Original Paper

ISSN: 2675-3626

Twenty Year review for Craniofacial Distraction in syndromic craniosynostosis

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Introduction: Craniosynostosis are cranial deformities resulting from the early closure of one or more sutures. Concomitant facial changes usually result from the involvement of multiple sutures, which may lead to restriction of cranial growth and brain expansion, ocular compression, and breathing difficulties. The distraction osteogenesis (DO)for frontofacial or posterior advancement have increased the safety of the procedure. The aim of this study was a critical evaluation of the use of DO in the surgical treatment of syndromic craniosynostoses, based on the experience of a single craniofacial team.
Methods: Retrospective review of third six patients operated from 2003 to 2022 by the same craniofacial team.
Results: 19 boys and 17 girls were operated, all with rigid distractors. Twenty-nine of them underwent monobloc frontofacial advancement. In seven patient a Le-Fort III/subcranial advancement was performed. A posterior craniofacial advancement was performed in 5. In 11 a fronto-orbital advancement was done in the first year of life. The mean age was 6.0 years (range, 6 months to 13 years). All patients presented with some degree of exorbitism, upper airway obstruction and signs of intracranial hypertension. Cerebrospinal fluid leakage was the most frequent complication (9 cases), pterygomaxillary disjunction was revised in 8, lateral rim orbital fracture in 3 cases, and 3 complication related to the device.
Conclusions: The evolution of surgical techniques allowed DO with rigid distractors to be an important tool for treating the craniofacial issues related to syndromic craniosynostoses. The multidisciplinary team's learning curve is critical to reducing complications associated with osteogenic distraction.

Keywords: Distraction osteogenesis; craniofacial surgery; Crouzon; Apert; Pfeiffer; pediatric neurosurgery; syndromic craniosynostosis

INTRODUCTION

The use of distraction osteogenesis (DO) in the treatment of craniofacial deformities started in 1992, by McCarthy (1). Since then, there have been outstanding advances in this field, both technological and surgical, leading to a more comprehensive management of complex defects and with good results overall. The occurrence of craniosynostosis, defined as the premature closure of calvarial sutures, inevitably produces a restriction in the growth vectors of the skull and ultimately leads to typical deformities of facial and cranial bones (2,3).

Syndromic craniosynostosis often have a retraction of both the frontal region, including skull base and the midface, and consequently show ophtalmological, intellectual and breathing disturbances (4-10). (Figure 1).

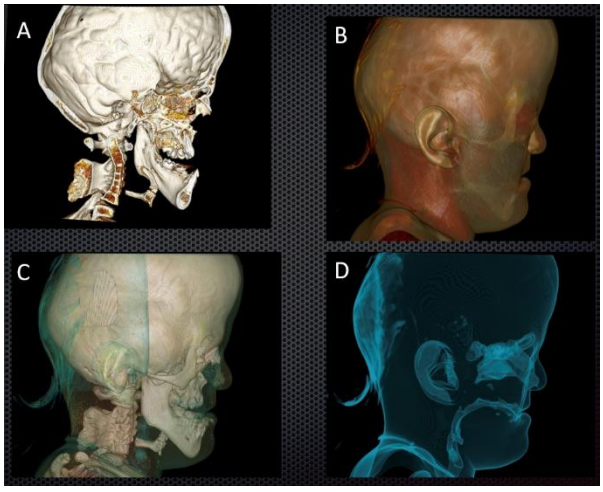

Figure 1- Faciocraniosynostosis typical findigs. A) Lateral view of 3D CT scan showing signs of intracranianal pressure in the inner part of skull; B) exorbitism; C) maxilar retrusion, D) obstructive sleep apnea; E) Fusion of images.

Received: 14 March 2023

Accepted: 01 May 2023

Surgical treatment of faciocraniosynostosis involves complex techniques which must address two issues: prevention of the cerebral damage secondary to craniosynostosis, optimal morphological correction of the facial retrusion and its consequences (exorbitism, and upper airways impairment) (11).

The frontofacial retraction that occurs in complex syndromic craniosynostoses is traditionally managed with advancements of both the upper (frontal) and midface (maxilla), in two steps (3-5,9, 11). In the past, standard craniofacial advancements had some technical limitations and high morbidity and mortality (3). Recently, the use of rigid devices for craniofacial distraction has become the gold standard for the management of these complex cases, producing a gradual expansion of the restrained structures (specially the frontal lobes), better compliance of soft tissues and progressive development of newly formed bone tissue (12).

The aim of this paper is to present our multidisciplinary experience in managing syndromic craniosynostosis with DO, with an emphasis on surgical techniques and pitfalls and perioperative complications.

METHODS

Patient population

Third-six patients were submitted to craniofacial DO between 2003 and 2022, in two different centers: Ribeirao Preto Medical School University of Sao Paulo and Sao Lucas Ribeirao Preto Hospital.

The same multidisciplinary craniofacial team underwent a total of 54 surgical procedures regarding their craniofacial deformities. Their ultimate treatment was based on craniofacial osteogenesis using rigid devices for distraction. Medical charts, surgical notes and other relevant data were assessed and analyzed individually for each patient. Frontofacial Monobloc Advancement (FFMA) was performed in 29 patients as their definitive treatment; a Le-Fort III osteotomy and /or subcranial for midface advancement was done in seven patient, and a posterior craniofacial advancement was performed in 5 cases.

Eighteen patients needed other surgical procedures before craniofacial distraction: eleven patients underwent fronto-orbital advancement (FOA), 5 needed a posterior fossa decompression, VP-shunt in 3 cases, and 2 cases a calvarial expansion due to intracranial hypertension.

Nineteen internal distraction devices (KLS Martin®, Germany), fifteen external devices (Synthes®, West Chester, PA, USA / RED device, KLS Martin®, Tuttlingen, Germany), and two external devices (Traumec®, Rio Claro, Brazil) were used (Figure 2).

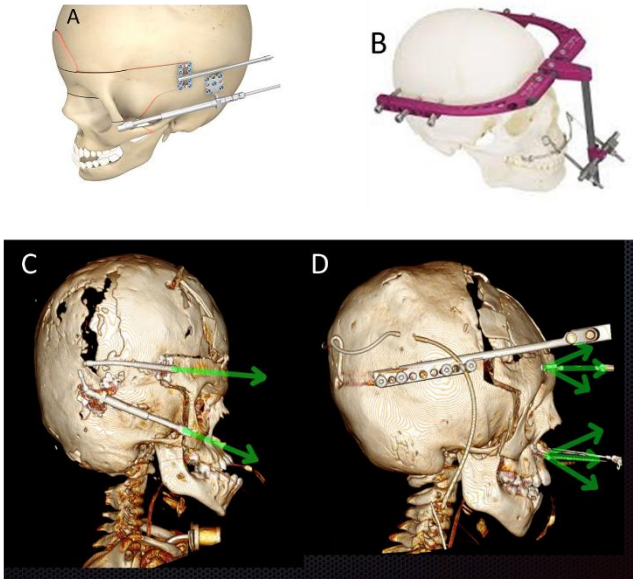

Figure 2- Illustration showing the internal (A) and external (B) devices for craniofacial distraction. C and D) 3D CT reconstruction for internal and external devices showing the vectors for distraction.

The monobloc advancement vector is defined intraoperatively and cannot be changed in the postoperative period for internal devices, unlike the external distraction that allow a refinement of the vectors according to the evolution of the frontal advancement.

All patients underwent pre-operative Computerized Axial Tomography (CT) scans, both for planning the surgery and to look for intracranial associated pathologies. Preoperative evaluation with the purpose of analyzing midface advancement a radiological free software (OsiriX Lite for Macintosh®, California, USA) was used. Using three dimensional reconstruction, we were able to measure the distance between a standard fixed anatomical landmark (the most anterior point of the anterior margin of the foramen magnum - AFM), the glabela and the maxillary point (the posterior most point in the concavity of the alveolar process of the maxilla bilaterally - MP), establishing the upper and midface advancement, respectively. Afterwards, the pre and post-operative images were fused and the advancement measurements were confirmed (Figure 3). For evaluation of exorbitism, a exophthalmometer was used. Measurements were obtained from the lateral orbital rim to the corneal apex. Third-two patients underwent a preoperative polysomnography.

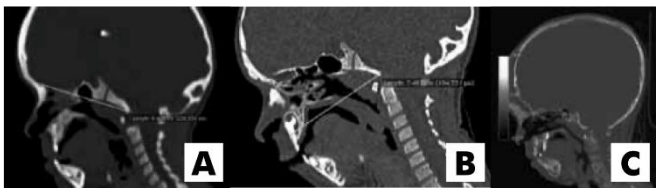

Figure 3- CT scan (sagittal reconstruction) images of a 5-year- old patient with Crouzon's syndrome. All images were produced using a radiological software (OsiriX®). A) measuring the distance between the AFM and the glabella; B) measuring the distance between the AFM and the MP; C) fusion of the pre and post-operative images, allowing a better visualization of the craniofacial advancement after distraction.

Surgical technique and distractors

Frontofacial monobloc advancement

A coronal approach and a subperiosteal dissection allowed: (i) to expose the fronto-orbital bandeau and the zygomatic arches, (ii) to perform the circumferential orbital dissection, (iii) to detach the temporal muscles from the pterional areas to access the pterygomaxillary disjunction. The craniectomy of the frontal flap was carried out with the

piezotome or the craniotome in case of thicker bone, and an extradural dissection of the anterior skull base was performed.

The basis of the nasal septum was disinserted, the osteotomy of the vomer and the pterygo-maxillary disjunctions were achieved using a curved osteotome. The osteotomies of the fronto-temporal region, the zygomatic arch and the lateral orbital wall were carried out. Secondly, the osteotomies of the orbital floors and those of the medial walls were performed, the latter being posterior to the lacrimal crest. Finally, the down-fracture of each hemi-maxilla, still attached to the ipsilateral orbito-zygomatic frame, was then carried out using a Rowe forceps.

The Le Fort III-type osteotomy

This procedure represents the highest level of maxillofacial osteotomy performed without a craniotomy. It separates the face from the skull with osteotomies across the orbital floor, lateral orbital walls, zygomatic arches, and nasofrontal area and with a pterygomaxillary disjunction

The orbital osteotomies go deep inside the orbit, behind the lacrimal system, and instead of going through the posterior hard palate—as originally described by Gillies—Tessier performed a disjunction between the maxillary tuberosities and the pterygoid plates (Figure 4) (13, 14).

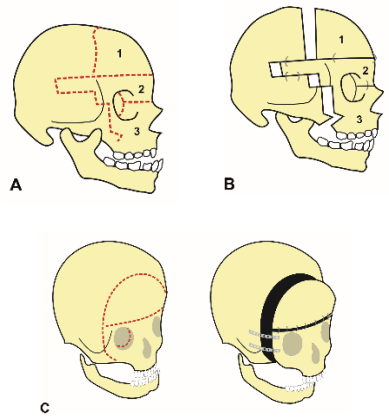

Figure 4- Illustration representing the osteotomies sites to perform the advancement of the midface. (A) Le Fort III and Le Fort IV type osteotomies, including the forehead; (B) advance in monobloc; (C) fixation and progressive advancement with rigid distractors

Around 18 months of age, an FFMA with DO is performed. It further decompresses the brain, improves respiratory function, and corrects exorbitism. Because we operate at such an early age, we favor internal over external distractors. We use 2 pairs of distractors. One pair is placed superiorly (Arnaud cranial distractors; KLS Martin, Tuttlingen, Germany) and distracts the forehead, whereas the other pair (Marchac-Arnaud cranial distractors; KLS Martin) is placed immediately above the zygoma and pushes the orbit forward.

We lowered the rate of distraction from 1 mm/d to 0.3 to 0.5 mm/d, starting at day 5. We leave the distractors in place for 4 to 6 months. Maintenance in pediatric ICU with intubation for at least 24–48 h to wait for diminution of initial swelling.

In older patients, DO is used in Le Fort III advancements and FFMA. We frequently use the rigid external distractors (REDs), which allow precise control over the rotation of the maxilla and are well tolerated after 5 years of age.

Results of treatment were categorized on the basis of the need for additional surgery and varied from no refinements necessary (category I) to major reduplication of the initial procedure (category IV) according to Whitaker et al., 1987 (15).

Statistical analysis

Statistical analysis was performed using SPSS for Macintosh, version 16.0 (SPSS, Inc.). The chi-square and Fisher exact tests were used to compare categorical data, and the nonparametric Mann-Whitney U test was used to compare independent groups. A probability value was deemed significant at less than 0.05.

RESULTS

Craniofacial distraction was completed in 36 patients from 2003 to 2022. In the same period 314 patients were submitted to craniosynostosis surgery correction. There were 19 males and 17 females. Of these patients, 19 (52.7%) had Crouzon syndrome, 10 (27.7%) had Apert syndrome and the last 7 (19.4%) Pfeiffer syndrome. Their mean age was 6 years, ranging from 4 months to 14 years. There were 19 (52.7%) boys and 17 (47.2%) girls.

All our patients presented with symptoms and signs of intracranial hypertension, including developmental delay. The mean pre-operative distance between the AFM and the glabella was 7.17 cm; post-operatively, it was 8.61 cm. Likewise, the mean distance between the AFM and the MP was 6.19 cm before surgery and 7.75 cm after. As a result, a mean advancement of 1.44 cm was obtained for the upper face and 1.56 cm for the midface.

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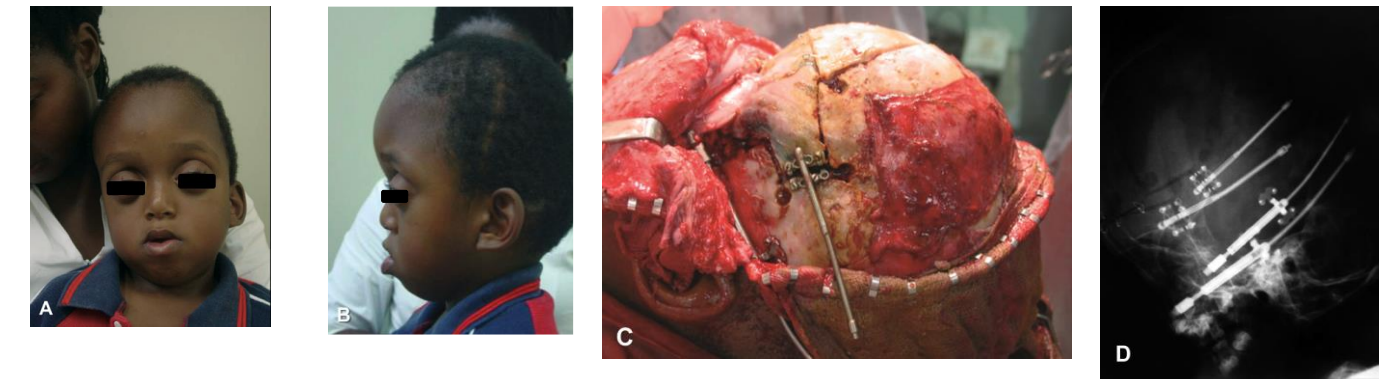

Figure 5- Monobloc frontofacial advancement in a 5-year old child with Crouzon's syndrome. A and B) preoperative photographs; C) intraoperative picture showing the placement of the internal device (fronto-orbital and maxilla - arrow); D) postoperative skull X-ray showing bilateral internal device in a correct postion before to start the distraction protocol. E) postoperative photographs after distraction showing a normalization for exorbitism and midface retrusion.

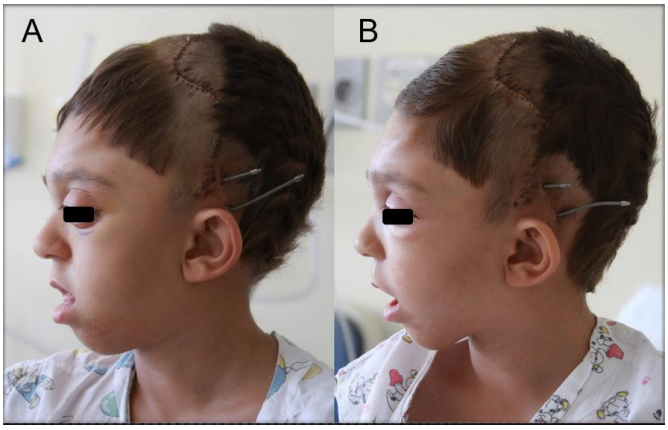

Figure 6 – Internal device for DO, A) Preoperative; B postoperative photographs

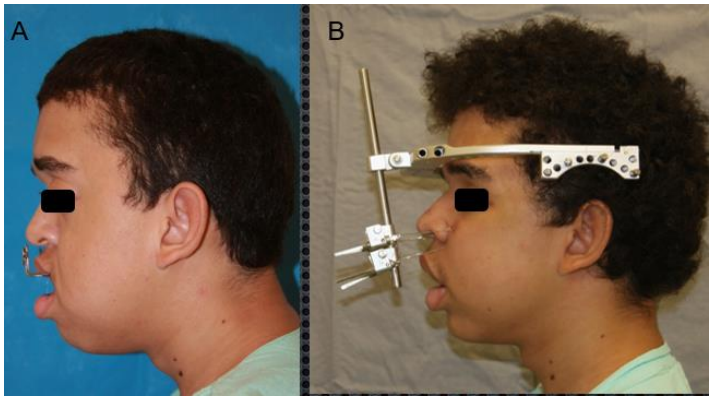

Figure 7 – External device for DO, A) Preoperative; B postoperative photographs

All patients underwent craniofacial distraction with rigid distractors. Twenty-nine of them underwent monobloc frontofacial advancement. In seven patient a Le-Fort III /subcranial advancement was performed. A posterior craniofacial advancement was performed in 5 cases. In 11 out of 36 a fronto-orbital advancement was done in the first year of life. The mean age for craniofacial distraction was 6.0 years (range, 6 months to 13 years). All patients presented with some degree of exorbitism and upper airway obstruction and also signs of intracranial hypertension pre-operatively. Craniofacial distraction surgery promoted an improvement in all patients. (Figures 5-7).

All patients had an exorbitism greater than 21 mm, 10 (27%) of them being greater than 30 mm (severe exorbitism). The mean pre-operative protrusion measurement was 27.4 mm. After the distraction process, all patients had a protrusion of 21 mm or less (mean 18.5 mm), resulting in a significant improvement in their proptosis (p=0.0065).

Twenty-five (69.4%) patients showed clinical features of severe upper airway obstruction, and six of them (16.6%) were even submitted to a tracheostomy. After surgery, all patents had a clinical improvement of their breathing pattern confirmed by polysomnography.

Complications were observed in 12 out of 36 (33%) patients. Nine (25%) cases presented a post-operative CSF leak; seven of them required a post-operative lumbar drain, resolving within no longer than five days; two of them had to be reoperated for dural repair. One case presented with a later recurrence of a CSF leak and a neuroradiological investigation showed a right frontobasal encephalocele by the cribriform plate. A endonasal surgery plus lumboperitoneal shunt were done to repair the skull base defect and CSF fistula. (Figure 8).

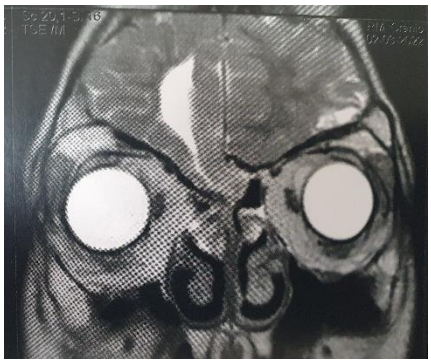

Figure 8- A 14 year-old boy with Apert syndrome presented with a later recurrence of a CSF leak. A coronal MR showing a right basal encephalocele treated by endonasal surgery and LP-shunt.

All cases required intra and post-operative blood transfusion. Five patients showed a perioperative hypovolemic shock that recovered with adequate blood transfusion.

Pterygomaxillary disjunction was revised in 8 cases. Three cases presented with a lower orbital rim fracture and/or fronto-zygomatic junction requiring surgical osteosynthesis, and 3 complication related to the device itself (cranial pin migration, exposition of cranial internal device, and another case showed a displacement of the zygomatic part of the internal distractor device, which needed to be repositioned). Figure 9.

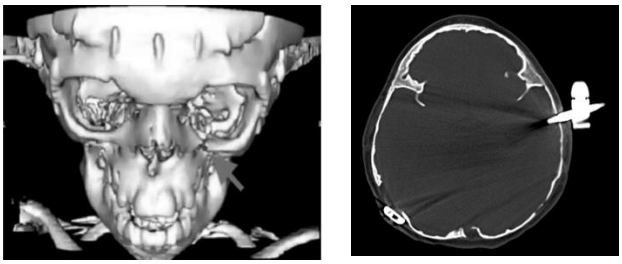

Figure 9- A) 3D reconstructed CT scan showing a post-operative zygomatic-maxillary fracture (red arrow) due to distraction in a patient with Crouzon's syndrome. B) Axial CT showing a intracranial migration by pin in an external device during the distraction process.

Three patients exhibited an orbital cellulitis, probably because of the communication between the ethmoid sinuses and the orbital compartment during facial osteotomies, that recovered after a course of antibiotics. We divided our series in two different period (2003-2014 / 2015-2022). The complication rate dropped to 39% to 23%. Figure 10.

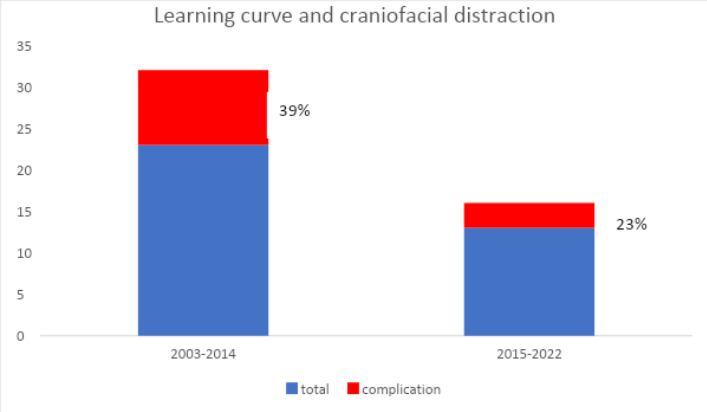

Figure 10- Bar Graph showing two different periods (2003-2014 / 2015-2022) for the craniofacial team related to complication rate.

Twenty-six patients were categorized as I and II (72.2%), four patients categorized as III, and six cases as IV according Whitaker's classification.

The follow-up ranged from 6 months to 20 years (mean follow-up 6.7 years).

DISCUSSION

In 1978, Ortiz-Monasterio developed a technique that enabled the advancement of both the upper and middle facial heights in a single procedure, thus sparing at least one surgery (16).

Albeit a huge development, it also brought a high morbidity that outweighed its benefits. The correction of craniofacial deformities associated with syndromic craniosynostoses was initially based on a two-staged approach, whose first step was the fronto-orbital advancement and the second one was the LeFort III osteotomy or the advancement of the midface (3,11,12). Indeed, although this technique provided some good results, it also carried out some substantial problems, especially regarding the need for more than one surgical procedure and the disadvantages of reoperations. It must be remembered that these patients already had to undergo a number of surgeries for the other issues related to their syndrome.

Craniofacial advancements with DO allow a progressive bone tissue formation between the separated bones without leaving a retrofrontal dead space (5,6). Therefore, good results can be achieved and the complications minimized. One might argue that the second operation required for the

removal of the distractor is a drawback, but we agree with Arnaud et al. (2) that, in this particular case, the benefits outweigh the disadvantages and that, in the future, the development of resorbable devices May solve this problem.

Many authors have published their experience with craniofacial DO 2,3,5,6,17-28]. It is now well established that gradual distraction reduces the risk of complications, preventing the occurrence of a retrofrontal dead space; also, it remotes a slowly progressive bone formation, with a smaller number of bony defects 3,5,6. In addition, there appears to be a consensus that monobloc frontofacial advancement is an adequate method to improve the breathing disturbances and the exorbitism related to faciocraniosynostosis, as well as intracranial hypertension (3).

Our experience has shown that the distraction interval between the separated bones provided by the distractors (15-30 mm) is enough to enlarge the intracranial, orbital and retropharyngeal and upper airway spaces, thus reducing intracranial pressure and improving the ocular proptosis and breathing obstruction that these patients present with.

As described by other authors (3,5,6,14,29) in order to prevent complications related to residual hematomas, we also perform an immediate small advancement (usually 3-5 mm). This maneuver stabilizes to bony flap as well.

It should also be stressed out that, for the use of the distraction techniques, a multidisciplinary team is mandatory, as well as the awareness that it involves a learning curve (30), which is clearly demonstrated in this series.

In a first period (2003-2014), we had a relatively high rate of complications, but as we became more familiar with this surgical technique, we were able to lower our complications to a minimum (in the last 13 cases, only three patients had some complication). No deaths were reported.

Significant blood loss occurs in most of the patients operated on, since these surgeries involve large osteotomies and soft tissue dissection; additionally, children do not tolerate blood losses as well as adults (5,6,22). The intraoperative hemorrhage related to craniofacial facial and FFMA poses a life threatening for patients, especially children. The risk of bleeding is associated to venous sinuses, venous hypertension due to raised intracranial pressure and also osteotomies of the pterygopalatine fossa (27). Blood transfusion is considered essential, and we believe it to be a safe practice. We have not observed any significant complications associated with this procedure."

CSF leaks are still a common complication, especially in patients that have already undergone previous craniofacial surgeries [3,14,22]. Nine of our patients (25%) needed additional procedures to correct CSF leaks; fortunately, all the leaks resolved and the distraction process did not need to be interrupted. The use of periosteal pedicled flaps to fill in the gaps in the anterior cranial fossa, ethmoid cells and orbital and nasal cavity may reduce the incidence of this complication 3,14.

In our series 18 out of 36 patients needed other surgical procedures before fronto facial craniofacial distraction. Eleven patients underwent FOA.

As per Wagner et al. (2023) (29), previous FOA appears to be linked to elevated rates of significant complications and dural tears in syndromic craniosynostosis patients undergoing fronto-facial surgery. Prior craniotomy results in challenging detachment of the dura mater and fibrosis, thereby raising the risk of cerebrospinal fluid (CSF) fistula.

Complications related to the distraction process itself may appear (30-33). Pterygomaxillary disjunction and midline skull base (the perpendicular lamina and vomer) are two main surgical steps in craniofacial advancement. Failure in these specific points may compromise frontofacial advancement. In the current series 8 cases needed a second-look surgery to review the pterigomaxillary fracture. The reinforce of the zygomatic maxillary junction is very important even in young children because of the fragility of the bone and the risk of fracture after distraction.

Close clinical and radiological follow-up is an important tool to prevent that, and confirmation that the distractors are well positioned and that the distraction vectors are synchronic is imperative.

Finally, simulations using haptic or virtual models, including augmented reality, have become essential tools in advanced surgical procedures for syndromic craniosynostosis (34). The utilization of such simulations in training and surgical programming has gained traction, with evidence showing a reduction in surgical time and, potentially, in complications (35). Our team has utilized virtual resources to program the FFMA in the most recent group of children (12).

Avoiding pitfalls and complications for craniofacial distraction:
• Systematic screening and multidisciplinary team.
• Preoperative assessment including neuroanestesiology and blood transfusion.
• Removal of forehead for safe approach to the base of the skull.
• Reinforcement in the fronto-zygomatic junction.
• Complete pterygo-maxillary disjunction, division of midline structures, and intra-operative positional control with Rowe forceps.
• Prevention of post-operative CSF leak by systematical lumbar evacuation and subsequently drainage in case of per-operatory evidenced CSF leak.
• Maintenance in ICU with intubation for at least 24–48 h to wait for diminution of initial swelling.
• Maintenance of the distractors in place for at least 4-6 months after the end of the distraction process

CONCLUSIONS

Craniofacial distraction osteogenesis has become a safe method for the correction of deformities of the craniofacial skeleton in children with syndromic craniosynostoses. It is important to discuss these potential complications with the team before undergoing craniofacial distraction, as well as any steps you can take to minimize the risk of complications. The use of these techniques encompasses a learning curve and a multidisciplinary team, and, with that in mind, the rate of complications can be minimized and the results turn out to be very satisfactory.

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, number: 3.335.080

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.

Funding

This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors

CONTRIBUTIONS

-Ricardo Santos de Oliveira: Conceptualization Conceptualization, Data curation, Formal Analysis, Investigation, Methodology, Writing – original draft, Supervision

-Matheus Fernando Manzoli Ballestero: Conceptualization, Formal Analysis, Writing – review & editing

-Paulo Barrero Marques-Netto: Investigation, Resources, Visualization

- André Andó: Investigation, Resources, Visualization

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