Comparing Surgical Advancement Outcomes of Retruded Maxilla in a Group of Egyptian Cleft Lip and Palate Subjects

Aida M. Mossaad 1,*, Mostapha A. Abdelrahman 1, Suzan A. Hassan 2, Hatem H. Al Ahmady 2, Nahed M. Adly 2, Wael A. Ghanem 3, Shadia A. Elsayed 2

1 Department of Orofacial Genetics, National Research Centre, Cairo, Egypt; 2 Department of Oral and Maxillofacial Surgery, Faculty of Dental Medicine for Girls, Al Azhar University, Cairo, Egypt; 3 Department of Plastic Pediatrics, Ain Shams University, Cairo, Egypt

Abstract

BACKGROUND: Cleft lip and palate (CLP) is one of the most common congenital deformities involving intervention in several sub-specialties.

AIM: The present study was conducted to investigate the amount of maxillary advancement obtained by three different methods.

METHODS: A retrospective comparative study was conducted on 24 CLP patients who were treated with three surgical maxillary advancement techniques: Group A was treated with Le Fort I (LFI) orthognathic surgery with bone grafting and rigid fixation (LFI). Group B was treated with intraoral maxillary bone distraction (MIDO). Group C was treated with orthodontic traction by facemask (orthodontic facemasks [OFM]) plus corticotomy. All pre-operative data were collected, which included intraoral and extraoral clinical photos and dental casts. Pre-operative radiographic assessment was compared with post-operative values using digital panorama, multi-slice computed tomography and lateral cephalometric X-ray measuring Sella-nasion-A point; point A-nasion-point B points, with a follow-up period of 6 months.

RESULTS: All approaches showed statistically significant success in maxillary advancement with p < 0.01. LFI has produced the highest advancement obtained with regard to the pre-operative advancement required (8.6 ± 1.4) and post-operative advancement achieved (7.8 ± 0.8). MIDO technique is an alternative method to LFI, but it gives less achieved post-operative maxillary advancement (6.25 ± 0.8) and is indicated for moderate cases. OFM gave the least advancement results; however, it has been the most convenient less-invasive method and was more suitable for unsevere cases.

CONCLUSIONS: The three approaches produced satisfactory results in rehabilitating deficient maxilla in cleft patients, although each technique has limitations and indications. Future research is recommended to assess the technique's long-term stability.

Introduction

One of the most common human congenital deformities is the cleft lip/or palate (CLP) with 1:700 occurrence of newborns [1]. Chewing, speaking, and breathing are severely impaired, and some psychological disorders as well as malnutrition are also aggravated by the problem. Clefts are either isolated or associated with syndromes such as Cleido-Cranial dysplasia, Crouzon syndrome, and Apert syndrome [2], [3], [4].

Patients with significant cleft maxillary deficiency are difficult to treat with traditional surgical approaches. During early childhood, primary CLP repair improves facial appearance, speech, and mastication, but affects maxillary development in comparison to mandibular growth [5]. Furthermore, scarring resulting afterward causes a series of secondary deformities, such as decreased maxillary growth in horizontal and vertical dimensions resulting in maxillary hypoplasia and limitation in arch expansion, as the patient grows up [6], [7].

Problems associated with maxillary retrusion are functional and esthetic as facial disfigurement, occlusal problems, temporomandibular joint pain, and psychological problems [8], [9]. The maxilla should be advanced to a position that catches up to the mandibular growth. Le Fort I (LFI) osteotomy followed by maxillary advancement and rigid fixation procedure is a classical technique commonly used to restore morphological and functional balance to achieve occlusal stability. It allows advancement of maxilla to the desired position up to 1 cm or more either in cleft patients or severe Class III occlusion [10], [11]. However, this technique is liable to post-operative relapse and might affect the velopharyngeal incompetence velopharyngeal insufficiency causing further hypernasality and phonation problems [12], [13].

Distraction osteogenesis is an alternative technique rather than classical orthognathic which achieves bone lengthening by gradual mechanical traction as defined by Ilizarov [14], [15]. Optimum preservation of extrasosseous and medullary blood supply, stable fixation, delay in distraction (latency),
distraction (rate) of 1 mm/day in regular small increments (rhythm), and consolidation time is factors that are critical for the formation of new bones, up to 4 months before device removal. Intraoral distractors provide gradual advancement of maxilla with simultaneous bone formation without the need for bone grafting and rigid fixation. Another advantage also is a less rate of relapse postoperatively; however, there is a limitation in the amount of maxillary advancement to <1 cm depending on the intraoral device [16], [17].

Lately, the use of orthodontic facemasks (OFM) has shown promising results as regard pulling the retruded maxilla outward either in Class III occlusion or cleft cases. The optimum results can occur in patients till the age of 16 years [18], [19]. The use of OFM following simple maxillary corticotomy helps in maxillary advancement without the need for complete maxillary separation or the use of bone grafting and internal fixation devices. However, it affects the quality of life of patients being worn all day and night. However, it is the most noninvasive method for maxillary advancement resulting in a better facial appearance [20], [21], [22]. The role of radiology is important in pre-operative and post-operative evaluation especially the lateral cephalometric X-ray anatomical points Sella-nasion-A point (SNA) and SNB which gives a good indication as regard the maxillary relation to the mandible and amount of advancement needed and achieved.

In our institute, we are practicing three different approaches to treat cleft patients with hypoplastic maxilla deficiency, so the present study aimed to compare between the surgical outcomes and the amount of maxillary advancement obtained after using LFI orthognathic surgery with bone grafting and rigid fixation (LFI) and maxillary intraoral bone distraction (MIDO) and facemask orthodontic traction (OFM).

Materials and Methods

Study design and population

This study included 24 cases age ranging from 12 to 18 years old males and females having non-syndromic complete bilateral CLP (BCLP) and suffering from retruded maxilla who attended the surgical clinic of the national center of research in the past 2 years.

In the present study, alveolar cleft grafting was performed in the majority of cases preoperatively, either with stem cell extract technique or traditional fresh autogenous cancellous graft, to restore the anterior maxilla into one piece, avoiding separation or fracture at the weak cleft area intraoperatively, as well as to aid the canine to erupt in its proper position in bone.

All of the patients had a Class III malocclusion with a seemingly normal mandible and an anterior cross bite, and none of them had previously had maxillary advancement surgery. To avoid the mixed dentition phase and witness the canine eruption, we chose to treat patients above the age of 11. Patients were allocated into three different groups according to the technique used for maxillary advancement.

Group A: Included eight patients who were treated by the orthognathic surgery using LFI osteotomy and rigid fixation. Group B: Included eight patients who were treated by maxillary distraction osteogenesis using intraoral bone distractor (MIDO). Group C: Included eight patients who were treated with orthodontic facemask traction extraorally (OFM) with intraoral miniscrews anchorage fixation.

All patients have been informed of all the aspects of the study and have signed an informed consent. The study was performed in compliance with the Helsinki Declaration of Clinical Research Study Guidelines and accepted by the Ethical Committee of the National Research Centre, Cairo, Egypt (# 12060204).

Surgical procedures

Surgical approaches of all were made under general anesthesia and complete aseptic conditions. Medical history was documented. Before each procedure, blood studies were performed for each patient, including hemoglobin level, bleeding profile, kidney functions (Urea and Creatinine), liver functions (SGOT and SGPT), blood glucose level, electrocardiogram, and chest examination.

Study groups and techniques

Group A

Classical LFI osteotomy was used with down fracture of maxilla and separation from pterygoid plates and nasal spine. Then mobilization of maxilla was performed and fixation with rigid miniplates (AO/ASIF, Maxillofacial, PA, USA) to the desired position in relation to the mandible after adjusting occlusion.

Group B

Classic Le Fort Osteotomy was used with elevation of full mucoperiosteal flap and placing intraoral bone distractor (Unidirectional Intra-Oral Distractor Liou by KLS Martin Germany) and fixation with mini screws to obtain callus formation at the osteotomy site. Distraction protocol after 7 days of latency period, starting the device activation with a rate of 1 mm/day and rhythm twice a day.
Group C

Patients treated through placing intraoral miniscrews plus corticotomy in the maxilla with placement of extraoral orthodontic facemask attached with elastics for daily traction of maxilla. All patients were administered effective antibiotics, analgesics, and anti-inflammatory medications with oral hygiene guidelines and a soft food regimen after the surgery (Figure 1).

This investigation. Radiographic evaluation included multislice computed tomography and digital panorama. Pre- and post-operative lateral cephalometry tracing measures for anatomical landmarks of the SNA and SNB, as well as the AP plane, were obtained and used to examine the differences between groups (Figure 3).

Methods of evaluation

Intraoral and extraoral photographs were used to assess each patient’s clinical condition (Figure 2), although soft-tissue measures were not taken in this investigation. Statistical analysis

All data were subjected into statistical analysis using descriptive and paired tests for pre- and post-operative values. Calculations were used using SPSS software program version 20.0 to compare measurements of mean and SD of pre and postintervention between groups. Comparisons between pre and postmeasures in each group were done using paired t-test and comparison between the two studied groups; pre and postintervention mean difference was done using independent t-test. Difference of p < 0.05 was considered significant.

Results

This study included 24 BCLP cases that suffered from maxillary retraction and required maxillary advancement. The patients were divided into three groups. Group (A) showed the highest statistically significant results with LFI osteotomy with regard to the pre-operative advancement required (8.6 ± 1.4) and post-operative advancement achieved (7.8 ± 0.8) with p = 0.01*. Mean/SD measurements of pre-operative SNA were 7.6 ± 3.7, while post-operative SNA was 83.5 ± 3.9 with p < 0.001* and pre-operative point A-nasion-point B points (ANB) was −2.1 ± 0.8, while post-operative ANB was 5.38 ± 2.1 with p < 0.001*(Figure 4).

Group B with intraoral distractor showed also statistically significant results in terms of pre-operative advancement needed (7.88 ± 1.1) and post-operative advancement achieved (6.25 ± 0.8) with p = 0.001*. Pre-operative SNA mean/SD was 74.5 ± 2.3, while post-operative SNA mean/SD was 79.75 ± 2.8 with p = 0.001* and pre-operative ANB mean/SD was
Discussion

This study included 24 patients who were divided into three groups where Group A treated with LFI orthognathic surgery that showed superior results in maxillary advancement and could be used for severe maxillary retrusion. However, Baker et al. [23] reported traditional LFI osteotomy as a technique for maxillary advancement, but noted that it had limitations such as high blood loss, delayed union, inadequate stabilization, and potential relapse.

Patients with maxillary hypoplasia caused by BCLP provided a greater challenge than normal cases in terms of lip scarring and palatal fibrosis, restricted blood supply, tooth crowding, and insufficient bone for putting distractors. Early multiple surgical interventions during childhood in the cleft patients cause scarring and limitations during adulthood orthognathic surgeries and sometimes resulting in relapse according to the Rachmiel [4]

Intraoral distraction osteogenesis (MIDO) is a successful alternative strategy for maxillary advancement in CLP patients requiring advancement <10 mm. It causes improvement of skeletal and soft-tissue profile. It can be used in moderate cases as in Group B with less tendency of relapse and without need for bone grafting. However, this technique needs a secondary surgical removal of the device[11], [24], [25]. In this investigation, we used bone-borne devices that anchored to the zygomatic bone, similar to Van Stickle et al. [26], who used a bone-borne distractor for anterior hypoplastic maxillary advancement in cleft patients to correct Class III malocclusion.

In 2004, Karakasis et al. [27] made the first attempt to use the bone-borne distractor in cleft patients with anterior maxillary segmental distraction for advancement of hypoplastic maxilla, who faced more challenges than normal patients due to soft-tissue tightness from the previous scars. Wang et al. [28] were also the first to use a tooth-borne device with anterior maxillary segmental distraction osteogenesis in cleft patients who depend on dental anchorage for hypoplastic maxilla advancement in 2009. Gateno et al., 2005 [29] also noticed in his studies, a clockwise rotation of the maxilla vertically correcting the existing open bite and also showed similar promising results using intraoral distractors in CLP patients. The Liou intraoral device was accepted by patients, generating neither bulkiness nor extraoral scars; however, it required pre-operative surgical planning with the use of a computer aided surgical model for prebending and customization of the device plates to minimize operative time.

De Clerck et al., 2009 [30], reported that facemask therapy is effective in maxillary advancement in Class III occlusion cases especially in late mixed or permanent dentition; however, it had a negative impact in roots proclination. In the present study, the use of miniscrews fixation with elastics helped in more stabilization of the maxillary segment and achieved the required advancement as shown in Group C [18], [31]. Some patients were rejecting the external appearance impeding their quality of life especially females who had

Table 1: Comparison among studied groups according to pre and post-operative measures

|                | pre       | post      | p value |
|----------------|-----------|-----------|---------|
| **Group A**    |           |           |         |
| ADV            | 8.63 ± 1.4| 7.81 ± 0.8| 0.01*   |
| SNA            | 76.0 ± 3.7| 83.5 ± 3.9| <0.001* |
| ANB            | -2.13 ± 0.8| 5.38 ± 2.1| <0.001* |
| **Group B**    |           |           |         |
| ADV            | 7.88 ± 1.1| 6.25 ± 0.8| <0.001* |
| SNA            | 74.5 ± 2.3| 79.75 ± 2.8| 0.001* |
| ANB            | -2.38 ± 1.1| 3.25 ± 2.4| 0.001* |
| **Group C**    |           |           |         |
| ADV            | 7.25 ± 1.0| 5.5 ± 0.7 | <0.001* |
| SNA            | 74.25 ± 2.1| 78.13 ± 2.6| <0.001* |
| ANB            | -2.75 ± 0.9| 2.13 ± 0.6| <0.001* |

*Statistically significant difference (p<0.05)

All approaches showed statistically significant success in maxillary advancement with p = 0.01. Group (A) with classic LFI osteotomy showed the highest results followed by group (B) with intraoral distraction and finally Group (C) with orthodontic facemask traction. All cases were satisfied with the results and no complications occurred (Table 2).

Table 2: Comparison among studied groups according to post and pre-operative measurements difference

|                | Group A | Group B | Group C | p value | p@ | p^ | p* |
|----------------|---------|---------|---------|---------|----|----|----|
| ADV            | -0.81 ± 0.7| -1.63 ± 0.7| -1.75 ± 0.6| 0.018*| 0.021*| 0.006*| 0.03*|
| SNA            | 7.5 ± 2.6| 5.25 ± 2.9| 3.88 ± 1.5| 0.02*| 0.072| 0.009*| 0.111|
| ANB            | 7.5 ± 2.6| 5.63 ± 2.7| 4.88 ± 1.1| 0.079| 0.26| 0.704| 0.513|

*Statistically significant difference (p<0.05)
to take it off for sometimes of the treatment, meanwhile, it was the most convenient non-invasive method; however, it showed the least achieved results in maxillary advancement in comparison to the other two groups with such limitations; it becomes more useful in unsevere cases.

For each technique used to correct maxillary hypoplasia, there were benefits and limitations. The maxillofacial cleft team should understand the best surgical outcomes that can be achieved from each approach and determine the chosen intervention on the basis of each individual condition of the cleft patient.

Conclusions

LFI gave superior results and is indicated for severe cases, MIDO technique with bone distraction is an alternative method to Group A with less liability to relapse, also the intraoral distractor gives less amount of maxillary advancement and its indicated for moderate cases. OFM gave the least advancement results; however, it has been the most convenient less invasive method despite being rejected with most of the cases because of its external appearance which interfere with their daily activities especially females and its limitations makes it more suitable for unsevere cases.

Recommendation

Future long-term research is needed to assess the differences in relapse and stability between various procedures for advancing the maxilla in cleft patients.

References

1. Altaweel AA, Lababidi AS, Abd-Ellatif EL-Patal M, Elsayed SA, Eldin MS, Dabbas J, et al. Outcomes of bifocal transport distraction osteogenesis for repairing complicated unilateral alveolar cleft. J Craniofac Surg. 2021;https://doi.org/10.1097/SCS.0000000000008260. PMID:34608012
2. Wakae H, Hanaoka K, Morishita T, Nakasima A. A clinical report on distraction osteogenesis applied for Apert syndrome. Orthod Waves. 2008;67:30-7. https://doi.org/10.1016/j.odw.2007.10.004
3. Kyprianou C, Chatzigianni A, Crouzon syndrome: A comprehensive review. Balk J Dent Med. 2018;22:1-6. https://doi.org/10.2478/bjdm-2018-0001
4. Rachmiao A, Aizenbud D, Peled M. Long-term results in maxillary deficiency using intraoral devices. Int J Oral Maxillofac Surg. 2005;34(5):473-9. https://doi.org/10.1016/j.ijom.2005.01.004. PMID:16053864
5. Mossaad AM, Ahmady HH Al, Ghanem WH, Abdelrahman MA, Abdelazim AF, Elsayed SA. The use of dual energy X-ray bone density scan in assessment of alveolar cleft grafting using bone marrow stem cells concentrate/platelet-rich fibrin regenerative technique. J Craniofac Surg. 2021;32(8):e760-3. https://doi.org/10.1097/SCS.0000000000007772. PMID:34727454
6. Pai BC, Hung YT, Wang RS, Lo LJ. Outcome of patients with complete unilateral cleft lip and palate: 20-year follow-up of a treatment protocol. Plast Reconstr Surg. 2019;143(2):359e-67. https://doi.org/10.1097/PRS.0000000000005216. PMID:30531628
7. Wu TJ, Lee YH, Chang YJ, Lin SS, Lin FC, Kim Y, et al. Three-dimensional outcome assessments of cleft lip and palate patients undergoing maxillary advancement. Plast Reconstr Surg. 201;143(6):1255e-65. https://doi.org/10.1097/PRS.0000000000005646. PMID:31136492
8. Wang DZ, Chen G, Liao YM, Liu SG, Gao ZW, Hu J, et al. A new approach to repairing cleft palate and acquired palatal defects with distraction osteogenesis. Int J Oral Maxillofac Surg 2006;35(8):718-26. https://doi.org/10.1016/j.ijom.2006.03.010. PMID:16690205
9. Ascherman JA, Marin VP, Rogers L, Prisant N. Palatal distraction in a canine cleft palate model. Plast Reconstr Surg. 2000;105(5):1687-94. https://doi.org/10.1097/00006534-200004050-00014. PMID:10809099
10. de Mol van Otterloo JJ, Tuinzing DB, Kostense P. Inferior positioning of the maxilla by a Le Fort I osteotomy: A review of 25 patients with vertical maxillary deficiency. J Cranio maxillofac Surg. 1996;24(2):69-77. https://doi.org/10.1016/0101-5182(96)80015-1. PMID:8773886
11. Marion F, Mercier JM, Ondri GA, Perrin JP, Longis J, Kün Darbois JD, et al. Associated relaps factors in Le Fort I osteotomy. A retrospective study of 54 cases. J Stomatol oral Maxillofac Surg. 2019;120(5):419-27. https://doi.org/10.1016/j.jormas.2018.11.020. PMID:30648606
12. Alaluusua S, Turunen L, Sairikko A, Geneid A, Leikola J, Heliovaara A. The effects of Le Fort I osteotomy on velopharyngeal function in cleft patients. J Cranio maxillofac Surg. 2019;47(2):239-44. https://doi.org/10.1016/j. joms.2018.11.016. PMID:30581082
13. Kloukos D, Fudalej P, Sequeira-Byron P, Karitos C. Maxillary distraction osteogenesis versus orthognathic surgery for cleft lip and palate patients. Cochrane Database Syst Rev. 2016;9(9):CD010403. https://doi.org/10.1002/14651858.CD010403.pub2. PMID:27689965
14. Scolozzi P. Distraction osteogenesis in the management of severe maxillary hypoplasia in cleft lip and palate patients. J Craniofac Surg. 2008;19(5):1199-214. https://doi.org/10.1097/SCS.0b013e318184365d. PMID:18812842
15. Van Sickels JE. Distraction osteogenesis: advances in the last 10 years. Oral Maxillofac Surg Clin North Am. 2007;19(4):565-74, vii. https://doi.org/10.1016/j.coms.2007.06.004. PMID:18088906
16. Spiegelberg B, Parratt T, Dheerendra SK, Khan WS, Jennings R,
Mossaad et al. Surgical maxillary advancement outcomes in cleft patients

Open Access Maced J Med Sci. 2022 Feb 18; 10(D):64-69.

17. Samchukov ML, Cope JB, Harper RP, Ross JD. Biomechanical considerations of mandibular lengthening and widening by gradual distraction using a computer model. J Oral Maxillofac Surg. 1998;56(1):51-9. https://doi.org/10.1016/s0278-2391(98)90916-8 PMid:9437982

18. Ahn HW, Kim KW, Yang IH, Choi JY, Baek SH. Comparison of the effects of maxillary protraction using facemask and miniplate anchorage between unilateral and bilateral cleft lip and palate patients. Angle Orthod. 2012;82(5):935-41. https://doi.org/10.2319/010112-1.1 PMid:22380632

19. Nevzatoğlu S, Küçükkeleş N. Long-term results of surgically assisted maxillary protraction vs regular facemask. Angle Orthod. 2014;84(6):1002-9. https://doi.org/10.2319/120913-905.1 PMid:24654941

20. Richardson S, Selvaraj D, Khandeparker RV, Seelan NS, Richardson S. Tooth-borne anterior maxillary distraction for cleft maxillary hypoplasia: Our experience with 147 patients. J Oral Maxillofac Surg. 2016;74:2504.e1-14. https://doi.org/10.1016/j.joms.2016.08.036

21. Richardson S, Krishna S, Khandeparker RV. A comprehensive management protocol to treat cleft maxillary hypoplasia. J Cranio-maxillofac Surg. 2018;46(2):356-61. https://doi.org/10.1016/j.jcms.2017.12.005 PMid:29305090

22. Zúñiga LR, Núñez EG. Management of a Class III malocclusion with facemask therapy anchored with TADs and orthodontic treatment. Case report. Rev Mex Ortod. 2017;5:e170-7. https://doi.org/10.1016/j.ro.2017.12.016

23. Baker DL, Stoelinga PJ, Blijdorp PA, Brouns JJ. Long-term stability after inferior maxillary repositioning by miniplate fixation. Int J Oral Maxillofac Surg. 1992;21(6):320-6. https://doi.org/10.1016/0901-5027(92)80752-0 PMid:1484197

24. Li H, Dai J, Si J, Zhang J, Wang M, Shen SG, et al. Anterior maxillary segmental distraction in the treatment of severe maxillary hypoplasia secondary to cleft lip and palate. Int J Clin Exp Med. 2015;8(9):16022-8. PMid:26629107

25. Menéndez-Díaz I, Muriel J, Cobo JL, Álvarez C, Cobo T. Early treatment of Class III malocclusion with facemask therapy. Clin Exp Dent Res. 2018;4(6):279-83. https://doi.org/10.1002/cre2.144 PMid:30603110

26. Van Sickels JE, Abadi B, Attisha R. Anterior segmental distraction for a Class III maxillary prosthetic defect in a cleft palate patient. J Oral Implantol. 2011;37(4):457-61. https://doi.org/10.1563/AOID-10-00010 PMid:20662670

27. Karakasis D, Hadjiotrou L. Advancement of the anterior maxilla by distraction (case report). J Cranio-maxillofacial Surg. 2004;32(3):150-4. https://doi.org/10.1016/j.jcms.2003.09.009 PMid:15113572

28. Wang XX, Wang X, Li ZL, Yi B, Liang C, Jia YL, et al. Anterior maxillary segmental distraction for correction of maxillary hypoplasia and dental crowding in cleft palate patients: A preliminary report. Int J Oral Maxillofac Surg. 2009;38(12):1237-43. https://doi.org/10.1016/j.ijom.2009.06.028 PMid:19720499

29. Gateno J, Engel ER, Teichgraeber JF, Yamaji KE, Xia JJ. A new Le Fort I internal distraction device in the treatment of severe maxillary hypoplasia. J Oral Maxillofac Surg. 2005;63(1):148-54. https://doi.org/10.1016/j.joms.2004.09.010 PMid:15635571

30. De Clerck HJ, Cornelis MA, Cevidanes LH, Heymann GC, Tulloch CJ. Orthopedic traction of the maxilla with miniplates: A new perspective for treatment of midface deficiency. J Oral Maxillofac Surg. 2009;67(10):2123-9. https://doi.org/10.1016/j.joms.2009.03.007 PMid:19761906

31. Buschang PH, Porter C, Genecov E, Genecov D, Sayler KE. Face mask therapy of preadolescents with unilateral cleft lip and palate. Angle Orthod. 1994;64(2):145-50. https://doi.org/10.1043/0003-3219(1994)064<0145:FMTOPW>2.0.CO;2 PMid:8010523