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Anthropometric Outcomes following Fronto-Orbital Advancement for Metopic Synostosis

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Abstract

Background—The authors' purpose is to present changes in anthropometric fronto-orbital dimensions after surgical correction of metopic synostosis.

Methods—The authors retrospectively analyzed craniometric dimensions in older patients with metopic synostosis corrected by fronto-orbital advancement performed by the senior author (J.B.M.). Preoperative and postoperative linear measures (frontal breadth, cranial width, and intercanthal distance) were taken by direct anthropometry. Interdacryon distance and width of the bandeau were also recorded intraoperatively, before and after widening. Follow-up anthropometric values were compared to age- and sex-matched normative data and standard (z) scores were calculated.

Results—Sixteen patients met the inclusion criteria. Syndromic diagnosis was documented in five of 16 patients. Average age at the last postoperative evaluation was 8.9 ± 3.8 years (range, 4 to 16 years). Mean frontal width z -scores decreased postoperatively from 0.82 to -0.32 ($p = 0.007$), indicating diminished growth in this dimension. The last measured frontal width strongly correlated with the breadth of the bandeau after surgical correction but not with preoperative values. Postoperative mean cranial width diminished significantly to a more normal value. Mean intercanthal distance was normal preoperatively and remained so but was significantly greater in syndromic than in nonsyndromic cases.

Conclusions—Frontal growth rate is diminished in the coronal plane after fronto-orbital advancement. The authors recommend primary techniques to overcorrect the width of the bandeau and frontal region, including zygomaticosphenoid osteotomies and interpositional cranial bone grafts to advance/widen the lateral orbital rim. Continued evaluation is required to assess whether overcorrection results in normal frontotemporal shape and breadth at skeletal maturity.

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DISCLAIMER

The content is the sole responsibility of the authors and does not necessarily represent an official view of the National Institutes of Health.

PATIENT CONSENT

Parents or guardians provided written consent for the use of patients' images.

Fusion of the metopic suture normally begins as early as 2 months of age and is completed by 9 months of age.^{1,2} The distinction between normal and pathologic early closure of the metopic suture is determined by clinical and radiographic signs of superolateral orbital/frontal hypoplasia, biparietal widening, and orbital hypotelorism.^{3–7} Patients with metopic synostosis have been classified into moderate and severe subtypes based on the severity of trigonocephaly.⁸ Improved awareness and the current trend to surgically treat all degrees of midline frontal ridging as pathologic trigonocephaly probably account for the reported increased incidence from 3 to 10 percent, to 23 to 28 percent of infants presenting with craniosynostosis.^{9,10}

The goals of operative correction for trigonocephaly are to normalize frontal contour and advance the superolateral orbital rims.^{11–13} Various technical maneuvers and modifications have been described in an attempt to improve appearance of the forehead.^{13,14} Unfortunately, bilateral frontotemporal depression and lateral supraorbital hypoplasia are often seen in the long term.^{15,16} These postoperative skeletal abnormalities are more likely in severe forms of trigonocephaly, wherein the deformity is progressive and worsens with age.^{13,15}

The purpose of this study was to use anthropometry (craniometry) to evaluate the senior surgeon's (J.B.M.) method for primary correction of patients with metopic synostosis. We focused on identifying intraoperative factors that contribute to successful anthropometric outcomes. A technical modification is described for primary correction of lateral orbital retrusion and narrowing.

PATIENTS AND METHODS

After approval by the committee on clinical investigation, we identified and reviewed the charts of all patients with a diagnosis of metopic synostosis treated by the senior author (J.B.M) from 1996 to 2005. Our assessment was limited to patients who were measured preoperatively and at least 3 years postoperatively. Data were collected for date of birth, age at the time of surgery, sex, syndrome, secondary operations, complications, and interval to the most recent evaluation.

Operative Technique

Fronto-orbital advancement includes division of the supraorbital bar in the midline and expansion with a wedge-like calvarial bone graft. Laterally along the bandeau, closing wedge osteotomies are used to modify the frontotemporal contours. Interdacryon width and bandeau length are measured before and after frontal widening. Special attention is paid to correct the posteromedial retrusion of the lateral orbital rims. An osteotomy is made along the zygomaticosphenoid suture (to the inferior orbital fissure), and an interpositional cranial bone graft is placed to advance and widen the frontal process of the zygoma (Fig. 1). The frontal bones are contoured using bone-benders or by radial osteotomies with homeotopic replacement. The fronto-orbital framework is stabilized using resorbable fixation (Fig. 2).

Preoperative and postoperative anthropometric measurements were taken by the senior author (J.B.M) with a spreading Vernier caliper (GPM, Zurich, Switzerland). Dimensions in

the study included: frontal width (ft-ft), cranial (biparietal) width (eu-eu), and intercanthal distance (en-en). The values were compared to age- and sex-matched normative data.¹⁷ Standard (z) scores were calculated using the formula $z = (x - \bar{x})/SD$, where x is the patient measurement, $\bar{x}$ is the mean normative value, and SD is the standard deviation of the mean normative data).

Statistical Analysis

A linear regression model determined the effects of both preoperative and postoperative changes to bandeau length-to-frontal width z -scores. All t tests were two-tailed. Statistical analyses were performed with IBM SPSS Version 22 (IBM Corp., Armonk, N.Y.). Values of $p < 0.05$ were considered to be statistically significant.

RESULTS

We identified 16 of 34 patients (10 boys six girls) with metopic synostosis who met the inclusion criteria. A syndromic diagnosis was determined in five of 16 patients: 9p deletion ($n = 2$), Baller-Gerold ($n = 1$), and unknown genetic disorder ($n = 2$). The average age at repair was 10.2 ± 2.0 months. Preoperative measurements were taken in the operating room, just before surgical correction (range, 7.2 to 15.4 months). Average age at the last postoperative measurement was 8.9 ± 3.8 years (range, 4 to 16 years).

Anthropometric measurements are summarized in Table 1. Mean forehead width z -scores decreased postoperatively from 0.82 to -0.32 ($p = 0.007$) (Fig. 3). There were no statistical differences in this dimension between syndromic and nonsyndromic patients ($p = 0.925$). Mean perioperative bandeau length ranged between 85 and 102 mm after correction. The estimated effect of each 1-mm increase in perioperative bandeau length on long-term frontal width z -score was 0.18 (95% CI, -0.02 to 0.38 per 1-mm increase). The overall regression model had an R^2 of 0.31 (Fig. 4). The estimated effect of bandeau width before fronto-orbital advancement was negligible (0.02 z -score increase per 1 mm). Neither preoperative ($p = 0.832$) nor perioperative ($p = 0.076$) transverse dimension of the bandeau achieved statistical significance in relation to long-term frontal width.

Mean cranial width z -scores decreased postoperatively from 0.9 to 0.2 in the 12 patients for whom both measures were available ($p = 0.003$). There were no significant differences in mean cranial width z -scores for patients with syndromic and nonsyndromic metopic synostosis ($p = 0.341$).

Mean perioperative interdacryon distance was 13.7 ± 1.9 mm (nonsyndromic, 13.7 mm; syndromic, 14.0 mm). Mean intercanthal distance was 26.5 ± 1.9 mm (nonsyndromic, 25.9 mm; syndromic, 27.6 mm). The average intercanthal z -score remained unchanged for all patients ($z = 0.10$), but was greater in syndromic ($z = 1.1$) than in nonsyndromic cases ($z = -0.4$, $p = 0.012$).

DISCUSSION

There are few published long-term outcomes in patients with corrected metopic synostosis, and most reports rely on subjective evaluation.^{13,16,18–20} Our results confirm the observations of other authors that frontal width exhibits decreased growth compared with the normal population, leading to bilateral fronto-orbital retrusion and temporal hollowing¹⁶ (Fig. 5). The recommendation is to overcorrect these contours, but quantitative guidance is lacking.^{13,15}

Frontotemporale, as defined by Farkas, is an anthropometric point along the temporal line of the frontal bone, corresponding with the level of the terminal point of the tail of the eyebrow.¹⁷ Difficulties in obtaining this measurement have been noted by both Farkas and Davenport.^{17,21} The temporal line of the frontal bone can be difficult to identify in a young child, and the frontotemporale point is more imprecise in patients with a craniofacial abnormality. The senior author chose the widest point of the forehead to measure frontal width. We found that the preoperative frontal width values were above age-matched normative data for 14 of 16 patients, with an average *z*-score of 0.82. Kolar and Salter documented a preoperative *z*-score of –0.48 for frontal breadth in their 50 patients.⁵ The difference in *z*-scores between these two reports highlights the difficulty in measuring this dimension. Compensatory biparietal widening is another possible explanation for increased preoperative frontal width in our patients. Cranial width was elevated preoperatively with a *z*-score of 0.9; a more lateral placement of the frontotemporale point could account for the difference from Kolar and Salter.

Selber et al.¹³ described five technical modifications for primary correction of trigonocephaly and assessed their outcomes. A lower number of reoperations and less postoperative temporal hollowing were seen in their patients treated with maximal expansion of the supraorbital bar and stabilization of the construction with strategic bone grafting and resorbable fixation. Our findings corroborate their conclusions. We found that for each 1-mm expansion of the bandeau, the frontal width *z*-score at latest evaluation increased by 0.18. The bandeau was widened in our patients using a midline osteotomy and a cranial bone graft. In addition, we now extend interpositional grafting to widen the forehead as well (Fig. 2).

Fearon et al. noted diminished bifrontal growth between the early (6 weeks) and late postoperative periods (4 years; range, 1.4 to 8 years); the *z*-score decreased from 1.00 to 0.49.¹⁵ This finding was more obvious in their patients with greater preoperative severity and earlier age at operation. The authors suggested overcorrection of frontal width at approximately 12 months of age at the time of operation, to more closely approximate the dimensions of a mature forehead. We agree with these recommendations. During fronto-orbital correction in late infancy, we now construct the bandeau and frontal region to closely match the dimensions of an older child. Our current recommendation is to build the frontal width to match the dimensions of a 6-year-old child (boy, 104 mm; girl, 103 mm) when cranial growth is 90 percent complete²² (Figs. 6 and 7). Continued long-term evaluation is required to assess whether overcorrection approaches normal frontotemporal shape and size at skeletal maturity.

Oh and colleagues, in addition to overcorrection and stabilization of the fronto-orbital construct, recommended two maneuvers to minimize bitemporal hollowing: (1) placement of full-thickness cranial bone grafts to fill the lower coronal defects; and (2) rotation of temporalis muscle, secure to the frontal bone to match the advancement, and reinsertion to the temporal line.²³ The upper coronal gap is filled with particulate bone graft and stabilized with fibrin glue.²⁴

Beckett et al. characterized patients with metopic synostosis as “moderate” or “severe” based on the fronto-orbital deformity.⁸ The severe group had significantly narrower fronto-orbital dimensions, whereas the moderate group did not differ from controls. They measured the mean interdacryon distance at age 5 months in the severe group to be 14.1 mm, similar to the values in our cohort (mean, 13.7 mm; range, 11 to 18).

Contrary to traditional descriptions of trigonocephaly, orbital hypotelorism was not documented in our study. Nguyen et al.²⁵ also did not find orbital hypotelorism in their 15 patients undergoing fronto-orbital advancement for trigonocephaly. They used indirect anthropometry from computed tomographic images at a mean age of 7 ± 1.7 months and found a mean preoperative intercanthal distance of 26.6 ± 1 mm ($z = 0.5$). Kolar and Salter reported the mean intercanthal distance z -score to be -1.75 in 50 patients with nonsyndromic metopic synostosis.⁵ The age range was from 2 to 69 months; however, no differences in z -scores were noted in patients evaluated before and after 6 months of age. Unlike the frontotemporale, the endocanthion is an easily located point that can be precisely measured. To improve accuracy, intercanthal and interdacryon dimensions in our study were measured at a mean age of 10 months in the operating room while the child was under general anesthesia.

It is controversial whether orbital hypotelorism needs to be corrected or can be corrected. Havlik et al. recommended inserting a cranial bone graft between the nasal bones, after outward “greenstick” fracture of the medial orbital walls.²⁶ They found that interorbital width increased between 1 and 4 mm on early postoperative computed tomographic scans. Use of interfrontal springs in patients with metopic synostosis has been reported to improve orbital hypotelorism.²⁷ Fearon and colleagues did not address hypotelorism during fronto-orbital correction, and they had found that the intercanthal z -score exceeded average rate of growth.²⁸ Furthermore, this improvement was greater in the more severely affected children. The mean intercanthal distance in our study remained unchanged in relation to normal values ($z = 0.1$). Intercanthal distance was significantly increased at the latest follow-up evaluation in patients with syndromic metopic synostosis compared with nonsyndromic patients. Our findings confirm that attempts to repair orbital hypotelorism are unnecessary.

Preoperative biparietal widening was noted in our patients, and normalized in time, although no effort was made to alter this dimension. Perhaps this cranial remodeling is analogous to the improvement in frontal bossing in patients with sagittal synostosis after posterior calvarial vault reconstruction.²⁹

Posteromedial retrusion of the lateral orbital rim is a common finding in infants with metopic synostosis; this finding can be used to differentiate between physiologic and

pathologic closure of the metopic suture.⁷ To prevent a palpable stepoff along the lateral orbital rim after frontal advancement, Marsh and Schwartz extended the lateral orbital wall osteotomy into the body of the zygoma during removal of the bandeau.¹¹ This technique involves more dissection and detachment of the lateral canthal tendon. Alternatively, we recommend an osteotomy along the zygomaticosphenoid suture and placement of a cranial bone graft to adjust the lateral orbital rim into a normal relationship to the advanced and widened zygomatic processes of the frontal bone (Figs. 1 and 2).

Endoscope-assisted strip craniectomy and postoperative orthotic therapy is increasing in popularity. Reported benefits include decreased operating time, fewer blood transfusions, shorter hospital stay, and reduced cost of treatment.^{30,31} Initially used for sagittal synostosis, this strategy has also been applied in patients with metopic synostosis.^{25,32–35} Appropriate concern has been raised that an endoscopic technique, which relies on brain growth and helmet therapy, does not achieve the same overcorrection as an open fronto-orbital repair.¹⁵ Early (1 year) postoperative outcomes have shown equivalent improvement in trigonocephaly between fronto-orbital advancement and endoscopic metopic osteotomy with orthotic therapy.²⁵ Longer term assessments are needed to assess whether growth in the frontal region will diminish in the same manner as with the open techniques.

CONCLUSIONS

There are limitations in this retrospective study. Anthropometric data were available for only 16 of 34 patients treated by the senior author. Syndromic diagnosis was identified in five of 16 patients, but not all 16 patients had genetic testing. The difficulty in identifying the frontotemporale, as noted above, especially in children with craniosynostosis, complicates comparison to other studies using this anthropometric point. Strengths of this study are that most of the patients were classified in the severe phenotypic category, and one surgeon performed all the procedures and direct anthropometry.

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Fig. 1.
(*Left*) Osteotomy in the right zygomaticosphenoid suture. (*Right*) Anterolateral advancement of the frontal process of the zygoma after insertion of calvarial bone graft.

Fig. 2.
(*Left*) Intraoperative anterior view after fronto-orbital advancement. (*Right*) Illustration showing operative details: expansion of the supraorbital bar and frontal bone in the midline with interpositional calvarial bone graft; placement of cranial bone graft in the osteotomy through the zygomaticosphenoid sutures to advance and widen the frontal process of the zygoma; and full-thickness cranial bone grafts to fill the lower coronal defects and particulate bone graft for upper coronal gap.

Fig. 3.

(*Above*) Female and (*Below*) male preoperative and postoperative frontal width compared to Farkas' normative data.

Fig. 4.
Relationship between last measured frontal width and breadth of the bandeau after surgical correction.

Fig. 5.
(*Left*) Nonsyndromic patient with metopic synostosis and a preoperative frontal width z -score of 0.6. The bandeau expanded from 72 mm to 85 mm. (*Right*) Postoperatively, at age 12 years; note bilateral frontotemporal narrowing. The frontal width z -score was -2.9 .

Fig. 6.
(*Left*) Patient with syndromic metopic synostosis (Baller-Gerold). Frontal width z -score of -0.1 . Bandeau expanded from 80 mm to 95 mm. (*Right*) Postoperatively, at age 7 years, frontal width z -score of -0.8 and intercanthal z -score of 1.5 (34 mm).

Fig. 7.
(*Left*) Nonsyndromic patient with metopic synostosis treated at 7 months of age. The bandeau expanded from 68 mm to 100 mm. (*Right*) View at 2 years postoperatively with frontal width z-score of 1.5 but with minor temporal hollowing and frontal irregularity.

Table 1

Preoperative and Postoperative Anthropometric Values

Measurements	All Patients (<i>n</i> = 16)		<i>p</i>	Nonsyndromic Patients (<i>n</i> = 11)		Syndromic Patients (<i>n</i> = 5)	
	Preoperative	Postoperative		Preoperative	Postoperative	Preoperative	Postoperative
Frontal width (ft-ft)	0.82	-0.32	0.007	0.95 ± 1.1	-0.33 ± 1.5	0.55 ± 1.1	-0.28 ± 0.9
Head width (eu-eu)	0.90	0.20	0.003	1.07 ± 1.5	0.61 ± 1.7	0.40 ± 0.8	-0.19 ± 0.9
Intercanthal distance (en-en)	0.10	0.10	0.996	-0.03 ± 0.7	-0.35 ± 1.0	0.37 ± 0.4	1.08 ± 0.5
Age, yr							
Mean	0.9	8.9		0.8	8.7	0.9	10.6
Range	0.6-1.3	4.1-16.4		0.6-1.3	4.1-16.4	0.8-1.0	7.2-12.8