Long-term 3D CT follow-up after endoscopic sagittal craniosynostosis repair

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OBJECTIVE Surgical treatment of sagittal synostosis involves various surgical modalities. Long-term follow-up issues include increased intracranial pressure, secondary sutural fusion, incomplete reossification, and suboptimal cosmetic appearance. The authors' objective in this study was to review their long-term endoscopic surgical results in children with sagittal synostosis using 3D CT.

METHODS The authors reviewed the long-term results of their first 38 patients who underwent endoscopic sagittal synostosis repair at age 16 weeks or younger. A standard vertex craniectomy with biparietal wedges was done in each case. After surgery, the children were fitted with a helmet, which they wore until 8 months of age. Patients were followed up for 5 years or longer, at which point a 3D CT scan was obtained. The authors examined data on the cranial index, area of bony defect, presence or absence of secondary sutural fusion, neosuture formation, and scalloping of the inner table of the skull.

RESULTS Thirty-two of 38 children met inclusion criteria. There was a small but significant recession of the cranial index after the completion of helmeting (from 0.772 after completion of helmeting to 0.755 at 5 years). Of 32 children, 14 had a bony defect area > 4 cm². Three children had secondary sutural fusion (two unilateral coronal, one bicoronal). Ten of 32 patients had partial neosuture formation.

CONCLUSIONS The authors report their experience with 32 of their first 38 children who underwent endoscopic sagittal synostosis repair at 16 weeks of age or younger. With a minimum duration of 5 years, this is the longest clinicoradiological follow-up utilizing 3D CT to date in children with sagittal synostosis treated with endoscopic surgery. The authors report detailed measurements of bony loss, adjacent sutural fusion, and neosuture formation.

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KEYWORDS craniosynostosis; sagittal synostosis; scaphocephaly; endoscopic; reossification; cranial index; craniofacial

CRANIOSYNOSTOSIS is the premature fusion of one or more cranial sutures. Sagittal synostosis is the most common form of craniosynostosis. There are many surgical approaches used to treat scaphocephaly. These are divided into open surgery and endoscopic surgery. Cosmetic outcomes are similar between endoscopic and open surgery.^{4,7,12,13,15,16,21,29,30,33,38,41} Modifications to the endoscopic technique, including barrel staves, spring-assisted devices, and cinching sutures, have been reported.^{17,35,40} Endoscopic surgery is associated with less blood loss, lower transfusion rates, decreased hospital length of stay (LOS), and decreased complication rates,^{5,11,18,41} as well as lower costs^{1,39} and less stress on parents.²⁰

The neurosurgical follow-up issues for children who

have had craniosynostosis surgery include cosmetic outcome, incomplete reossification, adjacent sutural disease, and elevated intracranial pressure (ICP). Cosmetic outcome and cranial index have been well studied following craniosynostosis surgery. In the short term, endoscopic surgery leads to cosmetic improvement. However, there is a recession of the cranial index over time toward the scaphocephalic phenotype.^{2,13,28} There are few studies addressing long-term surgical outcomes.

Methods

Institutional research ethics board approval was obtained for this study.

ABBREVIATIONS ICC = intraclass correlation coefficient; ICP = intracranial pressure; LOS = length of stay.

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This study was a single-center consecutive series of our first 38 children with isolated nonsyndromic sagittal synostosis in whom an endoscopic technique was used. Only children treated at age 16 weeks of age or younger were included. All children had a minimum 5-year follow-up duration. At 5 years or later, children underwent 3D CT scanning to characterize bony defects and skull morphology. The risks of radiation were discussed with families prior to booking the CT scan. Children who did not have clinical follow-up and a completed 3D CT scan were excluded.

All children were primarily operated on by a single surgeon (V.M.). Patients were positioned prone with a slight degree of head extension. Two 4-cm incisions were made, an anterior incision just posterior to the anterior fontanelle and a posterior incision just anterior to the lambdoid/sagittal junction. Following a vertex craniectomy (2.5 cm wide) with the assistance of an endoscope, small bilateral wedge craniectomies were completed. These were 1.5 cm in width and 3.5 cm in length and positioned at the anterior and posterior margins of the vertex craniectomy.

Within 4 weeks of surgery, the patients were sent for custom cranial orthotic fitting and began wearing the helmet 23 hours per day. They were followed monthly during the helmeting process, which took place until the patients were 8 months of age.

Following the conclusion of helmet therapy, each patient was followed up in the clinic yearly and measurements of head circumference and cephalic index were taken. At the 5-year follow-up visit, a 3D CT scan was offered; as stated, the risks of radiation were discussed with families.

Cranial indices were compared using one-way ANOVA. Paired t-tests were used to compare differences at each time point. Cranial indices were measured preoperatively, immediately following helmet removal, at the 1-year follow-up visit, and at the 5-year follow-up visit. IMPAX software was used for analysis of 3D CT scans. Cranial index, area of bony loss, evidence of neosuture formation, evidence of adjacent sutural fusion, and evidence of scalloping of the inner table of the skull were assessed.

Areas of bony loss were measured using the free-form region-of-interest tool. We defined incomplete ossification as any skull defect $> 0.5 \text{ cm}^2$. The two largest areas of bony loss $> 0.5 \text{ cm}^2$ were measured by two observers (V.M. and A.P.). The intraclass correlation coefficient (ICC) was computed with 95% confidence interval using SPSS software (version 24, IBM Corp.) to ensure interrater reliability of the measured data. Scalloping, neosuture formation, and adjacent sutural fusion were determined visually by both observers.

Results

We identified our first 38 patients who underwent endoscopic strip craniectomy at 16 weeks of age or younger. These patients had a minimum of 5 years of follow-up. Six patients were excluded for refusal to undergo 3D CT scanning (3 patients) or loss to follow-up (3 patients). Thirty-two patients were included (Fig. 1). The mean age at surgery was 12.5 ± 2.6 weeks. Twenty-five patients (78%) were male and 7 (22%) were female. The mean LOS was

FIG. 1. CONSORT diagram detailing operative numbers and patient inclusion/exclusion. Thirty-eight of 42 candidates met age inclusion criteria. Of these, 32 completed 5-year clinical/3D CT follow-up and were included in the study.

2.4 ± 0.5 days. The mean duration of helmeting was 4.7 ± 1.6 months, with a mean of 1.4 ± 0.5 helmets used during that period. The mean age at the time of helmeting therapy completion was 8.5 ± 1.8 months, and the mean age at the time of 3D CT scan acquisition was 5.8 ± 1.2 years (Table 1). No children had dural tears, sinus injury, or deep postoperative infections.

The mean preoperative cranial index was 0.670, which corrected to 0.772 at the time of completion of helmet therapy. The mean cranial index at the 1-year postoperative follow-up was 0.760, and the mean cranial index measured on the 3D CT scan at the 5-year follow-up was 0.755 (Table 2). This was comparable to clinical measurement at the time of the 3D CT scan (0.756), with no statistical difference between measurement methods ($p = 0.567$). The difference between groups was significant ($p < 0.001$) on one-way ANOVA. Paired t-tests between groups revealed significant differences when comparing preoperative and posthelmeting data ($p < 0.001$) and when comparing posthelmeting and 1-year follow-up data for the cranial index ($p = 0.003$). The paired t-test showed no statistically significant difference between the cranial index at the 1-year follow-up visit and that at the time of the 3D CT

TABLE 1. Demographic and follow-up data

Demographic & Clinical Data	Value
No. of patients	32
Age at surgery, wks	12.5 ± 2.6 (8.9–16)
Male, no. (%)	25 (78)
Female, no. (%)	7 (22)
LOS, days	2.4 ± 0.5 (2–3)
Helmet duration, mos	4.7 ± 1.6 (3–12)
No. of helmets worn	1.4 ± 0.5 (1–2)
Age at end of helmeting, mos	8.5 ± 1.8 (7.1–11.2)
Age at 3D CT, yrs	5.8 ± 1.2 (4.5–9)

Values are presented as the mean ± SD (range), unless otherwise stated.

scan ($p = 0.392$) (Fig. 2). The ICC of cranial index measurements was 0.85 (95% CI 0.82–0.88), which indicates good interrater reliability.

Twenty-seven of 32 children had some amount of bony defect as defined in *Methods* (Fig. 3). Fifteen of those 27 children had at least two areas of incomplete ossification. The range of bony defects was 0.6–13.5 cm². Of the smaller measured defect, the mean area was 0.96 cm². Of the larger measured defect, the mean area was 3.03 cm². Fourteen (44%) of 32 children had a total area of bony loss > 4 cm², and 9 (28%) of 32 children had a single skull defect > 4 cm² (Table 3). Two children had two separate areas of bony defect that were each > 4 cm². Five children had no single defect > 4 cm² but had a total area of the defect > 4 cm². The ICC values for bony area loss were 0.99 (95% CI 0.98–1) and 0.97 (95% CI 0.95–0.99) for the first and second areas, respectively, which indicates good interrater reliability. Among 14 children with > 4-cm² defect, consideration was given to surgical correction. Of these, only 3 patients underwent surgical repair of their cranial defect. This was done by split-thickness cranioplasty using autologous bone. There was a predilection of bony defects to occur along the vertex craniectomy rather than the wedge osteotomies.

Three children had adjacent sutural fusion (one left coronal, one right coronal, and one bicoronal) (Fig. 4). Ten children (31%) had at least partial neosuture formation at the sagittal suture. Only 2 children had scalloping of the inner table of the skull (Table 4). None of the children

exhibited clinical symptoms concerning for elevated ICP during follow-up. No children required a repeat operation due to poor cosmetic outcome, including those who had secondary sutural fusion.

Discussion

There have been few reports of long-term outcomes in endoscopic craniosynostosis surgery.^{2,6,13,28} Using clinical and imaging measures, Pickersgill et al.²⁸ found a small but significant regression of 0.017 in the cranial index and adjusted cranial index⁹ after helmeting. Isaac et al.¹³ also observed a greater regression in cranial index in an endoscopically treated group compared with an open surgically treated group. In the present study, we found a small regression, from a 1-year post-helmet therapy follow-up cranial index of 0.012, which was the peak, to a long-term follow-up cranial index of 0.017. This did not manifest clinically, and no patient needed a reoperation for a poor cosmetic outcome.

Secondary sutural fusion is a known sequela of craniosynostosis surgical repair.³ Three of 32 children had secondary sutural fusion seen on follow-up imaging. No child had a clinically significant cosmetic deformity or required surgery for repair of secondary sutural fusion. We did not test these children for genetic syndromes.

Elevated ICP occurs in 5%–10% of children as a late complication of craniosynostosis surgery.³⁷ In our series, no children were symptomatic with elevated ICP during clinical follow-up. Two children did have scalloping of the inner table of the skull, which was noted on secondary imaging review in 2 of 3 children with adjacent sutural fusion. We determined scalloping to be present because there were indentations diffusely along the inner table of the skull. This was beyond the usual convolutional appearance seen in other children. The significance of this finding is unclear. In these 2 children, there were no symptoms or signs of elevated ICP. These children were not poor responders to surgery, with a cranial index improvement of 0.085 and 0.117, respectively. In addition, they had no evidence of a head circumference growth that was static or below the 50th percentile. We will continue with clinical and ophthalmological follow-up of these children indefinitely. If any symptoms or signs of elevated ICP develop, then we will consider ICP monitoring.

Recently, partial or complete reformation of the sagittal

TABLE 2. Pre- and posttreatment age and cranial index values

Time Point	Age	Cranial Index	p Value (paired t-test vs preop)	p Value (paired t-test vs post-helmeting)	p Value (paired t-test vs 1-yr FU)	p Value (paired t-test vs 3D CT)	p Value (ANOVA)
Preop	2.1 ± 0.2 mos	0.670 ± 0.006					
Posthelmeting	8.5 ± 0.3 mos	0.772 ± 0.007	<0.001				
At 1-yr clinical FU	1.6 ± 0.1 yrs	0.760 ± 0.006	<0.001	0.003			
At 5-yr clinical FU	5.8 ± 0.2 yrs	0.756 ± 0.005	<0.001	0.003	0.401		
At 3D CT	5.8 ± 0.2 yrs	0.755 ± 0.007	<0.001	0.002	0.392	0.567	<0.001

FU = follow-up.

Mean values are presented ± SEM.

FIG. 2. Graph depicting cranial index at measured time points. *Error bars* represent standard error of the mean. One-way ANOVA showed a statistical change between time points ($p < 0.001$). Matched t-test showed no statistical difference between cranial indices at the 1-year follow-up and at the time of 3D CT ($p = 0.392$), with the remainder significantly different. The ICC was 0.85 (95% CI 0.82–0.88). Asterisks indicate statistical significance ($p < 0.05$) for the time points being compared. Figure is available in color online only.

suture, or so-called neosuture formation, has been reported,³² with an incidence of approximately 25%. The significance of the neosuture is uncertain. Ten patients (31.3%) had at least partial neosuture formation demonstrated on 3D CT scans in our series.

Few studies have addressed incomplete ossification following craniostomy surgery. Noordzij and colleagues reviewed the literature and found rates of incomplete reossification ranging from 0.5% to 18.2%.²⁶ They identified dural injury and infection as risk factors. Thenier-Villa et al. performed an analysis of 216 patients. In their series, 51.8% of patients who had undergone 3D CT had incom-

plete reossification. Their reoperation rate due to bone defects was 5.56%. Their findings were limited by the availability of the 3D CT scans (94 had recoverable scans, 101 had unrecoverable scans). They found larger postoperative defect and younger age to be risk factors.³⁶ Age has inconsistently been reported as a risk factor for bone loss.^{10,27,31,36}

We observed 27 children (84%) with at least a 0.5-cm² calvarial defect and 14 (44%) with a combined area of loss > 4 cm². Our prevalence of bony defects is higher than those previously reported. Three children (9.4%) underwent surgery for a skull defect, which is higher than the rate reported by Thenier-Villa et al.³⁶ The reoperation

FIG. 3. Representative 3D CT scans showing an insignificant bony defect (**A**; < 0.5 cm²), a bony defect (**B**; > 0.5 cm² but < 4 cm²), and a large bony defect (**C**; > 4 cm²).

TABLE 3. Cases of 3D CT–detected reossification failure

Bony Defect	No. of Children w/ Defect	Mean Defect Area, cm ²	No. of Children w/ Defect Area >4 cm ²
Defect 1	27	3.03 ± 0.51 (0.75–8.83)	9
Defect 2	15	0.96 ± 0.29 (0.55–6.77)	2
Total		3.98 ± 0.69 (0.75–13.53)	14

The defect area is presented as the mean ± SEM (range). Two children had two separate areas of bony defect, both > 4 cm². Five children had no single defect > 4 cm² but had a total area of defect > 4 cm².

rates for endoscopic suturectomy and open cranial vault remodeling are around 3% and 1.6%–3.7%, respectively,¹³ compared to our rates, which are higher. All patients were operated on at 16 weeks of age or younger. Our craniectomy size was relatively wide at 2.5 cm. Some authors have tended toward a smaller craniectomy size.^{8,25} The areas of incomplete ossification were mainly in the area of the strip craniectomy and not at the site of the wedge osteotomies. This potentially implicates the wide vertex craniectomy as a source of incomplete reossification.

We used 3D CT scans for the assessment of ossification. Some authors have argued that a defect not detectable by finger palpation is not significant.^{24,27} The positive predictive value of finger palpation is 50% for defects > 4 cm² and 71% for smaller defects. In a study of follow-up patterns for craniosynostosis surgery, 4 of 9 surveyed surgeons used CT scans as a follow-up tool.¹⁹ Each 3D CT scan of the head emits 0.57 mSv of radiation. Modifications exist to reduce this radiation dose.²⁴ A chest CT scan, which is considered to emit a safe radiation dose, has an exposure of 2 mSv, and the average yearly atmospheric exposure in Canada is 2–3 mSv/year. Though the imaging did not affect the clinical course of the majority of children, it did guide follow-up for children with large defects or with scalloping of the inner table of the skull. We will continue to offer 3D CT scans for this reason, though we are uncertain of their clinical relevance.

Postoperative duration of helmet therapy is not standardized. In most published series, the period of helmeting lasts between 8 and 12 months. The authors of one report described a shorter period of 6 months.⁷ Two studies have reported on the nonoperative treatment of craniosynostosis with helmet only.^{23,34} A recent study tracked the cranial index during helmet therapy. After the maximum cranial index had been achieved, there was no benefit to further helmet therapy.¹⁴ The maximum cranial index occurred between 7 and 9 months postoperatively.

We used a shorter duration of helmet therapy with a goal of 8 months of age. This resulted in a mean of 4.7 months of helmet therapy, when patients were a mean age of 8.5 months. Our reasoning for this was twofold. First, in Alberta there is a significant population spread with limited access to helmet therapists. A shorter duration of helmet therapy would potentially decrease parents' financial and travel burdens. Second, according to head growth models, there is a decrease in the velocity of cranial expansion after 7–8 months.²² Currently we continue helmet therapy for our children until they are 8 months of age.

FIG. 4. A and B: Examples of secondary sutural fusion demonstrating a unilateral right coronal fusion.

There are multiple limitations of this study. This is a single-center study. Our sample size was limited to long-term follow-up of 32 of our first 38 endoscopic sagittal synostosis cases (early in the series) performed by the senior surgeon (V.M.) of a total of more than 100 cases to date. We were unable to include 6 patients due to refusal to undergo long-term 3D CT follow-up scanning (3 patients) or loss to follow-up (3 patients). We did not include data for the 3 cases in which 3D CT scanning was refused, but there were no clinical issues with the long-term follow-up for these 3 children. We used the cranial index as the measure for our study. More recently, the adjusted cranial index has been used to account for altered euryon location in children.^{9,28}

The main purpose of this study was to review the long-term outcomes achieved with endoscopic sagittal synostosis repair. We have now considered narrowing our strip craniectomy and excluding the wedge osteotomies in our surgical technique. It is unclear if this will improve our rate of reossification failure. Another consideration is whether performing 3D CT scanning may impact individualized follow-up for these children. For children with good outcomes and no significant finding on 3D CT scans, further follow-up may be unnecessary. For children with secondary sutural fusion, scalloping of the inner table of the skull, or very large bony defects, continued follow-up may be necessary.

Conclusions

We report our long-term experience with 32 of our first 38 children who underwent endoscopic sagittal synostosis repair at the age of 16 weeks or younger. To our knowledge, this is the longest reported follow-up with 3D CT

TABLE 4. Other 3D CT–detected findings

Finding	No. of Patients (%)
Neosuture formation	10 (31.3)
Adjacent sutural fusion	3 (9.4)
Scalloping	2 (6.3)

to address cranial index, bone loss, adjacent sutural fusion, and neosuture formation. More research is needed to qualify these outcomes and to determine their relevance.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Mehta. Acquisition of data: Persad, Aronyk, Beaudoin. Analysis and interpretation of data: Mehta, Persad. Drafting the article: Persad. Critically revising the article: Mehta, Persad. Reviewed submitted version of manuscript: Mehta, Persad. Statistical analysis: Persad. Administrative/technical/material support: Mehta, Aronyk, Beaudoin. Study supervision: Mehta.

Supplemental Information

Previous Presentations

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