a plagiocephaly clinic. *Acta Paediatr*. 2009;98(9):1494–1499
8. Kennedy E, Majnemer A, Farmer JP, Barr RG, Platt RW. Motor development of infants with positional plagiocephaly. *Phys Occup Ther Pediatr*. 2009;29(3):222–235
9. Kordestani RK, Panchal J. Neurodevelopment delays in children with deformational plagiocephaly. *Plast Reconstr Surg*. 2006;118(3):808–809, author reply 809–810
10. Speltz ML, Collett BR, Stott-Miller M, et al. Case-control study of neurodevelopment in deformational plagiocephaly. *Pediatrics*. 2010;125(3). Available at: www.pediatrics.org/cgi/content/full/125/3/e537
11. Hutchison BL, Stewart AW, Mitchell EA. Deformational plagiocephaly: a follow-up of head shape, parental concern and neurodevelopment at ages 3 and 4 years. *Arch Dis Child*. 2011;96(1):85–90
12. Miller RI, Clarren SK. Long-term developmental outcomes in patients with deformational plagiocephaly. *Pediatrics*. 2000;105(2). Available at: www.pediatrics.org/cgi/content/full/105/2/E26
13. Steinbok P, Lam D, Singh S, Mortenson PA, Singhal A. Long-term outcome of infants with positional occipital plagiocephaly. *Childs Nerv Syst*. 2007;23(11):1275–1283
14. Bayley N. *Manual for the Bayley Scales of Infant and Toddler Development*. 3rd ed. San Antonio, TX: The Psychological Corporation; 2006
15. Leppig KA, Werler MM, Cann CI, Cook CA, Holmes LB. Predictive value of minor anomalies. I. Association with major malformations. *J Pediatr*. 1987;110(4):531–537
16. Hollingshead AB. *Four Factor Index of Social Status*. New Haven, CT: Yale University; 1975
17. Tobin MD, Sheehan NA, Scurrah KJ, Burton PR. Adjusting for treatment effects in studies of quantitative traits: antihypertensive therapy and systolic blood pressure. *Stat Med*. 2005;24(19):2911–2935
18. StataCorp. Stata Statistical Software; Release 12. College Station, TX: StataCorp LP
19. Squires J, Twombly E, Bricker D, Potter R. *Behavior—Ages & Stages Questionnaires*. 3rd ed. Eugene, OR: University of Oregon, Center on Human Development; 2009

(Continued from first page)

www.pediatrics.org/cgi/doi/10.1542/peds.2012-1779

doi:10.1542/peds.2012-1779

Accepted for publication Sep 17, 2012

Address correspondence to Brent R. Collett, PhD, Seattle Children's Research Institute, 2001 8th Ave, Suite 600, Mailstop CW8-6, Seattle, WA 98121. E-mail: bcollett@u.washington.edu

PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

FINANCIAL DISCLOSURE: *The authors have indicated they have no financial relationships relevant to this article to disclose.*

FUNDING: This publication was made possible in part by grant 1 R01 HD046565 from the National Institute of Child Health and Human Development (NICHHD) to Dr Speltz and grant 1 UL1 RR025014 from the National Center for Research Resources (NCRR), components of the National Institutes of Health (NIH). Its contents are solely the responsibility of the authors and do not necessarily represent the official view of NICHHD, NCRR, or NIH. Funded by the National Institutes of Health (NIH).

Development at Age 36 Months in Children With Deformational Plagiocephaly

Brent R. Collett, Kristen E. Gray, Jacqueline R. Starr, Carrie L. Heike, Michael L. Cunningham and Matthew L. Speltz

Pediatrics 2013;131;e109; originally published online December 24, 2012;
DOI: 10.1542/peds.2012-1779

PEDIATRICS®
OFFICIAL JOURNAL OF THE AMERICAN ACADEMY OF PEDIATRICS

Development at Age 36 Months in Children With Deformational Plagiocephaly

Brent R. Collett, Kristen E. Gray, Jacqueline R. Starr, Carrie L. Heike, Michael L. Cunningham and Matthew L. Speltz

Pediatrics 2013;131;e109; originally published online December 24, 2012;
DOI: 10.1542/peds.2012-1779