Suturectomies Assisted by Cranial Orthosis Remodeling for the Treatment of Craniosynostosis Can Be Performed Without an Endoscope

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Background: Minimally-invasive endoscopic strip-craniectomy (or suturectomy) for the repair of craniosynostosis combined with postoperative cranial orthotic molding has been widely adopted in the past 2 decades, proving itself as a safe and effective procedure. Over time the authors transitioned from performing an endoscopic strip-craniectomy, to performing the same surgery without the endoscope. The authors here describe our technique and compare its results to those published in the literature for endoscopic suturectomies.

Methods: A retrospective chart review was performed for patients with nonsyndromic craniosynostosis who underwent minimally-invasive nonendoscopic suturectomy between 2019 and 2020 at our institution.

Results: Thirteen patients (11 males; 2 females) were operated including 5 Metopic, 5 Sagittal, 2 coronal, and 1 lambdoid craniosynostosis. The average age at surgery was 4.35 months. The average length of surgery was 71 minutes. Averaged intraoperative estimated blood loss was 31.54 mL. Eleven patients received a blood transfusion (most before performing the skin incision) with a mean amount of 94.62 mL of blood transfused during surgery. The mean hemoglobin at discharge was 10.38 mg/dL. There was only 1 intraoperative mild complication. The mean intrahospital length of stay was 1.77 days with no postoperative complications noted. All patients initiated remodeling orthotic treatment following surgery. Long-term follow-up scans were available for 8 patients (5 metopic, 2 sagittal, and 1 lambdoid) with an average follow-up of 9 months. In all cases, there was a significant improvement in the skull width at

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- AO and LH-S have equal contribution to the manuscript.
- Author AK has a business relationship with Orthomerica Products, Inc in Mexico.
- The authors report no conflicts of interest.
- Supplemental digital contents are available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF versions of this article on the journal's Web site (www.jcraniofa-cialsurgery.com).

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ISSN: 1049-2275

DOI: 10.1097/SCS.000000000007943

the synostosis location as well as in the skull proportions and symmetry. The above outcomes are similar to those published in the literature for endoscope-assisted strip-craniectomies.

Conclusions: Suturectomies assisted with cranial orthosis remodeling for the treatment of all types of nonsyndromic craniosynostosis can be performed without an endoscope while maintaining minimal-invasiveness, good surgical results, and low complication rates.

Key Words: Craniosynostosis, minimally-invasive, orthotic helmet, strip-craniectomy, suturectomy

(J Craniofac Surg 2021;32: 2774-2778)

C raniosynostosis is a developmental defect that occurs in 1:2000 to 1:2500 live births and involves the premature closure of the calvarial sutures.¹

In the 1890s, Lannelongue et al² and Lane et al³ introduced the first reported strip craniectomy surgeries to release the fused sutures. They did not gain popularity due to high morbidity, mortality, and early re-ossification of the sutures.^{4,5}

From the 1960s to the 1990s, operative intervention for craniosynostosis evolved into extended craniectomies and extensive cranial vault reconstructions, typically utilizing long "ear-to-ear" bi-coronal skin incisions.⁵

In the late 1990s, Jimenez and Barone introduced the minimally invasive endoscopic strip craniectomy (or suturectomy) technique for the repair of craniosynostosis combined with postoperative cranial orthotic molding.^{6,7} Since then, minimally invasive techniques have been consistently developing and refined by the use of the endoscope for visualization with the addition of force therapy in the form of cranial molding helmets or springs.⁵

There is a lively debate among surgeons regarding the outcomes of minimally invasive versus open surgery techniques for the treatment and repair of nonsyndromic craniosynostosis. Outcome variables debated include magnitude and durability of head shape improvement, cost, neurodevelopmental trajectory, the burden of care to the patient, and intra- and post-operative complication rates.^{8,9}

Several studies have demonstrated favorable perioperative outcomes for endoscopic compared to open procedures. Two comprehensive reviews on endoscopic versus open repair for craniosynostosis, demonstrated an association of endoscopic repair with lower estimated blood loss (EBL), lower transfusion rates, shorter length of stay, shorter operative times, and lower costs.^{10–12} More recent studies continue to echo these findings.^{13,14}

Nonendoscopic suturectomies, assisted with cranial orthosis remodeling, have been performed within our group to treat craniosynostosis for the past 10 years. Though the endoscope was utilized initially, its use was abandoned in the early stages of the experience

The Journal of Craniofacial Surgery • Volume 32, Number 8, November/December 2021

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Received March 24, 2020.

Accepted for publication June 7, 2021.

as it was considered unnecessary. In other words, the exact same surgery could be performed in the same fashion with a headlight and loupe magnification. To our knowledge, since the introduction of the endoscopic suturectomy, a nonendoscopic suturectomy or strip craniectomy technique in the setting of cranial orthosis remodeling assistance has not been described in the literature.^{5,15,16} We believe that describing our experience with this technique is very important as many surgeons around the world may feel limited and avoid performing a suturectomy simply from their understanding that it cannot be performed without an endoscope.

We herewith describe our latest experience with the nonendoscopic strip-craniectomy technique for the correction of craniosynostosis followed by an orthotic molding helmet.

PATIENTS AND METHODS

A retrospective chart review was conducted of all patients undergoing primary strip-craniectomy repair of craniosynostosis at the Schneider Children's Medical Center of Israel (Petach-Tikva, Israel) between the years 2019 and 2020.

Preoperative and demographic data included age, sex, weight, and the type of craniosynostosis. Intraoperative data included surgery duration, EBL, blood transfusions, and intraoperative complications. Postoperative data included postoperative blood transfusions, hemoglobin level at discharge, length of stay in the hospital, and postoperative complications.

All surgeries were performed by the senior authors (AK and AO) as part of our institutional craniofacial program.

Suturectomies were offered and performed for infants that were 2 to 7 months old with a simple, single suture, metopic, sagittal, or coronal craniosynostosis.

An identical pre- and post-operative protocol were applied to each patient. Patients received intravenous antibiotics and tranexamic acid before conducting the skin incision. Pre- and intra-operative blood transfusions were given per the anesthesiologist's preference. A STARband Cranial orthosis (Orthomerica Products, Inc. [ORLANDO, FL, USA]) was used postoperatively; cephalic indices and head measurement scans were obtained using a "Smart-Soc" scanner (Orthomerica Products, Inc. [ORLANDO, FL, USA]).

Patients were scanned 1-week postoperatively, before the beginning of cranial orthosis treatment (3-weeks postsurgery) and at the end of the treatment, which varied among patients based on the physician's recommendation to discontinue the orthosis. The duration of the orthotic treatment was estimated to be around 6 to 8 months, with individual variations.

Outcome results were assessed by comparing various measurements taken from the initial scan, performed 1 week after surgery, and from the last follow-up scan available. We also compared these measurements to those taken manually during the same follow-up visits: medio-lateral (ML), anterior-posterior, and head circumference. The various measurements consisted of:

- the distance between 2 chosen points in the area of the deformity to be corrected (for metopic cases it was the ML distance at the level just above the orbits; for sagittal cases, the posterior parietal ML distance halfway between the selion/ tragion line and the apex; for coronal and lambdoid cases, the ML distance at the level of the plagiocephaly),
- Cranial vault asymmetry (the difference between 2 diagonal measurements taken from the forehead to the contralateral occipital area, both at 30 degrees from midline) and cranial vault asymmetry index (cranial vault asymmetry divided by the longer of the 2 diagonals), and
- 3. Cephalic ratio (CR) (width divided by length taken at midlines at the level of a head circumference measurement just above the eyebrows).

SURGICAL TECHNIQUE

All surgeries were performed using surgical loupes ($\times 2.5$ magnification) with a headlight. Following the anesthesia induction, at least 2 intravenous lines were placed. The preplanned skin incisions were infused with a tumescence solution of 0.9% normal saline with adrenaline (1:100,000 concentration). The scalp incisions were executed with a #15 blade scalpel followed by needle-tip electrocautery.

Sagittal Synostosis

Patients undergoing repair of sagittal synostosis were placed in a prone/modified sphinx position. Two separated 3 to 4 cm "Wshaped" horizontal skin incisions were made between the anterior bregma and the posterior lambda fontanelles (Fig. 1A). Blunt dissection through the subgaleal space was followed by stripping off the pericranial layer from the cranium. The planned bony suturectomy borders were marked bilaterally 1 cm lateral to the midline thus creating a strip-craniectomy with a width of 2 cm. After creating a plane between the dura mater and the skull at both open fontanelles (in most cases, even when seemingly close, we have found a small open space, and in rare occasions when not found, a small burr hole is drilled), Kerrison and Lexel rongeurs were used to initiate the first centimeter of the craniectomy. Then, the sagittal suturectomy was executed step-by-step (by subsequent square or rectangular-osteotomies) using an ultrasonic bone scalpel (bonescalpel, misonix) whereas the dura mater was protected by a malleable brain retractor (Figs. 2 and 3), first from posterior to anterior and then vice versa until the last piece of suture was removed at the midpoint between both incisions. During these steps, the bed was either raised or lowered to aid with visualization of the area of interest. Meticulous hemostasis was performed when needed, using bone wax, bipolar electrocoagulation, and absorbable gelatin sponges (gelfoam, pfizer) which were usually removed. Skin incisions were sutured primarily with subcutaneous 4-0 polyglactin 910 (vicryl, ethicon) sutures and the skin with 5-0 running poliglecaprone 25 (monocryl, ethicon) sutures dressed with antibiotic ointment (mupirocin 2%) for 1 to 2 days postoperatively.



FIGURE 1. Preoperative skin incisions planning and strip-craniectomy borders for (A) Sagittal suturectomy; (B) Metopic suturectomy; (C) Right unicoronal suturectomy; (D) Right lambdoid suturectomy.

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FIGURE 2. Intra-operative view of the sagittal suturectomy carried out in a stepby-step approach by subsequent square or rectangular-osteotomies using an ultrasonic bone scalpel while protecting the dura with a ribbon retractor (from left to right).

Coronal Synostosis

Patients undergoing repair of unicoronal synostosis were placed in a supine position, with the head tilted contralaterally to the synostosis, and a single horizontal "S-shaped" skin incision was made over the relevant midunicoronal suture at the temporal region, over the major curvature of the skull, such that both suturectomy trajectories, 1 toward bregma and 1 toward the orbito-zygomatic suture, would be relatively flat (Fig. 1B). Suturectomy was carried out as detailed above for sagittal synostosis.

Metopic Synostosis

Patients undergoing repair of metopic synostosis were placed in a supine position and a single horizontal "W-shaped" skin incision was made anteriorly to bregma and 1 cm behind the hairline (Fig. 1C). Metopic suturectomy was carried out as described above for the sagittal craniosynostosis in a step-by-step manner. For the nasofrontal region of the suture, the last 2 cm of suturectomy, a high-speed drill (medtronic legend) was used. Bone wax is always required for hemostasis in this part (the skull base) as the bone is thicker and more trabecular.

Lambdoid Synostosis

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Patients undergoing repair of lambdoid synostosis were placed in a prone/modified sphinx position. Two separated 3 to 4 cm "S-



FIGURE 3. 2×5.5 cm strip craniectomy of a fused sagittal suture removed using our described technique through 2 small incisions, under loop magnification, and a head light.

shaped" horizontal skin incisions were made between lambda (posterior fontanelle) and asterion (Fig. 1D). The lambdoid suturectomy was carried out as described above for the sagittal craniosynostosis in a step-by-step manner.

RESULTS

Thirteen patients (11 males; 2 females) were operated on between the years 2019 and 2020. The types of craniosynostosis included: 5 metopic, 5 sagittal, 2 coronal, and 1 lambdoidal (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C908). The average age at surgery was 4.35 months (from 2.5 to 7 months) with an average weight of 6.69 kg (range 5-8 kg). All patients underwent nonendoscopic suturectomy. The average length of surgery was 71 minutes (from 46 to 105 minutes.). Averaged intraoperative EBL was 31.54 mL (range 5-150 mL) equal to an estimated average of 6.15% of the blood volume. Twelve patients received a blood transfusion prior to the skin incision per the anesthesiologist's preference. The mean amount of blood transfused during surgery was 94.62 mL (range 0-150 mL) equal to an estimated average of 18.1% of the blood volume. The mean hemoglobin at discharge was 10.38 mg/dL (from 8.8 to 14.0). There was only 1 intraoperative mild complication due to a pinpoint puncture of the lateral part of the superior sagittal sinus during electrocautery assisted periosteal dissection from the skull, which caused an EBL of 150 mL, which required a blood transfusion, without any further consequences. The mean intra-hospital length of stay was 1.77 days (range 1-3 days). No postoperative complications were noted. In all the patients the remodeling treatment was initiated 3 weeks postoperatively with a custom-made cranial orthosis helmet.

The patients were seen regularly every 2 to 3 weeks at the cranial orthosis clinic and every 2 to 3 months at our craniofacial clinic.

Long-term follow-up scans were available for 8 patients (Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C909). Four were lost to follow up, 2 of which were tourists. The average follow-up was 9.0 months (range 5-13 months, a median of 9.5 months).

For the 5 metopic cases, which had an average of 8.8 months follow-up, the two-point distance in percentage increased on average 20.6% (Fig. 4A), compared to the manually measured growth of the ML axis at the midline of 10.3%. The cranial vault asymmetry index decreased on average from 4.02% to 2.47% and the CR remained stable at 78%.

For the 2 sagittal cases, who had 5 and 13 months of follow-up, the two-point distance in percentage increased by 21% and 18% respectively, compared to the manually measured growth of the ML axis at the midline of 7% and 14% respectively (Fig. 4B). The CR

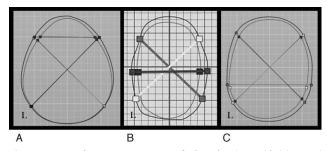


FIGURE 4. Pre- and post-treatment scans of selected patients with (A) metopic (B) sagittal and (C) right lambdoid craniosynosthosis. Red: pretreatment; blue: posttreatment; "L" represents the left side of the patient; The horizontal lines describe pre- and post-treatment "two-points measurements"; Head measurement scans were obtained using a "SmartSoc" scanner (Orthomerica Products, Inc. [ORLANDO, FL, USA]).

(which uses the ML more anterior to the biparietal narrowing) increased by 4% and 7.6%.

The lambdoid case had 10 months of follow-up at the time of the study. The two-point distance, measured at the occipital axis where the plagiocephaly was most evident, increased by 14.4 mm, equivalent to 16.25% (Fig. 4C). The manually measured growth of the anterior-posterior axis was 9 mm and in the ML axis 11 mm. The cranial vault asymmetry index improved from 5.4% to 0%. The CR increased from 81% to 88%.

In all cases, there was a significant improvement in skull width, symmetry, and proportion at the area of the synostosis.

DISCUSSION

The treatment of craniosynostosis has progressed dramatically over time. The benefits of endoscopic suturectomies have been broadly described, accepted, and widely adopted. These procedures are safe and effective with recognized decreased operating times, smaller amounts of blood loss, shorter hospitalization stays, lower costs, good cosmetic outcomes, and less need for secondary cosmetic surgeries at later ages, compared with the cranial vault reconstructions. Most publications have reported the surgery being performed around the age of 3 to 4 months old.^{1,17,18,21}

Within our group, this surgery has been performed for about 10 years. Initially, the endoscope was utilized, however, early in the experience its use was abandoned, replacing it with the above-described technique. The outcomes of the present small series of patients are the result of a refined technique over a decade and reflect similar outcomes to those published in the literature for endoscope-assisted strip-craniectomies.^{7,19,20,21}

Although the average EBL in our cohort was 31.5 mL, which is not concerning during surgery, almost all of the patients received blood transfusions. All transfusions were given per the anesthesiologist's preference at the beginning of the surgery. As described above our technique does not cause a significantly higher degree of blood loss than the endoscopic technique. However, it could be argued that the dural dissection from the skull, when performed with the endoscope, is done under direct vision, as opposed to being done blindly with a curved instrument. Most of the important hemorrhagic sources during the surgery are from attachment points between the superior sagittal sinus and the skull, that rupture during the dissection of the dura, or the craniectomized skull. Though the endoscopic technique allows to visualize such sources and possibly cauterize them under direct vision before removing the overlying skull, and thus, preventing more blood loss than tamponading them with gelfoam, or waiting to complete the craniectomy in that area in order to cauterize them under direct vision, it has not been our impression that the EBL in those occasions, warrants a blood transfusion in most cases. On the other hand, it can be argued that in those cases where bleeding does occur during the endoscopic dissection of the dura from the skull, a less significant amount of blood in a small space, blurs the endoscopic vision, delaying the hemostatic efforts.

Our incision pattern did not change during the transition from endoscopic to nonendoscopic. However, with the pass of the years, we learned to perform the surgery through smaller incisions, and we did change the direction of the "W" and "M" base toward the direction of the longer craniectomy side (Fig. 1A). This has been helpful to prevent skin rupture during inadvertent over-retraction, and if it already happens, in case the skin ruptures, it does it in continuation to the lateral edge of the incision and not perpendicular to the apex of one of the curves.

Although the endoscope-assisted surgical technique has many merits, it is not our purpose to demerit it, but rather to emphasize that an endoscope is neither necessary nor essential to be able to offer this surgery to patients anywhere in the world where there is access to cranial orthosis. This is of utmost importance as the use of neuroendoscopy is not available worldwide in terms of accessibility, training, and costs. The relatively short and easy learning curve and the satisfactory clinical results make this proposed method attractive. Unfortunately, clearing the endoscope from the equation to perform the suturectomy does not solve all accessibility and costs barriers, as cranial orthosis is also unavailable in many countries or medical centers. Solutions or alternatives for overcoming this problem are beyond the scope of this paper.

CONCLUSIONS

Suturectomies assisted with cranial orthosis remodeling for the treatment of all types of nonsyndromic craniosynostosis can be performed without an endoscope while maintaining minimal-invasiveness, good surgical results, and low complication rates.

ACKNOWLEDGMENTS

The authors extend their appreciation to Christina Hinton, C.P, Cranial Products Manager at Orthomerica Products, Inc., for her invaluable help in the acquisition of all measurement using the "SmartSoc" system; and to Mr Aharon Natan for his dedication and professionalism in conducting the cranial orthosis treatment in our clinic.

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Eternal flame.