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The Temporal Region in Unilateral Coronal Craniosynostosis: Fronto-orbital Advancement Versus Endoscopy-Assisted Strip Craniectomy

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Abstract

Objective: To compare postoperative temporal expansion in patients treated with fronto-orbital advancement or endoscopy-assisted craniectomy with cranial orthotic therapy.

Design: This is a retrospective, multicenter cohort study of patients with unilateral coronal craniosynostosis (UCS).

Setting: Computed tomographic (CT) scans were drawn from UCS patients treated at Boston Children's Hospital or St Louis Children's Hospital.

Patients: The study included 56 patients with UCS after fronto-orbital advancement (n = 32) or endoscopic repair (n = 24) and 10 age-matched controls.

Intervention: Fronto-orbital advancement entails a craniotomy of the frontal bone and superior orbital rim followed by reshaping and forward advancement. Endoscopic repair is the release of the synostotic suture and guidance of further growth of the cranium using a molding orthotic.

Main Outcome Measures: Measures included posterior temporal width, anterior temporal width, orbital width, and anterior cranial fossa area taken preoperatively and 1 year postoperatively. Linear regression was performed to assess 1 year postoperative improvement in symmetry; covariates included preoperative symmetry and type of surgery.

Results: Both treatments showed improvement in orbital width and anterior cranial fossa area symmetry 1 year postoperatively ($P < .001$), but no significant improvement in posterior or

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anterior temporal width symmetry. Linear regression revealed no difference between the 2 procedures in any of the 4 measurements ($.096 \leq P \leq .898$).

Conclusions: Fronto-orbital advancement and endoscopic repair show equivalent outcomes 1 year postoperatively in all 3 width measurements and anterior cranial fossa area. Neither procedure produced significant improvement in temporal width.

Keywords

unilateral coronal craniosynostosis; temporal; endoscopic; fronto-orbital advancement

Introduction

Unilateral coronal craniosynostosis (UCS) is the premature fusion of a single coronal suture in the developing cranium. Central features of this growth pattern include flattening of the frontal bone, elevation and retraction of the superior orbital rim, and hollowing of the temporal region, all on the synostotic side, as well as contralateral frontal protrusion (Figure 1A and B) (Marsh et al., 1986; Meara et al., 2003; Fearon et al., 2009; Stelnicki et al., 2009; El-Sadek, 2011; Mesa et al., 2011). A key difference in this phenotype as compared to other forms of craniosynostosis, such as sagittal and metopic, is that it results in asymmetric growth of the cranial and facial bones, making UCS particularly difficult to correct (Meara et al., 2003).

Fronto-orbital advancement is the most common treatment for UCS (Figure 1A). There is variation in the technique of this operation, but in general this procedure entails a craniotomy of the frontal bone and superior orbital rim followed by reshaping and forward advancement (Stelnicki et al., 2009). In fronto-orbital advancement, the surgeon will often attempt to overcorrect the depressed frontal region to minimize long-term regression (Fearon et al., 2009; El-Sadek, 2011; Mesa et al., 2011).

Endoscopy-assisted craniectomy with orthotic therapy has become an alternative to fronto-orbital advancement (Jimenez and Barone, 2013; Tan et al., 2013). The purpose of endoscopic repair is to release the synostotic suture and guide further growth of the cranium and skull base using a molding orthotic (Figure 1B) (Barone and Jimenez, 2004). Advantages of this procedure over fronto-orbital advancement include decreased operative time, hospital stay, blood loss, and scarring. In addition, no foreign material remains after surgery such as screws, plates, sutures, wires, etc (Stelnicki et al., 2009; Berry-Candelario et al., 2011; Shah et al., 2011; Vogel et al., 2014). However, the results are largely dependent on the skill of the cranial orthotist rather than the surgeon, and orthotic helmets must be worn until 1 year of age (Barone and Jimenez, 1999; Berry-Candelario et al., 2011; Proctor, 2012). The endoscopic procedure is performed in patients younger than 6 months (ideally 2–4 months), and in patients whose guardians are able to comply with orthotic therapy.

Fronto-orbital advancement is effective at addressing dys-morphology of the forehead and orbit (Steinbacher et al., 2011; Derderian et al., 2014; Taylor et al., 2015). However, based on anthropometric measures, Steinbacher and coworkers (2011) and Derderian et al. (2014) both concluded that temporal hollowing persists postoperatively. The purpose of this study is

to use craniometric analyses similar to those used in previous studies (Steinbacher et al., 2011; Derderian et al., 2014) to compare the outcomes of fronto-orbital advancement and endoscopy-assisted craniectomy with postoperative molding helmet therapy in achieving temporal expansion in patients with nonsyndromic UCS.

Methods

Patient Data

This is a retrospective, cohort study with institutional review board approval. We identified 95 patients who received fronto-orbital advancement and 25 patients who underwent endoscopic repair. Patients with inadequate computed tomographic (CT) data, excessive motion artifact, or scans that did not include necessary osseous landmarks were excluded from the study. Preoperative and postoperative CT scans were available for 56 patients with UCS treated at either Boston Children's Hospital or St Louis Children's Hospital. CT data for 10 normal control patients with no record of cranial trauma or deformity were also obtained (Table 1).

Craniometric Analysis

All measurements were obtained by a single operator (B.M.) using Analyze 12.0 (Mayo Clinic, Rochester, MN). CT scans were oriented using a modified version of the Frankfort Horizontal. Normally, the Frankfort Horizontal is established using the left inferior orbital rim and the two porions as points. For this study, the nonsynostotic orbital rim substituted for the left inferior orbital rim. The scan was then oriented again by setting left and right apex cornea in the same coronal plane. After orientation, a threshold for bone visualization was defined for each scan, and the coordinates of exterior osseous landmarks were measured. These landmarks included the posterior point of the pterion, the superior point of the sphenozygomatic suture (measured on the inner side of the orbit), the zygomaticofrontal suture, and the nasion (Figure 2A). Next, an endocranial view was created by removing all axial CT slices above a level located one-third the distance from the nasion to the apex of the skull. This cut point was the lowest level that consistently revealed a complete bird's-eye view of the anterior cranial fossa, which was commonly elevated on the synostotic side.

Linear measurements were based on the protocols of Steinbacher et al. (2011) and Derderian et al. (2014) with minor adjustments to the osseous landmarks. First, a midline was drawn along the anterior cranial fossa from the sella turcica to the anterior edge of the cribriform plate using the crista galli as a guide. Three widths were measured on each side of the cranium from midline to a lateral osseous landmark: posterior temporal width (midline to posterior intersection of pterion), anterior temporal width (midline to superior point of sphenozygomatic suture), and orbital width (nasion to zygomaticofrontal suture) (Figure 2A).

Next, the planar area of each side of the anterior cranial fossa was measured. Using the cranial base view described above, the medial border of the regions was established using the crista galli. Each side of the anterior cranial fossa was carefully outlined using the cross

section of the frontal bone, the lesser wing of the sphenoid, and the established midline as boundaries (Figure 2B).

Statistical Analysis

Intrater reliability was measured with the 2-way random single measures intraclass correlation coefficient. Repeat measures used for this assessment were made on 10 scans selected to reflect the proportion of patients who received fronto-orbital advancement or endoscopic repair, as well as the proportion of patients from the 2 participating institutions. The 4 intraclass correlation coefficients ranged from 0.6 to 0.98 (anterior temporal width and anterior cranial fossa area, respectively).

Student *t* test was used for comparisons between the synostotic and nonsynostotic sides, as well as pre- and postoperative measures. A linear regression model was used to determine if preoperative asymmetry or type of repair were significant predictors of improvement in the ratio between the synostotic and nonsynostotic sides. Age at surgery (and standard transformations of this variable) was not linearly related to the improvement and was excluded from the regression model.

The baseline threshold for significance for all results was $P = .05$. Significant *P* values in the tables and the text are shown in bold. The Holm-Bonferroni method of correction for multiple comparisons was applied. To determine the family-wise error rate for the Student *t* tests (shown in Tables 2–5), 3 families were categorized. These families and the number of comparisons are as follows: comparisons between synostotic and nonsynostotic measures (16 *P* values), pre and post measures and ratios (16 *P* values), and the threshold for the significance of covariates in the regression models (8 *P* values). All statistical analyses were performed using SPSS ver. 22 (IBM Corp, Armonk, NY).

Results

All CT scans were performed between 1993 and 2015. Table 1 shows the demographic characteristics of the patient population. Of the 56 UCS patients with preoperative and 1-year postoperative scans, 32 underwent fronto-orbital advancement at a mean age of 10.7 months (range, 8.1 to 18.8 months), and 24 underwent endoscopic repair at a mean age of 2.7 months (range, 1.9 to 3.4 months). Of the 10 unaffected control scans, 5 were age matched to the average age at preoperative scan and 5 to the average age at postoperative scan.

Table 2 shows the mean difference in width (mm) and area (mm²), pre- and postoperatively, between the synostotic and nonsynostotic sides for both surgical procedures. There was a significant difference ($P = .003$) between synostotic and nonsynostotic sides for all measures except posterior temporal width of the 1-year postoperative, fronto-orbital advancement group ($P = .310$). Postoperatively, both fronto-orbital advancement and endoscopic repair groups showed a significantly decreased difference between the 2 sides for orbital width and anterior cranial fossa area ($P = .002$), but not in posterior temporal width or anterior temporal width (.108 $P = .972$).

The ratio between the synostotic and nonsynostotic sides was used to assess cranial symmetry (Table 3). Control subjects showed excellent symmetry across all measures, ranging from 0.98 to 1.00. There was a significant improvement in the symmetry of orbital width and anterior cranial fossa area after both procedures ($P < .001$), but no significant difference between pre- and postoperative measures in posterior or anterior temporal width ratios ($.008 \leq P \leq .949$). In this case, $P = .008$ was not significant because of the constraints of the Holm-Bonferroni correction method.

Linear regression models were used to assess the effect of preoperative asymmetry and type of procedure on symmetric improvement (postoperative ratio – preoperative ratio) (Table 4). Based on the coding, for fronto-orbital advancement versus endoscopic repair, a positive value indicates that fronto-orbital advancement was more effective. Degree of preoperative asymmetry had a significant effect on symmetric improvement for the 3 width measurements ($P = .001$) but not in the anterior cranial fossa area ($P = .075$). For example, in regard to posterior temporal width, for every 1% increase in the preoperative asymmetry ratio a 0.56% increase in symmetric improvement is expected 1 year postoperatively. Type of procedure had no significant effect on symmetric improvement in any measurement ($.096 \leq P \leq .898$). Model strength was limited, with R -square values ranging from 0.06 to 0.44.

Discussion

The purpose of this study is to compare the efficacy of fronto-orbital advancement and endoscopic craniectomy with orthotic therapy in achieving temporal expansion in patients with UCS. It is unclear if the temporal hollowing in UCS is due to osseous or soft tissue abnormalities. Steinbacher and colleagues (2011) concluded that the superficial temporal fat pad does not play a role in hollowing, whereas the effect of the temporalis muscle was inconclusive. Derderian et al. (2014) added to these findings and determined that the temporal fossa and temporalis muscle volumes are significantly smaller on the synostotic side. Both studies found persistent osseous asymmetry between the synostotic and nonsynostotic sides. This investigation focused solely on osseous deformity because this factor was consistent across both studies (Steinbacher et al., 2011; Derderian et al., 2014).

Craniometric analysis showed that the 3 widths (posterior temporal, anterior temporal, and orbital) and the anterior cranial fossa area of the synostotic side were consistently smaller than those of the nonsynostotic side, and this remained true 1 year postoperatively for all measurements except the posterior temporal width (Table 2). This suggests some degree of normalization in the posterior temporal width for the fronto-orbital advancement (FOA) group, which is consistent with the results of Derderian et al. (2014), which demonstrated a nonsignificant difference in the sides of the skull for a similar measurement of the posterior temporal region in 15 UCS patients 1 year post-FOA. However, unlike the Derderian et al. study, the normalization observed with this cohort was not sufficient to produce a significant increase in symmetry as measured by the pre- and postoperative ratios of the synostotic and nonsynostotic sides shown in Table 3. The P value for the difference between the preoperative and postoperative ratios for the posterior temporal width was below $P = .05$ ($P = .008$), but was not significant because of the constraints of the Holm-Bonferroni correction method (Table 3). Increased sample size may demonstrate a significant increase in the

symmetry of this measurement post-FOA, but with the present data the increased normalization observed in Table 2 was not sufficient to produce significantly increased symmetry.

With regard to the other measurements, our results suggest that both procedures are effective at achieving some degree of improvement in the symmetry of the orbital width and anterior cranial fossa area ($P < .001$), but do not show improvement in the anterior temporal width ($P = .451$ for FOA, $P = .818$ for endoscopic) (Table 3). The use of the synostotic to nonsynostotic ratio as a measurement of symmetry was supported by the high degree of symmetry exhibited by the 10 control patients, indicating that this method was an accurate representation of cranial deformity in the temporal region (Table 3).

To complete the analysis of these measurements, linear regression was performed to compare the effectiveness of the 2 procedures and to assess the effects of preoperative asymmetry (Table 4). Preoperative asymmetry had a significant effect on the increase in symmetry for all width measurements in both procedures, but not for the anterior cranial fossa area ($P = .075$). This is likely because those with more severe asymmetry had more room for improvement. The linear regression showed no statistically significant difference between fronto-orbital advancement and endoscopic repair in any of the measurements. This result implies that at 1 year postoperation, these 2 procedures are equally effective at correcting orbital and anterior cranial fossa symmetry, and equally ineffective at correcting anterior and posterior temporal symmetry. The maximum R -squared value of the overall models was 0.44. This suggests that other factors explain the bulk of the variability in the morphometric improvement. In particular, because of the technical requirements of the analysis, age at surgery was excluded from all 4 regression models, limiting their power.

This analysis only included 1-year postoperative data because it is the policy of both participating institutions (Boston Children's Hospital and St Louis Children's Hospital) that repeat CT scans only be performed on children if clinically necessary. With regard to long-term outcomes of the 2 procedures, studies have suggested that patients who receive fronto-orbital advancement experience long-term regression. Becker and colleagues (2006) showed that patients followed to dentoskeletal maturity after fronto-orbital advancement had more asymmetry than those evaluated at 1 year postoperation. More recently, Taylor et al. (2015) found that patients followed 5 years or more after fronto-orbital advancement were likely to develop supraorbital retrusion and temporal hollowing. To address the challenge of long-term regression in FOA, Derderian and colleagues (2014) described a technical modification to the fronto-orbital bandeau that involves placing a bone graft for expansion and aggressively contouring the temporal limb of the bandeau to restore concavity of the temporal fossa. In contrast, there is a paucity of data regarding the long-term results of endoscopic repair. It has been suggested by Jimenez and Barone (2013) that patients with UCS continue to improve years after endoscopic repair in contrast to the regression seen after FOA; however, a true long-term analysis has not yet been performed, especially in the specific case of temporal hollowing. Future studies must focus on the long-term outcomes of endoscopic repair to effectively compare these 2 methods of treatment for UCS.

This study is limited by the small sample size of patients meeting inclusion criteria ($n = 56$). Furthermore, as a retrospective cohort study there may be bias in patient selection unaccounted for in the analysis. Additional limitations include variable gender distribution, age at surgery, variance between surgeons, the wider temporal range of the FOA group, and the exclusion of some older (pre-1995) FOA patients as a result of poor quality or incomplete CT scans. Another limitation of this investigation was achieving adequate precision in width measurements. The intraclass correlation coefficients for width measurements ranged from 0.60 to 0.86, whereas the area measurement was much more precise, with an intraclass correlation coefficient of 0.98. Future studies may be able to address this challenge by using volume and area rather than linear measurements to assess postoperative improvement of the temporal region. Additionally, the study included only 1-year CT scan follow-up because of the policies of the participating institutions. Future studies may require different methods of measurement, such as 3D photography, to obtain more long-term results.

Conclusion

Fronto-orbital advancement and endoscopy-assisted craniectomy with orthotic therapy show equivalent outcomes 1 year postoperatively with regard to cranial width and anterior cranial fossa area. Both procedures show consistent improvement in orbital width and anterior cranial fossa symmetry, but neither demonstrated significant temporal expansion. Long-term data suggest that regression is a common risk following FOA, whereas long-term data on endoscopic repair remain less clear. When deciding between these 2 methods of repair, it is important to consider the other risks and benefits involved in each procedure. According to a Vogel and colleagues, endoscopic repair had a decreased operative time (81 vs 166 minutes), decreased rate of blood transfusion (9.5% vs 100%), shorter hospital stay (1.1 vs 4.7 days), and lower cost (\$37,256 vs \$56,990) (Vogel et al., 2014). These advantages for endoscopic repair in the treatment of craniosynostosis have been cited in other studies as well (Stelnicki et al., 2009; Berry-Candelario et al., 2011; Shah et al., 2011; Jimenez and Barone, 2013). Disadvantages to the endoscopic approach include age restriction (younger than 6 months), need for postoperative orthosis until 1 year of age and availability of an orthotist trained in postoperative helmet therapy. Fronto-orbital advancement has the benefit of greater surgeon control, fewer age restrictions, and more room for variability and modification in technique. Future research, specifically in the area of temporal hollowing, is necessary to elucidate the long-term outcomes of endoscopic repair in UCS, but in the short term it appears that both procedures produce equivalent results.

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Figure 1.

Three-dimensional volume renderings from computed tomographic (CT) scans of a patient with (A) right unilateral coronal synostosis preoperatively (top row) and 1 year postoperatively (bottom row) after fronto-orbital advancement and (B) left unilateral coronal synostosis preoperatively (top row) and 1 year postoperatively (bottom row) after endoscopic repair. Neosuture formation and/or persistent craniectomy gap are common after endoscopic repair in patients with unilateral coronal craniosynostosis (UCS; Sauerhammer et al., 2014).

Figure 2.

Three-dimensional volume renderings from a preoperative computed tomographic (CT) scan of a patient with unilateral coronal craniosynostosis (UCS). (A) A red spot denotes the location of each osseous landmark. Blue arrows denote width measurements. SZ signifies the superior-most point of the sphenozygomatic suture; ZF signifies the zygomaticofrontal suture. All osseous landmarks were measured on the exterior of the skull, as shown in the images on the left. The midline was established along the cranial base from the sella turcica to the anterior end of the cribriform plate (right). (B) The anterior cranial fossa is shown in red on the synostotic side and blue on the nonsynostotic side. The boundaries of the anterior cranial fossa were defined as the edge of the lesser sphenoid wing posteriorly and the cross-sectional surface of the frontal bone anteriorly.

Table 1.

Patient Demographic Data.

	Fronto-orbital Advancement	Endoscopic	Total	Control
Age at preoperative scan, mo, mean $\pm$ SD	8.5 $\pm$ 7.4	2.0 $\pm$ 0.7	5.6 $\pm$ 6.5	4.2 $\pm$ 2.6
Age at postoperative scan, mo, mean $\pm$ SD	23.3 $\pm$ 9.7	16.0 $\pm$ 4.4	20.2 $\pm$ 8.6	25.7 $\pm$ 6.2
Age at surgery, mo, mean $\pm$ SD	10.7 $\pm$ 8.2	2.6 $\pm$ 0.7	7.2 $\pm$ 7.3	-
Institution, n, WUSM/BCH	32/0	8/16	40/16	10/0
Gender, n, M/F	15/17	6/18	21/35	3/7
Synostotic side, n, right/left	18/14	15/9	33/23	-

Abbreviations: BCH, Boston Children's Hospital; mo, month; SD, standard deviation; WUSM, Washington University in St Louis, St Louis, MO.

Tables 2.

Difference in Width (mm) and Area (mm²) Between Synostotic and Nonsynostotic Sides Before and After Repair for Each Procedure.^a

	Nonsynostotic and Synostotic	Nonsynostotic vs Synostotic (<i>P</i> Value)	Preoperative vs Postoperative (<i>P</i> Value)
Posterior temporal width (mm)			
Pre-FOA	2.14 ± 2.66	<.001	.108
Post-FOA	1.28 ± 3.21	.310	
Pre-Endo	1.95 ± 2.74	.002	.589
Post-Endo	2.31 ± 2.96	.001	
Anterior temporal width (mm)			
Pre-FOA	2.09 ± 1.93	<.001	.256
Post-FOA	1.48 ± 2.63	.003	
Pre-Endo	1.77 ± 2.45	.002	.972
Post-Endo	1.79 ± 1.67	<.001	
Orbital width (mm)			
Pre-FOA	5.23 ± 2.04	<.001	<.001
Post-FOA	3.37 ± 2.10	<.001	
Pre-Endo	5.56 ± 1.53	<.001	<.001
Post-Endo	3.54 ± 1.61	<.001	
Anterior cranial fossa area (mm ²)			
Pre-FOA	563.59 ± 192.35	<.001	<.001
Post-FOA	408.31 ± 237.80	<.001	
Pre-Endo	475.07 ± 145.37	<.001	.002
Post-Endo	378.18 ± 198.31	<.001	

Abbreviations: Endo, endoscopy-assisted craniectomy; FOA, fronto-orbital advancement.

^aPosterior temporal width: midline to pterion; anterior temporal width: midline to sphenozygomatic suture; orbital width: nasion to zygomaticofrontal suture. Significant *P* values are in bold.

Table 3.

Ratio of Synostotic to Nonsynostotic Width Before and After Repair for Each Procedure.

	Synostotic/Nonsynostotic Ratio		
	Preoperation	Postoperation	<i>P</i> Value
Posterior temporal width			
FOA	0.95 ± 0.06	0.98 ± 0.06	.008
Endoscopic	0.96 ± 0.07	0.96 ± 0.06	.949
Control	1.00 ± 0.01	0.98 ± 0.02	-
Anterior temporal width			
FOA	0.94 ± 0.05	0.95 ± 0.06	.451
Endo	0.95 ± 0.07	0.95 ± 0.05	.818
Control	1.00 ± 0.03	0.99 ± 0.01	-
Orbital width			
FOA	0.87 ± 0.05	0.92 ± 0.04	<.001
Endo	0.85 ± 0.04	0.91 ± 0.04	<.001
Control	1.00 ± 0.01	1.00 ± 0.01	-
Anterior cranial fossa area			
FOA	0.69 ± 0.09	0.80 ± 0.09	<.001
Endo	0.68 ± 0.07	0.79 ± 0.09	<.001
Control	0.99 ± 0.02	1.00 ± 0.04	-

Abbreviations: Endo, endoscopy-assisted craniectomy; FOA, fronto-orbital advancement.

^aPosterior temporal width: midline to pterion; anterior temporal width: midline to sphenozygomatic suture; orbital width: nasion to zygomaticofrontal suture. Ten controls, 5 age-matched to preoperative average and 5 to postoperative average were also included. Significant *P* values are in bold.

Table 4.

Linear Regression Measuring Degree of Symmetric Improvement in Relation to Type of Procedure and Preoperative Symmetry.^a

	B Coefficients	P Value	R-Squared
Posterior temporal width			
Open vs endoscopic	−0.024	.096	0.324
Initial severity	0.562	<.001	
Anterior temporal width			
Open vs endoscopic	−0.002	.898	0.408
Initial severity	0.717	<.001	
Orbital width			
Open vs endoscopic	0.004	.695	0.436
Initial severity	0.608	<.001	
Anterior cranial fossa area			
Open vs endoscopic	−0.008	.686	0.060
Initial severity	0.227	.075	

^aFor fronto-orbital advancement vs endoscopic repair, a positive value indicates that fronto-orbital advancement was more effective. Symmetric improvement increased as preoperative symmetry decreased for all measurements. Significant *P*-values are in bold.