Cleft Lip and Palate Midfacial Hypoplasia: Criteria to Choose the Treatment

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Abstract: A series of skeletal and dentoalveolar/occlusal criteria were proposed for choosing the treatment modality for the management of midface hypoplasia in cleft lip/palate patients, focusing on functional improvement, aesthetics, and minimizing the risk of recurrence and secondary alterations. For which, 42 patients with nonsyndromic cleft lip/palate, all with previous primary lip/palate surgeries and without previous osteotomies, were analyzed. Orthognathic surgery (OS) (n = 24) and maxillary distraction osteogenesis (n = 18) with anterior segmental osteotomies (segmental distraction osteogenesis [SD]), alveolar transport disc (TD), and midface total distraction osteogenesis (TDO) by modified Le Fort III osteotomy was done.

The average of maxillary advancement for OS was 5.58 ± 0.83 mm, for SD 9.4 ± 0.89 mm, for TD 8.00 ± 1.00 mm, and for TDO was 8.13 ± 1.55 mm.

In the presence of infraorbital and/or zygomatic hypoplasia, TDO was performed using skeletal anchorage, with the requirement of occlusal stability in dental cast in occlusion. In short maxillary arch without dental cast feasibility in occlusion, hypodontia/agenesis or absence of premaxilla, TD and SD was performed. There was only 1 mm of recurrence in 1 patient of each group. Changes in speech were detected in 2 patients in the OS group (8.3%). Orthognathic surgery can be indicated for advancements ≤7 mm not requiring orbito-zygomatic advancement, whereas distraction osteogenesis can be indicated for advances >8 mm with or without the need for orbito-zygomatic advancement, in addition with other dentoalveolar factors and velopharyngeal function.

Key Words: Cleft lip and palate, dentofacial abnormalities, dentofacial deformity, distraction osteogenesis, maxillary hypoplasia, maxillary osteotomy, midfacial advancement, midfacial hypoplasia, orthognathic surgery

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In cleft lip and palate (CLP) patients, the most frequent alteration in facial growth and development, is hypoplasia of the midface, meaning that a large percentage of patients require orthodontic and surgical procedures to correct it.[1] Commonly used procedures include orthognathic surgery (OS) and distraction osteogenesis (DO), which lengthens the bone and stimulates the formation of bone and soft tissue through osteotomies with gradual traction.[2] Abnormal facial morphology has mainly been attributed to 2 factors: deficiency in the intrinsic development of bone tissue and the main one, iatrogenic factors caused by surgical treatments in early stages.[3]

The purpose of this article is to show a case series with CLP and midface hypoplasia with different treatment modalities according to the clinical, occlusal, and dentoalveolar characteristics of each patient. The treatment protocols were OS and 3 kinds of osteogenic distraction: total distraction osteogenesis (TDO) via a modified Le Fort III, DO by transport disc (TD) and segmental distraction osteogenesis (SD).

MATERIALS AND METHODS

This study corresponds to a retrospective case series of 42 patients with CLP, all of them over 17 years old, who had midface hypoplasia. All of them were treated at the service of oral and maxillofacial surgery, Hospital del Salvador, Hospital San Borja Arriarán, and in the private practice of R.F, Santiago, Chile, between 2010 and 2018. This study was approved by the Ethics Committee of Hospital del Salvador.

The selection criteria for the sample were adult patients with primary lip and palate surgeries already done. All patients should not have corrective osteotomies of the jaws previously performed. The presence or absence of alveolar bone graft (ABG) and presence or absence of previous gingival periostioplasty was not a factor to consider to be included in the study, though it was considered to choose the treatment.

All patients underwent a preoperative CBCT imaging study and a dental cast study. The images were taken using a Kodak 9500 CBCT scanner (Carestream Health, Rochester, NY,), with settings of 90 kV, 10 mA, 0.2-mm^3 voxel size, and field of view of 20 × 18 cm. All these patients gave their detailed informed consent to participate in the study and the use of clinical and radiographic records.

The patients were divided into groups according to their individual characteristics, skeletal, dentoalveolar and occlusal

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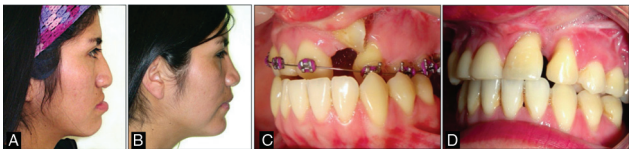

FIGURE 1. Orthognathic surgery with conventional Le Fort I maxillary advancement. (A) Profile before surgery; (B) profile after surgery; (C) preoperative intraoral view; (D) intraoral view after surgery (Patient number 9 of Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993).

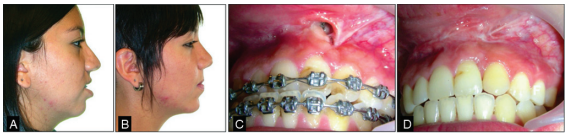

FIGURE 2. Orthognathic surgery with segmental Le Fort I osteotomy advancement with simultaneous bone grafting and gingival periostioplasty. (A) Profile before surgery; (B) profile after surgery; (C) preoperative intraoral view (with alveolar cleft); (D) intraoral view after surgery (Patient number 24 of Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993).

conditions, the treatment criteria were focused on functional and aesthetic improvement, and minimizing the risk of recurrence and secondary alterations.

To define skeletal characteristics and the severity of the maxillary hypoplasia, diagnostic cephalometric measures were used, such as Delaire's anterior abutment and McNamara's distance from A to the Nasion line. The maxillary advancement requirement was defined together with clinical and cephalometric analysis.

Dentoalveolar and occlusal characteristics analyzed were surgical feasibility in dental casts, the need of sagittal expansion of the maxillary arch, or the need of transversal expansion of the maxillary arch through the alveolar cleft.

Depending on the dentoalveolar/occlusal and skeletal characteristics mentioned above and other considerations as presence or not of ABG, width of alveolar gap, velopharyngeal function and the presence of pharyngeal flap, the patients were divided into 2 groups for treatment, the first one OS and the second group by DO.

The interventions indicated in the first group were OS using conventional Le Fort I osteotomy (Figs. 1 and 2) and DO in the second group by TDO by a modified Le Fort III[4] (Fig. 3), DO by TD (Fig. 4) and SD osteogenesis[5] (Fig. 5).

Orthognathic surgery was indicated for patients who needed 7 mm or less advancement at the Lefort I osteotomy, with adequate development of zygomas and infraorbital rims. Total distraction osteogenesis was indicated for patients with 8 mm or more of advancement and need of ZIH correction. According to dentoalveolar/occlusal criteria OS and TDO, patients had to be undergoing treatment of fixed orthodontics with surgical feasibility of dental casts. For TD and SD aligned arches were not necessary, in those groups of patients, the treatment was based on dentoalveolar

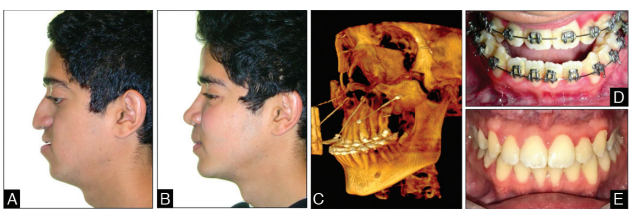

FIGURE 3. Modified Le Fort III osteotomy with total distraction osteogenesis with SARED technique. (A) Profile before surgery; (B) profile after surgery; (C) 3D imaging of Modified Le Fort III distraction osteogenesis; (D) intraoral view before surgery; (E) intraoral view after surgery. 3D, three-dimensional.

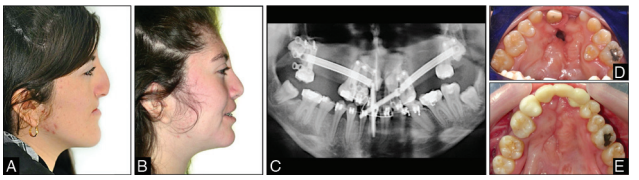

FIGURE 4. Transport disc osteotomy distraction osteogenesis. (A) Profile before surgery; (B) profile after surgery; (C) panoramic imaging of transport disc distraction osteogenesis with submerged distractors in the consolidation stage. (D) Preoperative occlusal view; (E) postoperative occlusal view with temporary restorations. Consider generation of premaxilla, the enlargement of the arch perimeter and the closure of the palatal cleft (Patient number 18 of Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994).

characteristics that do not allow adequate orthodontic treatment due to the small size of the maxilla, short maxillary arch, dental crowding, micro/hypodontia with the need of sagittal expansion of maxillary arch, wide unilateral or wide bilateral alveolar clefts, absence of premaxilla and pharyngeal flap with adequate velopharyngeal competence. Segmental distraction osteogenesis were indicated in cases with previous ABG, while TD in cases with very wide alveolar clefts.

Previous ABG was necessary for all TDO and SD patients. In OS patients, ABG could be there beforehand (Fig. 1), or done at the same time of Le Fort I osteotomy in cases where the maxilla can be manipulated as a 2-segment osteotomy (Fig. 2). In TD, ABG could not be necessary after distraction (Fig. 5), but if it was, it could be made after distraction (when the distractor devices were removed).

According to skeletal and dentoalveolar/occlusal criteria, 24 patients were treated with OS and 18 patients were treated with different techniques of DO (Fig. 6 and Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993 and Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994).

The other parameters assessed (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993 and Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994) were age, gender, diagnosis of the cleft, number of complementary surgeries required, the relapse rate after 1-year of surgery in point A, and speech changes after surgery using the Pittsburgh scale for the assessment of velopharyngeal incompetence (VPI), evaluated in all patients by the same operator.

Complementary surgeries for the correction of midface hypoplasia were rhinoplasty, genioplasty and bilateral sagittal split ramus osteotomy (BSSO). In all OS the complementary surgeries were done at the same time of the Le Fort I osteotomy. In TDO, TD, SD the complementary surgeries (rhinoplasty, genioplasty, and BSSO) were deferred to a second surgical time.

RESULTS

Of the 42 patients treated, 24 were treated with OS (as shown in Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993) and 18 with DO (as shown in Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994). All of the

FIGURE 5. Segmental osteotomy distraction osteogenesis with a hyrax screw device. (A) Profile before surgery; (B and C) preoperative intraoral view; (D) occlusal view of consolidation stage; (E and F) intraoral view of oral rehabilitation; (G) profile 3 years after surgery (Patient number 9 of Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994).

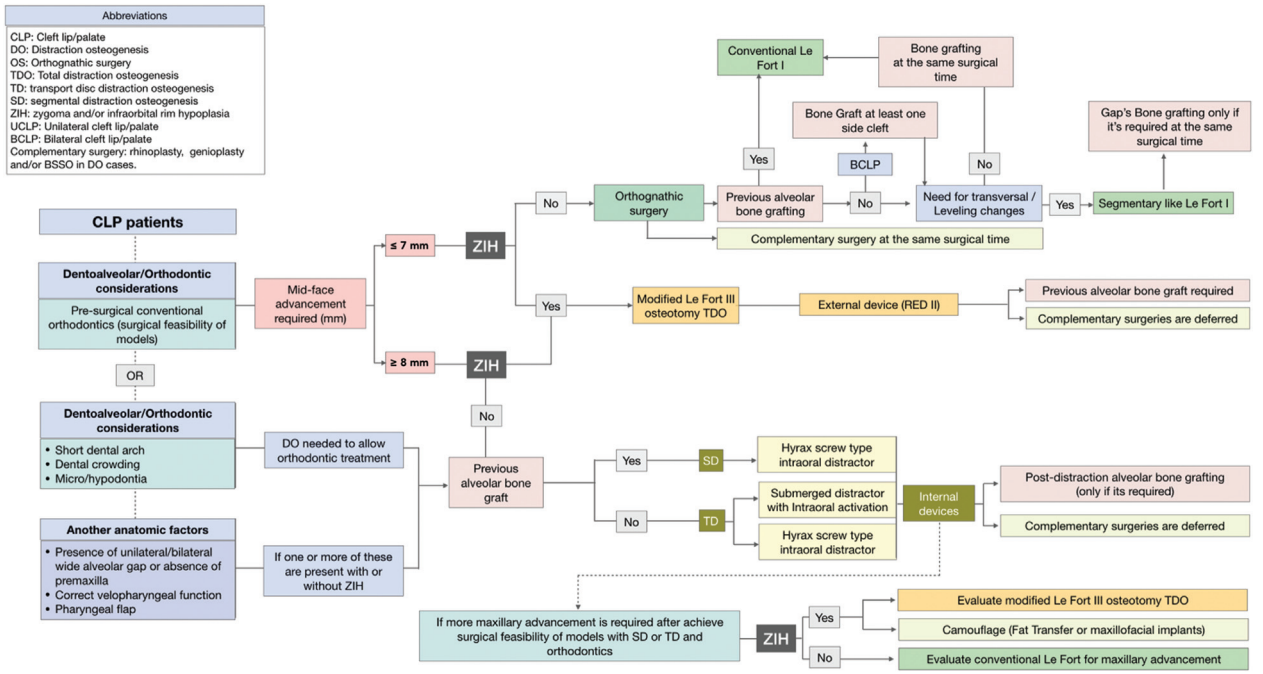

FIGURE 6. Skeletal and dentoalveolar criteria to evaluate the treatment of midface hypoplasia in cleft lip and palate patients.

surgeries were performed by the same surgeon (RF), with an average follow-up time of 3.16 ± 1.52 years. The patients' average age was 19.29 ± 1.95 years (minimum 17 years, maximum 27).

The distribution by gender was 18 men (42.8%) and 24 women (57.2%). A total of 54.78% of clefts were on the left side, 23.80% on the right side and 21.42% were bilateral.

The average maxillary advancement for TDO was 8.13 ± 1.55 mm, for SD 9.4 ± 0.89 mm, for TD 8.00 ± 1.00 mm, and for OS it was 5.58 ± 0.83 mm.

Regarding the ABG, 27 patients (64.28%) had a previous ABG before the maxillary hypoplasia treatment, 14 in the OS group and 13 in the DO group. In the OS group, the ABG was performed at the same surgical time of the Le Fort I ($n = 10$) on all patients without previous alveolar grafting (41.6%) (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993).

In the DO group, there was not ABG in 5 patients (all on the TD group), 2 of them had the ABG done in a delayed way in addition to genioplasty and rhinoplasty, at the same time of removing the distractors, while 3 patients did not require ABG, because the mobilization of the TD, closed the alveolar cleft. All the complementary surgeries were performed at the time of removing the distractors (Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994).

The complementary surgeries made were: only genioplasty: 14.3%; genioplasty + BSSO: 9.5%; genioplasty + BSSO + rhinoplasty: 26.2%; genioplasty and rhinoplasty: 50%. They were done at the same time of maxillary hypoplasia's correction in the case of OS (Supplementary Digital Content, Table 1, http://links.lww.com/SCS/C993) and were deferred in patients who underwent DO (Supplementary Digital Content, Table 2, http://links.lww.com/SCS/C994).

For the SD and TD techniques, submerged intraoral devices or Hyrax type were used. Both devices were used for unilateral as well as bilateral cases. Another aspect evaluated was the maxillary advancement required and the existence of hypoplasia on the level of infraorbital and/or zygomatic ridges (ZIH). The treatment for ≤ 7 mm of maxillary advancement without ZIH, was OS; on the other hand, in patients with hypoplasia of zygomas and infraorbital rims a TDO is recommended through a modified Le Fort III and a RED II device without needing bone grafts in the gap of the osteotomy site. Distraction osteogenesis was performed on severe cases of 8 mm or more, with the technique depending on the dentoalveolar and orthodontic considerations described above. The overall frequency of the type of osteotomy performed was 57% Le Fort I osteotomy, 19% TDO, 12% SD, and 12% TD.

In the present study we observed a 1 mm relapse out in 12 months follow-up, of a total advancement of 9 mm in only 1 patient 1 year after TDO, whereas in the OS group there was also a 1 mm relapse out of a total advancement of 6 mm, also in only 1 patient. No patient in the DO group showed changes in their speech after surgery, whereas 2 patients who had OS showed changes in their speech (as shown in Supplementary Digital Content, Tables 1, http://links.lww.com/SCS/C993 and 2).

The average follow-up period for patients with OS was 3.12 ± 1.48 years, whereas in the DO group was 3.22 ± 1.62 years.

DISCUSSION

Maxillary deficiency has been described as a sequel of surgical correction of the clefts,[6] in line with what has been described by Ortíz-Monasterio, Bishara, Mars, and Houston in previous studies.[7–9]

Several studies state that very early interventions, increase the incidence of maxillary hypoplasia.[3,10–12] In contrast, if the primary surgeries were made respecting growth and development agents (eruption of the deciduous teeth) and deferring the closure of the hard palate (6 months soft palate and 15 months hard palate), the effect on maxillary growth and development would be less harmful.[13–16] In our experience, primary surgeries that require large displacements in palatal closures and soft palate at the same time,

generate great scar flanges that restrict the growth and development of the maxilla.

The maxillary advancement of the patients in our study was 8.47 ± 1.32 mm for DO and 5.58 ± 0.83 mm for OS.

All patients ($n = 42$) had primary surgeries (cheilo-rhinoplasty and palatoplasty) and 3 patients in the SD group had a pharyngeal flap.

Regarding the correction of maxillary hypoplasia in CLP patients, 1 of the advantages of OS is the possibility of making transversal changes and to level the maxillary segments, with the cleft maxilla acting as a segmented Le Fort I. However, relapses are a common problem in this treatment modality, with a 5.5% to 20% relapse rate described in horizontal movements and 23% to 38% in vertical movements,[17] due to large amounts of scar tissue from previous surgeries and the need for big movements.[18]

Literature says that overcorrection during treatment planning can help to counteract this phenomenon, but according to our study's results, overcorrecting advancements are not necessary, given that stable occlusion and adequate overjet and overbite, associated with correct orthodontics, and intermaxillary elastics (class 3) are the main factors that decrease the relapse rate, which was 4.1% (1 out of 24 patients with 1 mm in the sagittal direction) in our series of OS cases. If any sign of relapse is observed shortly after surgery, it must be handled with traction devices and/or class III elastics to help counteract the maxillary retrusion movement.

As a Le Fort I osteotomy can only correct the occlusal discrepancy, without improving the projection of the zygomatic and/or infraorbital areas, in CLP patients with a severe deficiency in the midface,[19] Fariña et al[4] described DO by TDO in modified Le Fort III, is an ideal alternative to achieve maxillary advancement in these patients. DO is a better alternative to conventional OS,[20] because the newformed bone in the gap would allow better stability with fewer relapses, and sometimes with TD it does not need bone grafting in the gap.[21]

Since McCarthy et al[22] published the first work on DO in craniofacial deformities in 1992, numerous articles have been published on midface distraction using different types of devices.

Polley and Figueroa[23,24] used a rigid external distraction device (RED) to lengthen the entire maxilla for the treatment of severe maxillary deficiencies in growing cleft patients. The use of the RED I device has become an excellent treatment strategy, as it allows precise and controlled distraction of the maxillary osteogenesis.[25–27] RED II was later introduced to improve the control vector by means of an additional anchor to the zygomatic bone. All of these features have made the use of RED I and II an excellent alternative treatment for patients with severe maxillary hypoplasia. Fariña and Salinas[28] modified the anchorage technique, describing the "Skeletal Anchorage for a Rigid External Device", to apply force directly to the bone with a given vector, without intraoral splint nor other devices. This technique was used for the TDO with a modified Le Fort III osteotomy, where the osteotomy was performed using low morbidity approaches (transconjunctival and transoral approaches), a shorter surgery time and low blood loss, in contrast to conventional Le Fort III techniques. This technique avoids the use of grafts and osteosynthesis material.[4] On the other hand, by not including the nasal pyramid, the size, position and nasofrontal angle are maintained, with the possibility of a deferred rhinoplasty in the event that an adequate facial balance is not achieved. This technique was used on all the TDO patients in our study ($n = 8$).

Another DO method with intraoral devices, is a TD, that can be used to close wide clefts (uni- or bilateral) or even in the absence of premaxilla. Liou et al,[29] proposed interdental DO to create a segment of new alveolar bone and attached gingiva for the complete approximation of a wide alveolar cleft/fistula and the reconstruction of a maxillary dentoalveolar defect with tooth-borne device. Years later the same authors conducted a study in 21 patients using this technique with bone-borne internal distractor, approximating alveolar cleft and minimizing the risk of failure of the ABG afterwards.[30]

Segmental distraction osteogenesis technique using a Hyrax device allows traction along an anterior vector of a maxillary segment, allowing advancement as a block without compromising the relationship with the molars or the soft palate and hence without risk of VPI. Dolanmaz et al[31] reported the first case of segmental osteotomy in a noncleft patient with maxillary hypoplasia, using a Hyrax type palatine disjunction device with dental anchorage, but positioned in the anteroposterior direction. In 2004, Karakasis and Hadjipetrou,[32] performed the procedure on a cleft patient and observed that neither the posterior region nor the velopharyngeal function were affected in the maxillary advance, allowing it to be used in growing patients. Fariña et al[5] described SD in 4 CLP patients using a modified Hyrax device, increasing the size of the maxillary arch and advancing the anterior sector without altering speech or the molar relationship.

It is suggested that dental implants be installed in the neoformed bone when using the SD and TD techniques, or else locate teeth with significant crowding to prevent relapses. On the other hand, the open bite created with dental devices is resolved with temporary anchoring devices and intermaxillary elastics during the consolidation phase.[5] Other studies using this technique describe a 65% reduction in VPI post treatment, as a movement towards the back of the molars was detected and postdistraction speech even improved.[33–35]

Regarding postsurgery stability, there are authors who argue that OS as a treatment could be less stable when compared to DO,[36] describing a maxilla that suffers a severe relapse on the vertical and sagittal planes. A systematic review concluded that a maxillary advancement by means of DO is more stable in cleft patients with moderate and severe maxillary hypoplasia when compared to OS.[37] However, a recent review by Cochrane indicates that there are no conclusive studies with regard to which type of procedure to use. Regarding the changes in soft and hard tissues, 39 patients with maxillary advancements of between 4 and 10 mm were evaluated, revealing that there were no statistically significant differences between patients who received DO and OS after 2 years of follow-up.[38]

Andersen et al[39] performed a cephalometric analysis of maxillary advancements in 11 adult cleft patients following OS and DO procedures, with advancements of >8 mm. Their results showed a significant long-term difference in changes on vertical position of point A between the 2 groups, with a higher OS relapse rate. The horizontal and vertical relapses measured were 0.8 and 3.1 mm, respectively, which corresponds to a relapse rate of 10% and 61%. This contrasts with a previous study, in which they described an 8% horizontal relapse and a 19% vertical 1 after DO.[40] The varied results of these studies contrast with our study, in which we describe skeletal and occlusal criteria to decide each surgical technique to reduce relapses in both CO as well as DO, achieving a relapse rate of just 4.76% in 42 patients: a 4.1% ($n = 1$) in OS and 5% ($n = 1$) in DO, whose cephalometric change was just 1 mm of maxillary retrusion on the sagittal plane in each group in a period of 3.16 ± 1.52 years, without the need to overcorrect with CO or DO. As a protocol, all of the patients in our study required pre- and post-surgical orthodontics. The OS and TDO techniques required stability of models in a proper position of the dental casts. The only surgical procedures performed at the start of orthodontic treatment were SD and TD to allow the rearrangement of teeth. These 2 techniques were used in cases where there was a risk of postsurgical VPI, and in cases with a pharyngeal flap ($n = 3$) or with short maxillary arches. In patients with short dental arches, dental

crowding and micro-or hypodontia, these techniques allow to recover spaces lost due to agenesis and altered tooth shapes and sizes and relocating teeth in bad positions. These are also the indicated techniques for cases with a unilateral cleft or very wide bilateral cleft or patients lacking premaxilla. No patient subjected to DO required maxillary surgery later, only BSSO to bring the mandible to correct position.

There are authors who prefer modified OS instead of DO, where they perform stepped Le Fort I osteotomies with interpositional bone grafts in gaps and also installing onlay grafts to contour cheeks and maxilla for camouflage, however, surgical time and morbidity increase in these techniques.[41] Being the different DO techniques described a valuable alternative.

Regarding the presence of ABG at the time of consultation, it is known that the ideal time for this to be carried out is around 10 to 12 years of age, before the eruption of the permanent maxillary canine in the area of the cleft. Thus, allowing greater maxillary growth and adequate orthodontics before surgery.[42] However, if at the time of consultation, the patient has fixed orthodontics and an open cleft, it is preferable to perform gingival periostioplasty and ABG simultaneously with OS.

Finally, another one of the parameters that many surgeons evaluate when choosing between 1 technique or another has to do with VPI. Studies show that there are no statistically significant differences in the velopharyngeal function or hypernasality of either procedure (OS or DO) 17 months postintervention.[43,44] These results are consistent with the findings published in the Cochrane Group's review.[38] In our series, 2 patients (8.3%) who received OS for a 6 mm advancement registered postsurgery speech changes (from 2 to 3 and from 3 to 5 in Pittsburg's scale), whereas none of the patients in the DO group showed any changes.

The proposed treatment, based on the individual characteristics and requirements of the cleft patient, focusing on functional and aesthetic improvements, minimize the risk of relapse and secondary postoperative changes.

The limitations of the present study are the heterogenicity of the parameters related with the diagnosis, of the clinical characteristics, and the different treatment modalities. However, the objective of the present study is to take into account different common clinical characteristics seen in this type of patient, to guide the individual treatment decision, to get better functional and aesthetic results, with less morbidity and fewer surgical interventions.

CONCLUSIONS

For maxillary advances of 8 mm or more, DO is recommended, varying the technique depending on the case. Conventional OS can be reserved for advances $\leq$7 mm without the need for an orbito-zygomatic projection, in addition to considering other factors, of which the most important is occlusal stability. When the maxillary arch is short and with alterations that do not allow surgical feasibility, it is solved with SD or TD distraction, depending on the presence of previous ABG, the diameter of the cleft, velopharyngeal competence, among other characteristics.

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