Original Article

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Biparietal expansion with partial suturectomy of the sagittal suture and linear frontal osteotomies for the scaphocephaly correction in children over 12 months of age

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Introduction: Sagittal craniosynostosis (SC) has a prevalence of 1: 2500 live births and represents 40-60% of all craniosynostosis. The objective of this work is to share the preliminary results, with the use of the surgical technique mentioned above for the treatment of SC in children older than 12 months.
Methods: A retrospective study from April 2019 to September 2023. Four patients with a diagnosis of non-syndromic scaphocephaly were evaluated, who met the following inclusion criteria: (I) age over 12 months, (II) CT scan with 3D reconstruction, (III) scaphocephaly due to postoperative restenosis and (IV) patients operated on with this surgical technique.
Results: There were 2 females (50%) and 2 males (50%), with an average age of 39.25 months. All four patients had complete closure of the sagittal suture; there was no evidence of entrapment of the superior sagittal sinus by the stenosed suture. Papilledema was evaluated with the modified Frisén scale. All four patients had grade 2 bilateral papilledema preoperatively and this reversed 6 months postoperatively. The cranial index (CI) preoperatively was 64 (range 58-72) and postoperatively 72.25 (range 66-83). There was no need to reoperate on any patient and we had no complications
Conclusion: This surgical technique is presented as an option for the treatment of scaphocephaly in children older than 12 months, and in scaphocephaly due to postoperative restenosis, proving to be a reproducible and effective procedure to achieve adequate brain decompression and reach satisfactory aesthetic results.

Keywords: Craniosynostosis, Sagittal synostosis, Nonsyndromic Scaphocephaly, Neurosurgery

INTRODUCTION

Sagittal craniosynostosis (SC) or scaphocephaly, is a skull elongation due to premature closure of the sagittal suture, causing an ovoid-type cranial dysmorphism. It has a prevalence of 1: 2500 live births and represents 40-60% of all craniosynostosis [1]. There are three types of SC according to the affected suture segment: anterior, posterior and complete [2]. The elongated shape of the skull usually suggests the diagnosis; however, the gold standard for the diagnosis is three-dimensional (3D) computed tomography (CT) [3]. Despite being the most common form of craniosynostosis, its diagnosis is late and its treatment differs when the patient is less than 12 months old and when the patients is more than 1 year-old, with surgery being recommended before the first year of live [4]. The objective of this work is to share the preliminary results, with the use of the surgical technique mentioned above for the treatment of SC in children older than 12 months.

MATERIALS AND METHODS

A retrospective study was carried out on a series of cases, operated during the period from April 2019 to September 2023.

Four patients with a diagnosis of non-syndromic scaphocephaly were evaluated, who met the following inclusion criteria: (i) age over 12 months, (ii) CT scan with 3D reconstruction, (iii) scaphocephaly due to postoperative restenosis and (iv) patients operated on with this surgical technique.

The percentage of the sagittal suture closure was analyzed, if there was entrapment of the superior sagittal sinus by the narrowed suture, the degree of papilledema, the cranial index was measured and the satisfaction of parents and/or caregivers with the results of the surgery was reported. Follow-up was given in the outpatient clinic at

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1,3,6 and 12 months after surgery (range 12-38 months). Results are reported twelve months after surgery.

Surgical technique

The patient is in a dorsal position, a zigzag coronal retroauricular skin incision is made, exposing the periosteum until reaching the implantation of the temporal muscle, dividing the periosteum with a transverse incision at the level of the vertex, subperiosteal dissection is performed until the lambdoid suture and supraorbital edges are exposed. The temporal muscles are detached until the pterion and asterión are visualized. A craniotomy of the parietal bones is performed 1.5 cm from the midline (to avoid the superior sagittal sinus) and behind the coronal suture and in front of the lambdoid suture, at a lower level below the squamous suture of the temporal bone. Asterisk osteotomies are performed on the parietal flap to promote bone malleability. A partial suturectomy of the sagittal suture is performed, at its junction of the middle third with the anterior third. The frontal bone undergoes 6 to 8 linear osteotomies that cross the coronal suture, to reduce frontal bossing. The parietal flap is secured to the temporal portion of the craniotomy with two non-absorbable sutures. Subgaleal drainage is left and closed in planes (Fig. 1 and 2). All patients had a central venous access, arterial line, required blood transfusion during or after the procedure, and postoperative care in pediatric intensive care unit (PICU).

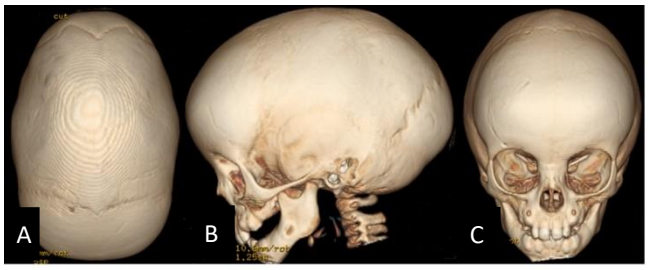

Figure 1- 3D CT scan of an 18 month-old female patient, showing total closure of the sagittal suture. A and B. The anteroposterior elongation of the skull can be seen in superior and left lateral views. C. Anterior view showing frontal bossing.

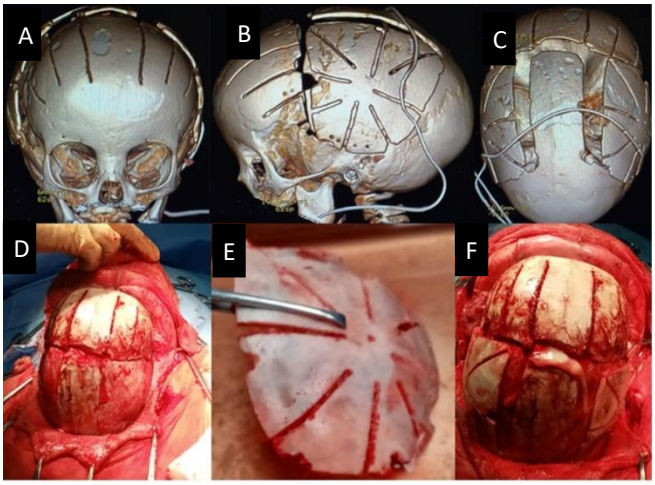

Figure 2- A, B and C. Postoperative CT scan showing the surgical procedure used. D, E and F. Show intraoperative images of cranial remodeling with adequate brain decompression.

RESULTS

Of the four patients operated on, three were operated on for the first time and one had previous craniosynostosis surgery performed at 4 months of age and developed postoperative restenosis. According to gender there were 2 females (50%) and 2 males (50%), the male/female ratio was 1:1, with an average age of 39.25 months (range 18-52 months). All four patients had complete closure of the sagittal suture; there was no evidence of entrapment of the superior sagittal sinus by the stenosed suture. To evaluate papilledema, the modified Frisén scale was used [5]. Table 1. All four patients had grade 2 bilateral papilledema preoperatively and this reversed 6 months postoperatively; there was no visual impairment in the ophthalmological follow-up. The cranial index (CI) (ratio of maximum cranial width/maximum cranial length) [6] preoperatively was 64 (range 58-72) and postoperatively 72.25 (range 66-83). Table 2. There was no need to reoperate on any patient and we had no complications. The stay in the PICU was 2-3 days and 2 days in the general ward until discharge. The patient´s parents and/or caregivers were satisfied with the results of the surgery (Fig 3).

Table 1- Modified Frisén Scale for Papilledema

Papilledema Grade
0 (Normal Optic Disc)
1 (Minimal Degree of Edema) C-shaped halo that is subtle and grayish with a temporal gap; obscures underlying retinal details
2 (Low Degree of Edema) Circumferential halo
3 (Moderate Degree of Edema) Obscuration of ≥ 1 segment of major blood vessels leaving disc
4 (Marked Degree of Edema) Total obscuration on the disc of a segment of a major blood vessel on the disc
5 (Severe Degree of Edema) Obscuration of all vessels on the disc and leaving the disc

Table 1. Modified Frisén Scale for Papilledema [a]

[a]Adapted from Ref. [5]

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Table 2. Results of the surgery in 4 children with scaphocephaly

Patient	Age (months)	Gender	Closed suture (%)	SSS entrapment	Preop MFS	Postop MFS	Preop CI	Postop CI	Previous surgery
1	18	F	100	No	2	0	67	73	No
2	39	F	100	No	2	0	59	66	No
3	52	M	100	No	2	0	72	83	Yes
4	48	M	100	No	2	0	58	67	No

F: Female, M: Male, SSS: Superior Sagittal Sinus, MFS: Modified Frisén Scale, CI: Cranial Index, Preop: Preoperative, Postop: Postoperative

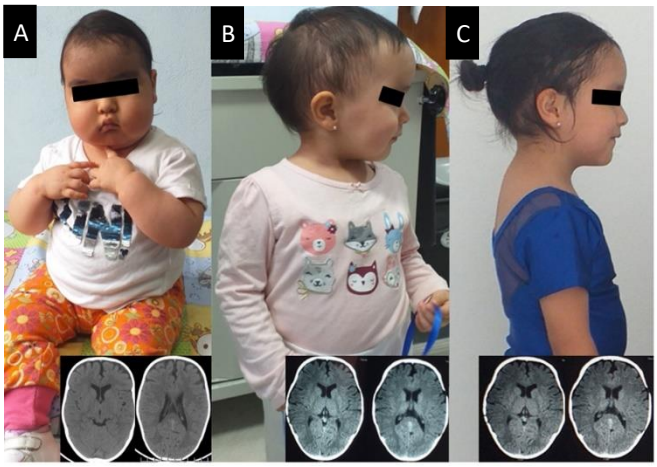

Figure 3- Clinical images with inset of the patient´s CT scan fig. 1 and 2. A. Patient in the preoperative period at 18 months of age. B. Patient 16 months after surgery. C. Patient 3 years after surgery.

DISCUSSION

The presence of children older than one year with scaphocephaly is usually due to a delay in diagnosis; however, there may be healthy children with a normocephalic skull and premature postnatal fusion of the sagittal suture in up to 3.3% [7]. In Mexico, alteration of FGFR1, FGFR2, FGFR3 and TWIST 1 genes has been identified in syndromic craniosynostosis (Crouzon, Pfeiffer, Apert, Saethre-Chotzen and Muenke) [8]. Some authors have found that the BMP, FGF and noncanonical WNT signaling pathways, which contain genes with multiple de novo mutations (SMAD6, SMURF1, DVL3, PSMC2, PSMC5, SPRY1 and SPRY4) and suggest that neurodevelopmental outcomes in nonsyndromic craniosynostosis patients may be driven more by mutational status than surgical technique [9,10].

Papilledema is an indirect sign of intracranial hypertension (ICH), however, it is not possible to correlate the degree of papilledema with the degree of ICH [11,12]. Thiele-Nygaard et al. [1] in a systematic review, 15 mmHg or less is considered the normal range of intracranial pressure in children and higher values are associated with ICH. They did not identify a correlation between ICH and the global neurocognitive condition in patients with scaphocephaly. Mitchell A et al. [13] in their study demonstrate that the presence of papilledema on fundoscopy is a strong indicator of ICH in children. Das S et al. [14] in patients undergoing ventriculoperitoneal shunt or endoscopic third ventriculostomy for the treatment of hydrocephalus, they identified a clear reduction in the degree of papilledema 7 days after surgery.

Regarding the surgical technique, we performed partial suturectomy instead of total resection of the sagittal suture, to avoid involvement of the superior sagittal sinus [15]. Linear osteotomies of the frontal bone were performed to reduce frontal bossing and improve cranial symmetry; we did not perform vertical craniectomies at the frontal level to avoid calvarial lacunae and aesthetic defects of the forehead [16]. We decided not to correct the occipital deformity to limit sinus bleeding related to the torcula dissection, as the patient´s hair hides the cosmetic defect.

Herlin et al. [17] operated on 9 patients over one year of age, performing parietal decompression and fronto-orbital bandeu with absorbable dual plates. The sings of ICH disappear at 4.2 months and an adequate cranial remodeling is achieved. Wagner W et al. [18] to correct frontal bossing performed vertical linear craniectomies of the frontal bone and drilling of the basal portion of the bony tongues, achieving good results in patients less than one year of age. Paternoster G et al. [19] operated on 10 patients older than one year with scaphocephaly, decompressed at the parietal level and corrected the frontal bossing by dividing the frontal bone into two parts and placing a coronal strip of bone between and performing a frontal advancement with absorbable plates, obtaining good results. Kang YS et al. [20] in 31 children older than 12 months, performed biparietal meander expansion and barrel-shape stave incisions frontally and occipitally to achieve adequate intracranial decompression and cranial remodeling. Arenas-Ruiz et al. [21] report 18 patients older than one year with scaphocephaly, performing resection of the sagittal suture and retrocoronal and prelamboidal osteotomies with osteotomies in asterisks in the parietal bones, mentioning adequate brain decompression and cranial remodeling.

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Gordillo-Dominguez et al. [22] performed biparietal transposition with craniectomy of the sagittal suture at the junction of the middle third with the anterior third, and rising sun osteotomies in the frontal bone, in children older than 24 months, achieving good aesthetical results. Vinchon M et al. [23] operated on 33 patients older than 9 months, performed cranioplasty by cutting the frontal and parietal bones, repositioned the frontal flaps and connected the frontal bandeu in lordosis. The parietal flaps are sutured above the squamosal suture. They achieve good results without the need to reoperate on any patient. Matthijs V et al. [24] operated on 65 patients between 4 and 8 months, performing total vault remodeling (TVR), which consisted of sagittal suturectomy, craniotomy with retrocoronal strip, creation of floating parietal bone flaps, and a bifrontal release with pterional decompression, achieving adequate results in long-term follow-up. Spazzapan P et al. [25] operated on 58 patients younger than 1 year, 28 with Renier´s "H" technique and 30 with TVR. They also included a bifrontal craniotomy, suturing the bifrontal bone flap to the supraorbital rim, finding better esthetical results with TVR.

Akai T et al. [26] in their comparatives series of syndromic and non-syndromic craniosynostosis, found better long-term results in patients operated on for non-syndromic craniosynostosis than in patients operated on with syndromic craniosynostosis. Proctor M et al. [27] review the management of craniosynostosis, concluding that surgery improves neurocognitive and aesthetic results in patients.

We did not use absorbable plates, bone distractors or helmets to achieve adequate cranial remodeling, which is an economic advantage for the management of scaphocephaly in low-or middle-income countries [28].

As has been demonstrated, there are various surgical techniques for the correction of scaphocephaly in children older than 12 months; most authors seek to achieve adequate biparietal decompression and reduce frontal bossing, according to clinical characteristics of each patient. Some authors perform cranioplasty starting at 9 months [23], and others perform TVR starting at 4 months of age [24], showing the lack of agreement of which surgery to perform and at what age? among pediatric neurosurgeons.

The disadvantage of our study is that it is a small case series, limited to a single medical center, has no control group, and does not evaluate neurodevelopment. Although our limited number of patients was affected by the Covid-19 pandemic, an increase in the number of scaphocephaly patients over the age of 12 months, who could not be treated during the pandemic and will require surgery is expected [29,30].

CONCLUSION

This surgical technique is presented as an option for the treatment of scaphocephaly in children older than 12 months, and in scaphocephaly due to postoperative restenosis, proving to be a reproducible and effective procedure to achieve adequate brain decompression and reach satisfactory aesthetic results.

ACKNOWLEDGMENTS

To all the physicians who, day after day, care for the most vulnerable and important part of our society: our children.

DISCLOSURES

Ethical approval

The study was conducted in compliance with the Declaration of Helsinki, Good Clinical Practice guidelines, and with the approval of institutional review boards and ethics committees. Written informed consent was obtained from parents or guardians before entry into the study.

Consent to participate

The patients gave consent to use their information and images for research purposes. *Consent for publication*

The patient gave consent to use his information and images for publication.

Conflict of interest

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

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Artificial intelligence

No artificial intelligence assistance were employed in the preparation of this manuscript.

CONTRIBUTIONS

-**Antonio Heredia-Gutierrez**: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

-**Maria Eugenia Carbarin-Carbarin**: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software,

Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

-**Ambar Heredia-Gutierrez**: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Project administration, Resources, Software, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing

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