Deformational brachycephaly: the clinical utility of the cranial index

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OBJECTIVE The incidence of deformational brachycephaly has risen since the “Back to Sleep” movement in 1992 by the American Academy of Pediatrics. Brachycephaly prevalence and understanding the dynamic nature of the pediatric skull have not been explored in relation to the cranial index (CI). The objective of the study was to determine the prevalence of brachycephaly, via the CI, with respect to time.

METHODS The authors conducted a retrospective review of 1499 patients ≤ 19 years of age who presented for trauma evaluation with a negative CT scan for trauma (absence of bleed) in 2018. The CI was calculated using CT at the lateral-most point of the parietal bone (cephalic width), and the distance from the glabella to the opisthocranium (cephalic length). Brachycephaly was defined as a CI ≥ 90%.

RESULTS The mean CI was 82.6, with an average patient age of 6.8 years. The prevalence of deformational brachycephaly steadily decreased from 27% to 4% from birth to > 2 years of life. The mean CI was statistically different between ages < 12 months, 12–24 months, and > 24 months ($F[2,1496] = 124.058$, $p < 0.0005$). A simple linear regression was calculated to predict the CI based on age; the CI was found to decrease by 0.038 each month. A significant regression equation was found ($F[1,1497] = 296.846$, $p < 0.0005$), with an R^2 of 0.140.

CONCLUSIONS The incidence of deformational brachycephaly is common in infants but decreases as the child progresses through early childhood. Clinicians can expect a significant decrease in mean CI at 12 and 24 months. Additionally, these regression models show that clinicians can expect continued improvement throughout childhood.

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KEYWORDS brachycephaly; cranial index; “Back to Sleep”; craniofacial

PREDOMINANTLY occurring in infants, brachycephaly is defined as symmetric flattening of the occiput typically caused by an external force pushing on the skull.¹ As a result, compensatory parietal widening occurs, thus creating a disproportionately short and wide head shape.^{1,2} The rise in the prevalence of brachycephaly and other deformational skull conditions can be a consequence of infant sleep positions.³ Specifically, the incidence of brachycephaly has increased since the American Academy of Pediatrics (AAP) initiated the “Back to Sleep” movement in 1992, recommending children sleep supine to reduce the risk of sudden infant death syndrome.^{3–5}

Since the inception of this campaign, it has been difficult to determine the prevalence of brachycephaly within the population.

The cranial, or cephalic, index (CI) is a metric designed to measure the effectiveness of cranial deformation correction methods in children, such as helmet use for brachycephaly.⁶ The CI is calculated by the following formula: (cephalic width/cephalic length) × 100.⁶ Cephalic width is the measurement from the most lateral point on either side of the skull, and cephalic length is the measurement between the most anterior and posterior points of the skull.⁷ The CI definition of brachycephaly is not standardized.

ABBREVIATIONS AAP = American Academy of Pediatrics; CI = cranial index; MCJCHV = Monroe Carell Jr. Children's Hospital at Vanderbilt.

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FIG. 1. Schematic representation of CI measurement. Figure is available in color online only.

The cutoff CI used to define brachycephaly has been a contested point in the literature, with a range of 81% to 95%;^{4–6} it is evident that there is no established guideline within the literature base.

Thus far, brachycephaly prevalence and understanding the dynamic nature of the pediatric skull have not been explored in relation to CI. This study measured CI in 1499 children who presented to Monroe Carell Jr. Children's

Hospital at Vanderbilt (MCJCHV), to gain insight into the condition's prevalence and progression. The authors investigated differences in CI with respect to age groups, and specifically within the first 2 years of life. We hypothesized that brachycephaly prevalence decreases with age, and that there is a natural correction of the CI with respect to time. This understanding could provide physicians with greater insight into the natural progression of brachycephaly, and more accurately inform families about resolution of skull asymmetry.

Methods

A cross-sectional review was conducted of pediatric patients evaluated for head trauma with a negative CT scan at MCJCHV between January 1, 2018, and December 31, 2018. The inclusion of study participants was based on the following criteria: 1) patients were ≤ 19 years of age at the time of CT; 2) patients were evaluated at MCJCHV; and 3) CT results were negative for posttraumatic focal or diffuse lesions at initial presentation. Exclusion of study participants was based on the following criteria: 1) history of shunted hydrocephalus; 2) radiographic presence of cranial trauma; or 3) history of craniosynostosis or skull surgery. After applying these criteria, 1499 patients were included in our retrospective review.

Patient data were retrospectively reviewed through electronic medical records at MCJCHV. The CI was calculated by measuring the cephalic width as the lateral-most point of the parietal bone, and the cephalic length as the distance from the glabella to the opisthocranium (Figs. 1 and 2). Patients were then grouped into three cohorts to determine changes in CI: < 12 months, 12–24 months, and > 24 months. While CI ranges for brachycephaly in the literature are between 81% and 95%, our study assessed prevalence using a cutoff of $\geq 90\%$.

Statistical analysis was performed using IBM SPSS Statistics software (version 23.0, IBM Corp.). ANOVA was used to compare CI between the age groups. A scatterplot with linear regression was performed and graphed to evaluate the relationship between age in months and CI.

FIG. 2. CT image representation of CI measurement. Figure is available in color online only.

TABLE 1. Patient demographics

Variable	Value (%)
Sex	
Male	851 (56.8)
Female	648 (43.2)
Race	
White/Caucasian	1007 (67.2)
Black/African American	248 (16.5)
Asian/Indian	24 (1.6)
American Indian or Alaska Native	6 (0.4)
Other/decline to answer	214 (14.3)
Ethnicity	
Hispanic/Latin American	138 (9.2)
Not Hispanic/Latin American	1181 (78.8)
Unknown or decline to answer	180 (12.0)

A Pearson chi-square test was used to determine the proportions of patients who met the definition of brachycephaly. Statistical significance was defined a priori at $p < 0.05$.

Results

From the total cohort of 1499 patients, the mean age was 6.8 years. Patient demographics are noted in Table 1. The mean CI across the total cohort was 82.6. A simple linear regression was used to examine how well age could predict CI. A scatterplot (Fig. 3) showed that the relationship between age and CI was negative and linear. The correlation between age and CI was statistically significant ($r = 0.165$, $p < 0.001$). The equation for predicting CI was found to be $y = 85.18 - 0.04x$ ($y = \text{CI}$, $x = \text{age in months}$). This relationship shows a decrease in CI by 0.038 each month. A significant regression equation was found

TABLE 2. Mean CI by age group

Age Group (mos)	Mean CI (95% confidence interval)	Test Statistic
<12, n = 482	85.047 (84.406–85.688)	$F(2,1496) = 124.058$, $p < 0.0005$
12–24, n = 139	84.211 (83.158–85.263)	
>24, n = 878	80.410 (79.985–80.835)	

($F[1,1497] = 296.846$, $p < 0.0005$), with an R^2 of 0.140, demonstrating a negative relationship between CI and age. Post hoc analysis revealed significant differences between age groups, with the younger cohorts having higher mean CI values ($F[2,1496] = 124.058$, $p < 0.0005$; Table 2, Fig. 4). The prevalence of brachycephaly (Fig. 5), as defined radiographically by a $\text{CI} \geq 90\%$, showed a consistent, significant decline across patient age groups: < 12 months (28.0%), 12–24 months (12.9%), and > 24 months (4.2%; $X^2 = 158.96$, $r = -0.322$, $p < 0.001$).

Discussion

This study investigated the incidence of deformational brachycephaly in children, and how it changes in relation to different ages. We hypothesized that brachycephaly prevalence decreases with age, and that there is a natural correction of the CI with respect to time. Cultural norms create a notion of what is considered “symmetrical,” and historically a dolichocephalic head shape was considered the norm.⁸ However, since the AAP released the Back to Sleep campaign, the incidence of plagiocephaly and brachycephaly has been steadily increasing.⁴ Thus, the accepted standard of skull shape has a dynamic definition as a result of sleeping position. The natural history of both

FIG. 3. Scatterplot of CI by age and CI severity. This graph demonstrates the negative linear relationship between age and CI.

FIG. 4. Graphic representation of mean CI by age groups.

deformational brachycephaly and plagiocephaly diminishes in adolescence due to brain growth and decreased force on the skull.⁴ Given that these conditions are benign in nature, the major concern is cosmetic acceptance. Ultimately, however, Feijen et al. demonstrated that head asymmetry is not a determinant in the self-evaluation of appearance during adolescence.⁴

There is no study to date that has evaluated the natural history of CI with respect to brachycephaly. The evolution, prevalence, and resolution of plagiocephaly have received more attention in the literature.^{9–12} The utility of CI has primarily been to determine the recovery progression of brachycephaly after helmet therapy, but the first-line treatment is repositioning therapy.⁵ Perhaps the modern notion of skull asymmetry represents a spectrum rather than individual domains. Plagiocephaly and brachycephaly may exist on a continuum.¹³ As such, the only study evaluating the natural resolution of plagiocephaly/brachycephaly was performed by Hutchison et al.,¹⁰ who observed a decrease in diagnosis rate from 16% to 3.3% in the first 2 years of life.

Our study has important clinical considerations. First, parents and clinicians should reasonably expect some degree of skull deformity in response to laying children supine. Second, the negative linear association between age and CI suggests that this form of deformity improves with age. Our study is the first to date showing that the degree of change is a decrease of 0.038 in the CI per month over the first 2 years of life. Our analysis demonstrated a statistically significant decrease in the mean CI at 12 and 24 months. From an overall prevalence standpoint, the rapid drop from 28% to 4.2% in the first 2 years of life also demonstrates an important trend. This knowledge should help curb any excess anxiety about skull asymmetry and also provide insight into what can be expected for the patient.

Furthermore, the mean CI in patients greater than 2 years of age was 80.4. As the trend in the overall pediatric population is for children to sleep on their backs as infants, the normative values of the CI might need to be adjusted as well. This is outside of the scope of this

FIG. 5. Percentage of patients with CI $\geq$ 90% across age groups. Figure is available in color online only.

investigation, but could be investigated with larger populations.

There are a few notable limitations to this study. First, the population is predominantly male and Caucasian/white, which limits the generalizability of the results to other races. Second, this study does not take into account the natural history of posterior positional plagiocephaly. Third, our study does not take into consideration patients who may have been treated via helmet therapy. Because this study focuses on an initial trauma evaluation, there is no follow-up and treatment records are unknown in the outpatient setting. Additionally, by virtue of using CT scans to determine brachycephaly diagnosis, there is no clinical correlation provided. Due to potential differences in skull shape, head positioning, and gantry angle, measurements may be subject to subtle variation.

Conclusions

The incidence of deformational brachycephaly is common in infants but decreases as the child progresses through early childhood. Clinicians can expect a significant decrease in mean CI at 12 and 24 months. Additionally, our regression models show that clinicians can expect continued improvement throughout childhood.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Ahluwalia, Shannon, Bonfield. Acquisition of data: Ahluwalia, Foster, Sherburn, Sellyn, Kelly. Analysis and interpretation of data: Ahluwalia, Foster, Sherburn, Sellyn, Ghani. Drafting the article: Ahluwalia, Foster, Sherburn, Sellyn. Critically revising the article: Ahluwalia, Foster, Sellyn, Shannon. Reviewed submitted version of manuscript: Ahluwalia, Foster. Approved the final version of the manuscript on behalf of all authors: Ahluwalia. Statistical analysis: Ahluwalia, Foster, Ghani. Administrative/technical/material support: Ahluwalia, Wiseman, Shannon, Bonfield. Study supervision: Ahluwalia, Wiseman, Shannon, Bonfield.

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