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

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