OPEN

PRS GLOBAL OPEN

ORIGINAL ARTICLE

Craniofacial/Pediatric

Combined Dynamic Osteotomies for Craniosynostosis

Vera Lúcia N. Cardim, MD, PhD
Geórgia M.C. Peres, MD
Alessandra dos S. Silva, MD

Background: In primary craniosynostosis, the premature fusion of one or more sutures prevents the perpendicular expansion of brain tissue (primary defect). Providing space for the brain to expand, the compensatory growth of unaffected sutures causes progressive skull deformation (secondary defect). Understanding the need to treat the osteogenic matrix responsible for the cranial vault's shape was essential to develop a novel surgical concept known as dynamic osteotomy. It uses springs to activate stenotic sutures and trigger dura-mater distension while flexibilizing compensatory osseous defects via helicoid osteotomy (nautilus technique), allowing for efficient bone expansion and remodeling in craniosynostosis.

Method: This case series describes patients with craniosynostosis treated with dynamic osteotomy utilizing structural transformation inductors such as springs and helicoid osteotomy (nautilus technique), operated on between July 2004 and January 2020 at a single center in Brazil.

Result: Dynamic osteotomy longitudinally achieved stable osseous remodeling during growth period while maintaining good vitality and continuity of the osteotomized cranial vault.

Conclusion: Dynamic osteotomy utilizing springs and nautilus technique, alone or in combination, is a successful treatment of craniosynostosis regardless of patient's age.
(Plast Reconstr Surg Glob Open 2023; 11:e5208; doi: 10.1097/GOX.0000000000005208; Published online 16 August 2023.)

INTRODUCTION

The experience of managing craniosynostosis cases since 1973 allowed the author to highlight the high rate of cranial deformity recurrence throughout the growth phase of patients undergoing the conventional "back-table" skullcap (BTS) remodeling technique.

Assuming that the pathophysiology of craniosynostosis is linked to the transmission of abnormal tensions from the fibrous tracts to the suture from the skull base deformity,[1] in the 1990s, Cardim started to perform a dura mater Z-plasty[2] on the dural fibrous bridle corresponding to the larger sphenoid wing observed in unilateral coronal stenosis, obtaining stable results throughout the expected skull growth process of the treated patients, even with the use of BTS (Figs. 1 and 2).

The springs proposed by Claes Lauritzen[3] showed that the modeling forces applied to the dura mater through the inserted cap produced the same distension effect as that in Z-plasty without the additional bone devitalization damage seen with the BTS technique, an effect proven by Persing in 2006.[4] In a further attempt to restore stenotic suture adaptability and correct compensatory cranial deformity during brain development/expansion, our group proposed to stretch and remodel the dura mater by placing springs in stenotic sutures and utilizing the nautilus technique.[6] A concept named dynamic osteotomy,[2] initially proposed by Cardim, maintains the dura mater adhered to the bone,[7] thereby preserving its malleability and allowing for remodeling (both compression and expansion) of the mature bone. Any technique that uses this concept, such as endoscopic osteotomies and the use of a helmet, would be considered dynamic osteotomy. In 1996, Lauritzen proposed cranial remodeling through the use of a technique that would keep osseous segments loose, allowing for progressive redirection through intrinsic forces that would result in further improvement of the surgical results.[8] The combination of techniques aiming to maintain the dura mater bone interface that allows for the transmission of shaping forces originating externally (eg, helmets), within the bone (eg, springs and external distractors), or internally (secondary to brain growth) is known as dynamic osteotomy and has been used by Cardim without age limits. The nautilus technique foments dynamic skull remodeling by inducing

From the Hospital Beneficência Portuguesa de São Paulo, Sâo Paulo, Brazil.

Received for publication August 10, 2022; accepted July 7, 2023.

DOI: 10.1097/GOX.0000000000005208

Disclosure statements are at the end of this article, following the correspondence information.

Related Digital Media are available in the full-text version of the article on www.PRSGlobalOpen.com.

Fig. 1. Dura mater Z-plasty postoperative aspect.

progressive skull matrix renovation through propagation of suture forces, while maintaining the natural metabogenic bone marrow properties during skull suture ossification.

Takeaways

Question: Are patients satisfied with the long-term results of craniosynostosis treatment?

Findings: This is a descriptive study of 108 cases of craniosynostosis treated with dynamic osteotomies that act on the dura mater and provide long-lasting natural cranial shape.

Meaning: The search for alternative surgical techniques that offer long-lasting cranial remodeling in craniosynostosis is associated with the understanding of its suspected etiopathogenesis: biochemical imbalances of growth factors acting on the dura mater. Dynamic osteotomies allow for use of the bone as a vector of remodeling forces applied to the dura mater.

The objective of this article is to describe the dynamic osteotomies and the long-term results of the first 131 cases of craniosynostosis treated with springs and nautilus technique, alone or in combination.

MATERIALS AND METHODS

This case series included all craniosynostosis cases operated on by the Advanced Plastic Surgery Center service from

Fig. 2. Long-term results of plagiocephaly operated on BTS and dura mater Z-plasty: A, Preoperative frontal view (2.5 years old); B, Frontal view, 7 years postoperative; C, Frontal view, 28 years postoperative. D, Preoperative basal view (2.5 years old). E, Basal view, 7 years postoperative. F, Basal view, 28 years postoperative.

July 2004 to January 2020 using dynamic osteotomies such as springs and helicoid osteotomy (nautilus technique).

All procedures were authorized and consented to by parents or guardians through an informed consent form. This study was approved by the research ethics committee of the Real e Benemérita Associação Portuguesa de Beneficência de São Paulo Hospital (Protocol number 777-12 of March 30, 2012), where the procedures were performed.

The indication for the use dynamic osteotomy techniques are as follows:

1. Springs [called structural transformation inductors (STIs)]: for volumetric cranial vault gain (presence of indirect sign of intracranial hypertension or Chiari malformation)
2. Nautilus technique: to remodel compensatory growth areas (secondary defect).

All procedures scheduled during this period for which diagnosis or request for surgery suggested craniosynostosis or craniofacial dysmorphisms were screened.

Inclusion Criteria

- Craniosynostosis diagnosis (including craniofacial stenoses).
- Surgical treatment performed with dynamic osteotomies for volumetric enlargement of the skull, using STI springs and secondary defect remodeling with helical osteotomies (nautilus), associated or not.

Exclusion Criteria

- Craniosynostosis patients who underwent surgical management without use of dynamic osteotomies.

All patients underwent surgery under general anesthesia and required intraoperative blood transfusion. The procedures were performed by the craniofacial surgery team, who were eventually accompanied by the pediatric neurosurgery team. The immediate postoperative follow-up was conducted in the intensive care unit.

Description of Dynamic Osteotomy Techniques

All dynamic osteotomies were performed with minimal dura mater detachment without removing any skullcap segment for out-of-field modeling.

STI Springs

The STI spring is an osteogenic skullcap distractor inspired in the proposal by Claes Lauritzen for skullcap decompression and remodeling in craniosynostosis that can be used for both expansion and compression (Fig. 3). In addition to the surgical time for its implementation, like any other distractor, the STI requires a second surgical time for removal.

The choice of STI spring thickness (0.8, 1.0, or 1.2) was determined by the patient's age or the degree of bone resistance: it was was 0.8 in children under one year old and/or with extensive lacunal skull, 1.0 for children over 1 year old, and 1.2 for adults and adolescents. The resistance level of the bone was determined empirically during the intraoperative period, according to the bone thickness and the degree of displacement during the application of the STI springs.

The STI springs remained for 4 to 5 months in infants less than 1 year of age, and 10 to 18 months in older children, based on the cessation of cefalic perimeter expansion (proven by two equal measures in two sequential clinic visits, 30 days apart), in addition to a plain skull X-ray demonstrating gap ossification.

Nautilus Technique

Patients requiring cranial remodeling who did not show sufficient bone flexibility to adequately respond to the distribution of forces propagated solely by STI springs inserted in the dural envelope were submitted to the nautilus technique in these compensatory defect areas.[6]

Data collection included demographic data, type of craniosynostosis, surgery performed, time to remove springs (when applicable), length of hospital stay, intraoperative/postoperative complications, and quality of results (assessed by Whitaker classification).

RESULTS

This case series analyzed 108 patients with varied craniosynostosis cases (42 syndromic and 66 nonsyndromic, either primary or secondary) treated in the past 16 years using dynamic osteotomies with minimal dura mater detachment. Twenty-three patients required two or three surgical interventions, totaling 131 procedures. Fourteen patients underwent initial surgical treatment of craniosynostosis in other centers, all of which were submitted to the BTS remodeling technique. The cases were stratified by affected suture and demographic distribution, as shown in Table 1.

The primary surgical aim in all cases was to activate the affected sutures with osteotomies directly on or parallel to them and implement STI springs on their borders. The secondary defect (product of the compensatory growth of the healthy sutures) was treated by making the bone flexible through helical osteotomies (nautilus technique) whenever the loss of bone flexibility due to age prevented spontaneous remodeling of the STI force distribution. These osteotomies were performed in the first surgical period or when the springs were removed. Dynamic osteotomy distribution are shown in Figure 4.

Sutural osteotomies (and para-sutural in scaphocephaly) went through the stenotic sutures and released resistance lines, such as skullcap curves and sphenoid wings, leaving an area of intact bone where the progressive bridging of the deformed skullcap was intended.

The scalp incision was bicoronal with retro-auricular extremities in a broken line in the region of the temporal muscles and straight in the coronal area. The detachment was subperiosteal to keep the periosteum continuous and vascularized in the flap that would cover the osteogenic distraction areas.

In sagital craniosynostosis, patients were positioned in the supine position. Whenever there was constriction of the frontal and occipital regions, two osteotomies were

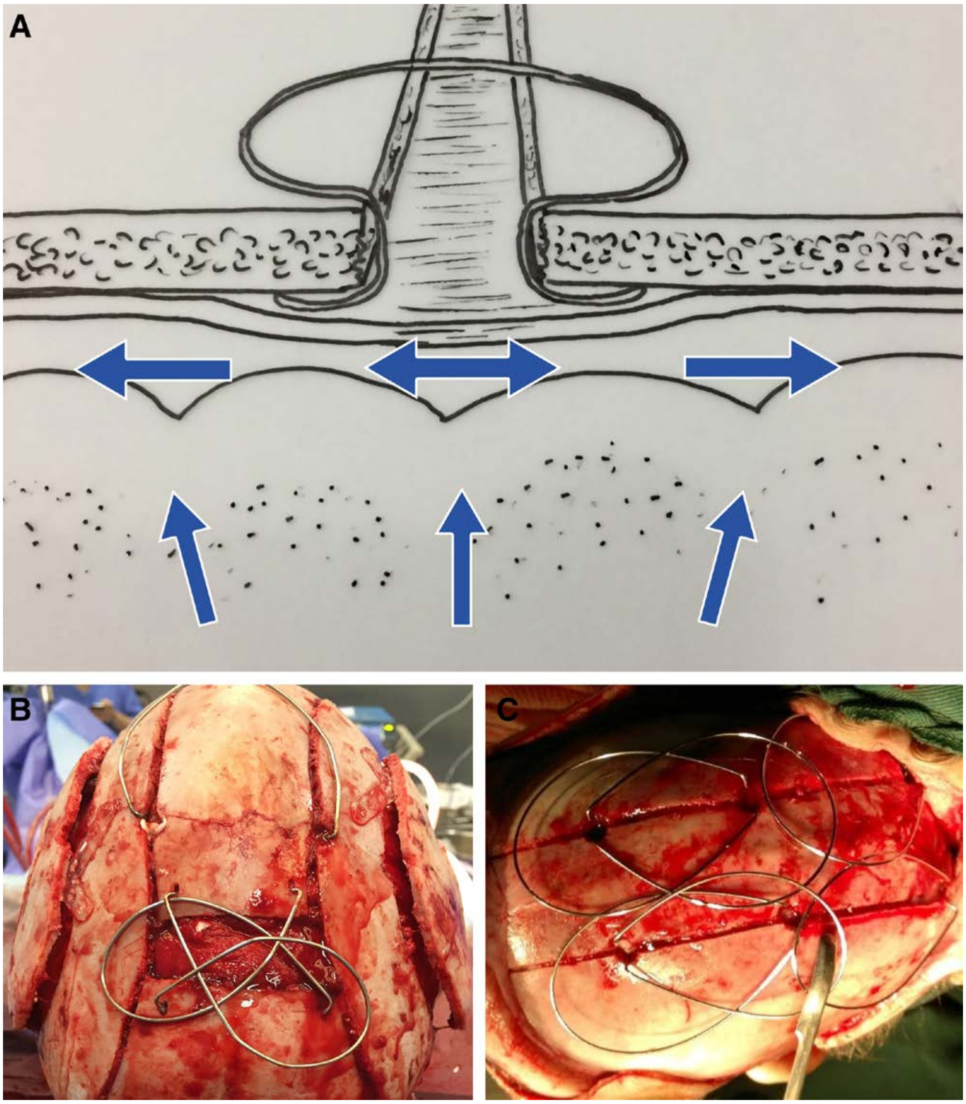

Fig. 3. Schematic and intraoperative photographs. A, Schematic representation of the expansion of the dura mater induced by the force applied to the bone. B, STI springs forming omega shape for expansion. C, Two STI springs for compression.

Table 1. Demographic Distribution of Cases

Affected Suture	No. Operations	Gender		Age (Mo)	
		Masculine	Feminine	Mean (Mo)	Range (Mo)
Sagittal	20	18	2	51,5	6–156
Metopic	11	9	2	27,33	6–108
Bicoronal	11	4	7	89,82	6–171
Unilateral coronal/ unilateral lambdoid	17	6	11	70,06	8–360
Multisuture	42	14	28	34,12	0–197
Other	7	3	4	42,29	20–92
Overall	108			52,35m	0–360

done in parallel to the sagital suture, prolonging beyond the coronal and lambdoid sutures. An osseous segment of 1–1.5 cm was also resected from the area in between the osteotomies, and constriction springs were positioned to reduce the anteroposterior cephalic diameter.

For correction of coronal craniosynostosis, the osteotomy was done, maintaining the frontal bone as a single segment. The osteotomy was done at the anatomic site of the coronal suture overlaying the orbital roof. Therefore, the orbital roof was osteotomized through a small craniotomy hole at the medial end of the orbital border with minimal dura mater detachment. For the sphenoid wing osteotomy with a curved chisel, the craniotomy hole in the orbitotemporal region (posterior to the frontal-zygomatic suture) had the required diameter to expose the anterior fossa in front of the wing and the middle fossa behind the wing.

For unilateral and metopic craniosynostosis, the same osteotomy runs along the roof and ascends in the medial aspect of the orbit, leaving a fulcrum in the frontal region that allows for the anterior rotation of the fronto-orbital osteotomized portion.

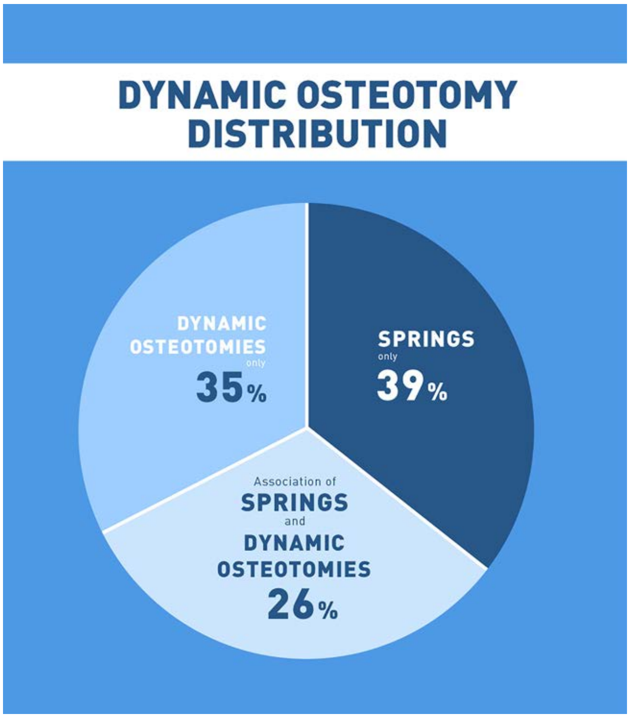

Fig. 4. Surgery distribution by type of dynamic osteotomy used.

For complex craniosynostosis, the same principle of expansion of large cranial portions is applied through circular osteotomies of the posterior regions, leaving the osseous segment only adhered to the dura mater and positioning the springs in a way to conduct the osseous flap in the desired direction.

In hump and pit areas of the cranium where the absence of osseous malleability would not allow remodeling, helicoid osteotomies were utilized to flatten the hump areas and permit expansion of the depressed areas, thus correcting the deformity.

The cranial shape obtained was clinically evaluated, and the Whitaker classification was utilized.[9] Although it may be a subjective and imprecise classification, it is still widely used, with the definitions as follows:

1. No refinements or surgical revisions considered advisable or necessary by the surgeon or the patient.
2. Soft-tissue or lesser bone-contouring revisions desirable, whether performed or not.
3. Major alternative osteotomies or bone-grafting procedures needed or performed.
4. Major procedure duplicating or exceeding in extent the original surgery necessary

The results were analyzed according to the Whitaker classification[9] (Fig. 5). The complications summary are shown in Table 2. One death occurred in a 6-week-old infant with severe craniosynostosis associated with syndromic oxicephaly, who was urgently operated on, given severe proptosis with globus oculares luxation. The death occurred on postoperative day 60, during exploratory craniotomy secondary to subgaleal fluid collection and suspected infection, although it was found to be sterile epidural hematoma. Two other deaths

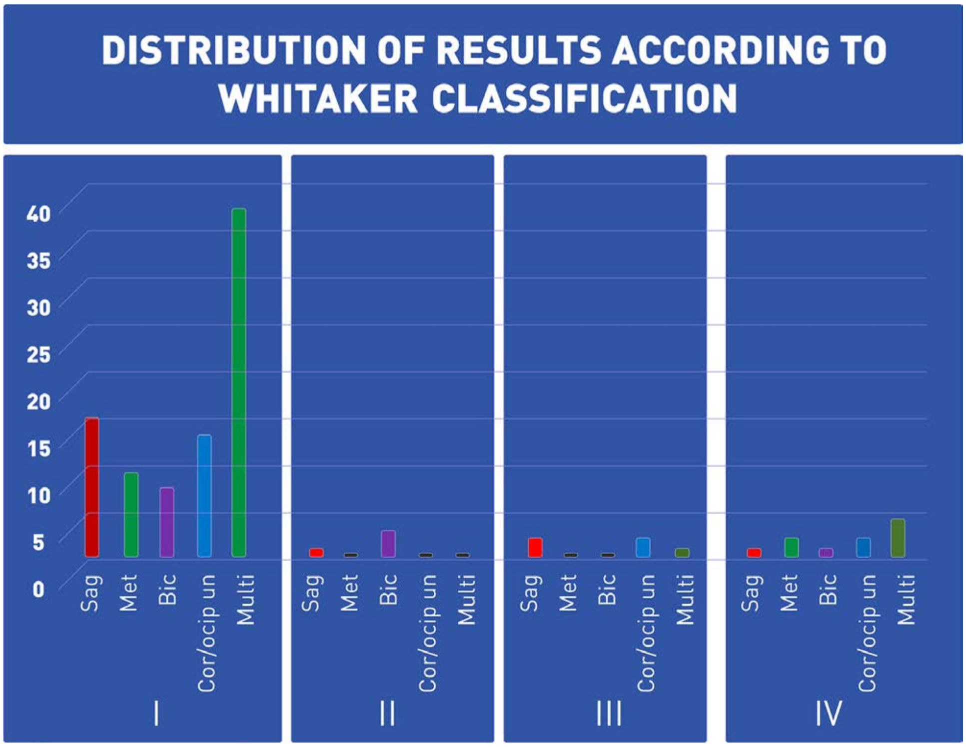

Fig. 5. Distribution of number of cases (vertical) according to Whitaker classification (horizontal—in I/II/III/IV) according of affected suture.

Table 2. Summary of Complications in Dynamic Osteotomies in 130 Patients (131 Operations)

Complication	Seroma/ Bruise	CSF Leaks	Infection	ITEs Dislodgement	ITEs Repositioning	Overcorrection	Hipovolemical Shock	Death
	4	6	2	6	3	0	2	1

CSF, cerebrospinal fluid.

occurred as a result of nonsurgical complications during the late (>6 months) postoperative period (not mentioned in Table 2). There were no intraoperative deaths.

DISCUSSION

Craniosynostosis prevents the natural brain growth and development in children, which will eventually impair their long-term neurocognitive function. Dynamic osteotomy techniques utilizing springs and nautilus have shown to improve brain development and growth in children with craniosynostosis from different etiologies by utilizing intrinsic bone marrow ossification process and dura mater expansion forces properties. The analysis of the cascade of events leading to craniosynostosis showed that sutural dura mater deformity precedes ossification process and skull deformity near the sutures, and is probably related growth factor imbalances.[10,11]

In normal skulls with normal internal volume and pressure, the normally shaped dural shell reproduces a normal skull in all stages of life. A skull deformed by synostosis and compensatory growth has a dura mater that reproduces this deformity as its final product. When undergoing BTS remodeling, the bone deprived of live cells will be re-inhabited by new osteocytes provided by this deformed matrix.

The most common treatment for craniosynostosis is skull decompression by BTS remodeling, which involves transforming the bone into a graft that will depend on the deformed osteogenic matrix (the altered dura mater that made it become deformed) to be integrated; this acts on the second to the last event of the craniosynostosis etiological cascade (Fig. 6 yellow arrow). Despite a recurrence of the deformity in children operated on after the age of 8 months, the positive and lasting results obtained by the BTS approach or endoscopic craniectomy when performed before 6 months of age and conducted with external modeling helmets demonstrated that the plasticity of very young bone allows for the external shaping force to act directly on the dural sac, correcting its shape.[12]

The osteogenic distraction proposals with devices (modeling helmets, springs, internal or external distractors, head, etc.) focus on the osteogenic matrix because the distractor (or helmet) strength is distributed in the dura mater through the bone that remains attached to it. Thus, when the treatment reaches the pathological process at a much earlier presentation stage (Fig. 6, blue arrow), the osteogenic matrix remodels itself.

According to Lauritzen,[7] the redistribution of the modeling forces of the dura mater act on the growing bone plates, which are still malleable and being remodeled according to the new standards. This signals the cessation of the compensatory growth response of the healthy sutures, normalizing the cranial shape as a whole. The distribution of spring forces in the calvaria changing the cranial base was experimentally documented.[13] Lauritzen,[7] who has published the largest case series on dymanic osteotomy treatment for craniosysnostosis, described that his cohort's age range was between 3.8 and 23 months, which is much younger than this study's age range (27.3–89.82 months). Furthermore, the majority of Laureitzen cohort are in an age range where BTS remodeling would be contraindicated. The lack of alternative surgical approach to treatment of the mature skulls leads to underappreciation of functional complaints such as migraines and learning disabilities, which end up being attributed to other etiologies. In parallel, the aesthetic alterations observed are neglected until adulthood when patients usually look for alternatives to minimize sequelae.

In mature skulls, the areas of the secondary defect received nautilus-shaped helical osteotomies[5] to also

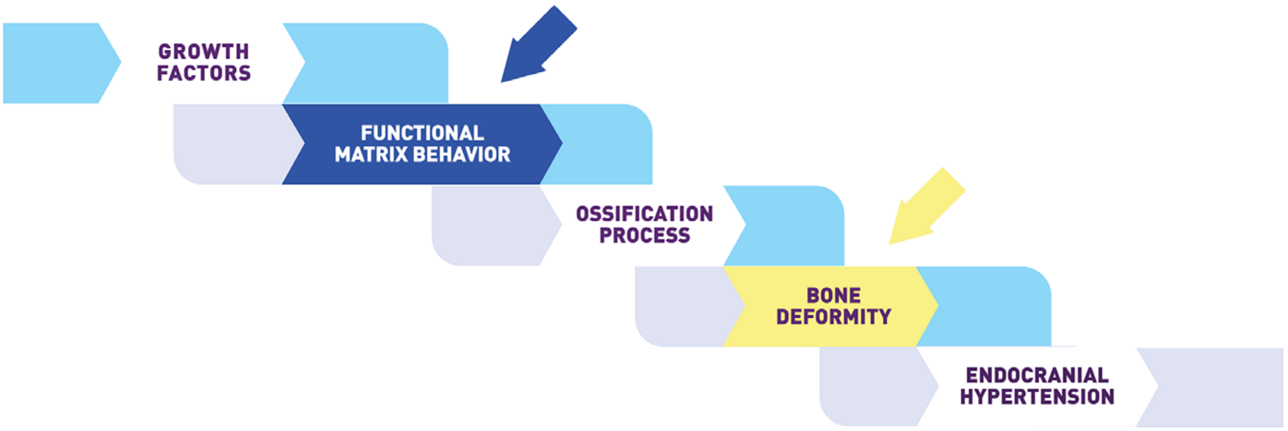

Fig. 6. Cascade of events leading to craniosynostosis intraoperative view.

remodel the force distribution. These osteotomies were already described by Salyer[14] and Salgado[15] as performed using the BTS technique on the deformed plates removed from the stenotic skulls. The bones that received helical osteotomies were modeled and immobilized with plates and screws, subsequently returning as grafts to the area to be corrected. Although with a similar design, dynamic osteotomy techniques differ from static grafts, as the former transforms the deformed cap into a malleable bone flap without devascularization, allowing for spring-induced volumetric expansion of the cranial vault.

In the areas of compensatory fossa (flattening), a nautilus technique was used for cranial expansion. Four to six triangular wedges with the base were fixed to the inner border of the helicoid steps and its vertex supporting the inner board of the outer border of the helicoid step without fixing. When the nautilus was made to sink the areas of the compensatory bossae, the osteotomy traces were enlarged using a wear drill, to reduce the surface during external postoperative compression. In retractor helicoids, the base of the wedges was fixed to the outer border of the helicoid steps, supporting the free end on the inner board of the inner edge of the step (Fig. 7). These wedges did not immobilize osteotomies; thus, it prevented unwanted movement during postural support of the head or bossae expansion in cases where area flattening was desired. In sagittal craniosynostosis, the combined action of contractile springs to decrease the sagittal band was responsible for ejecting brain content in the direction of the fragility created by the lateral nautilus.

The point of STI spring insertion defined the direction of the expansive force (tendency to open the extremities), which was perpendicular to the affected suture, to simulate the effect of a normal suture. A small slit made in the STI spring support points prevented its sliding and displacing on the osteotomy line.

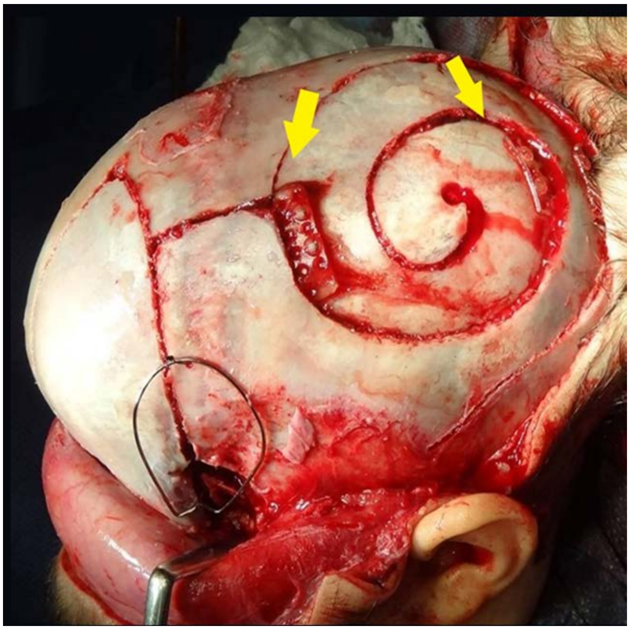

Fig. 7. Intraoperative view: detail of absorbable helicoid pads for compression (yellow arrows).

In cases where the expansion of the posterior fossa was indicated, the bone area bordered by osteotomy in the occipital region was wide enough for the vertical lateral lines to be placed in the temporal areas, allowing for the STI spring expansive force to be in the anteroposterior direction, repelling the entire bone window that carries the inserted dura mater and causing a "suction" of the posterior fossa contents upward and back.

The omega formed by the STI spring positioned in the osteotomy was individually curved to adapt it to the skullcap curvature. Whenever the area to be expanded by helical osteotomy included the insertion of the larger sphenoid wing, it was released.

As the STI spring forces act and propagate through the dura mater, interfering in the content/continent relationship of the cephalic segment, the nautilus technique provides indirect skull remodeling when associated with expanding springs and leading flattening zones to expand when associated with contraction springs Therefore, the two tools for the dynamic modification of the skull (Lauritzen springs and nautilus-shaped bone flaps) allowed for the expansion and remodeling of stenotic skulls at any age. [**See figure, Supplemental Digital Content 1,** which displays the evolution of oxycephaly treated by dynamic osteotomies (Spring plus Nautilus). A, Preoperative frontal view (6 months); B, Preoperative lateral view (6 months); C, Frontal view, 10 years postoperative; D, lateral view, 10 years postoperative. **http://links.lww.com/PRSGO/C741**.]

This retrospective study has several limitations. The data presented here were not collected in a systematic prospective fashion, and therefore, the innate retrospective data collection biases apply to this article, such as missing or incomplete data and other variables, which could impair critical analysis of the data presented. Nevertheless, descriptive studies like this are crucial in supporting alternative treatment techniques based on novel etiopathogenesis concepts that challenges the traditional culture, oftentimes impermeable to provocative innovation and paradigm shift. Recent studies on dural metabolism in craniosynestosis require us to adjust the treatment focus from the bone to the dura mater. Although we attempted to compare our results with those from other groups,[7,16] there is still a paucity of studies about dynamic osteotomy.

CONCLUSION

The use of dynamic osteotomy such as STI springs and nautilus techniques, alone or in combination, allowed for efficient indirect expansion and induction of skull remodeling in craniosynostosis cases, regardless of age.

Vera Lúcia N. Cardim, MD, PhD
Rua Augusta 2709/2705 Cj 42, CEP
São Paulo, SP 01413-100
Brazil
E-mail: vera@npa.med.br

DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

PATIENT CONSENT

Parents or guardians provided written consent for the use of the patients' images.

REFERENCES

1. Moss ML, Salentijn L. The primary role of functional matrices in facial growth. *Am J Orthod*. 1969;55:566–577.
2. Cardim VLN, Perez GMC, Silva AS, et al. Dynamic osteotomies in mature skulls - Cancum - 2017. Congresso mundial de CraniofacialE - Poster -. In: E - Poster XVII Congress of The International Society of Craniofacial Surgery. 2017.
3. Lauritzen C, Sugawara Y, Kocabalkan O, et al. Spring-mediated dynamic craniofacial reshaping. Case report. *Scand J Plast Reconstr Hand Surg*. 1998;32:331–338.
4. Persing JA, Jane JA, Shaffrey M, Virchow and the pathogenesis of craniosynostosis. *Plast Reconstr Surg*. 1989;83:738–742.
5. Nocchi Cardim VL, dos Santos Silva A, Salomons RL, et al. Nautilus-shaped dynamic craniotomy: a new surgical technique and preliminary results. *Rev Bras Cir Plást*. 2013;28:29–35.
6. de Faria R, Dornelles V, Lúcia V, et al. Spring-mediated skull expansion: overall effects in sutural and parasutural areas. An experimental study in rabbits [Expansão craniana com molas: efeitos globais nas áreas suturais e parassuturais. Estudo experimental em coelhos]. *Acta Chirurgica Brasileira*. 2010;25:169–175.
7. Lauritzen CGK, Davis C, Ivarsson A, et al. The evolving role of springs in craniofacial surgery: the first 100 clinical cases. *Plast Reconstr Surg*. 2008;121:545–554.
8. Lauritzen C, Friede H, Elander A, et al. Dynamic cranioplasty for brachycephaly. *Plast Reconstr Surg*. 1996;98:7–14; discussion 15.
9. Wes AM, Naran S, Sun J, et al. The Whitaker classification of craniosynostosis outcomes: an assessment of interrater reliability. *Plast Reconstr Surg*. 2017;140:579e–586e.
10. Greenwood J, Flodman P, Osann K, et al. Familial incidence and associated symptoms in a population of individuals with nonsyndromic craniosynostosis. *Genet Med*. 2013:1–9.
11. Greenwald JA, Mehrara BJ, Spector JA, et al. Regional differentiation of cranial suture-associated dura mater in vivo and in vitro: implications for suture fusion and patency. *J Bone Miner Res*. 2000;15:2413–2430.
12. Chou PY, Hallac RR, Patel S, et al. Three-dimensional changes in head shape after extended sagittal strip craniectomy with wedge ostectomies and helmet therapy. *J Neurosurg Pediatr*. 2017;19:684–689.
13. De Faria Valle Dornelles R, Cardim VLN, De Campos Fonseca Pinto ACB, et al. Skull base cephalometric changes in cranial expansion by springs. *J Craniofac Surg*. 2011;22:1496–1501.
14. SAlyer KE and, Bardach J. *Salyer & Bardach's Atlas of Craniofacial & Cleft Surgery*. Lippincott-Raven; 1999.
15. Solís-Salgado O, Anaya-Jara M. Remodelación craneal para craneosinostosis sagital mediante osteotomía en forma de espiral usando sistema de fijación con miniplacas y tornillos absorbibles. *Archivos de Neurociencias*. 2009;14:224–230.
16. Guimarães-Ferreira J, Miguéns J, Lauritzen C. Advances in craniosynostosis research and management. *Adv Tech Stand Neurosurg*. 2004;29:23–83.