The metopic index: an anthropometric index for the quantitative assessment of trigonocephaly from metopic synostosis

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OBJECTIVE Because the metopic suture normally fuses during infancy, there are varying degrees of severity in head shape abnormalities associated with premature fusion. A method for the objective and reproducible assessment of metopic synostosis is needed to guide management, as current methods are limited by their reliance on aesthetic markers. The object of this study was to describe the metopic index (MI), a simple anthropometric cranial measurement. The measurements can be obtained from CT scans and, more importantly, from palpable cranial landmarks, and the index provides a rapid tool for evaluating patients in both pre- and postoperative settings.

METHODS High-resolution head CT scans obtained in 69 patients (age range 0–24 months) diagnosed with metopic craniosynostosis were retrospectively reviewed. Preoperative 3D reconstructions were available in 15 cases, and these were compared with 3D reconstructions of 324 CT scans obtained in a control group of 316 infants (age range 0–24 months) who did not have any condition that might affect head size or shape and also in a subset of this group, comprising 112 patients precisely matched to the craniosynostosis patients with respect to age and sex. Postoperative scans were available and reviewed in 9 of the craniosynostosis patients at a mean time of 7.1 months after surgical repair. 3D reconstructions of these scans were matched with controls based upon age and sex.

RESULTS The mean preoperative MI for patients with trigonocephaly was 0.48 (SD 0.05), significantly lower than the mean values of 0.57 (SD 0.04) calculated on the basis of all 324 scans obtained in controls ($p < 0.001$) and 0.58 (SD 0.04) for the subset of 112 age- and sex-matched controls ($p < 0.001$). For 7 patients with both pre- and postoperative CT scans available for evaluation, the mean postoperative MI was 0.55 (SD 0.03), significantly greater than their preoperative MIs (mean 0.48 [SD 0.04], $p = 0.001$) and comparable to the mean MI of the controls ($p = 0.30$). In 4 patients, clinically obtained postoperative MIs by caliper measurement were comparable to measurements derived from CT ($p = 0.141$).

CONCLUSIONS The MI is a useful measurement of the severity of trigonocephaly in patients with metopic synostosis. This simple quantitative assessment can potentially be used in the clinical setting to guide preoperative evaluation, surgical repair, and postoperative degree of correction.

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KEY WORDS anthropometric indices; craniosynostosis; metopic synostosis; metopic index; trigonocephaly; craniofacial

METOPIC synostosis is the second most common type of craniosynostosis, and it can be nonsyndromic, occurring in isolation or with fusion of other sutures, or it may occur as part of a dysmorphic syndrome such as Crouzon or Saethre-Chotzen syndrome.^{12,15} The metopic suture is the first to fuse physiologically, with complete closure typically occurring by

9 to 11 months.^{16,17} Premature fusion is characterized by restricted growth of the frontal bones, resulting in a prominent midline ridge with a triangular forehead and bitemporal narrowing and occipitoparietal widening—the condition described as trigonocephaly. Compensatory changes in the remaining calvaria result in a characteristic orbital dysmorphism, with depression of the superolateral or-

ABBREVIATIONS EuD = eurion-eurion diameter; FPI = frontoparietal index; MFZ = midfrontozygomatic; MFZD = MFZ diameter; MI = metopic index.

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bital rims and ethmoidal hypoplasia, which manifests as orbital hypotelorism, epicanthal folds, and elevation of the lateral canthal angles.^{4,15}

Surgical repair of trigonocephaly, with the goals of restoring anterior cranial fossa volume and improving cosmesis, has evolved considerably over the past few decades, and is typically a collaborative effort between pediatric neurosurgeons and plastic craniofacial surgeons.¹³ However, up to this point, the assessment of metopic synostosis has been largely subjective and involves the use of aesthetic markers, which limits preoperative evaluation of trigonocephaly severity and postoperative assessment of correction. Indeed, one of the most widely used tools to assess craniosynostosis, the Whitaker classification, relies on the judgment of the patient, family, and surgeon to determine the need for surgical correction.¹⁹ Without an objective grading system, clinical determination of trigonocephaly that requires surgical intervention is challenging.⁸

The use of anthropometric cranial measurements has improved the precision and reliability of the assessment of other cranial deformities such as positional plagiocephaly, scaphocephaly, and brachycephaly.^{21,22} The objective of this study was to describe the metopic index (MI), a simple anthropometric cranial measurement that can be obtained both from CT scans and from palpable cranial landmarks, as a rapid tool for evaluating patients with metopic synostosis in both pre- and postoperative settings.

Methods

Clinical and radiographic information for 69 patients diagnosed with nonsyndromic single-suture metopic synostosis at our institution between 2003 and 2014 was retrospectively reviewed. All research protocols were approved by our Institutional Review Board for Human Research. Patients with other conditions affecting cranial size and shape, including hydrocephalus and intracranial masses, were excluded. Fifteen patients had preoperative CT scans with adequate slices to create high-resolution 3D reconstructions, and 7 of these patients had postoperative CT scans for comparison.

A total of 324 scans of 316 full-term infants without craniosynostosis or other abnormality affecting head cranial size or shape (age range 0 to 24 months) were reviewed; serial imaging studies were included only if the scans were performed at least 1 month apart. Two scans were included for each of 8 patients, and the remainder of the 292 scans were obtained in 292 distinct individuals. These scans were obtained between 2005 and 2012 in the course of clinical evaluations for trauma, headache, seizure, and change in neurological status. Patients who had a history of conditions affecting cranial size or shape, including hydrocephalus, prematurity, intracranial masses, skeletal diseases affecting the calvaria, and growth disorders, were excluded from the control group.

Using UltraVisual Advanced Visualization software (Emageon), high-resolution 3D reconstructions were created using bone windows. Superior projections were used for linear measurements between anatomical landmarks, which were made by means of the geometrical “polygon tool.” The metopic index (MI) consists of the midfronto-

zygomatic diameter (MFZD) divided by the eurion-eurion diameter (EuD). The MFZD is the minimum width between the midfrontozygomatic (MFZ) points, or the midpoint along the recess above the brow ridge between the anteroinferolateral frontal bone and frontal bone zygomatic process. The EuD was measured as the maximum cranial width (i.e., distance between the lateral extremes of the skull) (Fig. 1). The available high-resolution head CT scans were reviewed and measurements were performed by 2 observers (J.Y.W. and E.S.A.). Agreement between the 2 reviewers was assessed, and the intraclass correlation coefficient was found to be adequate at 0.82. The average of the measurements of the 2 reviewers were used for further analysis.

In the clinical setting, the corollary of the above measurement was determined with measuring calipers. Caliper points were placed in the recesses of the brow ridge that were visible from the superior vantage point. In infants with metopic craniosynostosis, these recesses were consistently pronounced and medially displaced compared with the position in infants without craniosynostosis, due to the lateral restriction of the forehead. Available measurements from clinical records were used to compare the clinically obtained measurements from those derived from CT scans.

EuDs, MFZDs, and MIs for patients with metopic synostosis were compared with measurements obtained in the complete set of 324 scans obtained in the control group as well as a subset of 112 scans obtained in exact age-matched controls using the unpaired Student t-test. For patients with pre- and postoperative head CT scans,

FIG. 1. Superior projection of a 3D reconstruction of a CT scan obtained in a child without craniosynostosis. Displayed are the midfrontozygomatic diameter (MFZD, 6.85 cm) and the eurion-eurion diameter (EuD, 11.67 cm). The metopic index (MI) is therefore 0.59, which is within the range of normal based upon measurements obtained in control subjects.

as well as to compare clinically obtained and CT-obtained measurements, MIs were compared using the paired Student t-test. Statistical significance was defined as $p < 0.05$. All statistical analyses were performed using Stata/IC 12 (StataCorp LP).

Results

The mean age of the 15 patients with metopic synostosis was 10.8 months (SD 3.9 months). The mean age of the 316 controls at the time the 324 scans were obtained was 11.5 ± 7.3 months. Of the 324 scans obtained in controls, 169 (52.2%) were obtained in male patients; in comparison, 13 (81.3%) of the 15 scans used for analysis of metopic synostosis were obtained in male patients. One hundred twenty-three (38.2%) of the scans obtained in controls were obtained in white patients, 161 (50%) were obtained in African American patients, and 38 (11.8%) were obtained in patients identified as being of another race. Twelve (80.0%) of the 15 patients with metopic synostosis were white, and 2 (13.3%) were African American (Table 1).

The mean preoperative EuD for patients with metopic synostosis was 121 mm (SD 8 mm), which was not significantly different compared with the mean EuD for controls (121 mm [SD 11 mm]). However, the mean preoperative MFZD for patients with trigonocephaly was 58 mm (SD 7

mm), significantly lower than that of the control cohort (69 mm [SD 5 mm]; $p < 0.001$). The EuD and the MFZD were used to calculate the MI (Table 1, Supplemental Table 1). The mean preoperative MI for patients with trigonocephaly was 0.48 (SD 0.05), compared with 0.57 (SD 0.04) in all 324 control scans ($p < 0.001$) and 0.58 (SD 0.04) in a subset of 112 scans obtained in age-matched controls ($p < 0.001$). Comparisons were also performed after the patients were stratified into 3 age groups (6–9 months, 9–12 months, and > 12 months); in each group, the mean MI for patients with trigonocephaly was significantly lower than that for the controls ($p < 0.001$, Table 2).

All 15 patients underwent open anterior craniofacial reconstruction for metopic synostosis. For the 7 patients with pre- and postoperative CT scans, the mean length of time between the surgery and the postoperative CT scan was 7.1 months (SD 8.5 months). The mean postoperative MI for these 7 patients was 0.55 (SD 0.03), significantly greater than the mean preoperative MI (0.48 [SD 0.04], $p = 0.001$) (Table 1). Additionally, the mean postoperative MI was comparable to the MI of the controls ($p = 0.30$). In Fig. 2A and C, preoperative photographs of a representative patient are displayed, and postoperative photographs are shown in Fig. 2B and D. The corresponding 3D reconstruction of preoperative CT and the MFZD and EuD measurements are displayed in the left panel of Fig. 3; the MI derived from these measurements is 0.45. The 3D reconstruction of the CT scan obtained on postoperative Day 1 and corresponding measurements are shown in the right panel; the corrected MI is 0.54. In the 4 patients with available clinical data, the mean MI obtained from clinical measurements (0.55 [SD 0.02]) was comparable to that derived from CT (0.59 [SD 0.03], $p = 0.141$) (Table 3).

Discussion

While once thought to account for only 3% to 4% of craniosynostosis cases, metopic synostosis is currently estimated to account for at least 10% of cases.¹⁵ There is a growing body of literature detailing techniques for surgical correction of trigonocephaly, but there is currently no standardized grading system for the degree of cranial deformity in metopic synostosis. The majority of studies focus on aesthetic markers, pre- and postoperatively.^{1,9,11,13,18} Although cosmetic parameters are a critical part of the clinical evaluation in trigonocephaly, the use of aesthetic markers in combination with objective measures may allow for more optimal preoperative evaluation, operative planning, and postoperative assessment of correction and need for further therapy. Many studies have described nor-

TABLE 1. Summary of clinical and demographic characteristics of 316 controls and 15 patients with metopic synostosis*

Characteristic	Controls	Pts w/ Metopic Synostosis
Age at presentation, mos	11.5 $\pm$ 7.3	10.8 $\pm$ 3.9
Sex		
Male	169 (54.2)	12 (80.0)
Female	155 (45.8)	3 (20.0)
Race/Ethnicity		
White	123 (38.0)	12 (80.0)
African American	161 (49.7)	2 (13.3)
Other	40 (12.3)	1 (0.7)
Baseline measurements		
EuD, mm	121 $\pm$ 11	121 $\pm$ 8
MFZD, mm	68 $\pm$ 5	58 $\pm$ 7
MI	0.57 $\pm$ 0.04	0.48 $\pm$ 0.05
Surgery		
Age, mos	NA	10.3 $\pm$ 3.7
Open procedure	NA	16 (100)
Follow-up imaging		
Time from treatment, mos	NA	7.1 $\pm$ 8.5
EuD, mm	NA	130 $\pm$ 9
MFZD, mm	NA	72 $\pm$ 7
MI	NA	0.55 $\pm$ 0.03

EuD = eurion-eurion diameter; MFZD = mid-fronto-zygomatic diameter; MI = metopic index; NA = not applicable; pts = patients.

* Values are given as mean $\pm$ SD or number (%). Two separate CT studies obtained at different time points were included for 8 of the patients in the control group. Thus the number of CT scans used for comparison is 324 and demographic data are given per scan rather than per patient.

TABLE 2. Age-matched comparison of metopic index values for controls versus patients with metopic synostosis

Age	Controls		Pts w/ Metopic Synostosis		p Value
	No. of Pts	MI	No. of Pts	MI	
6–9 mos	19	0.57 $\pm$ 0.02	5	0.46 $\pm$ 0.05	<0.001
9–12 mos	29	0.56 $\pm$ 0.04	7	0.49 $\pm$ 0.05	<0.001
>12 mos	64	0.56 $\pm$ 0.03	3	0.48 $\pm$ 0.03	<0.001

MI = metopic index.

FIG. 2. Preoperative (**A** and **C**) and 1-year postoperative (**B** and **D**) views of a patient with metopic craniosynostosis. The preoperative views demonstrate classic trigonocephaly with bifrontal narrowing. In a trigonocephalic patient, the MFZD can be easily measured between the palpable recesses above the brow ridges bilaterally (**A** and **C**, red circles). In the clinical setting, caliper points can be placed in the recesses of the brow ridge that are visible from the superior vantage point (**C**). This recess is pronounced and displaced medially in infants with trigonocephaly.

mative anthropometric cranial measurements and proportion indices in the pediatric population.^{3,5} Although 2D anthropometric measurements may not be able to fully convey the 3D cranial deformities that exist in craniosynostosis, such measurements constitute a valuable tool for the accurate and reliable assessment of craniofacial disorders. Anthropometric indices have been successfully applied to assessing cranial deformity in Down syndrome, positional plagiocephaly, and brachycephaly.^{6,21,22}

Indeed, previous studies have demonstrated that anthropometric indices can be successfully used in the assessment of metopic synostosis. Kolar and Salter¹⁰ assessed 24 anthropometric measurements in patients with trigonocephaly and calculated 11 cranial indices. They found that the cephalic index (CI), the EuD taken over the glabella-opisthocranium diameter, did not differ significantly between controls and patients with trigonocephaly. However, they also described the frontoparietal index (FPI), calculated from the frontotemporal diameter taken over the EuD, which was significantly lower in patients with metopic synostosis. Beckett et al.² found that the endocranial bifrontal angle varied significantly between patients with trigonocephaly and controls, and defined 2 severity grades. Wilbrand and colleagues²⁰ described their percentile-based assessment of craniosynostosis, comparing values obtained in patients against normative values for cranial circumference, length, and width, cranial index, as well as sagittal and transversal circumference. Our goal was to describe an anthropomorphic index that would afford similar reliability and would allow for rapid calculation from both CT scans and clinical examination using values obtained with measuring calipers. Studies

FIG. 3. Superior projections of 3D reconstructions of CT scans obtained in the patient in Fig. 2 preoperatively (**left**) and 1 day after surgery (**right**). The MI was 0.46 (MFZD 6.30 cm/EuD 13.59 cm) preoperatively and significantly improved to 0.54 (MFZD 7.34 cm/EuD 13.68 cm) postoperatively. The postoperative image demonstrates interval bifrontal contouring with rounding and widening of the MFZD.

TABLE 3. Comparison of postoperative clinically obtained versus CT-obtained cranial measurements*

Case No.	CT-Obtained			Clinically Obtained		
	MFZD	EuD	MI	MFZD	EuD	MI
7	74.5	137	0.54	84	134	0.63
10	72.5	130	0.56	80	134	0.60
11	62.0	116	0.54	68	124	0.55
15	66.6	118	0.57	71	122	0.58

MI = metopic index.

* Measurements in millimeters.

have suggested that not all cases of single-suture craniosynostosis require CT scans for evaluation.⁷ An anthropometric index that can be calculated from palpable cranial landmarks would not only be cost-effective but would also minimize potential side effects from ionizing radiation. In this study, we compared measurements obtained through CT and through physical examination in 4 patients and found no significant differences (Table 3). With the exception of 1 patient, the measurements were nearly the same with both modalities.

The metopic index (MI) was conceived by taking into account the characteristic forehead dysmorphology of patients with trigonocephaly. Compared with the FPI, we believe the MFZD more accurately captures the severity of the abnormalities of the frontoorbitozygomatic complex. In addition, the MFZ recess is readily visible and is also a more easily palpable cranial landmark, particularly in patients with trigonocephaly. While the EuD was comparable between control patients and patients with trigonocephaly, the MFZD was significantly different and accounted for the difference between the 2 groups with respect to the MI. The MI is a potentially useful tool that can be used to assess the degree of cranial deformity, but clinical judgment remains paramount in determining surgical candidates, with understood variability in measuring techniques and other patient characteristics. In particular, there remains a potential for interrater variability in determining cranial measurements in young infants by physical examination. Recently, Rinkinen and colleagues¹⁴ described a set of temporal morphomic indices (TMIs), anthropometric measurements of the temporalis muscle and temporal fat pad, that can also be derived from CT scans. Although craniosynostosis is a disorder of the cranium, the authors proposed that compensatory changes to the surrounding soft tissue are reflected in TMIs, which appear to also be specific to each subtype of craniosynostosis.¹⁴ While we believe that the MI has value in part due to its ability to be calculated from the clinical examination, in the future, assessment of cranial deformity with anthropometric cranial indices and aesthetic parameters with quantification soft tissue changes will allow for a more comprehensive evaluation of trigonocephaly severity.

Study Limitations

Although a large sample of control patients was used to define normative values for the MI, the current study is limited by the size of the metopic synostosis cohort. Furthermore, data from patients with trigonocephaly of a

wider age range with multiple patients in each age group would improve generalizability. Further refinement of the MI and definition of mild, moderate, and severe subclasses of trigonocephaly will require validation in a larger cohort. Additionally, a limited number of patients received postoperative CT scans of adequate resolution to create 3D reconstructions, limiting the pre- and postoperative comparison of MI values. All patients in the trigonocephaly cohort underwent open surgical correction, but postoperative evaluation of patients after endoscopic repair should also be studied in the future. Despite the above limitations, this study describes a potentially useful cranial index for the assessment of metopic synostosis.

Conclusions

Because the metopic suture normally fuses during infancy, there are varying degrees of severity in head shape abnormalities associated with premature fusion. Objective and reproducible assessment of metopic synostosis is imperative to ensure appropriate patient selection for surgical repair and to aid in operative decision making and postoperative evaluation of correction and need for additional therapy. Current methods of assessing craniosynostosis are limited by their reliance on aesthetic markers, which, while important for ensuring optimal cosmesis, remain highly subjective. The metopic index (MI) is an anthropometric index that can be derived from CT scans and clinical examination of palpable cranial landmarks and is a potentially useful tool for the evaluation of trigonocephaly from metopic synostosis.

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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: Ahn, Dorafshar. Acquisition of data: all authors. Analysis and interpretation of data: Ahn, Wang, Dorafshar. Drafting the article: Ahn, Wang. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Ahn. Statistical analysis: Wang.

Supplemental Information

Online-Only Content

Supplemental material is available with the online version of the article.

Supplemental Table 1. <http://thejns.org/doi/suppl/10.3171/2016.2.PEDS15524>.

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