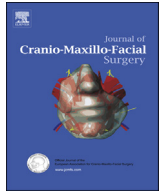




Contents lists available at ScienceDirect

Journal of Cranio-Maxillo-Facial Surgery

journal homepage: www.jcmfs.com

Modified Le Fort III osteotomy: A simple solution to severe midfacial hypoplasia

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ARTICLE INFO

Article history:

Paper received 1 March 2017

Accepted 13 March 2018

Available online 21 March 2018

Keywords:

Midfacial hypoplasia
Maxillary retrognathia
Maxillary hypoplasia
Class III malocclusion
Le Fort III osteotomy
Distraction osteogenesis

ABSTRACT

Purpose: There are multiple conditions that may affect the development of the middle third of the face and with varying degrees of severity. The surgical treatment alternatives for major midfacial sagittal deficiencies consist in Le Fort I, II, or III with conventional osteotomies or with distraction osteogenesis (DO). Both techniques have advantages and disadvantages that should be evaluated specifically in each case. The aim of this report is to present a group of patients with severe hypoplasia of the middle third of the face, with different origins, and their treatment with a Modified Le Fort III osteotomy and distraction osteogenesis, using a minimally invasive surgical approach.

Materials and methods: The surgical technique was performed in a group of patients with severe hypoplasia of the middle third of the face, through a transconjunctival approach with lateral canthotomy and a trans-oral approach. The osteotomy consisted of a Le Fort III without the nasofrontal component. A rigid external distractor (RED) type II or internal distractor was installed. The amount of distraction, surgical time, blood loss, and complications were evaluated.

Results: A total of 7 patients underwent operation, 5 men and 2 women with an average age of 20.8 (range 11–41) years; 3 patients with Crouzon syndrome, 2 with Pfeiffer syndrome, 1 patient with cleft lip and palate sequel, and 1 with a severe non-syndromic class III. The average follow-up was 3.14 years. All patients achieved stable occlusion without postoperative changes, positive overbite and overjet, without relapse in the skeletal position. The average advancement was 14.7 (± 4.07) mm, in 1.1 incisors, and 15.2 (± 3.19) in point A. The average time of surgery was 2.78 (± 0.64) hours, with an average blood loss of 240 (± 48.6) ml. Four patients required a rhinoplasty in a secondary surgery.

Conclusion: This technique shows a surgical approach with low morbidity, short surgery time, and low blood loss. It allows optimal resolution of severe hypoplasia of the middle third of the face with long-term stability. It avoids the use of grafts and osteosynthesis material. By not including the nasal pyramid in the osteotomy design, the size, position, and nasofrontal angle in patients with adequate facial balance is maintained. If nasal correction is necessary, a second surgery may be done. In cases of asymmetrical hypoplasia of the middle third, this osteotomy shows great versatility and can be done unilaterally and/or simultaneously combined with other distractions.

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1. Introduction

There are multiple causes of hypoplasia in the middle third of the face, which can be of congenital or acquired origin. The congenital disorders include syndromic craniosynostosis, non-syndromic conditions, neurocristopathies, facial clefts (lip-palate

or central), and other syndromes like Binder syndrome. Among the acquired alterations, class III malocclusion is a frequent side effect of complex facial trauma (Jabaley and Edgerton, 1969).

Syndromic craniosynostosis is characterized by the early closure of one or more cranial sutures and growth alteration of the cranial base, causing abnormal secondary growth in the facial skeleton. In this range of malformations, the Apert, Crouzon, Pfeiffer and Saethre Chotzen syndromes stand out, in which there is brachycephaly, bilateral coronal synostosis, hypoplasia of the infra-orbital rims and a short anterior cranial base in the sagittal direction. A class III skeletal and dental malocclusion, severe hypoplasia of the middle third, micrognathia, nasal deformity, maxillary compression, posterior crossbite, inverted bite and open anterior bite can be observed (Buchanan et al., 2014). These skeletal abnormalities produce functional alterations such as intracranial hypertension, ocular disorders and obstruction of the upper airway. The surgical sequence for these patients is determined depending on the severity of these functional alterations (Ghali et al., 2002).

In patients with sequelae of cleft lip and/or palate, a significant reduction in sagittal and transverse growth of the maxilla is observed, due to a mechanical limitation caused by the scars from primary surgery on the lip, and hard and soft palate. It is common to observe hypoplasia of the middle third, inverted bite and maxillary compression, often associated with velopharyngeal insufficiency in many degrees (Figuroa et al., 1999; Iannetti et al., 2004).

1.1. Surgical alternatives for sagittal hypoplasia of the middle third

The surgical treatment alternatives for major midfacial sagittal deficiencies consist of Le Fort I, II or III with conventional osteotomies or with distraction osteogenesis (DO). Gillies and Harrison (1950) performed the first Le Fort III osteotomy with net advancement using external fixation, with poor results in terms of stability over time. Tessier then incorporated the placement of bone grafts between osteotomies, significantly improving the stability of the advancement (Tessier, 1971). In the 1980s, the concepts of rigid osteosynthesis were applied to Le Fort III advancements and improved levels of stability over time (Marchac and Arnaud, 1999). However, the skeletal advancement that conventional osteotomies allow is limited, and the use of osteosynthesis can alter skeletal development in growing patients. Skeletal distraction emerges as an alternative for patients requiring a major advancement and in patients who are still growing.

In patients with cleft lip and palate with a severe middle third deficiency, a Le Fort I osteotomy can correct only the occlusal discrepancy, without improving the sagittal projection of the zygomatic area (Denny et al., 2003; Hettlinger et al., 2013). In these patients, DO presents a lower risk of velopharyngeal insufficiency (Taha and Elsheikh, 2016).

1.2. Le Fort III osteotomy

Initially Gillies used the classic Le Fort III osteotomy for middle-third advancement in patients with post-traumatic side effects. Tessier modified it into types (TI, TII, and TIII), differentiating them by the projection of the malar bone and the superciliary arches, optimizing the advancement areas depending on the needs of each case (Nout et al., 2008). These osteotomies require a coronal approach and modify the nasofrontal angle, but not always with a proper balance and esthetic. In response to this, Obwegeser described the Le Fort III osteotomy in a “Butterfly” design, which combines the classic Le Fort I and III osteotomies without involving the nasal component, describing a coronal approach for its performance (Obwegeser, 1969; Marchac and Arnaud, 1999; Nout et al., 2008).

Table 1 Patient characteristics.

Patient no.	Age (y)	Sex	Diagnosis	Kind of surgery/mm of movement in tooth 1.1/point A/distractor type	Time of surgery: hours/Blood loss (ml)	Complications	Rhinoplasty/genioplasty	Follow-up (y)
1	17	M	CLP, left zygoma hypoplasia	Right segmental distraction osteogenesis internal	2.5/200	No	Yes/yes	3
2	14	F	Severe class III non-syndromic	Left Modified Le Fort III/15/17/internal Modified Le Fort III/15/15/RED II	2.5/250	No	No/no	6
3	20	M	Pfeiffer syndrome	Modified Le Fort III/10/13/RED II	3/200	No	No/no	3
4	21	M	Crouzon syndrome	Modified Le Fort III/20/18/RED II	2/180	No	Yes/yes	2
5	41	F	Pfeiffer syndrome	Modified Le Fort III/11/13/RED II	3/250	No	No/yes	3
6	22	M	Crouzon syndrome	Modified Le Fort III/12/11/RED II	2.5/300	2 cranial pins got loose	Yes/yes	2.5
7	11	M	Crouzon syndrome	Modified Le Fort III/20 RED II, simultaneous Bilateral horizontal mandibular distraction/18/20/internal	4/300	No	Yes/yes	2.5
Average	20.85	5M/2F	Crouzon, 3; Pfeiffer, 2; CLP, 1; non-syndromic, 1	Modified Le Fort III: 14.4 (SD 3.69)/15.2 (SD 3.19)	2.78 (+/-0.64)/240 (+/-48.6)		Rhinoplasty, 4; genioplasty, 5	3.14

CLP, cleft lip and palate; F, female; M, male; RED, rigid external distractor.

The aim of this report is to present a group of patients with severe hypoplasia of the middle third of the face, with different causes, and its treatment with a Modified Le Fort III osteotomy using a minimal surgical approach, similar to the one previously described by (Kademani and Tiwana, 2015), but using distraction osteogenesis.

2. Materials and methods

All patients were treated in the private practice of Dr. Rodrigo Fariña and Maxillofacial service of Hospital del Salvador, Santiago, Chile.

This study was approved by the Hospital del Salvador Ethics Committee (number CEC 13082017) and has been carried out in accordance with the Declaration of Helsinki.

The report includes 7 patients: one with cleft lip and palate sequel, 1 with severe non-syndromic skeletal class III, 2 patients with Pfeiffer syndrome, and 3 with Crouzon syndrome. The average age was 20.8 years. There were 2 women and 5 men with at least 24 months of follow-up (Table 1).

All these patients had a severe midfacial hypoplasia and severe class III malocclusion, and needed a midfacial advancement of 10 mm or more (measured in point A). Two patients still growing (patient 2, 14 years old; patient 7, 11 years old).

The surgical technique was performed only through a transconjunctival approach with lateral canthotomy and cantholysis (which allows an appropriate approach of the lateral orbit and the zygomatic bone), in addition to a trans-oral approach.

The osteotomy begins in the medial region of the infraorbital rim just lateral to the lacrimal sac via the transconjunctival approach, spreading downwards towards the lower nasal meatus. Laterally, the osteotomy extends along the anteroinferior portion of the orbital floor up to the lateral wall of the eye socket to the level of the root of the frontal process of the zygomatic bone (the height is customized according to the needs of each patient). Then it descends vertically along the body of the zygomatic bone (Fig. 1)

The medial osteotomy performed from the eye socket is completed with a transoral approach on the latero-base level of the

nose. The vomer is then detached, and the pterygomaxillary disjunction is performed.

The osteotomy can be performed with a reciprocating saw, as well as a cylindrical bur or piezoelectric saw. The full mobility of the osteotomized segment is verified with a Rowe forceps.

A rigid external distractor (RED) type II was then installed in 6 patients with double skeletal anchoring (SARED), 2 at the level of each infraorbital rim and 2 others on the bilateral paranasal base level (Fariña and Salinas, 2017).

Internal distractors were used in 1 patient (cleft lip and palate sequel with a large alveolar cleft), where a unilateral Le Fort III osteotomy was performed, associated with contralateral segmental distraction osteogenesis (Figs. 2–7).

The imaging studies of each patient were analyzed before and after surgery. The sagittal differences of the right central incisor (tooth 1.1) and point A were established in the teleradiography and cephalometry, determining the amount of sagittal advancement required, according to the visual treatment objective [VTO] individualized for each patient. Blood loss in the operating room was measured, and the surgery time was recorded for each patient.

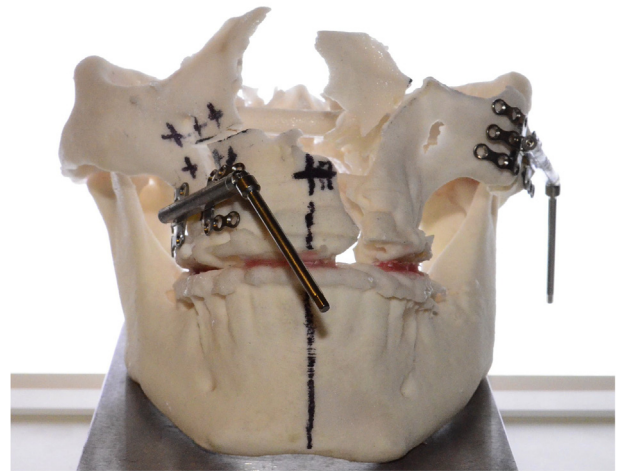


Fig. 2. Biomodel planification of segmental distraction in right side with hemi Le Fort I, and left side with Modified Le Fort III. Large alveolar cleft can it observed on left side (patient 1).

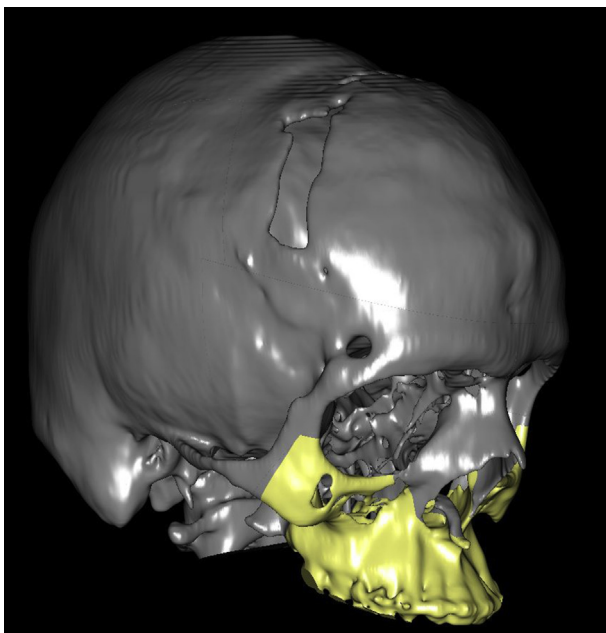


Fig. 1. Design of modified Le Fort III (patient 4).



Fig. 3. Cone beam computed tomogram showing the internal distractor device on left hemi Le Fort III at the end of distraction (patient 1).



Fig. 4. Axial view before surgery in cleft lip and palate patient with maxillary hypoplasia. Left side shows a severe midfacial and zygoma hypoplasia (patient 1).

3. Results

The operative group comprised 7 patients, 5 men and 2 women, with an average age of 20.8 years (11–41 years). Of them 3 had Crouzon syndrome, 2 Pfeiffer syndrome, 1 patient had cleft palate, and 1 patient had severe non-syndromic skeletal class III. The average sagittal advancement of the right upper central incisor was 14.4 mm (8–20 mm; SD 3.69), and the A point advanced a mean of 15.2 mm (11–20 mm; SD 3.19), measured in the sagittal plane perpendicular to the Frankfurt plane.

The average surgical time took 2.78 h (range 2–4 h; SD 0.64), the average blood loss was 240 ml (range 180–300 ml; SD 48.6), and



Fig. 5. Axial view 3 years after segmental distraction in right hemi Le Fort I and left Modified Le Fort III distraction (patient 1).



Fig. 6. Occlusal view before surgery in cleft lip and palate patient with maxillary hypoplasia (patient 1).



Fig. 7. Occlusal view 3 years after segmental distraction in right hemi Le Fort I, and left modified Le Fort III distraction (patient 1).

the mean follow-up 3.14 years (range 2–6 years). All patients achieved stable occlusion, positive overjet and overbite, without relapse of skeletal position (measured clinically by their occlusion). One of the patients, with a tracheostomy since birth, could be successfully decannulated after maxillomandibular distraction (patient 7). Four patients required a primary rhinoplasty in a second surgery, and 5 patients required advancement genioplasty. A complication occurred in 1 patient, when 2 cranial anchoring pins came loose and had to be repositioned (patient 6). None of the patients experienced overcorrection or needed a second surgery for their midfacial hypoplasia (Figs. 4–14).

Skeletal anchoring was easily removed without the need for local or general anesthesia (2–2.5 months after the end of distraction).

4. Discussion

Le Fort III osteotomy was originally described by Gillies and Harrison (1950) for facial correction of craniosynostosis, to achieve a zygomatic and maxillary advancement in a patient with oxycephaly. For this, they made multiple facial approaches on the nasofrontal, paranasal and the inferior palpebral regions, to allow access to the osteotomies areas. Gillies found that the technique's main disadvantage was long-term relapse, showing edge-to-edge



Fig. 8. Facial view before surgery in Crouzon syndrome patient (patient 4).

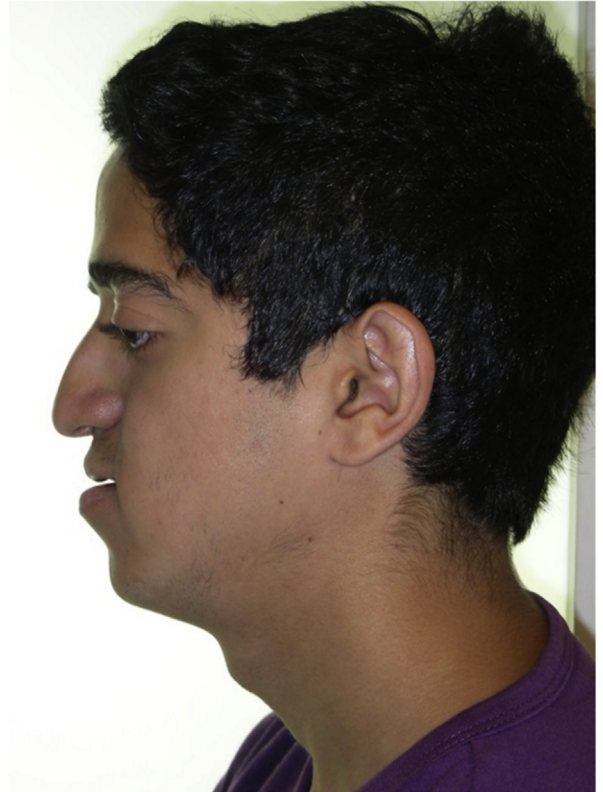


Fig. 10. Lateral view before surgery in Crouzon syndrome patient (patient 4).



Fig. 9. Facial view 2 years after Modified Le Fort III distraction, rhinoplasty, and genioplasty (patient 4).



Fig. 11. Lateral view 2 years after Modified Le Fort III distraction, rhinoplasty, and genioplasty (patient 4).



Fig. 12. Occlusal view before surgery in Crouzon syndrome patient (patient 4).



Fig. 13. Occlusal view 2 years after Modified Le Fort III distraction, rhinoplasty and genioplasty (patient 4).

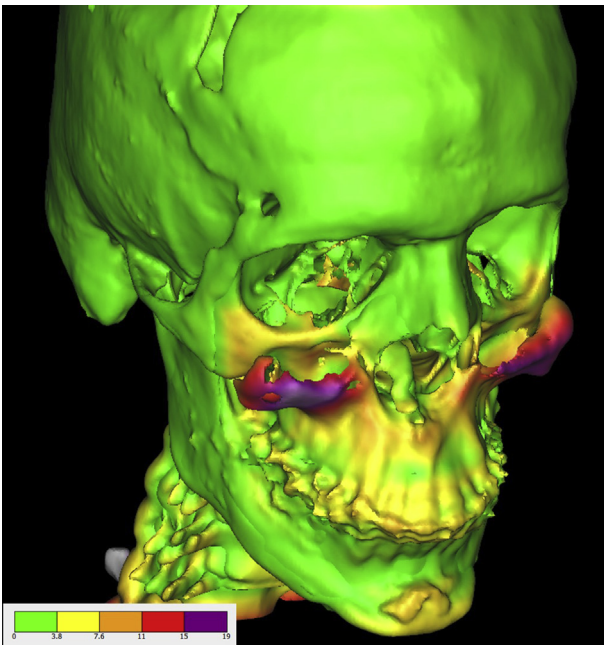


Fig. 14. Superposition of 3-dimensional computed tomography scan, before and 2 years after surgery (patient 4).

bite results after 7 years of follow-up (Gillies and Harrison, 1950). Because of this Tessier (1971) proposed the interposition of bone grafts to improve the results of long-term stability, 10 years later he published the results in 7 patients. He observed that growth of the nasal region continued unchanged in patients who received surgery while still growing. These patients showed poor occlusal stability results, compared with patients who received surgery in adulthood (Tulasne and Tessier, 1986).

One of the first authors to describe the modified Le Fort III osteotomy was Kufner (1971) for the treatment of maxillary retusion, then Epker and Wolford (1975) modified the conventional Gillies osteotomy, avoiding frontonasal mobilization. Many other authors published similar osteotomies. Tiwana and Turvey (2004) described a subcranial Le Fort III osteotomy in midface deficiency. They considered an extracranial approach for surgical correction of the deformity and the use of bone grafts harvested from the cranium or iliac crest (Tiwana and Turvey, 2004). More recently Vu and Tiwana (2016) present a case of Modified Le Fort III osteotomy and mentioned that the subconjunctival access as an option to the traditional coronal approach. They also use bone grafts to stabilize the advancement. García and Sanchez (García et al., 2015) showed a specific design of a surgical guide for modified oblique Le Fort III to make a stable symmetrical bilateral pterygomaxillary cut, decreasing the possibility of injury on vascular structures during the osteotomy, and shorten the surgical time. Later the same authors presented a mathematical formula that gives the angulation needed for moving the midface complex in a simultaneous vertical and sagittal direction. Once given the correct angulation for the desired oblique movement in a stereolithographic model, they made custom surgical guides to achieve the desired movement during surgery (García et al., 2017).

The modification used by us (based on Miloro's (Miloro et al., 2004) and (Kademani and Tiwana, 2015)) has several advantages over the conventional osteotomy: on the one hand, the simplicity and lower morbidity of the approaches (transconjunctival and transoral); on the other hand, it does not alter the position of the nasal pyramid or the nasofrontal angle, contributing to a better facial balance, which could be optimized with a nasal correction afterwards, if necessary.

Due to the need to make major advancements, some authors and also us, propose a distraction osteogenesis to improve the levels of stability and increase the displacement ranges in severe deficiencies, avoiding the use of bone grafts, with all of their associated morbidities and costs (Nout et al., 2008) (Saltaji et al., 2014).

The distraction process requires a high level of cooperation from patients and families. Submerged distractors require a second surgery to remove them. In some cases, managing the distraction vector can be complicated, depending on the type of distractor (internal or external) and the anchorage used. RED-type II distractors allow greater control over the distraction vectors in the 3 directions of space, making it easier to modify them, during the distraction process (Meling et al., 2006). This is the biggest advantage compared to internal distractors, as they do not allow the vectors to be modified, in addition to the fact that their removal does not require a second surgery (Kuroda et al., 2005; Fariña and Salinas, 2017). The RED requires patients tolerance and cooperation for its successful use, in addition to higher postoperative precautions, especially during the consolidation period (Kuroda et al., 2005), during which it can be replaced by a front traction mask and the use of intermaxillary elastics. One of the counter-indications of external distractors is the need for adequate cranial anchoring, which could affect the stability of the device and, therefore, alter the complete distraction process (for example, cranial defects from previous cranioplasties). In these cases the use

of distraction with internal devices and planning with stereolithographic models for the correct vector design must be considered (Nout et al., 2006; Cheung and Lo (2006)) illustrate a case of nasomaxillary hypoplasia treated with Le Fort II osteotomy using intraoral distractors achieving good esthetic and occlusal results. They agree that the use of biomodels enables to calculate the distraction vector, decide where the bone cut should be made and where the distractors should be placed (Cheung and Lo, 2006).

The anchors used for distraction of the middle third of the face have been a subject of dispute among different authors. The first anchor described by (Figueroa et al., 1999) in cleft patients with severe maxillary hypoplasia was of the dental type. It showed certain drawbacks compared to skeletal type anchoring. They included handling the vertical vector, which in many cases causes an open anterior bite and discomfort at the level of the upper lip. On the other hand, the dental anchor can be difficult to use when there are lost teeth or in temporary teeth. At the same time, the rigidity of the intraoral appliance and its connection to the external device is basic, and usually it gets deformed reducing the magnitude of the traction and it increases the distraction time to achieve the desired position as planned (Kobayashi et al., 2012).

In our group of patients, skeletal anchoring was used. This was achieved with screws and wire ties, allowing better distribution of the distraction forces, thus improving the handling of vectors. It also offers greater comfort for the patient and facilitates their removal (Fariña and Salinas, 2017).

In relation to the morbidity of the approaches that we propose, we observed a reduction of the surgical time and the blood loss compared with other published reports in the literature. Meling et al. reported an average surgical time of 5.88 h and 1251.4 ml of blood loss (Meling et al., 2006). Hollier and Kelly reported an average surgical time of 3.5 h and 266 ml of blood loss (Hollier and Kelly, 2002). In our series of patients, we had an average surgical time of 2.78 h and 240 ml of blood loss. None of these patients needed mechanical ventilation after surgery.

Within the methodological limitations of the present study, we must mention that this group is small and very inhomogeneous; therefore future studies should be done with a larger number of patients. On the other hand, this study only measures dental and skeletal parameters in point A for the quantification of the advance of the middle third. Other skeletal parameters should be considered for future studies.

5. Conclusions

This technique shows a surgical approach with low morbidity, short surgery time and low blood loss. It allows optimal resolution of severe hypoplasia of the middle third of the face with long-term stability. It avoids the use of grafts and osteosynthesis material. By not including the nasal pyramid in the osteotomy design, the size, position, and nasofrontal angle in patients with adequate facial balance is maintained. If nasal correction is necessary, a secondary surgery may be done.

In cases of asymmetrical hypoplasia of the middle third, this osteotomy shows great versatility and can be done unilaterally and/or simultaneously combined with other distractions.

Funding

Dr. Fariña has nothing to disclose. No funding was received for this article.

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