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PRS GLOBAL OPEN

ORIGINAL ARTICLE

Pediatric/Craniofacial

Multidirectional Cranial Distraction Osteogenesis with Simplified Modifications for Treating Sagittal Synostosis

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Background: Multidirectional cranial distraction osteogenesis (MCDO) is a procedure of ours developed earlier for treating craniosynostosis. However, the numerous bone flaps led to prolonged operative time and occasional bone detachment from dura. We have since simplified the osteotomy design. In treating sagittal synostosis, required bone flaps have been reduced to 11 (from ~20).
Methods: In a 2-year period (2014–2015), 5 boys with sagittal synostosis underwent MCDO using our simplified and fixed-form osteotomy. Mean age at surgery was 9.4 months (range, 8–11 months). Pre- and postoperative cranial morphology was assessed by cephalic index and by mid-sagittal vector analysis.
Results: Improved cranial shape was confirmed by 3-dimensional CT scans and by mid-sagittal vector index. Mean preoperative cephalic index (68.7) progressively increased to means of 78.5 immediately after distraction device removal, 75.2 at postoperative month 6, and 75.1 at 1 year postoperatively. There were no major complications, although transient cerebrospinal fluid leakage and loosening of anchor pins occurred in 1 patient.
Conclusions: Simplified MCDO has a number of advantages over conventional distraction procedures such as discretionary reshaping/expansion of cranium and predictable osteogenesis and is a valid treatment option for patients with sagittal synostosis. *(Plast Reconstr Surg Glob Open 2017;5:e1536; doi: 10.1097/GOX.0000000000001536; Published online 26 October 2017.)*

INTRODUCTION

Corrective surgical procedures for sagittal synostosis remain controversial. One-stage cranioplasty, endoscopically assisted strip craniectomy, and spring-mediated cranioplasty are widely used,[1] but distraction osteogenesis is also a valid option.[2]

Although superior to other procedures in various aspects, distraction osteogenesis is clearly limited in terms of the direction of bone flap movement. Because flaps can only be moved in 1 direction (along axis of distraction device), some cranial deformities (e.g., frontal bossing) may be difficult to correct.[3] To surmount such problems, we developed a new method of treating craniosynostosis—multidirectional cranial distraction osteogenesis (MCDO).[4]

Initially, it was our contention that better cranial shape would result from smaller bone flaps. Thus, roughly 20 bone flaps were originally required to treat sagittal synostosis. However, in creating so many small flaps, operative time increased substantially and unexpected detachment of bone flaps from dura occasionally occurred. We have subsequently amended our approach by using a fixed-pattern of osteotomy design and reducing the number of required bone flaps to 11, which is the minimum for correcting cranial deformities in sagittal synostosis.

Herein, we present a series of 5 patients treated for sagittal synostosis, using this modified MCDO. In doing so, frontal bossing and retrocoronal constriction were well improved through short consolidation periods, and

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Received for publication March 24, 2017; accepted August 17, 2017.

DOI: 10.1097/GOX.0000000000001536

Disclosure: *The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors.*

Supplemental digital content is available for this article. Clickable URL citations appear in the text.

Table 1. Summary of Patients (All Patients Were Male)

Patient No.	Age (mo)	Operative Time (min)	Blood Transfused (ml/kg)	Complications	Activation Period (d)	Consolidation Period (d)	Hospital Stay (d)	Follow-Up (mo)
1	11	236	10.8	None	10	41	15	31
2	11	264	23.5	None	11	33	16	35
3	8	241	44.6	Transient CSF leakage, anchor pins loosening	10	22	15	30
4	8	263	54.5	None	10	41	15	29
5	9	249	62.9	None	7	44	12	17
Average	9.4	250.6	39.3		9.6	36.2	14.6	28.4

CSF, cerebrospinal fluid.

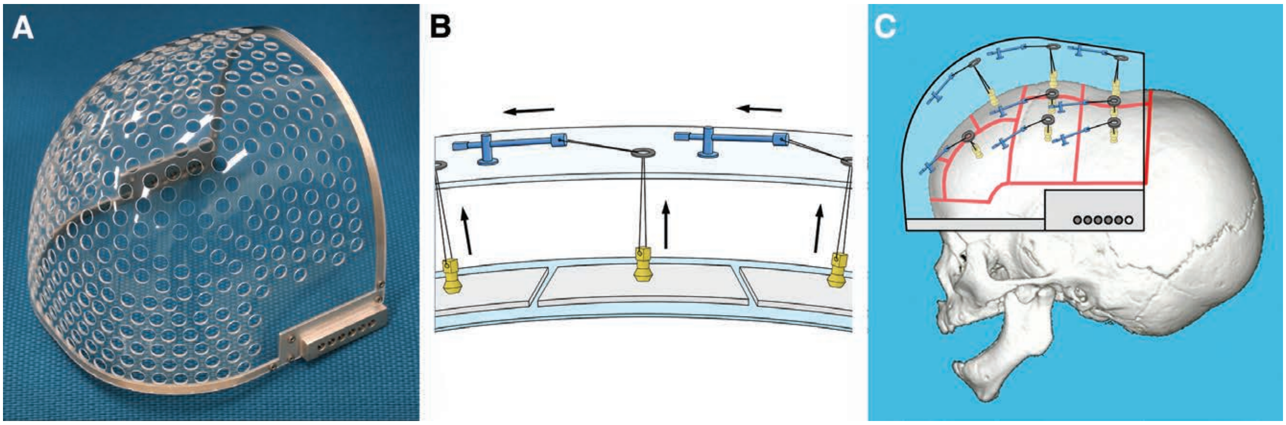

Fig. 1. A, MCDO. B, C, The frame of MCDO. Schemas of MCDO. Vertical distraction of each bone flap enables remodeling of skull to desired shape.

modified MCDO was validated as an attractive treatment option for sagittal synostosis.

PATIENTS AND METHODS

In a 2-year period (2014–2015), 5 male patients with sagittal synostosis underwent simplified, fixed-form MCDO (Table 1). Patient ages ranged from 8 to 11 months (mean, 9.4 months). All were diagnosed clinically, confirming isolated sagittal synostosis through preoperative 3-dimensional CT imaging.

Surgical Technique

The original MCDO surgical procedure has been previously described.[4] Cranial osteotomies are performed, creating multiple rectangular or triangular pieces. An ultrasonic bone scalpel (Sonopet; Stryker, Kalamazoo, Mich.) is used to prevent dural tears. In modified MCDO, the number of bone flaps was reduced to 11, and a fixed-pattern of osteotomy design was used (see below for details). Individual bones are not dissected from dura, thereby preserving vascular supply. Traction pins are fixed in each bony piece. After closure of wounds, a helmet-type frame is fixed by anchor pins in the temporal bones. Four or 5 anchor pins were needed on each side to obtain the stability of the frame. The frame is made of acrylic resin, and its weight is approximately 170 g. Wires secured in traction pinholes are then passed through holes in the frame, ultimately fixing the wires to frame-mounted distractors (Fig. 1).

Five days after surgery, distraction is initiated at a rate of 1.5 mm/d. The traction rate was later modified depending on the shape of the cranium, which was evaluated by a 3-dimensional CT scan on postoperative day 12.

Desired skull shapes generally are achieved within a 10-day activation period. Because the distance separating bony pieces is minimal, bone formation/fusion quickly ensue. The frame and all pins may thus be removed under sedation after a consolidation period of 4–6 weeks.

Design of Osteotomy Line

Frontal bone is divided into 5 pieces: 2 triangular pieces at the temple (medial edges passing through maximum curvature of frontal bossing) and 3 rectangular pieces divided laterally. Two rectangular bone flaps are made in parietal region of the skull. Finally, temporal cranium is divided vertically into 2 long bone flaps. The flaps are pulled up by wires perpendicularly to each bone flap, with exception of the 2 frontal rectangle flaps. The 2 rectangles recede (without wires) to adequate position through active traction of adjacent flaps (Fig. 2). This osteotomy design has allowed us to improve frontal bossing, raise parietal region, and expand temporal area.

Quantitative Assessment

Adhering to routine clinical protocol, all patients underwent 3-dimensional CT scans preoperatively, just after device removal (postoperative month 0), at postoperative month 6, and 1 year postoperatively.

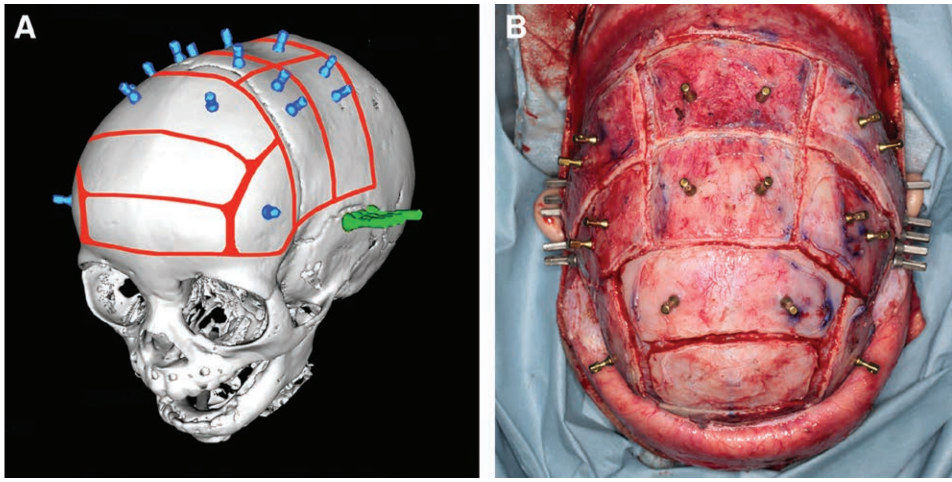

Fig. 2. A, Design of osteotomy lines. This fixed design enables improvement in frontal bossing and retrocoronal constriction. Traction pins are shown in blue and anchor pins in green (B). Intraoperative photograph after osteotomy and fixing the pins.

All measurements were made by DICOM image viewer (OsiriX; Pixmeo, Bernex, Switzerland). Pre- and postoperative cranial morphology was assessed by cephalic index and by mid-sagittal vector analysis.[5] Cephalic index was calculated by dividing the largest biparietal width of the cranium by the anteroposterior length and multiplying by 100. Mid-sagittal vector analysis was undertaken as follows: (1) mid-sagittal plane of cranium was first delineated by defining the plane traversing central vertex, midpoint of sella, and midpoint of nasofrontal suture; (2) based on this mid-sagittal image, a radial set of digital vector indicators were created, originating from a fixed point at apex of dorsal sellar summit; (3) vectors were prescribed from origin to outer table at 10-degree increments, beginning at nasofrontal suture (V0) and extending 180 degrees dorsally (V1-V18); and (4) ratios between lengths of V0 and Vn (n = 1–18) were calculated, setting length of V0 at 1.

RESULTS

There were no major complications within follow-up time. Transient cerebrospinal fluid leakage occurred in 1 patient during the activation phase but resolved with conservative therapy (Table 1). Loosening of anchor pins occurred in the same patient, resulting in accelerated removal of the devices. Operative time ranged from 236–264 minutes (mean, 250.6 minutes), and transfused blood volume ranged from 10.8–62.9 ml/kg body weight (mean, 39.3 ml/kg). Mean postoperative hospital stay was 14.6 days (range, 12–16 days). The planned distraction program was completed in all patients, with activation phase ranging from 7 to 11 days (mean, 9.6 days), and consolidation period ranging from 22 to 44 days (mean, 36.2 days).

Cranial shapes were improved in all cases (Fig. 3; **see figure, Supplemental Digital Content 1**, which pre- and postoperative 3D computed tomographic images of patient 2, ***http://links.lww.com/PRSGO/A570***; **see figure, Supplemental Digital Content 2**, which displays pre- and postoperative 3-dimensional computed tomographic images of patient 3, ***http://links.lww.com/PRSGO/A571***).

Cephalic index increased from baseline (mean, 68.7), averaging 78.5 immediately after device removal, 75.2 at postoperative month 6, and 75.1 at 1 year postoperatively. Although having a 35% relapse in the first 6 months, cephalic index maintained from postoperative 6 months to 1 year and good cranial shape was achieved at 1 year. The anteriposterior length and width of the cranium changed from baseline (mean, 16.6 and 11.4), averaging 16.2 and 12.8 immediately after device removal, 16.9 and 12.7 at postoperative month 6, 17.0, and 12.8 at 1 year postoperatively (Fig. 4). Mid-sagittal vector analyses confirmed reduced frontal prominence and increased height of vertex as well as sustained profile improvement, with no postoperative relapses (Fig. 5).

Patient 1

This 11-month-old boy with sagittal synostosis showed frontal bossing and retrocoronal constriction preoperatively (cephalic index, 72.6). MCDO with simplified and fixed-form osteotomy (described above) was performed (operative time, 236 minutes), and postoperative course was uneventful. Distraction began 5 days after surgery and ceased on postoperative day 15. All devices were removed 41 days later. Frontal bossing had improved, and cephalic index (79.0) increased substantially by 1 year postoperatively (Figs. 3, 4).

Patient 2

This 11-month-old boy with sagittal synostosis showed frontal bossing and an elongated skull preoperatively (cephalic index, 66.3). The same procedure was done (operative time, 264 minutes), with uneventful postoperative course. Distraction began 5 days after surgery and ceased on postoperative day 16. All devices were removed 33 days later. Frontal bossing and retrocoronal constriction improved, and cephalic index (75.4) was higher at 1 year postoperatively (**Supplemental Digital Content 1**).

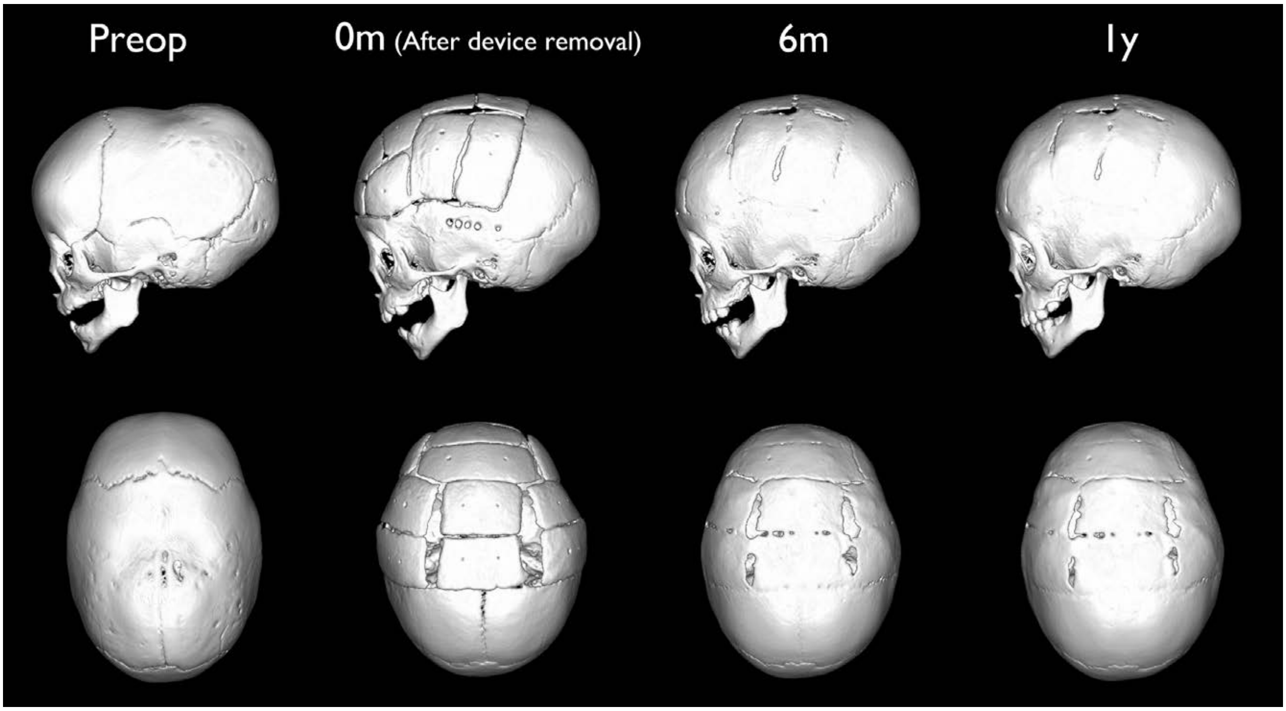

Fig. 3. Pre- and postoperative 3-dimensional computed tomographic images of patient 1.

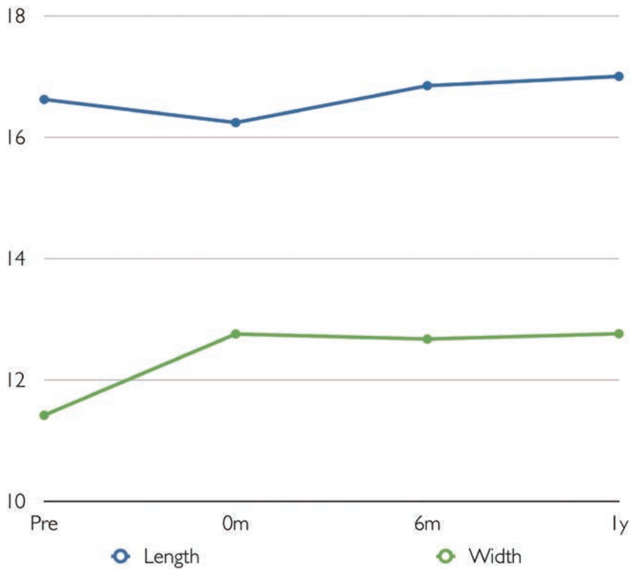

Fig. 4. Pre- and postoperative anterioposterior length and width of the cranium.

Patient 3

This 8-month-old boy presented with sagittal synostosis, for which the same procedure was used (operative time, 241 minutes). During the activation period, cerebrospinal fluid leakage occurred at the pinhole of vertex. Surrounding scalp was compressed by stuffing a sponge between frame and scalp for 3 days (while continuing distraction), and the leakage stopped. Distraction ceased on postoperative day 15. During the consolidation period, anchor pins loosened, forcing device removal 22 days after completing distraction. However, resultant cranial shape was good and was stable at 1 year postoperatively (**Supplemental Digital Content 2**).

DISCUSSION

Surgical goals for sagittal synostosis are to improve the length/width ratio of the cranium and to correct other regional deformities such as frontal bossing, retrocoronal constriction, and occipital protrusion. Many procedures have been described for the treatment of sagittal synostosis, although many of them remain controversial.

The results of endoscopically assisted strip craniectomy, frequently performed before 6 months of age,[6,7] rely on brain growth and postoperative helmet-molding therapy. Moreover, resultant morphologic correction reportedly has proved inferior to that of 1-stage cranioplasty in twins with sagittal synostoses.[8] Spring-mediated cranioplasty is a less invasive procedure for actively expanding the width of cranium. The procedures call for strip craniectomy, applying 2 or 3 springs across craniectomy edge.[9,10] However, surgeons cannot control distance or rate of advancement, and correction of bone deformities, such as frontal bossing. One-stage cranioplasty does offer good cosmetic results and is the mainstay procedure for treating sagittal synostosis.[1] In particular, the Melbourne method[11] seems capable of fully correcting severe deformities, not only frontal bossing and occipital protrusion but also displacement of vertex. Still, the high level of invasiveness and the general complexity of this surgical procedure have prompted craniofacial surgeons to seek alternative methods.

Distraction osteogenesis was introduced in 1998 as an alternative to 1-stage cranioplasty in treating craniosynos-

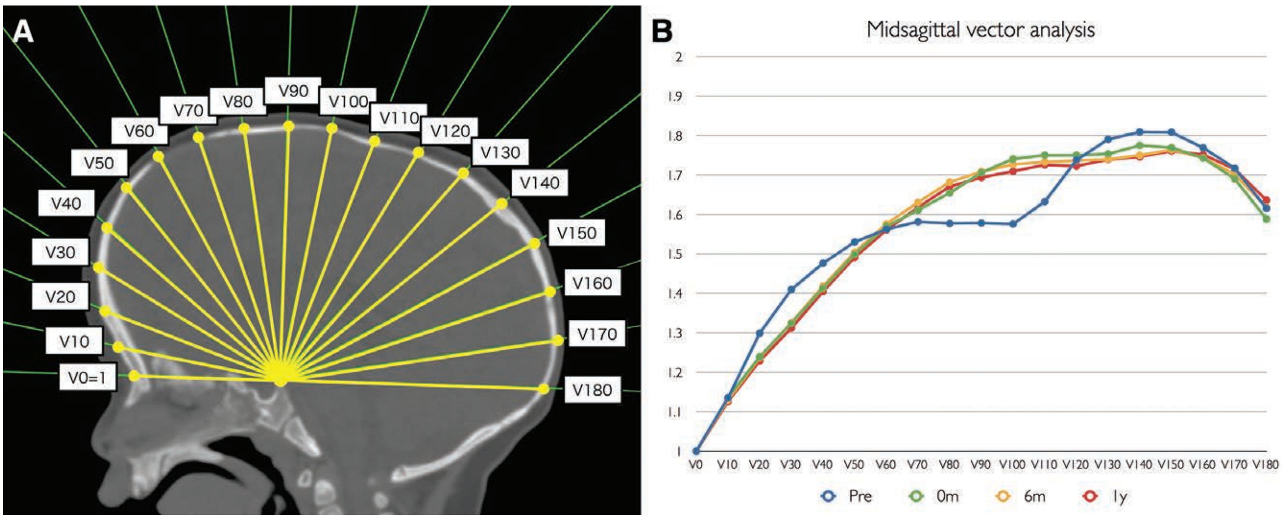

Fig. 5. A, Mid-sagittal vector analysis. B, Postoperative reduction in frontal prominence and increased height of vertex (without relapse) were demonstrated.

tosis,[12] and it has been widely used in East Asia.[3,13–15] It has several advantages over 1-stage cranioplasty. Gradual expansion of the cranium and the surrounding soft tissue allows greater increase of the intracranial volume than 1-stage cranioplasty. Moreover, because bone flaps are not dissected from dura and blood supply to bone flaps is preserved, early bone formation and good cranial growth as well as reduced risk of extradural abscess[16] are anticipated after surgery.

MCDO is even more advantageous than conventional distraction osteogenesis. Each bone flap may be moved in any desired direction, as opposed to 1 direction (along axis of distractor) via conventional distraction osteogenesis. Thus, the remodeling of deformed cranium is quite discretionary. For instance, retrocoronal constriction at vertex is correctable by vertical traction through MCDO but not through other distraction methods. Also, the distances separating bone flaps after distraction are comparatively less in MCDO, contributing to earlier bone formation/fusion and shortening the consolidation period. Finally, the distraction devices of MCDO are easier to remove than the internal distractors conventionally applied. Removal is done under sedation, without any incision and within 10 minutes.

As reported here, we modified the original MCDO procedure by reducing the required number of bone flaps and instituting a fixed osteotomy design. This simplified method produced more consistent results with less surgical exposure. Despite these modifications, operative time and transfused blood volume appeared to be still higher, compared with other recent data on distraction osteogenesis.[2,15,17] The time needed to create so many flaps may explain the disparity. Eleven bone flaps are raised in our fixed-form osteotomy, whereas other distraction methods require only 2 to 4. In addition, the gently-powered ultrasonic scalpel that we use to ensure safer osteotomy is certainly more time-consuming than a craniotome.

In this series of patients, surgical intervention was limited within the anterior two-thirds of cranium, and the occipital region was not treated. Although Khechoyan et al. reported that spontaneous frontal remodeling occurred with sagittal synostosis following mid- and posterior cranioplasty,[18] surgical intervention for the posterior cranium is much more invasive than that for the anterior cranium. Therefore, we applied MCDO to the anterior two-thirds of cranium in sagittal synostosis unless patients have prominent occipital protrusion.

The major limitation of this investigation is the small number of patients involved. Although satisfactory results were obtained in all 5 subjects, a greater number of patients and long-term monitoring are essential to validate our approach.

CONCLUSIONS

Simplified MCDO has a number of advantages over conventional procedures such as discretionary reshaping/expansion of cranium and predictable osteogenesis and offers a viable alternative for treatment of sagittal synostosis.

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