School-Aged Anthropometric Outcomes After Endoscopic or Open Repair of Metopic Synostosis

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abstract

BACKGROUND AND OBJECTIVES: Metopic craniosynostosis can be treated by fronto-orbital advancement or endoscopic strip craniectomy with postoperative helmeting. Infants younger than 6 months of age are eligible for the endoscopic repair. One-year postoperative anthropometric outcomes have been shown to be equivalent, with significantly less morbidity after endoscopic treatment. The authors hypothesized that both repairs would yield equivalent anthropometric outcomes at 5-years postoperative.

METHODS: This study was a retrospective chart review of 31 consecutive nonsyndromic patients with isolated metopic craniosynostosis treated with either endoscopic or open correction. The primary anthropometric outcomes were frontal width, interfrontal divergence angle, the Whitaker classification, and the presence of lateral frontal retrusion. Peri-operative variables included estimated blood loss, rates of blood transfusion, length of stay, and operating time.

RESULTS: There was a significantly lower rate of lateral frontal retrusion in the endoscopic group. No statistically significant differences were found in the other 3 anthropometric outcomes at 5-years postoperative. The endoscopic group was younger at the time of surgery and had improved peri-operative outcomes related to operating time, hospital stay and blood loss. Both groups had low complication and reoperation rates.

CONCLUSIONS: In our cohort of school-aged children with isolated metopic craniosynostosis, patients who underwent endoscopic repair had superior or equivalent outcomes on all 4 primary anthropometric measures compared with those who underwent open repair. Endoscopic repair was associated with significantly faster recovery and decreased morbidity. Endoscopic repair should be considered in patients diagnosed with metopic craniosynostosis before 6 months of age.

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WHAT'S KNOWN ON THIS SUBJECT: Traditionally, metopic craniosynostosis was treated with open fronto-orbital advancement. More recently, minimally invasive endoscopic strip craniectomy has gained favor and has been shown to have equivalent anthropometric outcomes, with decreased morbidity at 1 year. School-aged outcomes are unknown.

WHAT THIS STUDY ADDS: When evaluated at school age, endoscopic correction of metopic craniosynostosis leads to equivalent anthropometric outcomes as open repair with significantly decreased morbidity. Early referral to a craniofacial center by the pediatrician is crucial in maintaining eligibility for the endoscopic repair.

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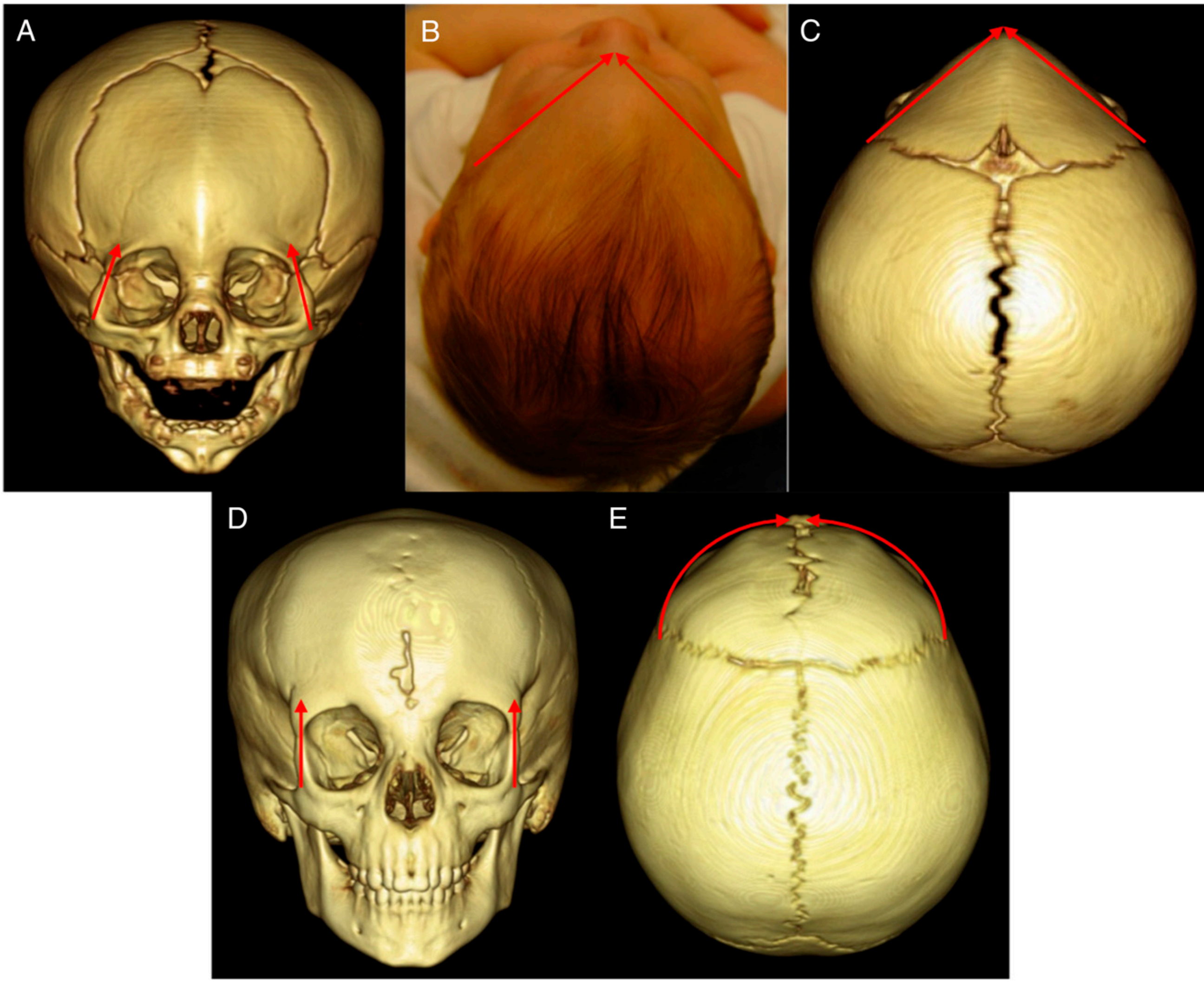

FIGURE 1

Clinical and radiographic findings suggestive of metopic craniosynostosis and their correction after endoscopic repair. A, Upsloping superior orbital rims. B and C, Straight frontal bones. D, More vertically oriented orbital rims at 5 years postoperative. E, More rounded contour of frontal bones at 5 years postoperative.

Metopic craniosynostosis is the premature closure of the metopic suture. The incidence is rising relative to other forms of synostosis, with some estimates as high as 27.3%.[1] For pediatricians and surgeons alike, it is important to distinguish metopic synostosis from benign metopic ridge. Unfortunately, diagnosing metopic synostosis is challenging because physiologic closure can occur as early as 2 months of age.[2–4] In a 2013 retrospective review, Birgfeld et al[2] showed that the "classic triad" of keel-shaped forehead (trigonocephaly), biparietal widening, and hypotelorism was present in only 14% of patients with metopic synostosis. They identified that straight lateral frontal bones and narrow orbits with upsloping superior orbital rims are highly predictive of the condition (Fig 1A–C).[2] Our institution requires upsloping superior orbital rims on a computed tomography (CT) scan for the diagnosis of metopic synostosis (Fig 1A). The most consistently cited physical examination finding of metopic synostosis was lateral frontal retrusion, which describes the "pinching" seen in the lateral forehead.

Traditionally, metopic synostosis has been treated by open cranial vault expansion with fronto-orbital advancement when the infant reaches 6 to 12 months of age.[5–13] Recently, minimally invasive endoscopic strip craniectomy with postoperative helmet therapy has gained favor; however, only young infants <6 months of age (ideally 2–4 months) are suitable candidates because it relies on early rapid brain growth to achieve normocephaly.[14,15]

Current evidence has revealed that anthropometric outcomes after endoscopic and open repairs of metopic synostosis are equivalent at 1-year postoperative.[16] However, regression of head shape, with lateral frontal retrusion, has been documented after open repair in older children, resulting in the need for secondary procedures.[8,10,17] In the absence of data, there is concern that head shape may also regress in patients after endoscopic repair subsequent to completion of helmet therapy. In this article, we aim to report anthropometric and peri-operative outcomes after both methods of repair in school-aged children, which we define here as ≥5 years of age. These findings will be used to inform pediatricians regarding the treatment options and timing of referral to a craniofacial center.

METHODS

Study Design and Population

After institutional review board approval, a retrospective review was performed of consecutive nonsyndromic patients with isolated metopic synostosis treated by endoscopic or open (fronto-orbital advancement) technique between 2006 and 2014. The diagnosis of metopic synostosis was made by the craniofacial team using a CT scan and clinical examination. Only patients with follow-up data available at school age (defined here as ≥5 years of age) were included. Those with multisuture craniosynostosis, syndromic diagnosis, or incomplete follow-up data were excluded.

CT data from age- and sex-matched children without craniofacial deformity served as postoperative controls. Control subjects presented to our institution between 2007 and 2011 and were scanned for reasons including trauma and seizures. Patients with conditions affecting cranial growth were excluded.

TABLE 1 Peri-Operative Variables

	Endoscopic (n = 15)	Open (n = 16)	P
Male sex, n (%)	12 (80)	10 (63)	.24
Age at surgery, mo, mean ± SD	3.3 ± 1.3	10.4 ± 3.5	<.001
Age at 5-y scan, y, mean ± SD	5.3 ± 0.5 (n = 10)	5.5 ± 0.3 (n = 11)	.60
OR time, min, mean ± SD	65 ± 18	389 ± 49	<.001
Length of stay, nights, mean ± SD	1.0 ± 0.0	3.7 ± 0.8	<.001
Estimated blood loss, mL, mean ± SD	52 ± 44	378 ± 215	<.001
Blood transfusion, n (%)	1 (7)	15 (94)	<.001

OR, operating room.

Surgical Techniques and Postoperative Care

We offer both endoscopic and open repair to patients <6 months of age regardless of the severity of synostosis. A shared decision-making process is used, and the family decides on the treatment based on their preference and ability to comply with postoperative helmet therapy. Patients who present after 6 months of age are offered open repair at 10 to 12 months of age.

During endoscopic repair, a 2.5 cm incision in the hairline is used to create a 1-cm–wide craniectomy from the anterior fontanelle to the nasofrontal junction, as previously reported.[16] Postoperatively, these patients are admitted to the hospital floor. All patients were discharged on postoperative day 1. Helmet therapy is initiated within 1 to 3 days of discharge and continued until 1 year of age. During this period, the helmet is worn 23 hours per day and adjusted to guide cranial shape by an orthotist with additional training in postoperative molding (Supplemental Fig 9).

During open repair, a coronal incision is used to advance and reshape the supraorbital bandeau and forehead. Interpositional calvarial bone grafts, temporalis muscle rotation flaps, and/or resorbable fixation are performed as appropriate. These techniques are similar to those described by other high-volume craniofacial centers.[5–8,12,13]

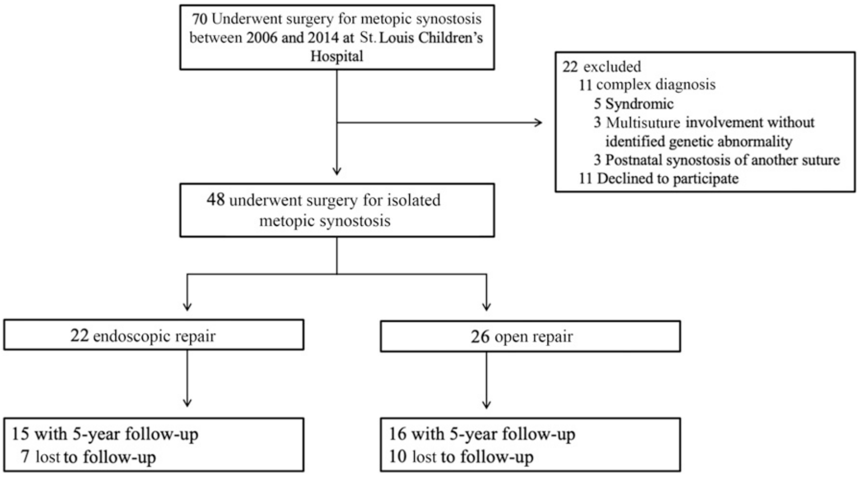

FIGURE 2
Patient enrollment and follow-up. Thirty-one total patients met the inclusion criteria. Seventeen patients were lost to follow-up because of families feeling no medical reason to return and the financial burden of follow-up visits.

Postoperatively, these patients are admitted to the ICU. On postoperative day 1, they are transferred to the floor and discharged from the hospital between postoperative days 3 and 5.

Data Collection and Outcome Measures

Four primary anthropometric outcome variables were selected to evaluate changes in cranial shape: frontal width, interfrontal divergence angle (IFDA), the presence of lateral frontal retrusion, and the Whitaker classification.

The most common clinical measure of frontal morphology is frontal width.[18] To measure frontal width, all CT scans were rendered in 3 dimensions and oriented to the Frankfort horizontal plane by using Analyze 12.0 (Mayo Clinic, Rochester, MN). The straight-line distance in millimeters between the right and left frontotemporal on the coronal plane in the bone threshold was recorded as the frontal width (Supplemental Fig 10).

IFDA measurements were obtained by using the protocol described by Wood et al[19] (Supplemental Fig 11). Analysis was conducted by using Materialise Mimics Innovation Suite 19 (Materialise NV, Leuven, Belgium). IFDA is currently the most accurate measure of forehead contour (trigonocephaly) and can be used to differentiate metopic synostosis from the normal population. Wood et al[19] found an accuracy in the diagnosis of 95.4% for values <134.2°. All frontal width and IFDA measurements were determined by a single operator (A.Y.H.).

The Whitaker classification is a qualitative measure to assesses morphology by stratifying postoperative patients into 4 groups: class I, not requiring any revisions; class II, requiring soft tissue or minor bony revisions; class III, requiring major bony revisions less extensive than the original procedure; and class IV, requiring major bony revisions equal to or more extensive than the original procedure.[20] Lastly, the presence or absence of lateral frontal retrusion was determined from clinical photographs that included frontal and vertex views. These 2 qualitative outcome measures were made by an independent, blinded board-certified plastic surgeon with fellowship training in pediatric plastic surgery.

TABLE 2 Linear Regression Analysis of 5-Year Frontal Width (*n* = 21)

Covariate	Model R^2 = 0.109		
	B	95% CI	*P*
Procedure (reference category = open)	3.02	−3.22 to 9.26	.32
Preoperative frontal width, mm	0.01	−0.35 to 0.38	.93
Age at 5-year scan, y	1.93	−4.89 to 8.75	.55

B, unstandardized coefficient; R^2, coefficient of multiple determination.

Head circumference was included from the medical record to ensure appropriate cranial volume growth and measured in clinic in centimeters. These measurements were then converted to a percentile by using the World Health Organization growth chart for head circumference.[21] Perioperative variables, including sex, age at surgery, procedure duration, length of stay, estimated blood loss, need for blood transfusion, and rates of complication and reoperation were also collected.

Statistical Analysis

Descriptive statistics were calculated. Comparisons between groups were made by using Student's *t* tests or one-way analysis of variance for scalar variables and Fisher's exact test for categorical variables. In

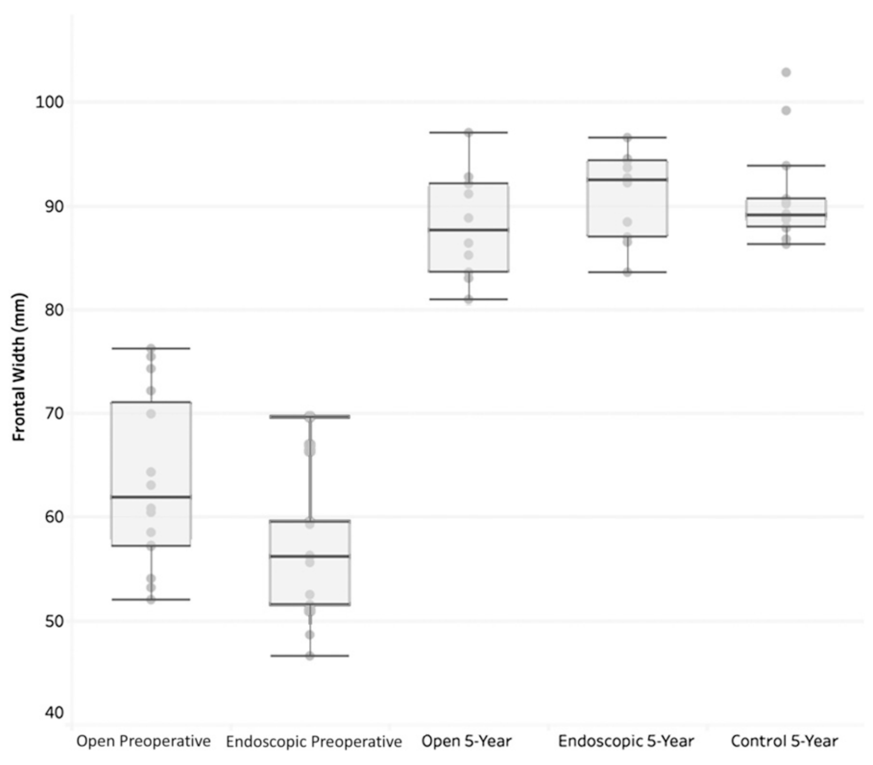

FIGURE 3
Box and whisker plot of frontal widths of preoperative, 5 year, and 5-year controls between open and endoscopic repair.

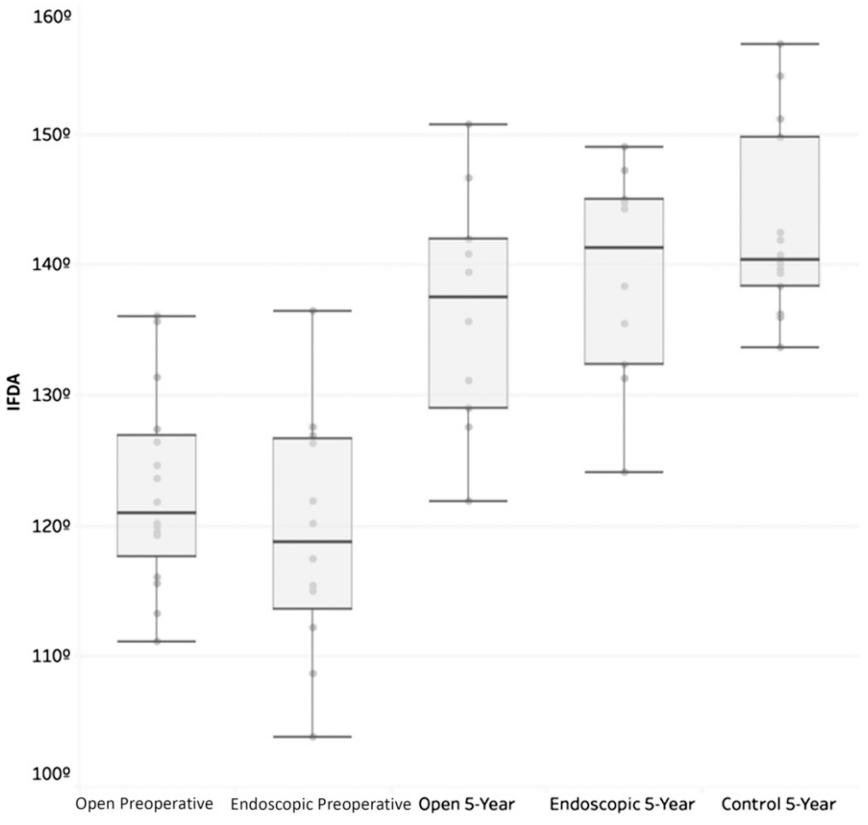

FIGURE 4
Box and whisker plot of IFDA of preoperative, 5 year, and 5-year controls between open and endoscopic repair.

further analysis, we used forced entry linear regression analysis to account for possible confounding covariates. Significance was set a priori at *P* =. 05. The intrarater reliability of quantitative outcome measures was determined with the interclass correlation coefficient (ICC) by using the single measures two-way random effect consistency model. Repeat measures were obtained for 12 scans as follows: one-half from patients who had endoscopic repair and one-half from those who had open repair. Each of those sets was equally divided between preoperative and 5-year scans. R-3.3.2 (R Foundation, Vienna, Austria) was used for Fisher's exact testing. SPSS version 26 (IBM SPSS Statistics, IBM Corporation) was used for all other statistical analysis.

RESULTS

Fifteen consecutive patients who underwent endoscopic repair and 16 consecutive patients who underwent open repair between 2006 and 2014 met the inclusion criteria. During this time period, 70 total patients underwent surgery for metopic synostosis. Eleven were excluded because of syndromic or multisuture diagnoses, 11 declined to participate in research, and 17 were lost to follow-up (Fig 2). Results are presented as mean ± SD. 95% confidence intervals (CIs) are also presented.

Peri-Operative Variables

The endoscopic group was significantly younger at the time of surgery (3.3 ± 1.3 months versus 10.4 ± 3.5 months; *P* <.001; CI of difference 5.1 to 9.0) (Table 1). The endoscopic group spent a significantly shorter amount of time in the operating room (65 ± 18 minutes versus 389 ± 49 minutes; *P* <.001; CI of difference 296 to 351) and had significantly lower estimated blood loss (52 ± 44 mL vs 378 ± 215 mL; *P* <.001; CI of difference 210 to 443), which led to a decreased requirement for blood transfusions (7% vs 94%; *P* <.001; CI of odds ratio <0.001 to 0.11). Furthermore, patients treated endoscopically had a significantly shorter length of stay (1.0 ± 0.0 nights versus 3.7 ± 0.8 nights; *P* <.001; CI of difference 2.3 to 3.1).

Both groups had low complication and reoperation rates. One patient in the open group underwent incision and drainage on postoperative day 3 for surgical wound infection. One patient in each group underwent cranioplasty with autologous particulate bone graft or hydroxyapatite for minor calvarial defects.

Primary Anthropometric Outcomes

The intrarater reliability of both frontal width (ICC = 0.995 [CI 0.982–0.999]) and IFDA (ICC = 0.958 [CI 0.862–0.988]) measurements was excellent.

The mean preoperative frontal width was 56.9 ± 7.2 mm for the endoscopic group and 63.3 ± 8.1 mm for the open group (*P* = .04; CI of difference 0.5 to 12.3; Fig 3). At 5 years, the mean frontal widths were

TABLE 3 Linear Regression Analysis of 5-Year IFDA (*n* = 21)

Covariate	Model R^2 = 0.089		
	B	95% CI	*P*
Procedure (reference category = open)	4.22	−5.56 to 14.01	.37
Preoperative IFDA, degrees	0.01	−0.54 to 0.57	.97
Age at 5-y scan, y	−3.06	−15.63 to 9.51	.61

B, unstandardized coefficient; R^2, coefficient of multiple determination.

TABLE 4 Postoperative Assessments

	Endoscopic (n = 15), n (%)	Open (n = 16), n (%)	P
Complications, surgical wound infection	0 (0)	1 (6)	>.99
Reoperations	1 (7)	2 (13)	.56
Minor cranial defect	1 (7)	1 (6)	
Surgical wound infection	0 (0)	1 (6)	
Whitaker class			.09
I	7 (47)	2 (13)	
II	7 (47)	9 (56)	
III	1 (7)	2 (13)	
IV	0 (0)	3 (19)	
Lateral frontal retrusion	5 (33)	12 (75)	.03

90.9 ± 4.2 and 88.1 ± 5.1 mm, respectively (P = .20; CI of difference −7.2 to 1.6). In linear regression analysis, frontal width did not differ significantly on the basis of repair type when controlling for preoperative frontal width and age at 5-year CT scan (P = .32; Table 2).

The IFDA was similar in the endoscopic and open groups preoperatively (119.3 ± 9.1° vs 122.6 ± 7.4°; P = .30; CI of difference −3.1° to 9.7°) and at 5 years (139.1 ± 8.2° vs 136.4 ± 9.1°; P =.49; CI of difference −10.8° to 5.4°; Fig 4). Linear regression analysis revealed that 5-year IFDA did not differ significantly on the basis of repair type when controlling for preoperative IFDA and age at 5-year CT scan (P = .37; Table 3). The increase in IFDA from preoperative to school age was statistically significant for both endoscopic (P <.001; CI 14.3° to 30.8°) and open (P = .02; CI 2.4° to 23.5°) groups.

CT scans of 14 age- and sex-matched children without craniofacial deformity were identified and included as postoperative controls. In analysis of variance comparisons between the open, endoscopic, and control groups, we found no significant differences in 5-year frontal width (P = .31) or IFDA (P = .17; Figs 3 and 4).

Average preoperative head circumference percentiles were 29 ± 30 for the endoscopic group and 31 ± 27 for the open group (P = .85, CI of difference −20 to 24). At 5 years, they were 67 ± 34 and 54 ± 32, respectively (P = .33; CI −40 to 13). The increase in head circumference over time was significant for both the endoscopic (P = .001) and open (P = .01) groups. Linear regression analysis accounting for preoperative head circumference did not reveal a significant impact of procedure type on head circumference at school age (procedure B [unstandardized coefficient] = 10.5 and P = .39; preoperative head circumference B = 0.56 and P = .01; model R^2 [coefficient of multiple determination] = 0.274 and P = .03).

Endoscopic patients were classified as Whitaker I, II, and III, whereas patients in the open cohort ranged from Whitaker I to IV (Table 4). Examples are shown in Figs 5–8 and

FIGURE 5
Preoperative and 5.5-year postoperative follow-up photographs of a patient who underwent endoscopic repair of metopic craniosynostosis at 2 months of age. This patient was classified as Whitaker I and does not have evidence of lateral frontal retrusion.

FIGURE 6
Preoperative and 6-year postoperative follow-up photographs of a patient who underwent endoscopic repair of metopic craniosynostosis at 4 months of age. This patient was classified as Whitaker III and has evidence of lateral frontal retrusion.

Supplemental Figs 12–15. Differences in the 2 groups' Whitaker classification proportions did not reach statistical significance (*P* = .09). The proportion of patients treated endoscopically with evidence of lateral frontal retrusion at school age (33%) was significantly lower than the proportion treated by open repair (75%; *P* = .03; CI of odds ratio 0.03 to 0.98).

DISCUSSION

We report 5-year anthropometric and peri-operative outcomes after endoscopic and open repair of patients with metopic synostosis. In this cohort of children with isolated metopic synostosis, endoscopic repair was associated with significantly faster recovery and decreased morbidity. At school age, multiple objective and subjective anthropometric outcomes were similar in the 2 groups, except for a lower rate of lateral frontal retrusion in patients treated endoscopically.

Traditionally, metopic synostosis is corrected by open cranial vault expansion with fronto-orbital advancement.[5–8] There have been numerous technical modifications documented in literature to counteract regression toward trigonocephaly presenting as lateral frontal retrusion.[8,10,13] Fearon et al[10] note in their series of 24 patients with metopic synostosis that, although frontal width and head circumference were corrected to normal values at 6 weeks postoperatively, these values decreased significantly at 4-year follow-up. Similarly, Wes et al[8] report in their series of 147 patients with isolated metopic synostosis that patients with >5 years follow-up were significantly more likely to exhibit stigmata of the condition, such as lateral frontal retrusion. These observations have led multiple groups to advocate for overcorrection of the deformity at time of initial operation.[12,13] At our institution, we employ a combination of barrel-stave osteotomies, interpositional bone grafts, and/or temporalis muscle flaps to overcorrect frontal width to that of a 6-year-old child.[13] Our techniques are similar to those described by other high-volume centers.[5–8,12,13] A few groups have noted poorer anthropometric outcomes with earlier operations at or before 8 months of age.[10–12] Although there is institutional variability in the timing of open repair, it is the authors' view that any time between 10 and 14 months of age is acceptable. Many large craniofacial centers report the average age at open repair to be between 10 and 12 months.[8–13]

Introduced by Jimenez et al,[14] endoscopic release of the fused metopic suture with postoperative helmeting therapy offered a paradigm shift in the treatment algorithm for this group of patients. Several recent reports, including a systematic review, provide convincing evidence

FIGURE 7
Preoperative and 5-year postoperative follow-up photographs of a patient who underwent open repair of metopic craniosynostosis at 10 months of age. This patient was classified as Whitaker I and does not have evidence of lateral frontal retrusion.

that endoscopic repair leads to decreased peri-operative morbidity.[15,22,23] This finding is not surprising because dissection and intraoperative calvarial reshaping is limited in endoscopic surgery. The improvement in cranial shape and frontal expansion after endoscopic strip craniectomy is driven by rapid brain growth within the first year of life that is guided by helmet therapy; overcorrection of the deformity at the time of surgery is not possible.[10,13,24] Whereas there is concern for impaired postoperative growth after open cranial vault remodeling,[8,10] evidence suggests that brain growth continues along expected isocurves in endoscopically treated children.[25] In our cohort, head circumference increased to normal range at school age for both groups. In the endoscopic group, efficacy of repair persists into 5 years postoperative, with correction of upsloping superior orbital rims into a more vertical orientation and straight lateral frontal bones into a more rounded contour, implying that appropriate frontal expansion has occurred (Fig 1 D and E).

The frontal width is a direct and easy-to-obtain anthropometric outcome measure for metopic synostosis. However, obtaining consistent and reliable frontal width measurements is challenging.[26,27] Nonetheless, frontal width represents a sensitive and somewhat specific measure of metopic synostosis: in Birgfeld et al's[2] study, 89% of patients with metopic synostosis had a narrow forehead compared with only 32% of patients with metopic ridge. We found that the frontal widths of both open and endoscopic repair groups were statistically equivalent to those of the 5-year control group (Fig 3).

The Whitaker classification is a subjective outcome measure determined by the surgeon evaluating the need for additional procedures to correct residual deformity.[20] Despite its subjectivity, it is commonly used, and its use can impact a patient's postoperative course. In our cohort, as assessed by an independent, blinded reviewer, a large majority (88%) of the patients who underwent open repair were classified as Whitaker class II or higher because of calvarial defects requiring cranioplasty or evidence of lateral frontal hypoplasia and pterional constriction. In comparison, almost one-half (47%) of the patients who underwent endoscopic suturectomy were classified as Whitaker I. Similarly, only 5 of 15 in the endoscopic group had evidence of lateral frontal retrusion compared with 12 of 16 in the open group.

A notable limitation of this study is the small sample size, which limits the power of the study to detect differences between the two groups. Seventeen of 48 (35%) patients who underwent surgery for isolated metopic synostosis were lost to follow-up. Our group has previously noted similarly high attrition rates from the multidisciplinary craniofacial team clinic because of a variety of factors, most commonly because of families feeling no medical

FIGURE 8
Preoperative and 6-year postoperative follow-up photographs of a patient who underwent open repair of metopic craniosynostosis at 11 months of age. This patient was classified as Whitaker III and has evidence of lateral frontal retrusion.

reason to return and the financial burden of follow-up visits.[28] The development of more uniform criteria for the diagnosis of metopic synostosis and objective assessment of anthropometric outcomes, along with larger prospective registries evaluating long-term outcomes at skeletal maturity, are needed. Incorporation of patient-reported and neurodevelopmental outcomes would further strengthen our understanding. These were not included in the current report because they were not consistently documented in our retrospective review of the medical record.

Considering that anthropometric outcomes at school age are equivalent for endoscopic and open repairs and peri-operative outcomes are better for the endoscopic repair, it is our view that endoscopic repair should be strongly considered in all patients diagnosed with metopic synostosis. Because early evaluation before 6 months of age (ideally 2–4 months) is important for endoscopic repair, any suspicion for metopic synostosis on the part of the pediatrician should prompt a referral to a craniofacial center offering both endoscopic and open techniques. Although there has been a global increase in the awareness and diagnosis of single suture craniosynostosis,[1,29] studies have revealed that referral before 3 months of age remains low.[30] Notably, one of the risk factors to delayed referral was prespecialist appointment imaging. Because the multidisciplinary team will evaluate the patient by clinical examination and CT scan as necessary, referrals should not be delayed to obtain imaging by the pediatrician. Other studies suggest that underrepresented minorities and patients with multisuture synostosis experience a longer delay in referral to craniofacial care.[30,31]

Relative contraindications to endoscopic treatment in patients <6 months of age include concerns related to postoperative helmet compliance, such as a long travel distance to a cranial orthotist trained in postoperative orthotic therapy. Helmet therapy is a critical component of the endoscopic treatment, and collaboration with an experienced orthotist is essential to achieving a successful outcome (Supplemental Fig 9). Patients usually require 2 to 4 helmets during the treatment period. Despite the additional cost of postoperative helmet therapy, we have shown that endoscopic treatment is significantly less expensive than open treatment ($50 840 vs $95 588).[32]

CONCLUSIONS

We compared anthropometric outcomes after endoscopic and open repairs of metopic craniosynostosis in school-aged children. We found a significantly lower rate of lateral frontal retrusion in the endoscopic group. Endoscopic repair is associated with significantly faster recovery and decreased morbidity with equally low complication and reoperation rates when compared to open repair. Early referral to a craniofacial center that offers both endoscopic and open repairs by the pediatrician is crucial and should not be delayed by imaging or to establish a definitive diagnosis.

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ABBREVIATIONS

CI: confidence interval
CT: computed tomography
ICC: interclass correlation coefficient
IFDA: interfrontal divergence angle

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