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LONGITUDINAL, THREE-DIMENSIONAL ANALYSIS OF HEAD SHAPE IN CHILDREN WITH AND WITHOUT DEFORMATIONAL PLAGIOCEPHALY OR BRACHYCEPHALY

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Abstract

Objectives—To assess three-dimensional (3D) changes in head shape in infancy and at age 18 months in children with and without plagiocephaly or brachycephaly.

Study design—Using a longitudinal design, we evaluated head shape using 3D surface imaging. We compared the head shapes of children with (1) diagnosed deformational plagiocephaly or brachycephaly (cases; n=233); (2) unaffected controls, with no evidence of dysmorphology (n=167); and (3) affected controls, who despite having no previous diagnosis demonstrated skull dysmorphology on 3D surface imaging (n=70).

Results—Cases had greater skull flattening and asymmetry than unaffected controls at both time points, as did controls with skull dysmorphology. In all groups, head shapes became less flat and more symmetric over time. Among cases, symmetry improved slightly more for those who received orthotic treatment.

Conclusions—Although head shape improves over time for children with deformational plagiocephaly or brachycephaly, skull dysmorphology persists relative to unaffected controls. Further research is needed to clarify the extent to which these differences are detectable to clinicians and lay observers.

Keywords

dysmorphology; infant; toddler; skull

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The increased prevalence of posterior skull flattening and asymmetry among infants in the United States and other countries is well-documented.¹⁻⁴ Skull deformation most commonly results in plagiocephaly (“oblique head”⁵) or brachycephaly (“short head,” or a decreased skull length:width ratio⁵). Infants can experience either form of deformation in isolation, or they can exhibit a combination of plagiocephaly and brachycephaly.⁶

Despite the prevalence of these conditions, little is known about the natural course of plagiocephaly or brachycephaly, particularly in the post-infancy period. The few existing studies suggest that head shape “normalizes,” even without treatment or with repositioning interventions alone.⁶⁻¹¹ For example, in a study of children with brachycephaly or plagiocephaly in the first year of life, Hutchison et al.⁹ found that by ages 3-5 years, 60% no longer had these conditions based on objective shape measurements. However, this study did not include controls for comparison, and it is unclear whether the cut-off scores used in infancy are suitable for older children. Further, most studies have relied on two-dimensional (2D) imaging or clinical impressions, which may be insensitive measures of some aspects of abnormalities in head shape.

We assessed changes in head shape in infants with and without deformational plagiocephaly and/or brachycephaly over an interval of approximately 12 months by using previously established three-dimensional (3D) imaging procedures.¹² We sought to determine whether the group differences in head shape observed in infancy persisted at age 18 months, and we explored whether results differed for cases with isolated brachycephaly (i.e., without asymmetry), orthotic helmet use, and history of torticollis.

METHODS

Participants were enrolled after informed consent was obtained using procedures approved by the Institutional Review Board at Seattle Children’s Hospital and in compliance with Health Insurance Portability and Accountability Act (HIPAA) standards.

Infants with diagnosed plagiocephaly and/or brachycephaly (cases) were recruited at the time of diagnosis in the Seattle Children’s Hospital Craniofacial Center. Infants were eligible for participation in this group if they had been diagnosed with plagiocephaly or brachycephaly by a craniofacial specialist and were between the ages of 4-11 months. Exclusions were: (1) history of prematurity (< 35 weeks gestation), (2) a diagnosed neurodevelopmental condition (e.g., Down syndrome), brain injury, or significant hearing or vision impairment; (3) presence of a major malformation or ≥ 3 minor malformations;¹³ (4) diagnosis of craniofacial microsomia; (5) a non-English speaking mother; (6) adoption or out-of-home placement; and (7) family plans to move out of state before project completion. Between June 2006 and February 2009, we recruited 235 infants with plagiocephaly or brachycephaly, representing 52% of all eligible case subjects. Participants were similar to non-participants with regard to demographic characteristics and severity of cranial deformation.¹⁴

The first 8 infants without diagnosed plagiocephaly or brachycephaly (controls) enrolled in the study were identified through pediatric practices. Remaining controls were identified from a participant pool consisting of families in King and Snohomish counties in Washington State who agreed to be contacted for research participation when their child was born. By phone, we contacted families with a child in the target age range and screened interested parents for eligibility. In addition to the exclusions listed for cases, control infants were excluded if they had been diagnosed with plagiocephaly, brachycephaly, or any other craniofacial condition. We selected controls who were similar to cases with regard to the distribution of infants’ age and sex, and family socioeconomic status (SES) and ethnicity.

Two hundred thirty-seven control infants were recruited between June 2007 and February 2009, representing 90% of those screened and determined eligible.

3D cranial images were de-identified, randomly sorted, and rated by 2 dysmorphologists (MC and CH) who were unaware of case status. Raters documented the degree of skull flattening, asymmetry, and brachycephaly using previously published 4-point severity scales (Cranial Technologies Inc. 2002 Rev 01®). Raters then used this information to rate the overall severity of cranial deformation 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 plagiocephaly or brachycephaly using the overall severity of cranial deformation scale) was excellent ($\kappa = 0.80$), and exact agreement for each of the 4 severity categories was adequate (weighted $\kappa = 0.72$). The mean of the two raters' scores was used to represent the severity of each participant's cranial deformation. Cases were included in group comparisons if they had at least mild cranial deformation detected by one of the raters (rating ≥ 0.5) and controls were included if no plagiocephaly or brachycephaly was detected (rating = 0). Controls without previously diagnosed deformation, who nonetheless had ratings indicating some degree of skull dysmorphology, were retained in a separate group.

We used these ratings to form three groups: *Cases* (clinical diagnosis, confirmed with expert ratings), *Unaffected Controls* (no previous diagnosis, no evidence of skull dysmorphology with expert ratings), and *Affected Controls* (no previous diagnosis, though detectable skull dysmorphology based on expert ratings of 3D images).

Study Procedures

Children were first assessed in infancy (Time 1). For cases, the Time 1 visit was scheduled within 4 weeks of diagnosis. We aimed to schedule participants' Time 2 visit within 2 weeks of the child's 18-month 'birthday' and set an upper age limit of 30 months. 3D cranial images were obtained for all participants by using the 12-camera 3dMDcranial™ active stereo photogrammetry system.

Interviews were completed with mothers at Time 1 to collect data on the child's medical history and demographic characteristics, including family SES and ethnicity. Interviewers also asked mothers if their infants had suspected or diagnosed torticollis. An abbreviated interview was completed at Time 2 to document whether participants had received orthotic helmet or band treatment. Mothers whose child received orthotic treatment a helmet or band were asked "How well do you think you followed through with suggestions for [child's name]'s helmet treatment for his/her plagiocephaly?" rated on a 5-point scale (very poor, poor, adequate, good, very good).

Image analysis

Procedures for generating automated head shape measures are described in detail in Atmosukarto et al.¹² In brief, measures of posterior cranial flattening and asymmetry were derived by first constructing 2D histograms of azimuth-elevation angles of 3D surface normal vectors. The value of each histogram bin is the percentage of surface normal vectors with a particular azimuth-elevation angle combination. Because the surface normal vectors of points that lie on a flat surface are almost parallel, they will have similar azimuth-elevation angles, leading to high valued bins or peaks in the 2D histogram (Figure, B; available at www.jpeds.com).

We used values from the bins representing similar azimuth-elevation angle to generate left and right posterior flatness scores (LPFS and RPFS, respectively). We selected the greater of the LPFS and RPFS to determine the PF score (Figure). We generated an Absolute

Asymmetry Score (AAS), representing the absolute difference between the LPFS and RPFS (Figure).

We generated a score that approximates the Oblique Cranial Length Ratio (aOCLR).¹⁵ We used a top view 2D snapshot of the 3D head shape and measured the cross-diagonal length of the head contour to calculate the ratio of the longer to the shorter length (ie, skull asymmetry).

We estimated brachycephaly using the Cephalic Index (CI) by measuring the width of the head at the widest point divided by the length, using the same 2D snapshots used for the aOCLR.

To aid interpretation, we converted participants' scores to standardized z-scores. Separately for each head shape measure, we subtracted the mean for unaffected controls at Time 1 from each participant's score and divided by standard deviation for the unaffected controls at Time 1. By definition, this resulted in a mean of 0 and standard deviation of 1 for unaffected controls on all measures at Time 1. We repeated this at Time 2, again standardizing each participant's scores using the group mean and standard deviations for unaffected controls at Time 1. On all measures, higher scores reflect greater asymmetry or flattening.

Data Analyses

We summarized the clinical and demographic characteristics of participants by calculating means, standard deviations, or frequencies separately for cases and controls. To examine potential attrition bias, we compared Time 2 participants to Time 2 non-participants (i.e., those who were lost to follow-up or withdrew from the study) with respect to their demographic characteristics and Time 1 measures of skull deformation.

We calculated descriptive statistics, including means, standard deviations, and percentile scores on all head shape measures separately for all cases, cases with isolated brachycephaly, unaffected controls, and affected controls. We estimated case-control group differences and 95% confidence intervals at each time point by fitting linear regression models for z-scores on each of the four head shape indices, adjusted for children's age (in months), sex, race/ethnicity (White and Non-Hispanic versus Non-White or Hispanic), and family SES (continuous scores from the Hollingshead composite¹⁵). These comparisons were made for cases versus unaffected controls, and for affected controls versus unaffected controls. We also examined outcomes separately for cases with "isolated brachycephaly" versus unaffected controls. Based on Time 1 data, and using previously established clinical thresholds,^{6,12} cases with isolated brachycephaly were defined as having a CI above the clinical threshold and AAS and aOCLR scores below clinical thresholds.

To examine changes in head shape over time, we used a generalized estimating equations (GEE) approach to fit linear regression models, with each of the four head shape indices as a dependent variable and visit (1 or 2) as the independent variable. We performed these analyses separately for cases, unaffected controls, and affected controls. Again, we repeated these analyses for cases with isolated brachycephaly.

Among cases only, we used GEE analyses to evaluate the interaction between time and orthotic helmet treatment. Outcome variables included the AAS, PF, CI, and aOCLR scores. Orthotic treatment was used as the independent variable, after excluding children whose parents reported poor or very poor treatment compliance. Finally, we used GEE analyses to examine the interaction between time and the presence of suspected or confirmed torticollis in infancy among cases. Suspected or confirmed torticollis was used as the independent variable. These analyses controlled for baseline head shape, using the AAS, PF, aOCLR,

and CI scores at Time 1, and for demographic characteristics (i.e., age at Time 1, sex, socioeconomic status, ethnicity).

To avoid missing potentially important associations and group differences in this exploratory study (i.e., Type II errors), we did not adjust p-values for multiple comparisons. Rather than viewing p-values as dichotomous outcomes (i.e., statistically ‘significant’ versus ‘non-significant’), we interpreted adjusted group differences and p-values as estimates of the magnitude and precision of group differences.^{17,18}

We completed all analyses using the STATA SE 10.0 software package.¹⁹

RESULTS

Children in the case and control groups were predominately male, of white race, and of middle to upper SES (Table I). At Time 1, the mean age for cases was 7.2 months (SD = 1.6) and for controls was 6.8 months (SD = 1.7). At Time 2, the average age in both groups was approximately 18.5 months old. Cases were more likely than controls to have torticollis, to be a twin, and to have received care in the NICU. Between Time 1 and Time 2, 79 cases (35%) received orthotic helmet therapy and 1 case (0.4%) received orthotic band therapy. Based on clinicians’ reviews of subjects’ 3D images at Time 1, there were 2 cases (0.9%) with no discernible deformation, 103 (45.4%) with “mild” skull dysmorphology, and 122 (53.7%) with “moderate” or “severe” dysmorphology. One hundred sixty-seven (70.5%) controls had no discernible deformation, 68 (28.7%) had “mild” dysmorphology, and 2 (0.8%) had “moderate” to “severe” skull dysmorphology. 3D images were available for all 235 cases and 237 controls at Time 1. Clinician’s ratings were moderately correlated with Time 1 computer-based measures ($r = 0.52$ to 0.68).

Nearly all cases and controls returned for a Time 2 visit (95% cases and 97% controls). Another 4 cases and 3 controls could not be seen for 3D imaging but completed an update interview by phone or in a home visit. Compared with families followed at Time 2, families who were lost to follow-up had lower average SES and were more likely to be non-White or Hispanic, though they were similar in their average severity of deformation at Time 1.

Group Differences at Time 1 and 2

Descriptive statistics for head shape measures at Time 1 and Time 2 are presented in Table II_[MH1] for all cases, cases with isolated brachycephaly, affected controls, and unaffected controls. Cases and unaffected controls differed on all head shape measures at Time 1, with adjusted differences ranging from 1.19 to 3.21 (all p-values < 0.001) (Table III). At Time 2, these differences were slightly reduced, with adjusted differences of 1.18 to 2.32 (all p-values < 0.001). Cases with isolated brachycephaly scored higher on average than unaffected controls on the PF and CI at Time 1 (adjusted differences = 1.32 to 3.77; p-values < 0.001) (Table III), but did not score appreciably differently on measures of asymmetry. These comparisons yielded similar results at Time 2, except that cases with isolated brachycephaly also had slightly higher scores on the AAS. Compared with unaffected controls, affected controls scored higher on average on all measures at both Time 1 (adjusted differences = 0.43 to 1.20; p-values < 0.001 to 0.183) and Time 2 (adjusted differences = 0.28 to 1.13; p-values < 0.001 to 0.028).

Within-group Changes in Deformation over Time

On average, for all groups, there was a trend for head shape to become less flat, less brachycephalic, and more symmetric over time. Time 1-Time 2 differences across the various measures for cases ranged from -0.71 to -1.67 (all p-values ≤ 0.001). In cases with isolated brachycephaly, differences over time were limited to the CI and were minimal on

other indices. For unaffected controls, changes ranged from -0.19 to -0.43 points (p -values < 0.001 to 0.052). Affected controls showed inconsistent change across measures, with (1) reduced posterior asymmetry based on the AAS, but not the aOCLR (differences = -0.45 and -0.31 , respectively; p -values = 0.026 and 0.319 , respectively), (2) reduced brachycephaly based on CI (difference = -0.53 ; $p < 0.001$), and (3) minimal reduction in PF scores (difference = -0.17 ; $p = 0.299$).

Head Shape as a Function of Helmet Use and Torticollis

Of the 79 children treated with orthotic helmets, 71 parents (89.9%) reported 'adequate' or better compliance. Change over time in PF and CI scores differed little for children who underwent helmet treatment versus those who did not (interaction coefficients = -0.08 and 0.03 , respectively). Changes in AAS and aOCLR scores were larger for those in the helmet group (interaction coefficients = -0.92 and -0.47 , respectively; p -values = 0.004 and < 0.001 , respectively). Shape index changes over time differed little for children with versus without a history of torticollis (interaction coefficients = -0.32 to 0.11). On the PF, children with torticollis showed slightly larger improvements than those without torticollis (interaction coefficient = -0.43).

DISCUSSION

This study followed 3 groups of children recruited in infancy: (1) cases: infants diagnosed with plagiocephaly and/or brachycephaly in a specialty clinic, confirmed by experts "blind" to case status; (2) unaffected controls: infants drawn from a research participant pool without skull dysmorphology on 3D images rated by the same experts; and (3) affected controls: participants from the same pool but with previously undiagnosed (and mostly mild) skull dysmorphology that was detectable on 3D images by experts. Consistent with previous studies,⁶⁻¹¹ head shape became increasingly rounded and symmetric with time. However, cases differed from unaffected controls on head shape measures taken in infancy and again at age 18 months. Similarly, affected controls differed from unaffected children at both time points. As might be expected, children with isolated brachycephaly differed from unaffected controls on measures of posterior flattening and brachycephaly at both time points, and shared similar scores for measures of asymmetry.

Among cases, we examined the association between head shape, torticollis, and the child's participation in orthotic treatment on head shape. Torticollis has a relatively high rate of co-occurrence with plagiocephaly, and restricted neck motion may increase the probability of persistent skull deformation. However, we observed only minor differences in skull dysmorphology between infants with and without torticollis.

Orthotic helmets and bands are common treatments for deformational plagiocephaly,²⁰ yet there have been few methodologically rigorous studies of their effect on head shape,²¹ and follow-up in these studies has been mostly short-term. Consistent with a recent study by Lipira et al.,¹¹ we observed greater improvements in skull symmetry among infants receiving orthotic treatment than among cases without such treatment. We additionally measured skull flatness and brachycephaly, and differences in both aspects were only minor. Based on our measures, these observations suggest that these molding treatments improve only some dimensions of plagiocephaly by age 18 months.

The latter caveat is important as shape is a multi-dimensional construct, ultimately requiring not only automated, objective measurements like those used in this study, but also subjective analyses, including judgments by trained individuals (e.g., dysmorphologists or clinicians) and judgments by parents and other lay adult caregivers. The present findings speak only to specific objective aspects of shape measurement. Facial appearance may be influenced more

by overall skull symmetry than by isolated posterior skull flattening, and this may be important for lay perceptions of change associated with orthotic treatment. Studies involving such subjective evaluations are critical, as clinicians and parents currently have little empiric information upon which to make treatment recommendations or decisions. Furthermore, there has been no formal risk-benefit analysis to aid in such decisions; and these treatments typically are not covered by health insurance and can be costly for families. In the meantime, internet sites continue to claim that long-term cosmetic disadvantage is likely among untreated children with plagiocephaly or brachycephaly despite the virtual absence of rigorous and systematically collected data to support these assertions.

Our study has limitations. Future studies would benefit from larger groups of treated children and use of more detailed, “real-time” reporting of daily helmet use. Similarly, infants’ history of torticollis was assessed only by parents’ interview reports rather than more direct assessment. This may have increased measurement error and reduced the study’s sensitivity to detect associations between time-related head shape changes and torticollis. Finally, the shape descriptors we used in this study were developed in infants, by using 3D skull images of young infants with little scalp hair. At age 18 months, we used snug-fitting wig caps to minimize the effects of hair and to allow for better assessment of skull shape. The wig caps may also introduce measurement error. Such error, if existent, should affect cases and controls similarly and would likely influence group comparisons by biasing the estimate towards the null.

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ABBREVIATIONS

(3D)	Three dimensional
(2D)	Two dimensional
(SES)	Socioeconomic Status
(PF)	Posterior Flatness
(LPFS, RPFS)	Left and Right Posterior Flatness Scores
(AAS)	Absolute Asymmetry Score
(aOCLR)	Approximate Oblique Cranial Length Ratio
(CI)	Cephalic Index
(GEE)	Generalized Estimating Equations

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Figure.

A_[MH2], 3D mesh images of the head viewed from the top, arrow indicating the forehead. Illustration of the average mesh image for severity scores 0='none', 1='mild', 2='moderate', 3='severe' from expert clinician ratings. B, Surface normal vectors of points that lie on a rounded surface have a wider distribution of angles as illustrated for the head on the extreme left, and those that lie on a flat surface tend to have similar azimuth and elevation angles as illustrated for the head on the extreme right. C, Posterior flattening and average (standard deviation) z-scores for the PF, AAS, CI and aOCLR for the 3D mesh images above, now viewed from the back. Posterior flattening is depicted using a color map where high score values are represented by warm colors (red, orange, yellow), and low score values correspond to cool colors (blue, cyan, green). A representative unaffected control participant is shown at the extreme left. All index scores for this infant are near 0, shown by the cool coloring at the posterior. A case infant is shown at the extreme right, with elevated index scores and flattening severity depicted by red coloring on the affected side.

Table 1

Demographic and clinical characteristics for children diagnosed with plagiocephaly or brachycephaly (cases) and children without previously diagnosed plagiocephaly or brachycephaly (controls).

	All Cases (n=235)	Isolated Brachycephaly (n=21)	Controls (n=237)
Age in months at Time 1 (mean, standard deviation)	7.2 (1.6)	7.6 (1.9)	6.8 (1.7)
Age in months at Time 2 (mean, standard deviation)	18.5 (0.8)	18.8 (0.9)	18.5 (0.9)
Sex			
Male	153 (65%)	14 (67%)	140 (59%)
Female	82 (35%)	7 (33%)	97 (41%)
Race/Ethnicity			
White	159 (68%)	13 (62%)	145 (61%)
Asian/Pacific Islander	14 (6%)	1 (5%)	13 (5%)
Black	0 (-)	0 (-)	6 (3%)
Hispanic	28 (12%)	5 (24%)	30 (13%)
Mixed Race/Other	34 (14%)	2 (10%)	43 (18%)
Socioeconomic Status ^a			
I (high)	83 (35%)	7 (33%)	61 (27%)
II	90 (37%)	8 (38%)	100 (42%)
III	37 (17%)	4 (19%)	49 (20%)
IV	18 (7%)	1 (5%)	21 (9%)
V (low)	7 (4%)	1 (5%)	6 (3%)
Clinical Characteristics ^b			
Torticollis ^c	100 (43%)	3 (14%)	5 (2%)
Twin	27 (12%)	1 (5%)	2 (1%)
NICU	27 (12%)	1 (5%)	12 (5%)

^a Categorized using the Hollingshead four factor scoring system

^b Reflects the number and proportion of children with each characteristic

^c Includes suspected and confirmed torticollis

Table 2

Time 1 and Time 2 descriptive statistics for head shape measures (expressed as z-scores^a) among: children with diagnosed plagiocephaly or brachycephaly, confirmed on 3D imaging (all cases); children with brachycephaly, but not plagiocephaly (isolated brachycephaly); children without diagnosed plagiocephaly or brachycephaly, with some dysmorphology detected on 3D imaging (affected controls); and children without previously diagnosed plagiocephaly or brachycephaly and confirmed absence of cranial dysmorphology on 3D imaging (unaffected controls).

	Time 1			
	All Cases n=233	Isolated Brachycephaly Cases n=21	Affected Controls n=70	Unaffected Controls n=167
Posterior Flatness (PF)				
Mean (SD)	2.15 (1.43)	1.29 (0.96)	0.47 (1.20)	<i>Ref</i>
5 th percentile	-0.09	-0.43	-1.68	-1.63
25 th percentile	1.15	0.68	-0.34	-0.66
50 th percentile	2.11	1.32	0.42	-0.08
75 th percentile	2.96	1.92	1.13	0.68
95 th percentile	4.50	2.52	2.54	1.49
Absolute Asymmetry Score (AAS)				
Mean (SD)	3.19 (2.17)	0.31 (0.93)	0.56 (1.47)	<i>Ref</i>
5 th percentile	-0.32	-1.01	-1.18	-1.17
25 th percentile	1.70	-0.32	-0.49	-0.70
50 th percentile	3.13	-0.05	0.27	-0.23
75 th percentile	4.61	1.18	1.61	0.46
95 th percentile	6.84	1.51	3.07	1.92
Oblique Cranial Length Ratio (aOCLR)				
Mean (SD)	1.17 (1.22)	-0.08 (0.40)	0.35 (2.35)	<i>Ref</i>
5 th percentile	-0.41	-0.61	-0.59	-0.68
25 th percentile	0.40	-0.41	-0.29	-0.49
50 th percentile	1.01	-0.09	-0.02	-0.28
75 th percentile	1.74	0.15	0.42	0.25
95 th percentile	3.09	0.56	1.69	1.15
Cephalic Index (CI)				
Mean (SD)	2.48 (1.46)	3.66 (0.80)	1.19 (1.33)	<i>Ref</i>
5 th percentile	0.16	2.71	-1.02	-1.69
25 th percentile	1.40	2.88	0.14	-0.69
50 th percentile	2.52	3.81	2.25	0.06
75 th percentile	3.62	4.13	2.10	0.71
95 th percentile	4.79	4.96	3.34	1.69

Time 2

	Time 1			
	All Cases n=233	Isolated Brachycephaly Cases n=21	Affected Controls n=70	Unaffected Controls n=167
	All Cases n=221	Isolated Brachycephaly Cases n=20	Affected Controls n=68	Unaffected Controls n=161
Posterior Flatness (PF)				
Mean (SD)	1.36 (1.12)	1.0 (1.30)	0.32 (0.95)	-0.30 (0.94)
5 th percentile	-0.42	-1.64	-0.83	-1.68
25 th percentile	0.72	0.22	-0.46	-0.80
50 th percentile	1.33	0.98	0.23	-0.41
75 th percentile	2.07	1.99	0.69	0.25
95 th percentile	3.31	2.95	2.24	1.26
Absolute Asymmetry Score (AAS)				
Mean (SD)	1.52 (1.62)	0.40 (1.35)	0.12 (1.12)	-0.19 (0.85)
5 th percentile	-0.83	-0.87	-1.11	-1.14
25 th percentile	0.20	-0.65	-0.61	-0.87
50 th percentile	1.44	-0.15	-0.10	-0.33
75 th percentile	2.46	1.37	0.47	0.31
95 th percentile	4.15	2.97	2.22	1.30
Oblique Cranial Length Ratio (aOCLR) ^b				
Mean (SD)	0.94 (0.84)	-0.19 (0.41)	0.05 (0.98)	-0.23 (0.35)
5 th percentile	-0.49	-0.69	-0.57	-0.62
25 th percentile	0.37	-0.50	-0.39	-0.49
50 th percentile	0.92	-0.14	-0.15	-0.28
75 th percentile	1.51	-0.01	0.26	-0.04
95 th percentile	2.38	0.71	0.73	0.39
Cephalic Index (CI)				
Mean (SD)	1.76 (1.31)	2.32 (0.73)	0.65 (1.27)	-0.46 (0.88)
5 th percentile	-0.13	1.18	-1.02	-1.76
25 th percentile	0.86	1.88	-0.43	-1.10
50 th percentile	1.68	2.26	0.62	-0.43
75 th percentile	2.53	2.80	1.37	0.09
95 th percentile	3.97	3.66	3.02	1.00

^aScores are expressed in z-score units (mean = 0, standard deviation = 1) using Time 1 scores for unaffected controls as the reference group (e.g., z = observed score - Time 1 unaffected control group mean / Time 1 unaffected control group standard deviation).

^bThe aOCLR could not be used for 1 case at Time 2 due to poor 3D photo quality. This did not affect other measures.

Table 3

Adjusted z-score^a differences in head shape at Time 1 and Time 2 among: children with diagnosed plagiocephaly or brachycephaly, confirmed on 3D imaging (all cases); children with brachycephaly, but not plagiocephaly (isolated brachycephaly); children without diagnosed plagiocephaly or brachycephaly, with some dysmorphology detected on 3D imaging (affected controls); and children without previously diagnosed plagiocephaly or brachycephaly and confirmed absence of cranial dysmorphology on 3D imaging (unaffected controls).

Time 1						
Measure	All Cases vs. Unaffected Controls		Isolated Brachycephaly vs. Unaffected Controls		Affected Controls vs. Unaffected Controls	
	Adj. Diff ^b (95% CI)	p-value	Adj. Diff ^b (95% CI)	p-value	Adj. Diff ^b (95% CI)	p-value
Posterior Flatness (PF)	2.19 (1.95, 2.43)	<0.001	1.32 (0.89, 1.75)	<0.001	0.43 (0.11, 0.75)	0.009
Absolute Asymmetry Score (AAS)	3.21 (2.89, 3.53)	<0.001	0.29 (-0.15, 0.73)	0.194	0.58 (0.21, 0.96)	0.002
Oblique Cranial Length Ratio (aOCLR)	1.19 (0.98, 1.40)	<0.001	-0.06 (-0.30, 0.18)	0.615	0.45 (-0.22, 1.12)	0.183
Cephalic Index (CI)	2.59 (2.35, 2.83)	<0.001	3.77 (3.41, 4.13)	<0.001	1.20 (0.85, 1.55)	<0.001
Time 2						
Measure	All Cases vs. Unaffected Controls		Isolated Brachycephaly vs. Unaffected Controls		Affected Controls vs. Unaffected Controls	
	Adj. Diff ^b (95% CI)	p-value	Adj. Diff ^b (95% CI)	p-value	Adj. Diff ^b (95% CI)	p-value
Posterior Flatness (PF)	1.73 (1.52, 1.93)	<0.001	1.38 (0.79, 1.96)	<0.001	0.68 (0.42, 0.95)	<0.001
Absolute Asymmetry Score (AAS)	1.75 (1.49, 2.01)	<0.001	0.66 (0.06, 1.25)	0.032	0.35 (0.04, 0.67)	0.028
Oblique Cranial Length Ratio (aOCLR) ^c	1.18 (1.05, 1.31)	<0.001	0.06 (-0.13, 0.24)	0.547	0.28 (0.05, 0.51)	0.017
Cephalic Index (CI)	2.32 (2.09, 2.55)	<0.001	2.83 (2.46, 3.19)	<0.001	1.13 (0.80, 1.46)	0<0.001

^a Differences are expressed in z-score units (mean = 0, standard deviation = 1) using scores for unaffected controls at Time 1 as the reference group (e.g., z = observed score - Time 1 unaffected control group mean / Time 1 unaffected control group standard deviation).

^b Adjusted for race/ethnicity, gender, age (months), and socioeconomic status (scored as a continuous variable, using the Hollingshead composite score).

^c The aOCLR could not be used for 1 case at Time 2 due to poor 3D photo quality. This did not affect other measures.