

Twenty Year review of a Single Surgeon's Experience for craniosynostosis surgery

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Introduction: Nonsyndromic craniosynostosis (NC) is more commonly encountered than syndromic cases in pediatric craniofacial surgery. Affected children display characteristic phenotypes according to the suture or sutures involved. This study aims to report a single surgeon experience with surgery for primary craniosynostosis correction.

Methods: A retrospective review was carried out including all cases performed by the same surgeon between 2001 and 2021. The clinical data of 356 patients (371 surgeries) with primary craniosynostosis that have undergone surgery were retrospectively analyzed.

Results: There were 232 (65.1%) males and 124 (34.9%) females, with most of the male preponderance occurring due to the trigonocephaly (86%) and scaphocephaly (84.5%) groups. A female predominance was observed only in brachycephaly group (64.8%). There was a statistically significant male predominance between non-syndromic versus syndromic cases ($p=0.0001$). The most frequent forms in non-syndromic forms were scaphocephaly in 136 (51.5%), trigonocephaly in 48 (18.1%), anterior plagiocephaly in 47 (17.8%), and brachycephaly in 35 (13.2%). A named syndrome was present in 57 (16%). Genetic syndromes presented more commonly than expected with bilateral synostosis, the Kleeblattschädel deformity, and multiple suture synostosis ($p=0.001$). Complications were observed in 15 (4.2%). Intraoperative hemorrhage and CSF leakage were the most common ones. Morbidity was significantly associated with monobloc fronto-facial advancement. The mean follow-up was 3.7 years (9 months to 22 years). Morphological results in NC were classified in category I in 210 (70.2%), II in 82 (27.9%), and III in 7 (1.9%) cases for NC.

Conclusions: Non-syndromic craniosynostosis can be corrected with good outcomes and relatively low morbidity and mortality. For syndromic cases a multidisciplinary team is fundamental to achieve satisfactory results.

Keywords: craniosynostosis, craniofacial, pediatric neurosurgery, skull deformity, genetics

INTRODUCTION

Skull deformity in infants continues to be a diagnostic and therapeutic challenge. Craniosynostosis, defined as premature fusion of cranial sutures, was first described in 1830 by Otto [1]. In 1851 Virchow created a classification system for the types of skull deformity observed in craniosynostosis and made the important observation that premature suture fusion resulted in compensatory growth in other areas of the skull [2]. Since Virchow, multiple theories have been proposed to explain the pathogenesis of abnormal suture fusion, with recent studies focusing on genetic regulation.

Over the past decade, there has been significant progress in understanding mesenchymal stromal cell differentiation in the context of suture development and genetic control of suture pathologies, such as craniosynostosis [3,4].

Craniosynostosis may present with single versus multiple sutures involvement and/or syndromic versus nonsyndromic craniosynostosis [5]. Functional and morphological problems vary according to the type of craniosynostosis. Isolated craniosynostoses usually pose different problems than syndromic cases. In particular, facio-craniosynostosis carry out much more difficult problems, both in functional and morphological aspects [6].

In isolated craniosynostoses, the goal of surgical correction is (i) to expand the intracranial volume, thereby allowing cerebral growth; and (ii) to improve the cosmetic appearance of the child.

In this field, progress came from multidisciplinary teams involving neurosurgeons, plastic surgeons, pediatricians, geneticists, ophthalmologists, otorhinolaryngologists, psychologists and radiologists. A retrospective review of a

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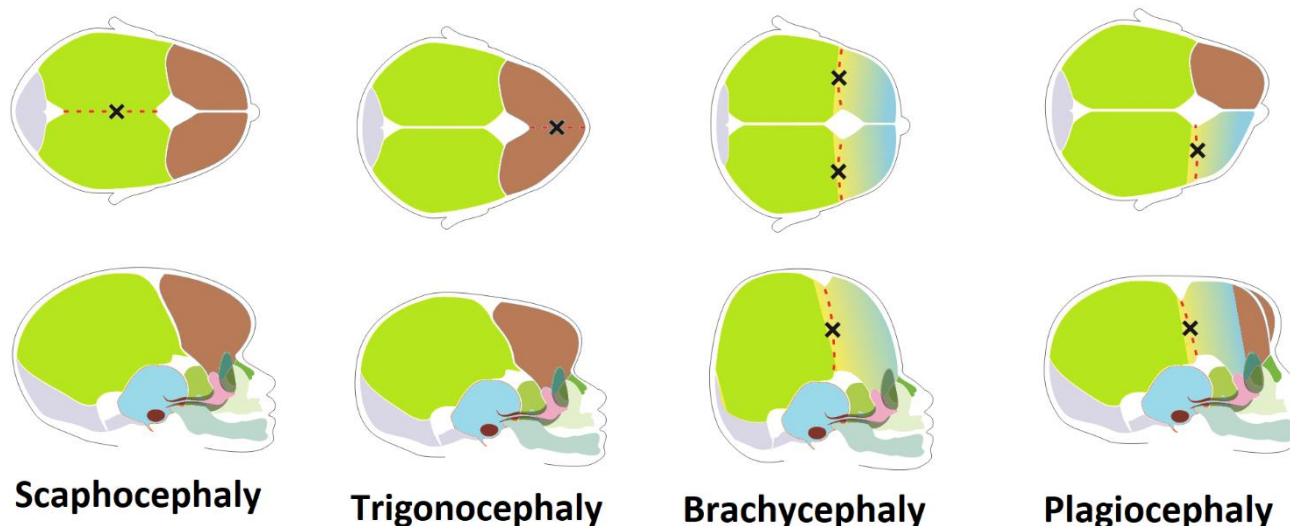


Figure 1- Illustration demonstrating the most common different types of non-syndromic craniosynostosis with the respective cranial sutures involved. (A) scaphocephaly (sagittal); (B) trigonocephaly (metopic); (C) brachycephaly (bilateral coronal suture), (E) anterior plagiocephaly (unilateral coronal).

single surgeon experience with surgery for primary craniosynostosis correction was undertaken assessing multiple preoperative and operative variables.

METHODS

We retrospectively reviewed the cases of 356 children (371 surgeries) who were admitted consecutively to our service with primary craniosynostosis from January 2001 to December 2021. For the purposes of analyzing the patients' outcome, medical charts, imaging findings, and operative notes were reviewed.

This study was approved by the local ethical committee. Patients were classified based on the type of synostosis present: sagittal (scaphocephaly), metopic (trigonocephaly), unilateral coronal (anterior plagiocephaly), bilateral coronal (brachycephaly), true lambdoidal synostosis, oxicephaly, and multiple-suture synostosis (complex). (Figure 1). We also divided the patients in non-syndromic craniosynostoses (NC) and syndromic craniosynostoses (SC) (faciocraniosynostoses). Skull deformation due to positional plagiocephaly and/or any secondary skull deformities were excluded in this series. In this present study, the surgical techniques presented will be related to non-syndromic craniosynostosis.

Operative Technique and timing for non-syndromic craniosynostosis

Operative procedures were customized for each patient with the following techniques and principles typically being followed. After 2002, a bicoronal incisions are frequently used for exposure and access to the craniofacial skeleton. A zigzag design was often used to camouflage the resultant

scar and the osteotomies were made using Daniel Marchac templates. (Figure 2).

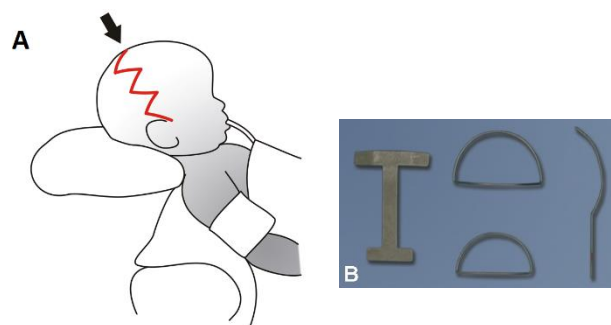


Figure 2- Illustration showed (A) a typical zigzag bicoronal surgical incision (arrow). (B) Specialized surgical instruments. Metal templates performed by Daniel Marchac for craniofacial surgeries.

Scaphocephaly: A complete remodeling of the entire vault (total vertex craniectomy) was performed in only 7 cases in 2001 and abandoned, and then simple craniectomies were performed along the sagittal suture and around the parietal region in less severe cases, especially those seen early (before 6 months of age). Frontal and occipital barrel staving osteotomies with recontouring were typically performed in the most severe cases as well. (Figure 3).

Biological glue was used for final bone assembly. In scaphocephaly, we generally do not use absorbable plates.

In general, after one year of age, we used the cranial morcellation technique. If there is persistence of an important frontal bossing, we correct it with a new frontal.

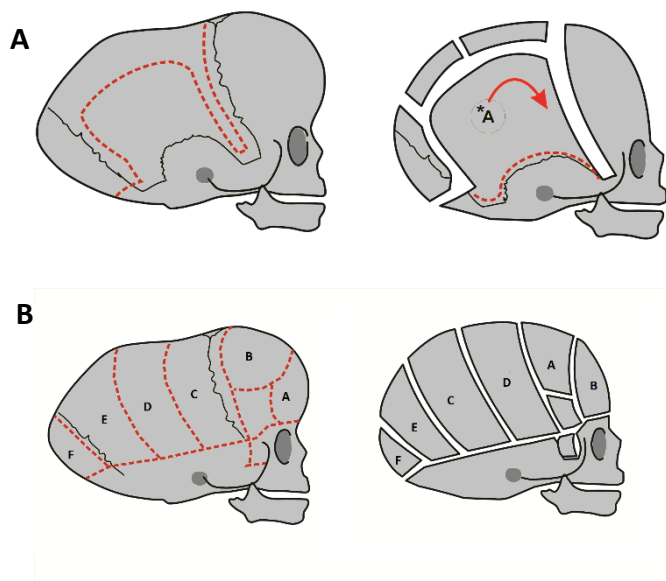


Figure 3- (A) Illustration demonstrating the cranial remodeling technique for correction of scaphocephaly, before 1 year of age (preferably between 3 and 6 months). In detail (asterisk), the “green stick” fracture can be seen to correct the lateral retraction; (B) Illustration demonstrating the surgical technique in late scaphocephaly. It consists of morcellation and frontal remodeling in cases diagnosed after the first year of life. Complete reconstruction of the skull.

Trigonocephaly: The V-shaped supraorbital bar was straightened and a new upper forehead was reconstructed using a bone flap taken from somewhere else on the vault, usually the posterior part of the forehead. (Figure 4).

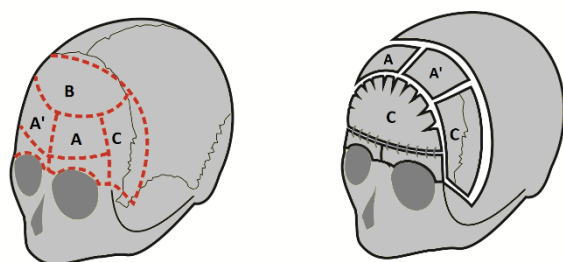


Figure 4- Illustration demonstrating the technique for correcting trigonocephaly, through complete remodeling of the frontal region with the making of the supra-orbital bar and resection of the frontal stenosis (forehead reconstruction).

Anterior Plagiocephaly. The supraorbital bar is lifted with a tenon taken in continuity in the temporal fossa on the recessed side. It is then brought to a normal shape by incomplete cuts at the posterior surface and fixed in the correct position with advancement of the temporal tenon. A new upper forehead is selected on the vault and wired to the supraorbital bar. (Figure 5)

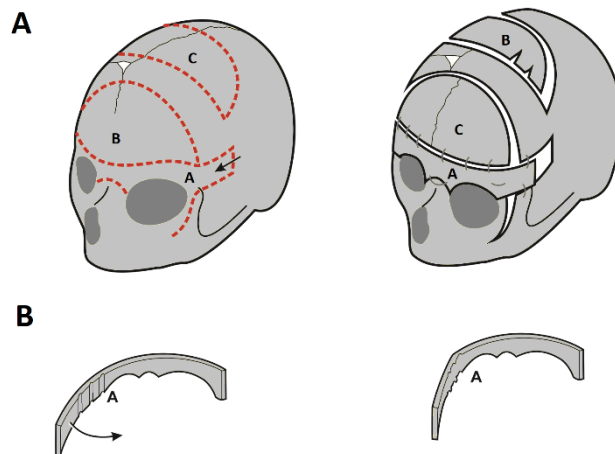


Figure 5- (A) Illustration demonstrating the technique for correcting anterior plagiocephaly, through complete fronto-orbital remodeling with unilateral advancement. (B) In detail, the remodeling of the supraorbital bar, by performing osteotomies to obtain the final shape. Forehead reconstruction for right plagiocephaly.

Brachycephaly: These patients were treated with removal of both sides of the supraorbital bar with tenon extensions. The bulging of the temporal fossa was corrected by lifting a small bone flap and putting it back with the concavity outward. For unilateral coronal synostosis, a supraorbital bar with tenon extensions was used either unilaterally or bilaterally, depending on the attending surgeon's assessment of the degree of deformity. (Figure 6).

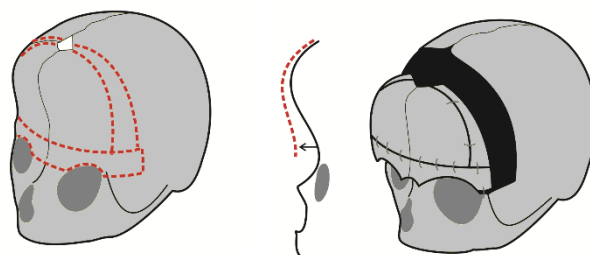


Figure 6- Illustration demonstrating the brachycephaly correction technique, through frontal remodeling and bilateral fronto-orbital advancement. Floating forehead advancement for brachycephaly.

Lambdoidal stenosis was corrected via bilateral parietal-occipital craniotomies. The bone flaps were then reconfigured to produce the best posterior expansion.

A posterior horizontal expansion can be performed, mostly as a first step in the youngest children. A prone position of the patient is necessary to allow direct access to the posterior part of the skull. A posterior coronal incision is performed, and the occiput is exposed. This region is then mobilized and careful extra-dural undermining performed in front of the posterior and lateral venous sinuses. A large posterior bony flap is raised according to a bilateral tongue-in-groove design. (Figure 7).

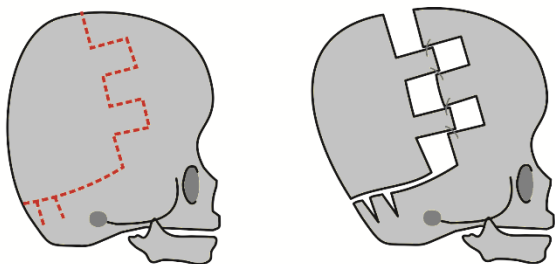


Figure 7- Illustration demonstrating the surgical technique for correcting true posterior plagiocephaly ("tongue and groove"). Posterior expansion of the skull.

Oxycephaly. The supraorbital bar is lifted and repositioned with an anterior rotation. The fixation is provided by a bony Z-plasty performed in the temporal fossae. The upper forehead is reconstructed using a bone flap taken from the upper part of the previous forehead and transposed. (figure 8)

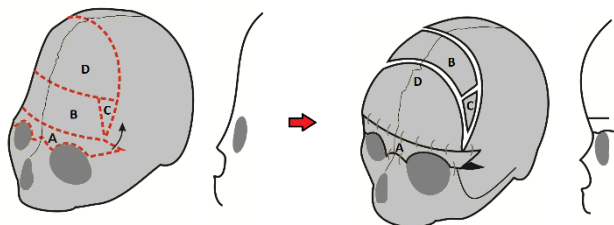


Figure 8- Illustration of the technique for reconstruction of the fronto-orbital region in oxycephaly. Forehead reconstruction for oxycephaly.

The purpose of the frontocranial remodeling is to restore the normal anatomy of the forehead and cranial vault and to allow normal growth of the brain. The forehead is considered to be composed of an inferior part, the supra-orbital bar, and a superior part, the remainder of the forehead up to the coronal region. These two parts are treated separately. If necessary, the rest of the vault behind the coronal region is also remodeled.

The fixation of the fronto-orbital bar (*bandeau*) and new frontal changed according the time in this series and was made from steel wires, absorbable wires and recently the introduction of absorbable plates.

Analysis of Operative Results

The Whitaker scale [7] to asses aesthetics results was performed in non-syndromic craniosynostosis. This is a scale ranging from I for excellent; II for incomplete, soft tissue or lesser bone contouring revisions desired; III for poor, necessitating surgical correction and IV for failure (reoperation mandatory). Statistical analysis was performed using SPSS for Macintosh® version 22.0 software (SPSS,

Inc.). We compared categorical data using the chi-square test and the Fisher exact test, and the nonparametric Mann-Whitney U test between independent groups. A probability value was deemed significant at less than 0.05.

RESULTS

The mean age of the 299 (83.9%) non-syndromic patients at the time of surgery was 7 months $\pm$ 1.4 years (range, 32 days – 9.1 years) and in the 57 (16.1%) syndromic forms it was 1.6 years $\pm$ 2.3 years (range, 24 days-10.9 years) ($p=0.034$). The trigonocephaly (86%) and scaphocephaly (84.5%) groups were responsible for a male preponderance in non-syndromic cases, with a male/female proportion of 6:1 and 5:1, respectively ($p=0.0001$). A female predominance was observed only in the brachycephaly group (64.8%). There was a statistically significant sex difference between non-syndromic versus syndromic cases ($p=0.0001$). According to the suture involvement the group was composed of sagittal (scaphocephaly) in 136, metopic (trigonocephaly) in 48, unilateral coronal (anterior plagiocephaly) in 47, bilateral coronal (brachycephaly) in 35, sagittal and bilateral coronal (oxycephaly) in 15, lambdoid in 10, complex in 11, Kleeblattschadel (cloverleaf) in 10, and pansynostosis in 7. Syndromic cases and no classified sutures were 37 cases. (Figure 9).

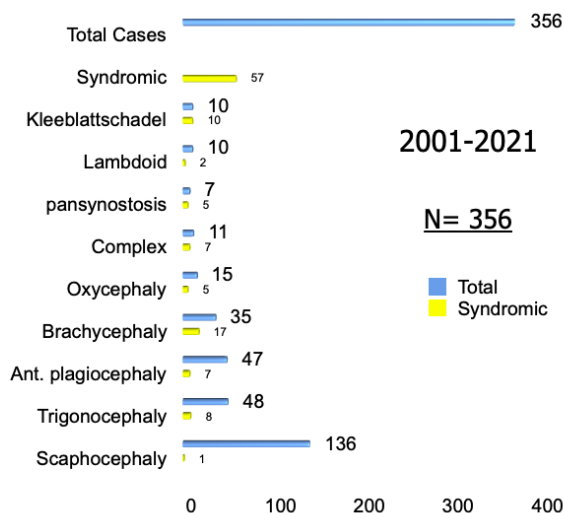


Figure 9- Bar graph representing the different types of craniosynostosis in non-syndromic and syndromic cases.

For syndromic forms Crouzon syndrome was the most frequent, occurring in 21 (36.8%), Apert in 15 (26.3%), Pfeiffer in 11 (19.2%), Saethre Chotzen in 5 (8.7%), and other in 5 (8.7%).

Figure 9 represents each form of craniosynostosis in syndromic and non-syndromic group. Operative procedures were customized for each patient. For the sagittal synostosis group a total calvarectomy was performed in 7 (5.1%) patients in 2001, and then simple craniectomies were performed along the sagittal suture and around the frontal

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and parietal region in 108 (73.5%) patients. A complete frontocranial remodeling was performed in 21 (15.4%) cases.

For the anterior plagiocephaly and brachycephaly cases, the advancement of supraorbital bar was done uni or bilaterally, respectively. Complex cases with multiple synostosis sutures were treated by total cranial vault remolding associated with fronto-orbital advancement. It is striking that the 10 cases of Kleeblattschädel (cloverleaf) malformation were operated on at earlier age (median 92 days) than any other group. Due to the total sutural involvement and high risk of resultant damage to the central nervous system from increased intracranial pressure, these patients were treated quite urgently. Genetic syndromes presented more commonly than expected with bilateral synostosis, the Kleeblattschädel deformity, and multiple suture synostosis /pansynostosis ($p=0.001$).

In this series, 312 (87.6%) of patients received intraoperative and/or perioperative blood transfusion in pediatric intensive care unit. The mean rate of blood transfusion was 11-20 ml/kg in 250 patients (80.1%). We did not observe any major complication associated with blood transfusion.

Complications were observed in 15 out of 356 (4.2%) patients. Intraoperative hemorrhage with hypovolemic shock (5) and CSF leakage (6) were the most common ones. Morbidity was statistically significantly associated with monobloc fronto-facial advancement ($p=0.05$). There was only one death in the all series related to perioperative period. There was a syndromic brachycephaly in Apert case presented with a larynx malformation and consequent severe respiratory problems.

The mean follow-up was 3.7 years (9 months to 22 years). Morphological results in NC were classified in category I in 210 (70.2%), II in 82 (27.9%), and III in 7 (1.9%) cases for NC.

In non-syndromic craniosynostosis reoperations for craniofacial deformity were necessary in only 4 cases (1.3%). The mean follow-up was 3.7 years (9 months to 20 years).

The hospital mean length of stay in our series was total of 3.2 days for NC and 1.3 days in pediatric ICU.

DISCUSSION

The optimal timing of craniosynostosis repair depends on a multitude of factors, which include the presence of elevated intracranial hypertension, the operative technique employed, and surgeon preference. The aim of surgical intervention in craniosynostosis is to excise the prematurely fused suture and correct the associated deformities of the calvaria [6, 8, 9].

Although it is currently considered that surgery is indicated mainly for cosmetic reasons in isolated craniosynostoses, the functional aspects of the treatment must not be underestimated. Increased intracranial hypertension is more frequent in craniosynostoses affecting several sutures. Uncorrected synostosis quite frequently is associated with an increase in intracranial pressure [8].

It is now generally accepted that operations for craniosynostosis should be done as early as possible. It is established that the craniofacial population are at risk of neurocognitive deficits, including lower generalized intelligence quotient, learning disabilities, language delays, and behavioral difficulties, compared to the general population [11]. According to the literature surgery for NC should be done before one year of age to prevent to prevent impairment of their mental development [12].

In the current series in NC the mean age at surgery was 7 months whilst in syndromic patients it was 1.6 years. Our preference is to perform surgery as soon as possible to prevent the effects of intracranial hypertension in the developing brain. In our series we found a striking male preponderance among the sagittal and metopic synostosis patients. That finding is not well understood.

In non-syndromic cases, the standard surgical techniques published by others was sufficient to obtain good results in late outcome [6, 9, 13, 14].

A frequently raised issue in craniofacial surgeries in the pediatric population is blood loss. Consequences of perioperative blood loss and subsequent blood-product administration can be grave, from hemodynamic instability to transfusion-related reactions and their sequelae. However, throughout history there has been a significant advance in surgical and anesthetic techniques, in addition to safety in blood transfusion.

Indeed tranexamic acid (TXA) reduced blood loss and transfusion requirement in patients undergoing surgery for craniosynostosis [15]

In the present series 81.1% of patients received blood transfusion without any problems related. Following the aforementioned intent of minimizing blood loss, a skin incision using low-intensity electrocautery. The monopolar opening of the epidermal and dermal layers using scissors, allowing the hemostatic control of the emissary vein, consequently avoiding unnecessary galeal bleeding.

In our series almost 70.2% of patients reached an excellent morphological result with only one surgical procedure. Most craniosynostoses were treated by frontocranial remodeling, including some cases of scaphocephaly. In cases of harmonious craniosynostoses

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with intracranial hypertension a simple decompression was performed, using free flaps.

The purpose of frontocranial remodeling was to restore the normal anatomy of the forehead and cranial vault and to allow normal growth of the brain. For selected cases posterior fossa decompression should be performed [16].

Our global complication rate was 4.2% and recurrence as pansynostosis was observed in four (1.3%) patients, who needed a second surgery. Pearson et al. [17] reported an institutional total major reoperation rate in 22.9% and 19.5% in nonsyndromic rate. This number is higher than the comparative data of McCarthy et al. [14] (13%) and Williams et al. (5%). [18].

These authors also point out that rigid fixation with metallic wires, plates, and screws could be associated with a high rate of complications in children and described that all other ways of fixation had an increased risk of major reoperation (n=49; 28%) when compared with resorbable plating (n=23; 17%). We disagree partially with these authors because, in our series, we used wires to fix a supra-orbital bar in the forehead in a large number of patients, and in only one patient we needed to remove. In the current series we did not use metallic plates or screws. Resorbable plates are an excellent option to stabilize fronto-orbital advancement.

The cost of this material has fallen over time, thus expanding its use in public hospitals. The low complication and mortality rates may be attribute to improved care provided by multidisciplinary pediatric teams, stressing the importance of these teams in caring for children with craniosynostosis [19].

In this large series the functional and the cosmetic results are better after early surgery and operative risks are not higher in infants than in older children independent of which suture is stenosed. The principles of the surgical treatment of single suture craniosynostosis have practically remained unchanged in our center. On the other hand the management of faciocraniosynostosis has greatly changed. It was in fact greatly modified by the introduction of the distraction techniques to craniofacial surgery. The complexity of the theoretical and practical armamentarium nowadays necessary for treating craniosynostoses emphasizes the referral of patients to specialized centers with multidisciplinary craniofacial teams which include geneticists, pediatric neurosurgeons, maxillofacial surgeons, head and neck specialists, intensive care pediatricians, oculoplastic surgeons and psychologists, in order to assure that the child with craniosynostosis is receiving the best available treatment.

CONCLUSIONS

Non-syndromic craniosynostosis can be corrected with standard craniofacial techniques with good outcomes and

relatively low morbidity and mortality. For syndromic cases a multidisciplinary team is fundamental to achieve satisfactory results.

DISCLOSURES

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki. Approval was granted by the local Ethics Committee (Hospital das Clínicas de Ribeirão Preto e da Faculdade de Medicina de Ribeirão Preto), number: 4.829.130

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 declare no conflicts of interest with respect to the content, authorship, and/or publication of this article.

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Figures were adapted from reference 20.

CONTRIBUTIONS

Ricardo Santos de Oliveira: Conceptualization; Data curation; Formal Analysis; Investigation; Methodology; Project administration; Writing – original draft; Writing – review & editing

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