Single Sutural Craniosynostoses: Surgical Outcomes and Long-Term Growth

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Background: This study was designed to examine long-term skull growth following single sutural synostosis corrections and to evaluate surgical outcomes.
Methods: A retrospective outcome assessment of all children treated with a single-stage, open-remodeling procedure for sagittal, metopic, unilateral coronal, and unilateral lambdoid synostosis was performed. Long-term growth was assessed from serial anthropometric measurements taken up to 11 years postoperatively (mean, 4 years).
Results: Of 296 consecutive patients, complete records were available for 248 operative procedures. The mean surgical age was 12.3 months (range, 2.5 months to 8 years), with 36 patients older than 12 months. Transfusion rates decreased from 81 to 19 percent following the institution of blood conservation strategies. The average hospitalization was 2.5 days. There was one nonsurgically treated infection (0.4 percent), and no major complications or deaths. Five patients underwent secondary remodeling procedures (2 percent). Serial anthropometric measurements, obtained in 75 patients, showed normalization of cranial indices 6 weeks postoperatively, but subsequent measurements revealed statistically significant diminished growth. Earlier surgical treatment of metopic synostosis (4 months) was associated with significantly more growth inhibition than seen in those treated at an older age (12 months), with the other synostoses showing similar tendencies.
Conclusions: Treatment of single sutural synostosis was extremely safe with very low reoperative rates, but subsequent calvarial growth was abnormal, with a tendency toward recapitulation of the primary deformity. Growth was less diminished in procedures performed in older infants. Surgeons treating single sutural craniosynostosis should consider expanding treatment goals beyond normalization to an overcorrection of the abnormal skull shape. (*Plast. Reconstr. Surg.* 123: 635, 2009.)

It is generally recognized that infants born with multiple sutural syndromic craniosynostoses are not expected to have normal skull growth and will require sequential operations for enlargement of the calvaria, to prevent chronically raised intracranial pressure, as well as to normalize appearance. In contrast, families of patients with nonsyndromic single sutural synostosis are typically counseled that a single surgical procedure will successfully treat their child's condition, implying that subsequent growth should be normal. Considering that the surgical treatment for the single sutural synostoses typically occurs during infancy, few craniofacial surgeons have the opportunity to see patients after a substantial period of growth has occurred, 15 or 20 years after these repairs. This raises a number of questions: how good are our repairs, and do they stand the test of time? Do the single sutural synostoses have normal growth following repair? How often do significant residual asymmetries arise, and when these do

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occur (Fig. 1), are they the result of an inadequate initial repair, poor postoperative growth, or a combination of the two? Our center recently published data showing that following sagittal synostosis correction, growth of the skull is not normal and has a tendency toward recapitulation of the initial scaphocephalic skull shape.[1] This current retrospective outcome assessment was designed to expand our assessment of both surgical outcomes and long-term skull growth for all four of the single sutural synostoses.

PATIENTS AND METHODS

Exemption approval was obtained from the Institutional Review Board at Medical City Dallas Hospital before beginning this retrospective outcome assessment. The clinical group was obtained from a database containing all patients who presented to our center between 1990 and 2007 (software was designed exclusively for the Craniofacial Center by M.A. Herbert, Ph.D., Medical City Dallas Hospital). All patients were clinically diagnosed as having nonsyndromic single sutural synostoses (sagittal, metopic, unilateral coronal, or unilateral lambdoid). Exclusion criteria included initial surgery at an outside center and multiple sutural involvements. A total of 296 consecutive children were identified as meeting these criteria. Of those, completed records of 248 surgically treated patients were retrospectively reviewed (89 sagittal, 80 metopic, 64 unilateral coronal, and 15 unilateral lamboid). Data were collected including sex, age at time of surgery, length of surgery, hospital length of stay, blood loss, blood transfusions, and complications.

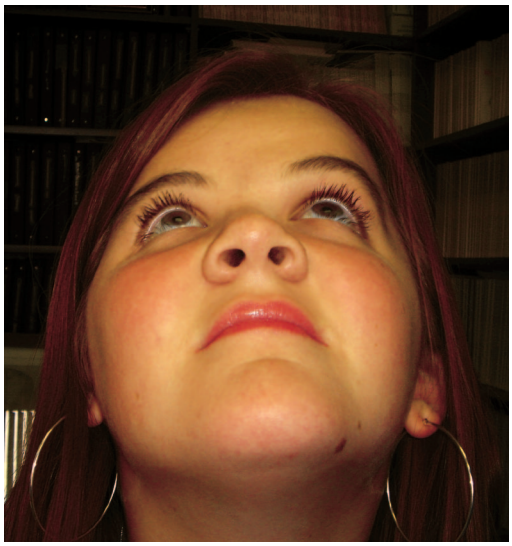

Fig. 1. Thirteen years after a right plagiocephaly correction, recession of the involved supraorbital rim is present. Is this residual deformity the result of an inadequate correction, poor growth, or a combination of the two?

Our standard treatment protocol included a detailed preoperative anthropometric evaluation as part of the patient's initial craniofacial team assessment. At approximately 6 weeks postoperatively, after a majority of the postoperative swelling had resolved, a second series of anthropometric measurements was taken to document the postsurgical alterations in cranial morphology. These postoperative measurements not only gauged how successful the operative procedure was at restoring normal anatomy, but they also served as the basis from which to assess subsequent postoperative growth. Additional measurements were obtained annually until 4 years of age and then biennially until puberty to follow any changes in the cranial vault morphology over time. Our clinical anthropologist, Dr. John Kolar, obtained all patient measurements in this series. At each evaluation, standard anthropologic protocols were followed, with multiple measurements obtained for each dimension, and these measurements were then averaged to reduce measurement error. All patients without preoperative and at least two postoperative measurements were excluded from this analysis. As a result, our final data analysis on long-term growth is based on 75 of the 248 patients we reviewed for surgical outcomes data (24 metopic, 21 unicoronal, 29 sagittal, and one unilateral lambdoid). The mean length of follow-up was 4 years, with a range of 1 to 11 years. Four specific measurements were chosen for this analysis, for they most accurately defined the dimensions of the cranial vault that are involved in the single sutural synostoses. Those dimensions were minimum frontal breadth (ft-ft), head circumference, maximum cranial length (g-op), and maximal cranial breadth (eu-eu). All anthropometric findings were compared with sex- and age-matched normal standards and converted to standard (Z) scores for comparative purposes using the formula $Z = X - \bar{X}/SD$, where X is the patient measurement, $\bar{X}$ is the sex- and age-matched normal mean value for that measurement, and SD is the SD of the mean. The pooled standardized data for the preoperative and both postoperative examinations, as well as the mean change between postoperative examinations 1 and 2 (ΔZ), were analyzed using a single-sample t test to evaluate deviations from the norm ($\bar{X} = 0$). All surgical procedures were performed at Medical City Dallas Children's Hospital from 1990 to 2007 by our senior author (J.A.F.) and one of five pediatric

neurosurgeons. All patients were treated with a single cranial vault remodeling procedure secured with absorbable suture osteosynthesis.[2] The preferred age for treatment of sagittal synostosis was 4 months of age (to prevent the development of significant frontal bossing), and for patients with unicoronal, metopic, and lambdoid synostosis, 9 months of age (old enough to achieve adequate stability of the bandeau sufficient to permit the desired overcorrection, yet young enough to ensure dural reossification of advanced bony gaps). All surgical corrections focused on the areas of the skull that were closest to the fused suture to minimize the extent of the procedure (i.e., no frontal remodeling was directly performed in the sagittal synostoses, and no total calvarial vault remodelings were performed for any of the other single sutural synostoses). In 2001, our center instituted a blood conservation protocol that entailed the use of preoperative erythropoietin administration and intraoperative blood recycling.[3,4]

All remodeling procedures were performed under general anesthesia by a pediatric anesthesiologist, with two large-bore venous catheters, an arterial line, and a Foley catheter. A first-generation cephalosporin was administered before the initiation of the surgical incision (now discontinued after one postoperative dose). Access to the calvaria was made utilizing a scallop-patterned coronal incision, which more recently has been restricted in length.[5] It is our belief, and others have shown, that the eventual coronal scar is narrower when the initial incision is made with a scalpel instead of a microneedle.[6] The osteotomy designs utilized for the various cranial vault remodeling procedures, and the fixation techniques with resorbable sutural osteosynthesis, were performed as has been previously described.[1,2] For patients over 10 months of age, an effort was made to fill in all resultant skull defects with split autogenous bone harvested from the diploic spaces of all the removed bone segments, including the bandeau. Before closure, the entire surgical area was irrigated with gentamicin solution (80 mg/liter). No drains were placed and the incision was closed in two layers using absorbable sutures. No dressings were applied, and the patients were extubated in the operating room and transferred directly to the pediatric intensive care unit for overnight observation. Over the course of this series, there was some evolution of treatment, not only with respect to more of a focus on blood conservation, but also there was a gradual shift toward achieving an overcorrection of the observed deformity, especially after recognizing the impairment in postoperative growth among our sagittal synostosis repairs.[1]

RESULTS

The surgical age ranged from 2.5 months to 8 years (mean, 12.3 months), with 36 of 248 patients over 12 months of age. After the institution of the blood conservation protocol (2001), the allogenic blood transfusion rate decreased from 81 percent to below 19 percent. The average hospitalization was 2.5 days. There were no deaths or major complications. There was one nonsurgically treated infection (0.4 percent). Five patients underwent secondary remodeling procedures (2 percent): two with unilateral coronal synostoses, two with metopic synostoses, and one with sagittal synostosis.

Analysis of the anthropometric measurements revealed that all patients had normalized cranial indices postoperatively. Subsequent postoperative measurements, however, revealed that growth was not normal after the repairs. The results of the statistical analysis are presented in Tables 1 through 13.

Trigonocephaly

In the metopic synostosis group (Table 1), minimum frontal breadth Z scores fell (with 1.0 being normal) from 1.003 postoperatively to 0.490, indicating statistically significant diminished bifrontal growth ($p < 0.05$). Similarly, the Z scores for head circumference (Table 2) fell from 1.049 to −0.193; again showing significantly diminished overall growth ($p < 0.001$). When we

Table 1. Metopic Synostosis: Growth in Minimum Frontal Breadth (ft-ft) (Z Scores) (*n* = 24)

	Preop	Surgery	Postop 1	Postop 2	ΔZ_2
Mean	−0.359		1.003	0.490	−0.425
SD	0.895		1.127	0.767	0.822
t	1.924		4.268	3.064	2.480
p	NS		<0.001	<0.01	<0.05
Age, yr					
Mean	7.2	7.7	11.3	49.1	
Range	2–25	3–25	5–28	17–97	

Table 2. Metopic Synostosis: Growth in Head Circumference (Z Scores) (*n* = 24)

	Preop	Postop 1	Postop 2	ΔZ
Mean	0.985	1.049	−0.193	−1.242
SD	1.481	1.274	1.214	1.299
t	3.258	4.034	0.779	4.585
p	<0.01	<0.001	NS	<0.001

Table 3. Metopic Synostosis: Postoperative Growth by Severity (*n* = 16)

	Most Severe		Least Severe			
Measure	**Mean**	**SD**	**Mean**	**SD**	***t***	***p***
ft-ft	−0.528	0.830	−0.289	1.008	0.517	NS
circ	−1.540	1.800	−1.089	1.079	0.608	NS

Table 4. Metopic Synostosis: Postoperative Growth by Age at Surgery (*n* = 16)

Measure	**Mean**	**SD**	**Mean**	**SD**	***t***	***p***
ft-ft	−0.940	0.726	0.214	0.577	3.518	<0.01
circ	−1.594	1.552	−1.236	1.383	0.486	NS

NS, not significant.

compared the eight patients with the narrowest minimum frontal breadth (the most severely affected) with the eight patients who had the least narrowed minimum frontal breadth (the mildest), the postoperative growth was more than twice as diminished in the more severely affected group (−0.528 versus −0.289), but this difference was not significant (Table 3). When we examined postoperative growth according to the age of surgery (Table 4), those eight patients operated on at the earliest age (4 months) showed more than four times the growth impairment seen in the eight oldest patients at the time of surgery (12 months), with Z scores of −0.940 and 0.214, a statistically significant difference ($p < 0.01$).

Plagiocephaly

Unilateral coronal synostosis repaired children (Table 5) showed findings similar to those who underwent metopic repairs; there was a statistically significant reduction in growth in head circumference, with Z scores falling from 0.879 to 0.030 ($p < 0.001$). When we compared the seven youngest patients to undergo surgical correction (5 months) with the oldest seven patients (14 months), growth in head circumference was more than twice as impaired in the earlier treatment group (−0.877 versus −0.420); however, this difference was not statistically significant (Table 6).

Scaphocephaly

Sagittal synostosis repairs also showed significantly diminished growth in head circumference (Table 7), with Z scores falling from 2.714 to 1.099 ($p < 0.001$). In this group, maximum cranial length and breadth were likewise diminished (Tables 8 and 9), with Z scores falling from 1.085 to 0.526 ($p < 0.01$) and 0.922 to −0.137 ($p < 0.001$), respectively. Notably, the restriction in growth in width was twice that of the restriction in length, leading to a regression of the cephalic index with a tendency toward a recapitulation of the initial scaphocephalic skull shape. Minimum frontal breadth (one marker for frontal bossing) showed very slightly diminished growth (Table 10), which was not significant. There was no difference in growth for any of the measured indices based on the severity of the initial presentation (Table 11). When we compared the seven patients treated at the youngest age (2.5 months) to the seven patients treated at the oldest age (15 months), the growth in head circumference and maximal cranial breadth (Table 12) were more than twice as impaired in those patients operated on at an earlier age (−2.341 versus −1.106, and −1.231 to −0.553); however, this difference was not significant.

Table 5. Unicoronal Synostosis: Growth in Head Circumference (Z Scores) (*n* = 21)

	Preop	**Surgery**	**Postop 1**	**Postop 2**	**ΔZ**
Mean	0.726		0.879	0.030	−0.772
SD	1.451		1.111	1.347	0.736
T	2.293		3.623	0.102	4.690
p	<0.05		<0.01	NS	<0.001
Age, yr					
Mean	7.9	8.9	12.0	60.5	
Range	4–26	4–26	7–28	34–125	

NS, not significant.

Table 6. Unicoronal Synostosis: Postoperative Growth in Head Circumference by Age at Surgery (*n* = 14)

	Early (mean = 5 mo)		Late (mean = 14 mo)			
Measure	**Mean**	**SD**	**Mean**	**SD**	***t***	***p***
circ	−0.877	0.677	−0.420	0.542	1.187	NS
ft-ft	0.146	1.247	0.057	1.248	0.223	NS

NS, not significant.

Posterior Plagiocephaly

Only one patient with lambdoid synostosis had serial measurements, and these measurements did show diminished postoperative growth in both head circumference and cranial breadth, with negative ΔZ scores between the first and second postoperative measurements (Table 13).

DISCUSSION

The final result of any single sutural craniosynostosis repair performed during infancy will not be fully realized until growth is complete. Yet,

Table 7. Sagittal Synostosis: Growth in Head Circumference (Z Scores) (*n* = 29)

	Preop	Surgery	Postop 1	Postop 2	ΔZ
Mean	3.661		2.714	1.099	−1.614
SD	1.226		1.367	1.411	1.489
t	16.081		10.692	4.194	5.837
p	<0.001		<0.001	<0.001	<0.001
Age, yr					
Mean	6.2	6.7	10.6	65.7	
Range	1–34	2–35	4–43	40–116	

Table 8. Sagittal Synostosis: Growth in Maximum Cranial Length (g-op) (Z Scores) (*n* = 29)

	Preop	Postop 1	Postop 2	ΔZ
Mean	2.201	1.085	0.526	−0.549
SD	0.777	1.042	1.336	0.992
t	15.255	5.607	2.120	2.980
p	<0.001	<0.001	<0.05	<0.01

Table 9. Sagittal Synostosis: Growth in Maximum Cranial Breadth (eu-eu) (Z Scores) (*n* = 29)

	Preop	Postop 1	Postop 2	ΔZ
Mean	0.407	0.922	−0.137	−0.772
SD	0.839	1.158	1.393	0.736
t	2.612	4.288	0.530	4.690
p	<0.02	<0.001	NS	<0.001

Table 10. Sagittal Synostosis: Growth in Minimum Frontal Breadth (ft-ft) (Z Scores) (*n* = 29)

	Preop	Postop 1	Postop 2	ΔZ
Mean	1.786	1.324	1.273	−0.051
SD	0.792	1.128	1.061	1.155
t	12.144	6.321	6.461	0.238
p	<0.001	<0.001	<0.001	NS

Table 11. Sagittal Synostosis: Postoperative Growth by Severity (*n* = 14)

	Most Severe > 3 SD		Least Severe < 1.5 SD			
Measure	Mean	SD	Mean	SD	*t*	*p*
circ	−0.831	1.436	−1.879	1.195	1.430	NS
ft-ft	−0.239	0.925	−0.156	1.430	0.129	NS
g-op	−0.324	0.934	−0.634	0.918	0.626	NS
eu-eu	−0.551	1.217	−1.490	0.832	1.684	NS

NS, not significant.

Table 12. Sagittal Synostosis: Postoperative Growth by Age at Surgery (*n* = 14)

	Early (mean = 2.4 mo)		Late (mean = 15.3 mo)			
Measure	Mean	SD	Mean	SD	*t*	*p*
circ	−2.341	1.628	−1.106	1.379	1.532	NS
ft-ft	−0.541	0.708	−0.160	0.363	1.268	NS
g-op	−0.974	1.116	−0.616	0.922	0.655	NS
eu-eu	−1.231	0.520	−0.553	1.158	1.415	NS

NS, not significant.

Table 13. Unilateral Lambdoid Synostosis: Postoperative Changes (Z Scores)

	Preop	Postop 1	Postop 2	ΔZ
Cranial breadth	−0.05	0.47	−1.07	−1.54
Cranial length	−2.80	−3.26	−3.19	0.07
Head circumference	−2.46	−1.88	−2.98	−1.10

until most surgeons approach retirement, it is more likely that they would see a patient 10 to 15 years after a craniosynostosis correction performed by someone else, rather than one of their own patients. It is a natural tendency to assume that any unfavorable outcome observed, following a repair by another surgeon, is the result of an inadequate initial repair. When patients present with suboptimal results, how can we assess whether the observed asymmetries are from an inadequate surgical correction or the result of poor growth following a perfect repair? There are only a few published studies that have examined outcomes following cranial vault remodeling for single sutural synostosis, and most have limited sample sizes or short-term follow-ups.[7–12] Through this retrospective review of 248 single sutural craniosynostosis repairs, we sought to critically evaluate our long-term outcomes, as well as to analyze skull growth following correction. In this series, our patients were followed for up to 11 years postoperatively, and 75 of 248 had the requisite series of anthropologic measurements required to assess whether or not identifiable changes in skull configuration were the result of surgical technique or altered postoperative growth. We found that the surgical procedures performed to correct the deformities associated with a single sutural synostosis resulted in successful normalization of measured cranial indices with no deaths, and with very low infection (0.4 percent) and reoperative (2 percent) rates. Given that the average length of follow for all the patients in this series was approximately 8 years, we recognize that reoperative rates could potentially rise with longer follow-up. The senior author's current criterion for reoperation is to only consider treatment for those patients sufficiently concerned with their appearance that they, or their parents, desire an operative procedure to correct the deformity. Nevertheless, these data compare very favorably with previous studies, which have reported complication rates varying between 2.5 and 13 percent, and reoperation rates

as high as 12 to 20 percent.[12–18] Furthermore, our blood conservation protocol, which was instituted in 2001, has reduced our blood transfusion rate to below 19 percent, which is significantly lower than that in any other published series.[3,4,19–21] We believe that a high volume of patients, which influences the entire team's level of experience, may have contributed in part to our extremely low complication rates; however, advances in anesthesiology and critical care have certainly also played a role in these favorable outcomes.

The assessment of aesthetic results following cranial vault remodeling is a challenging endeavor. Other authors have relied on various subjective analyses, including photographic reviews, parental satisfaction surveys, and classification schema that are based on the need for subsequent surgical interventions.[10,11,14–16,18,19,22] In an attempt to bring greater objectivity into the postoperative analysis of all the single sutural synostoses (and to avoid using computed tomography data, which would result in potentially unnecessary brain irradiation), we chose to rely on direct surface anthropometric measurements.[23] Although anthropometry can provide reproducible data, this methodology is limited to particular surface landmarks, and we found that our measurements were not able to assess the more subtle asymmetries. Despite these minor shortcomings, we were able to demonstrate that with a single, regionally limited surgical procedure (i.e., no total vault remodeling procedures were performed), normalization of established anthropologic measurements was achieved in all patients. Moreover, we found statistically significant impaired postoperative growth for all the single sutural synostoses, aside from lambdoid synostosis. Among our lambdoid synostosis patients, we have noticed clinically impaired growth (Fig. 2); however, only one of our patients had a complete set of serial measurements (which, nevertheless, did show abnormal postoperative growth). Although it might appear that the surgical procedures performed in this series were successful, given our measured postoperative indices, our observation that growth is not normal following correction, as well as our 2 percent secondary surgical procedure rate, suggests that our results could be further improved. These findings of impaired postoperative growth raise the question: is the observed growth disturbance a result of the primary process leading to sutural fusion, sequelae of the surgical procedure, or a combination of both? An analysis of our data shows that the greatest impairment of growth appears to be perpendicular to the fused suture,

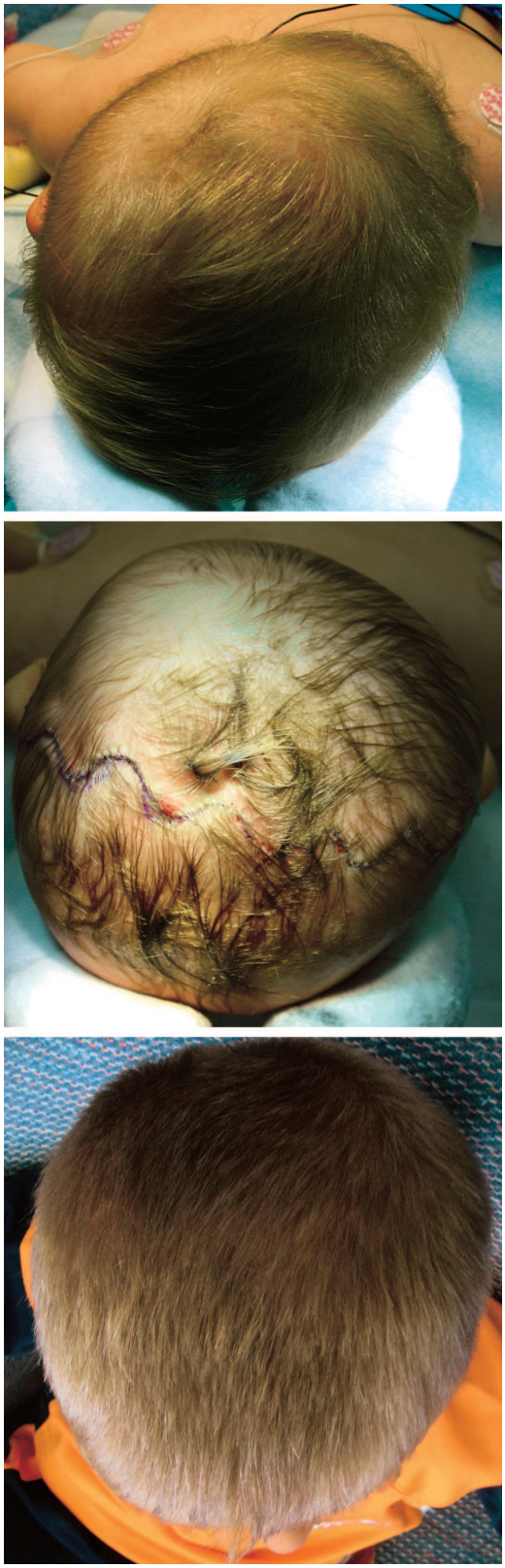

Fig. 2. (*Above*) Preoperative view of an infant with right lambdoid synostosis; (*center*) immediate postoperative view, showing a slight overcorrection; (*below*); 2 years postoperatively, a subtle recurrent flatness can be appreciated on the corrected side.

which implicates either the primary process that led to the sutural fusion or the inability to surgically recreate a functioning suture. With subsequent growth, we have found that there is a tendency for the skull to recapitulate the presenting deformity (Fig. 3). We believe, but were unable to show with statistical significance, that growth inhibition correlates with the severity of the deformity. We did, however, discover a correlation between operations performed at an earlier age and poorer postoperative growth (this finding was statistically significant only for metopic synostosis corrections, although our data appear to show this might also hold true for unilateral coronal and sagittal synostosis corrections). It seems intuitive that if growth is not normal following correction, later corrections will be more likely to achieve a reconstruction that more closely approximates that of a fully mature skeletal anatomic position. The findings of impaired growth following single sutural craniosynostosis corrections should not be surprising. If the observed skull deformity with craniosynostosis is secondary to a nonfunctioning suture, and the surgical procedure performed is incapable of creating a functioning suture, why should one expect normal growth postoperatively? Nevertheless, our data do not eliminate the possibility that surgery might have some negative effect on growth, which keeps open the possibility that earlier operations might contribute to a longer period of subsequent growth impairment.

How can surgeons use these findings to improve their results? If growth is not normal following a single sutural synostosis correction, then any correction performed with the goal of normalizing cranial shape will likely result in a long-term undercorrection. Given our findings, a significant overcorrection is required if the surgical goal is to normalize appearance with a single procedure, and the degree of overcorrection should be greater in younger children, who have more growth ahead of them. Furthermore, we believe that these data raise questions about endoscope-assisted strip craniectomies, which have a significant reliance on postoperative molding therapy to achieve corrections in skull shape.[24] Is it possible to attain the necessary overcorrection with helmets or banding therapy? Another unanswered question concerns the timing for surgical intervention for the treatment of the single sutural synostoses. We have noted that later corrections appear to result in better postoperative growth (or less of a recapitulation of the presenting deformity), which suggests that delaying surgery may result in better long-term outcomes with respect to appearance. How long should surgeons delay operative treatment for the single sutural synostoses? This decision must certainly take into account concerns for potential impairment of brain function and development that might result from the de-

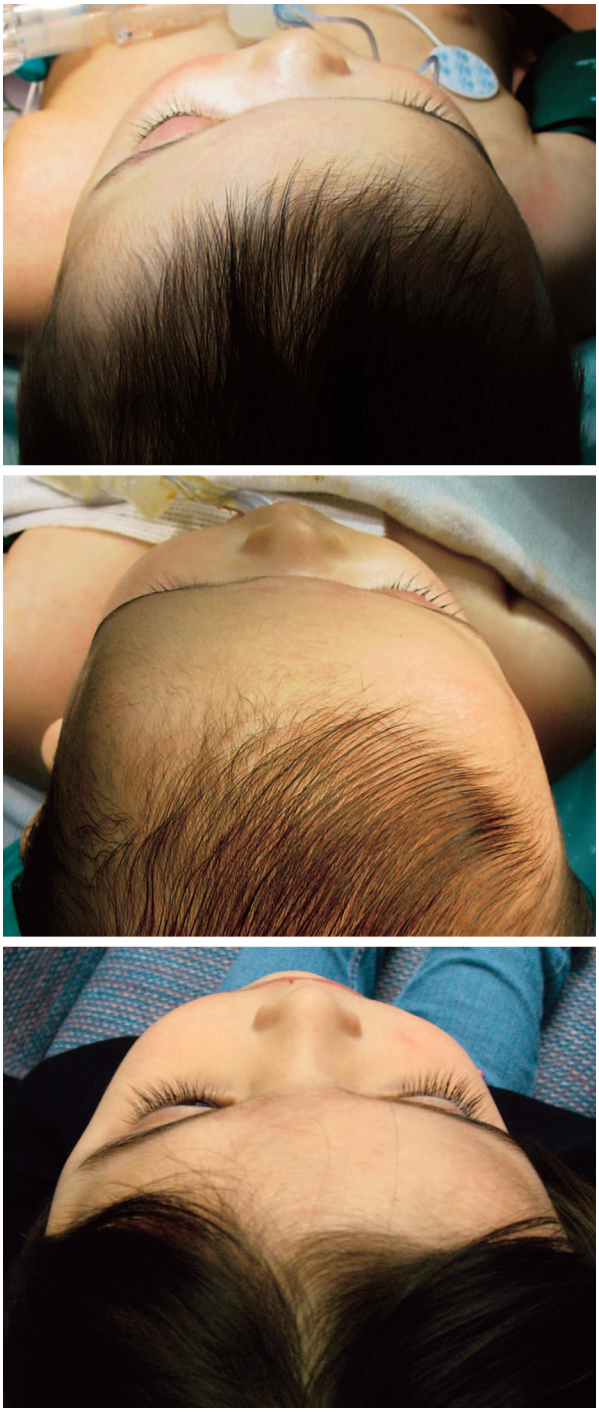

Fig. 3. (*Above*) Preoperative view of an infant with left coronal synostosis; (*center*) immediate postoperative view, showing a slight overcorrection of the affected side; (*below*) 3 years postoperatively, a slight left-sided recurrence is evident.

creased skull compliance caused by sutural fusion, as well as the need to fully reconstruct any advanced skull defects in children over 10 months of age because the dura will not regenerate bone as readily, and balance these concerns with long-term aesthetic goals.

CONCLUSIONS

This retrospective outcome analysis of children with nonsyndromic single sutural synostosis, all treated with a single regional cranial vault remodeling procedure, showed normalization of cranial indices with low reoperative rates and exceptionally few complications. We found that growth is not normal following surgical correction, with a tendency for the calvaria to revert toward the primary deformity. Surgeons treating patients with single sutural craniosynostosis should expand their treatment goals beyond normalization to an overcorrection of the abnormal skull shape.

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