Case Report

Distraction Osteogenesis: Role and Clinical Application in the Maxillofacial Region

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Abstract

Distraction osteogenesis or callostasis is a technique for new bone formation by gradual separation of bony fragments. The method was first developed for limb lengthening but recently this process has been widely applied in the cranio-maxillofacial bone. The application of this technique included bone lengthening and reconstruction of segmental defects. Several designs of extraoral and intraoral distraction devices were invented to suit different areas of the craniofacial bone. Nevertheless, intraoral distractors have several advantages including minimal scarring and being less cumbersome. Clinical cases using intraoral distraction osteogenesis technique as the alternative treatment for conventional surgical procedures for maxillo-mandibular lengthening and reconstruction of the alveolar segmental defect after tumor resection and before implant installation are presented and discussed in this study.

Key words: Distraction application; Distraction osteogenesis; Maxillofacial region

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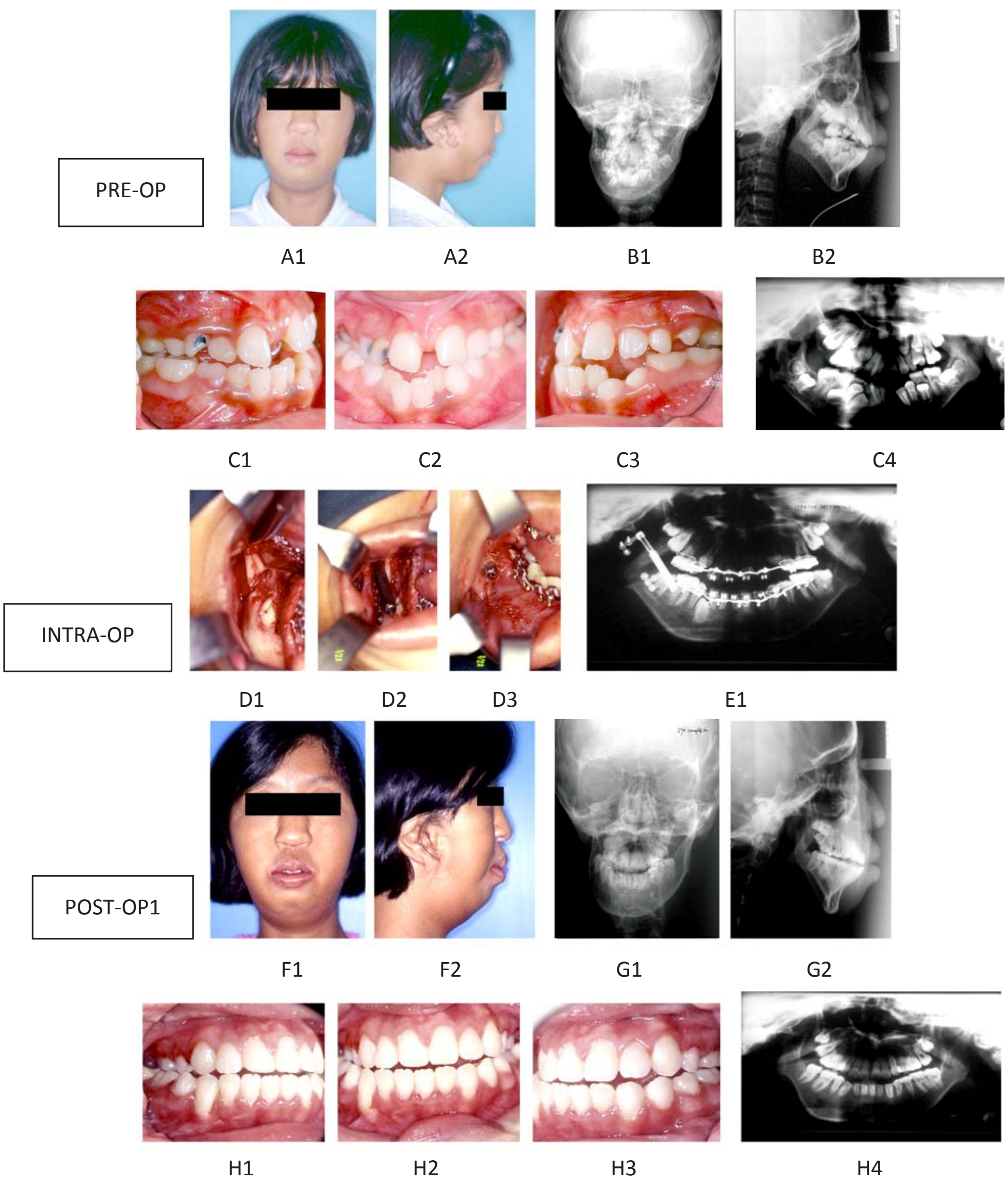
Introduction

Distraction osteogenesis is a method of producing living bone directly from a special osteotomy by controlled mechanical distraction. The regenerated fibrovascular tissues in the distraction gap align themselves parallel to the distraction vector. Then the osteoid tissue is lay down and fill with the mineralize tissue. The new bone spontaneously bridges the gap and rapidly remodels to a normal macrostructure local bone.[1-3] Distraction osteogenesis is also called Callostasis (generating new bone by stretching the callus, as in a fracture). This concept of bone lengthening was first described by Codivilla in 1905[4], who used it to elongate a femur by repeated pulling forces. Other investigators also applied this technique but it remained undeveloped because of associated complications such as nonunion, nerve damage, local edema, skin necrosis, and pin track infection.[5,6] However, the technique of bone lengthening by gradual distraction was further developed and refined by Russian orthopeadic surgeon G.A. Ilizarov in 1952.[1] Since distraction osteogenesis used local host tissue to regenerate new bone, it offers many potential advantages over bone grafting. Sources for autografts are limited and may leave local morbidity at the donor site. Allografts may transmit unknown antigens, bacteria or even viruses. As dead foreign bodies, allografts may not be desirable in infected wound. The use of distraction osteogenesis in the craniofacial skeleton was first reported by Snyder *et al.*[7] who used monofocal distraction to lengthen the canine mandible. Successful clinical bone lengthening in craniofacial surgery was first described by McCarthy *et al.* in 1992.[8] Using extraoral distraction devices; McCarthy lengthened the congenital hypoplastic mandible in four children with Nager's syndrome. The result was satisfied with new bone formation without any relapse being found. Since then several clinical reports with a variety of devices and techniques are available to lengthen segments or entire maxillary or mandibular arches.[9-12] Although the application of the Ilizarov technique to the maxillofacial skeleton showed promising outcome, its use has not been widespread. Extraoral appliances have been effective in clinical cases, but their use has been hampered by many complications.[8,13] These included skin or bone necrosis, pin track infection, scarring, facial nerve and inferior alveolar nerve injury, and poor predictability.[8,14] Michieli and Miotti addressed these concerns by the use of a specially fabricated intraoral tooth-borne appliance to provide the necessary distraction. The development of intraoral appliances occurred in several centers and authorities as reported by Guerrero[15], McCarthy *et al*[16], Chin and Toth[9] and Diner *et al.*[17-18] Potential benefits of internal devices included 1) elimination of skin scars caused by translation of transcutaneous fixation pins, 2) improved patient compliance during the fixation or consolidation phase because there is no external component, and 3) improved stability of the attachment of the device to the bone. The following cases report demonstrated the use of intraoral distraction devices to correct a variety of maxillofacial skeletal deformities in four patients. These included mandibular lengthening in hemifacial microsomia and severe mandibular deficiency patients, maxillary distraction for hypoplastic maxilla in cleft lip and palate patient and interdental distraction of posterior maxilla in patients with benign odontogenic tumor after tumor resection.

Cases illustration

Case I

An 11-year-old female presented with the diagnosis of left hemifacial microsomia type II b. Hypoplastic face and ear deformities were noticed on the left side of the face (Fig. 1: A1, A2). The radiographic examination revealed hypo-development of the right ramus and condyle with chin deviation to the affected side (Fig. 1: B1, B2). Severe malocclusion and canting of the occlusal plane to the right side were demonstrated (Fig. 1: C1 - C4). Mandibular ramus distraction osteogenesis was done using an intraoral partially submerged mandibular distraction device. The osteotomy was performed and the distraction device was installed in the planed position leaving the activation rod outside the mucosal cuff (Fig. 1: D1 - D3). After the 3-day latency period the bone stumps were gradually separated by 0.5 mm twice a day to obtain a total bone lengthening of 14 mm (Fig. 1: E1). After a consolidation period of 8 weeks the device was removed without any complication. The facial asymmetry of the patient dramatically improved (Fig. 1: F1, F2). The radiographic study demonstrated a normal cortico-medullary pattern of the distraction gap indistinguishable from the adjacent native bone. The chin deviation was significantly improved by ramus lengthening (Fig. 1: G1, G2). The satisfied occlusion and less canting of the occlusal plane were obtained following the post-distraction orthodontic treatment (Fig. 1: H1 - H4). One year later, Phase II surgical correction was performed including right intraoral vertical ramus osteotomy (IVRO), left sagittal split ramus osteotomy (Lt SSRO) and advancement genioplasty to correct the residual deformities and obtain the optimum facial profile balance (Fig. 1: I1 – I3; J1, J2). The stable occlusion and canting of the occlusal plane was corrected (Fig. 1: K1 – K3; L1)



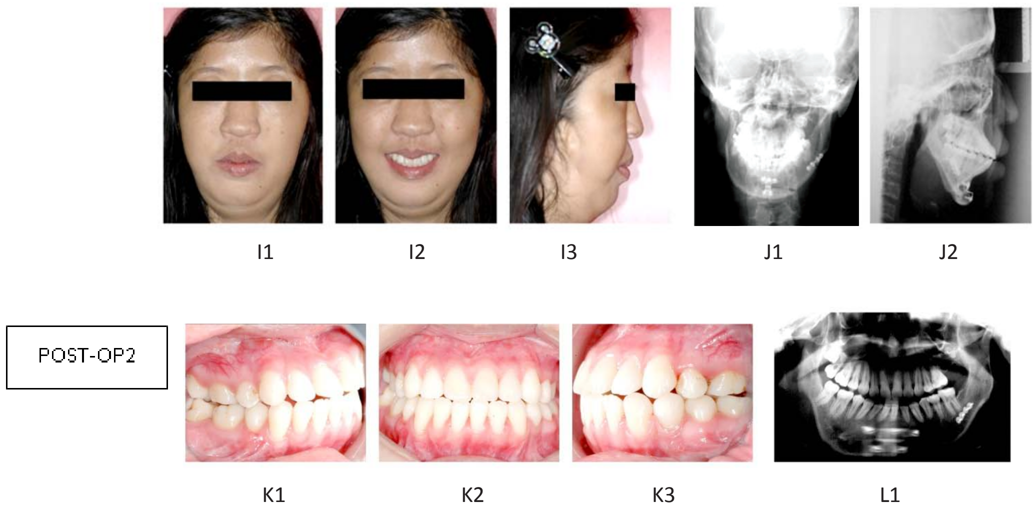
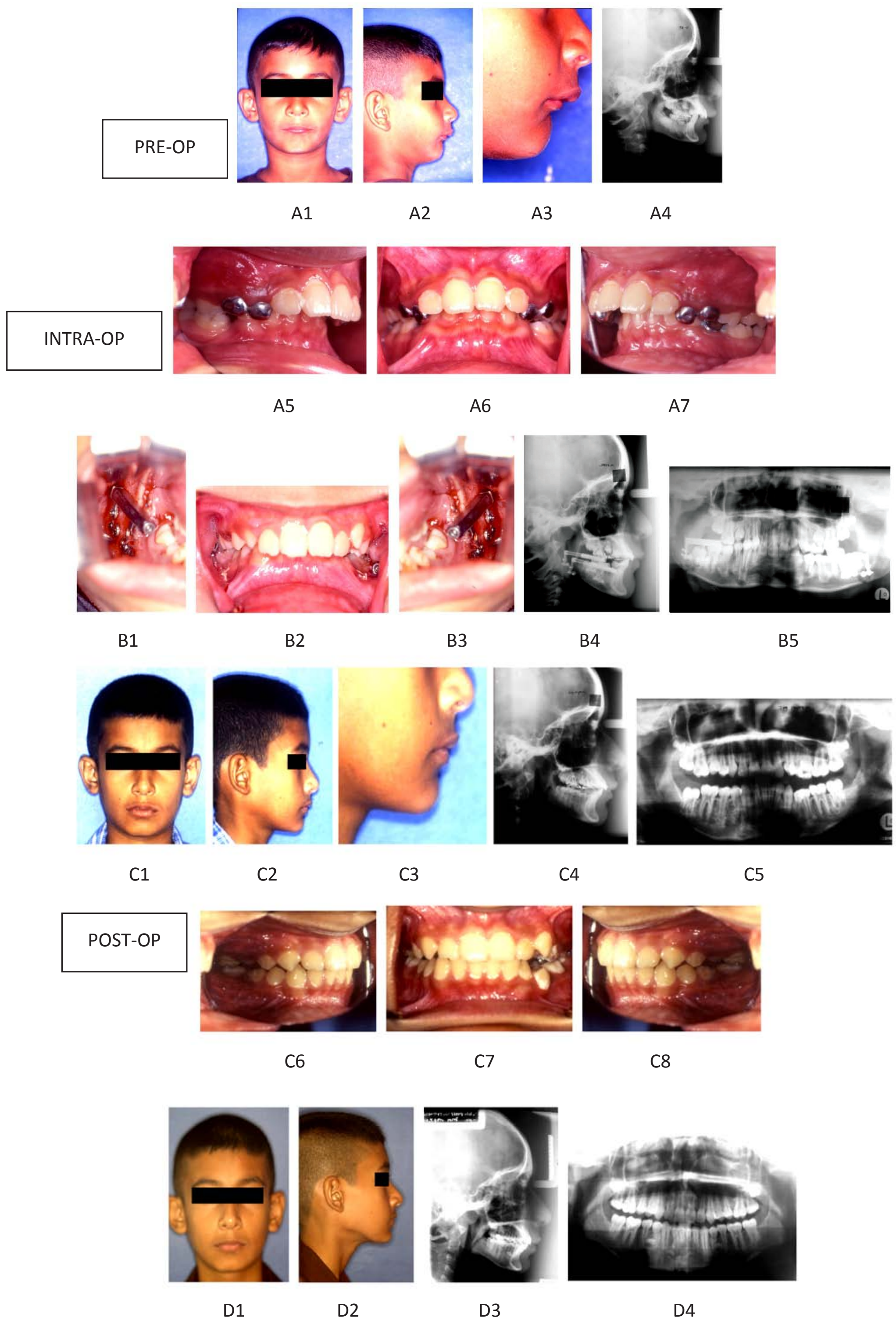


Figure 1 *Clinical case of an 11-year-old girl with hemifacial microsomia who underwent right mandibular ramus distraction osteogensis.*

A1 - A2	*Pre-operative view showed left hypoplastic face and ear deformities.*
B1 - B2	*Hypo-development of right ramus and condyle with chin deviation.*
C1 - C4	*Severe malocclusion and canting of the occlusal plane to the right side.*
D1 - D3	*A distraction device was fixed to the osteotomized bone and the surgical wound was closed leaving the activation rod exposed to the oral cavity.*
E1	*A 14-mm-distraction gap was achieved.*
F1 - F2	*Improvement of the hypodevelopment on the right side of the face.*
G1 - G2	*A distraction gap filled with radiographically normal bone and the lengthening mandible resulted in improvement of the deviated chin.*
H1 - H4	*Satisfactory occlusion and less canting of the occlusal plane.*
I1 - I3	*Clinical appearance of the patient after Phase II surgical correction including Rt IVRO, Lt SSRO and advancement genioplasty.*
J1 - J2	*The bone gap was completely healed and the facial profile was improved.*
K1 - K3, L1	*Stable occlusion and canting of the occlusal plane was corrected.*

Case II

An 11-year-old male presented with a marked retrusive chin caused by a non-syndromic severe mandibular deficiency (Fig. 2: A1 - A7). The functional orthodontic treatment to enhance mandibular growth was not successful after two years of treatment. Bilateral mandibular distraction osteogenesis was planned using 2 intraoral partially submerged mandibular distraction devices. The osteotomy line was cut just anterior to the mandibular angle. After complete bone separation, the distractor was placed in position guided by prediction tracing. A similar procedure was duplicated on the other side. The activation rods were left uncovered in both buccal vestibular areas (Fig. 2: B1 - B3). Gradual distraction of 0.5 mm twice a day was performed after a 3 days latency period. The vector of the distraction movement was controlled by the preformed occlusal splint with a total distance gain in both sides of approximately 14 mm (Fig. 2: B4, B5). After the consolidation period of 8 weeks, the distraction devices were removed without any complication. The clinical facial profile was significantly improved with a straight profile and better chin prominence (Fig. 2: C1 - C3). The radiographs showed a 14 mm gain with normal cortico-medullary pattern of new bone in the previous distraction gap similar to the adjacent normal bone (Fig. 2: C4, C5). The final class I occlusion was achieved (Fig. 2: C6 - C8) after postoperative orthodontic treatment. The occlusion was stable without relapse after 3 years follow up (Fig. 2: D1 - D7).



POST-OP 3 YR

D5 D6 D7

Figure 2 *Clinical case of an 11-year-old male with a severe retrusive chin receiving mandibular lengthening by bilateral distraction osteogenesis of mandibular body.*

A1 - A7 *Clinical appearance of the patient showed marked retrusive chin and malocclusion from severe mandibular deficiency.*

B1 - B3 *The distractor were placed after mandibular osteotomy on both sides and the mucosa was closed leaving the activation rod exposed in both buccal vestibules.*

B4 - B5 *14-mm-distraction gap was created on both sides of the mandibular body.*

C1 - C3 *A dramatically improved of the facial profile with normal chin projection postoperatively.*

C4 - C5 *A normal radiographic pattern of mature bone filled in the distraction gap.*

C6 - C8 *Class I occlusion was achieved after post-distraction orthodontic treatment.*

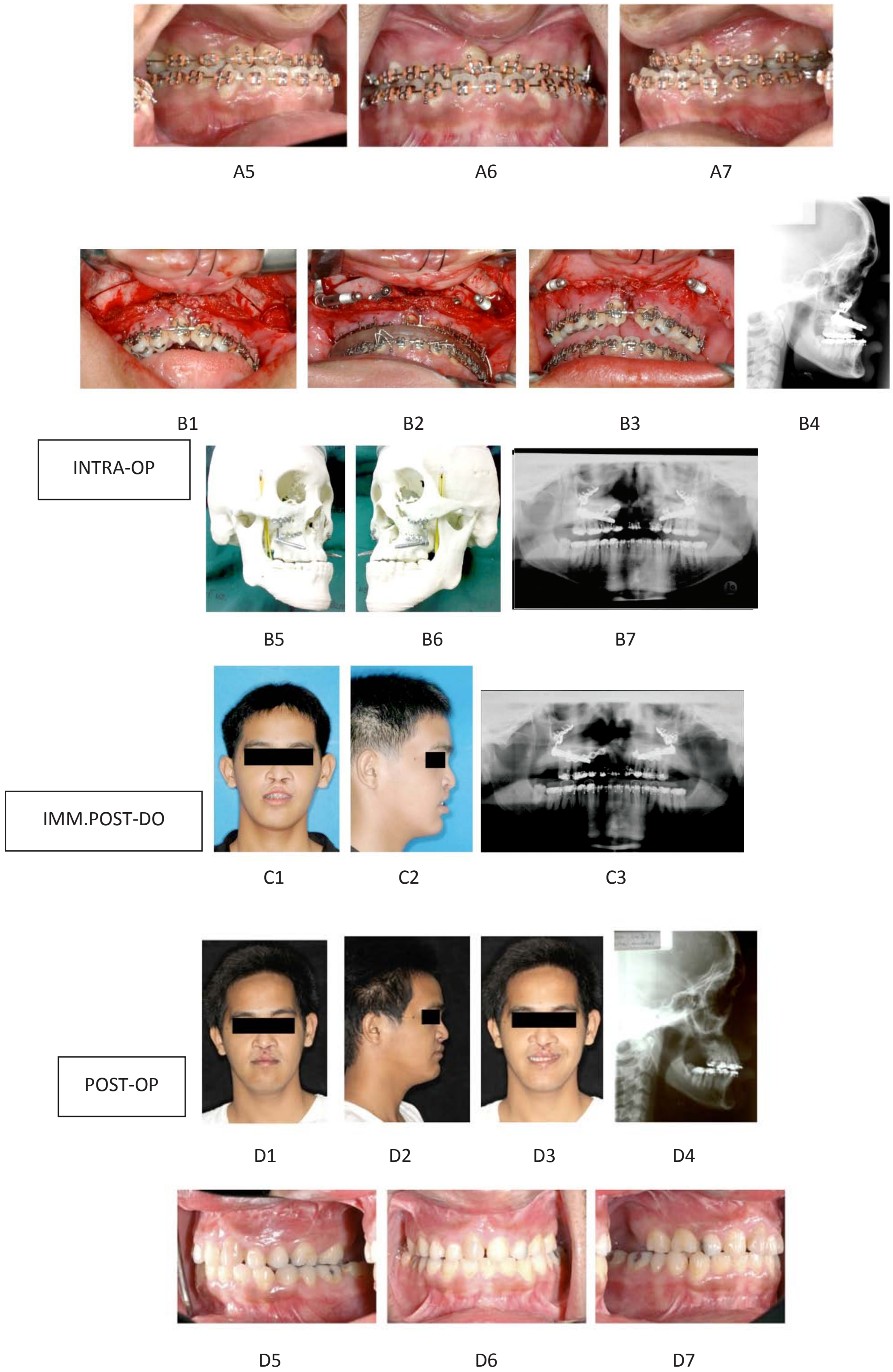
D1 - D7 *Final stable occlusion was stable without any relapse was detected after 3 years follow up.*

Case III

An 18-year-old male born with a bilateral complete cleft lip and palate had undergone surgery for lip repair in infancy, and his cleft palate was repaired in early childhood. Alveolar bone grafting had been performed two years prior the operation with satisfactory results. The residual problem was severe hypoplasia of the maxilla resulting in a concaved facial profile with large negative anterior overjet (Fig. 3: A1 - A7). Maxillary distraction osteogenesis was planned using an intraoral bone borne maxillary distractor. Le Fort I level osteotomy was performed, and the maxilla was partially mobilized to facilitate the distraction movement. The distraction devices were placed according to the planned direction according to the lateral cephalometric film prediction tracing on both sides of the maxilla and fixed on the zygomatic buttress and subapical areas with titanium screws (Fig. 3: B1 - B7). The activation was done intra-operatively to confirm the possibility of the maxillary bone movement vector and stability. The surgical wound was closed leaving the activation rod uncovered in the anterior vestibular area. After a latency of 3 days, activation of both maxillary distracters commenced at 1 mm per day in 2 rhythms. An advance of the maxilla by 12 mm was achieved. Eventually the dramatically improvement of the facial profile and stable class I occlusal relationship were obtained (Fig. 3: C1 - C3). Quality of life was achieved from both significantly improved in facial profile and good occlusion after the postoperative orthodontic treatment without any relapse (Fig. 3: D1 - D7).

PRE-OP

A1 A2 A3 A4


A5
A6
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B1
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D1
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__Figure 3__ Clinical courses of an 18-year-old male born with bilateral cleft lip and palate who underwent maxillary distraction osteogenesis for correcting cleft maxillary hypoplasia.

A1 - A7 A clinical examination and radiographic study revealed a concaved facial profile from severe maxillary deficiency and marked negative overjet.

B1 - B4 Le Fort I level osteotomy was performed and minimum mobilized. The distraction devices were fixed on both sides. The exposed activation rods were located at the anterior vestibule.

B5 - B7 Excellent position of the devices were achieved by pre-bending of the distractor in the individual fabricated stereo-model .

C1 - C3 Clinical appearance of the patient after 12 mm gradual maxillary advancement, the improved facial profile and stable class I occlusion were achieved.

D1 - D4 Significant improvement of the facial profile and normal radiographic pattern of bone remodeling with normal relationship of both jaw bones.

D5 - D7 Class I occlusion was achieved after postoperative orthodontic treatment with stable occlusion without any relapse.

Case IV

A 51-year-old female patient presented with a painless slow growing mass at the right posterior maxilla with pathological diagnosis of cementoblastoma (Fig. 4: A1 - A5). The tumor with the adjacent bone including the molar teeth and maxillary tuberosity were surgically removed by partial maxillectomy (Fig. 4: B1 - B3). A vertical interdental osteotomy line between the maxillary canine and first premolar teeth for a subapical bone osteotomy was performed. The transported dento-osseous segment comprised of two premolar teeth and its alveolar process was completely mobilized leaving only the palatal pedicle as the source of blood supply. The mandibular ramus distraction device was modified and placed on the buccal surface and fixed to the transported segment and apical bone above the canine area (Fig. 4: B4 - B7). The vector of the distraction was adjusted to accord with the posterior maxillary arch form and lower dentition. Following a 3 days latency period, the distractor was activated, 0.5 mm per time, twice daily for 13 consecutive days. A distraction gap of approximately 13 mm was achieved between the right maxillary canine and the first premolar (Fig. 4: C1 - C3). The distraction device had good stability during the distraction procedure and throughout the 6 weeks consolidation period. The transported segment was moved posteriorly along the planned direction until the maxillary second premolar occluded on the mandibular second molar without any occlusal interference. The regenerated tissue in the distraction gap eventually healed with normal contour and covered with soft tissue similar to the adjacent mucosa (Fig. 4: D1, D2). The radiographic and histological study obtained from the tissue in the distraction area demonstrated newly formed bone in the distraction gap with normal gingival tissue coverage (Fig. 4: D3 - D5). Six months after the consolidation period, the distracted gap was reentry and dental implant was inserted into the normal appearance regenerated bone. Eventually the final dental prosthesis was successfully constructed with fully function.

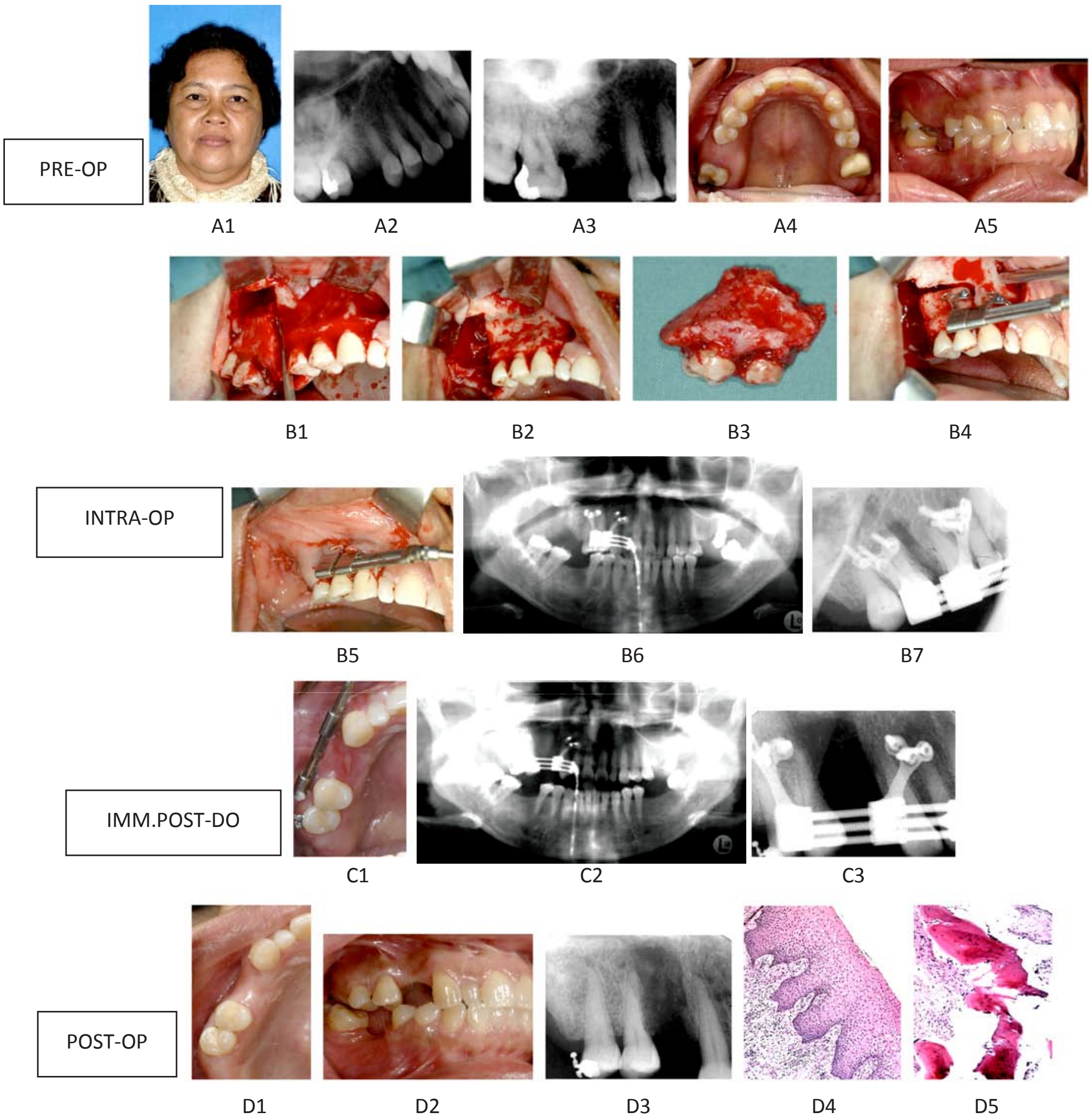

Figure 4 *A clinical case of a 51-year-old female patient with diagnosis of cementoblastoma underwent interdental transport distraction osteogenesis.*

A1 - A5 *A painless slow growing mass at the right posterior maxilla with diagnosis of cementoblastoma.*

B1 - B3 *A right posterior partial maxillectomy was performed.*

B4 - B7 *An interdental osteotomy line between the maxillary canine and the first premolar teeth was created to mobilize the transported segment then the distraction device was fixed to secure the segments.*

C1 - C3 *Following gradual distraction, a distraction gap of approximately 13 mm was achieved.*

D1- D2 *Regenerated tissue in the distraction gap eventually healed with normal contour and covered with intact mucosa.*

D3 - D5 *Radiographic and histological study showed new bone formation in the distraction gap with normal gingival tissue coverage.*

Discussion

The technique of distraction osteogenesis involves creation of new bone by gradual movement of two or more bony fragments following their surgical division. This technique can provide required amounts of regenerated bone in the skeleton that still has the potential of fracture healing. Distraction osteogenesis was first developed in the field of orthopaedic surgery and was principally utilized in limb lengthening. Knowledge from long bone lengthening provided the major basis for development of the distraction osteogenesis in the membranous bone of the craniofacial skeleton. Nevertheless, application of extraoral distraction devices in the maxillofacial region does not seem to be popular with the majority of the patients because of the cumbersome appearance, scar tissue formation and social adjustment. The innovation of the intraoral distraction devices provided treatment that is more acceptable in the mainstream practice of maxillofacial surgery. The intraoral approach for device placement can avoid skin incision that resulted in reducing the risk of injury to the nerves such as marginal mandibular branch of facial nerve. The other potential benefits include no skin scars caused by translation of transcutaneous fixation pins, improved compliance during the long consolidation phase, and no maintenance is required because the submerged placement. Nevertheless the drawbacks of this technique are the expensive sophisticated device and multiple operation including device installation and removal. In addition, in some cases the adjunction surgical procedure must be performed to obtain the optimum result. In the present study demonstrated four clinical cases with different deformities using an intraoral distraction device to correct the skeletal deficiencies with satisfactory outcomes.

Mandibular lengthening in the present report was performed in an early stage of life in hemifacial microsomia patient. This deformity should be treated as early as possible since mandibular asymmetry becomes worse over time in comparison to the normal growth of the non-affected, contralateral side. This then leads to secondary malformation of the maxilla, nose and orbit. A progression of facial deformity and psychological problems may occur with time.[19] Therefore, the mandible should be operated upon early in order to support the growth of the adjacent structures and to avoid or minimize secondary deformities. In addition, in hemifacial microsomia, lengthening of the hypoplastic mandible will create an ipsilateral posterior open bite. The provided space will allow occlusal canting correction by orthodontic means over a short period by gradual selective grinding on the occlusal splint since there still has vertical growth of the maxilla in children when it is released from the constriction effect of the mandible and soft tissue.[20] By this protocol of treatment, the maxillary surgery can be avoided when comparing to the delay treatment in the adult stage with the conventional surgical operation.

Major advancement of the maxilla is one of the unstable procedures since the movement of the large skeletal fragment of maxilla typically requires overcoming significant resistance from the soft tissue envelopes. This situation seems worse when the advancement of maxilla is performed in the cicatrized tissue from a previous operation in cleft palate patients. Incremental movement using distraction mechanics allows displacement of fragments over large distances because the soft tissue is allowed to accommodate slowly. The elongation of muscles, ligaments, vessels, nerves, subcutaneous fat, and skin that can not be achieved by other more radical procedures is one of the main advantages of this method.[21] By combining the conventional Le Fort I osteotomy with postoperative gradual distraction, a correction of severe overjet of approximately 12 mm was achieved in case III. Eventually a soft tissue facial profile and stable positive overjet occlusion was obtained without any detected relapse during the follow up period.

In addition to the application of distraction osteogenesis to craniomaxillofacial defects, this modality of treatment could be modified to use in the dentoalveolar region. Interdental transport distraction osteogenesis

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was demonstrated in case IV. The transported dento-osseo segment comprised of two premolars, and their adjacent subapical bone was gradually moved posteriorly to close the distal defect created by the partial maxillectomy for tumor resection. The vector of movement was planned and controlled by distraction device to conform to the posterior maxillary arch, and lower dentition resulted in acceptable post-distraction occlusion. The regenerated tissue in the distraction gap eventually healed without any complications. The newly formed bone in the distraction alveolar segment and normal mucosal coverage could be demonstrated by both radiographic and histological studies. The transported segment retained good stability after distractor removal without detectable relapse and provided sufficient bone volume for implant placement. According to the satisfactory outcome, interdental transport distraction osteogenesis offers an alternative treatment for segmental defect reconstruction. The procedure could be performed in a single operation where the tumor was resected without additional donor site morbidity.

Distraction ostegenesis demonstrates a significant role and provides a variety of applications in the maxillofacial region with acceptable results. The development of an intraoral distractor holds the promise of allowing the patient to enjoy a more normal life during the course of treatment.[22,23] Further development including design, miniaturization of the intraoral devices and multidirectional vectors are essential to reconstruct various parts for complex cranio-maxillofacial malformation. Long-term follow up is necessary to monitor the post-distraction growth potential and possible relapse.

References

1. Ilizarov GA. The tension-stress effect on the genesis and growth of tissue: Part I. The influence of stability of fixation and soft-tissue preservation. ***Clin Orthop Relat Res*** 1989;238:249-81.
2. Ilizarov GA. The tension-stress effect on the genesis and growth of tissues: Part II. The influence of the rate and frequency of distraction. ***Clin Orthop Relat Res*** 1989;239:263-85.
3. Ilizarov GA. Clinical application of the tension-stress effect for limb lengthening. ***Clin Orthop Relat Res.***1990;250:8-26.
4. Codivilla A. On the means of lengthening in the lower limbs, the muscles and tissues which are shortened through deformity. ***Am J Orthop Surg*** 1905;2:353-69.
5. Abbott LC. The operative lengthening of the tibia and fibula. ***J Bone Joint Surg (Br)*** 1927;9:128-52.
6. Abbott LC, Saunders JB. The operative lengthening of the Tibia and Fibula: a preliminary report on the furhter development of the principles and technic. ***Ann Surg*** 1939; 110:961–91.
7. Snyder CC, Levine GA, Swanson HM, Browne EZ Jr. Mandibular lengthening by gradual distraction. ***Plast Reconstr Surg*** 1973;51:506-8.
8. McCarthy JG, Schreiber J, Karp N, Thorne CH, Grayson BH. Lengthening the human mandible by gradual distraction. ***Plast Reconstr Surg*** 1992;89:1-10.
9. Chin M, Toth BA. Distraction osteogenesis in maxillofacial surgery using internal devices: review of five cases. ***J Oral Maxillofac Surg*** 1996;54:45-54.
10. Block MS, Chang A, Crawford C. Mandibular alveolar ridge augmentation in the dog using distraction osteogenesis. ***J Oral Maxillofac Surg*** 1996;54:309-14.
11. Altuna G, Walker DA, Freeman E. Rapid orthopedic lengthening of the mandible in primates by sagittal split osteotomy and distraction osteogenesis: a pilot study. ***Int J Adult Orthodon Orthognath Surg*** 1995;10:59-64.
12. Rachmiel A, Levy M, Laufer D. Lengthening of the mandible by distraction osteogenesis: report of cases. ***J Oral Maxillofac Surg*** 1995;53:838-46.
13. Molina F, Ortiz Monasterio F. Mandibular elongation and remodeling by distraction: a farewell to major osteotomies. ***Plast Reconstr Surg*** 1995;96:825-42.
14. Aronson J. Experimental and clinical experience with distraction osteogenesis. ***Cleft Palate Craniofac J*** 1994;31:473-82.
15. Guerrero CA, Bell WH, Contasti GI, Rodríguez AM. Intraoral mandibular distraction osteogenesis. ***Semin Orthod*** 1999;5:35-40.
16. McCarthy JG, Staffenberg DA, Wood RJ, Cutting CB, Grayson BH,

Thorne CH. Introduction of an intraoral bone-lengthening device. ***Plast Reconstr Surg*** 1995;96:978-81.

17. Diner PA, Kollar E, Martinez H, Vazquez MP. Submerged intraoral device for mandibular lengthening ***J Craniomaxillofac Surg*** 1997;25:116-23.
18. Diner PA, Kollar EM, Martinez H, Vazquez MP.Intraoral distraction for mandibular lengthening: a technical innovation. ***J Craniomaxillofac Surg*** 1996;24:92-5.
19. Murray JE, Kaban LB, Mulliken JB. Analysis and treatment of hemifacial microsomia. ***Plast Reconstr Surg*** 1984;74:186-99.
20. Ortiz Monasterio F, Molina F, Andrade L, Rodriguez C, Sainz Arregui J. Simultaneous mandibular and maxillary distraction in hemifacial microsomia in adults: avoiding occlusal disasters. ***Plast Reconstr Surg*** 1997;100:852-61.
21. Sawaki Y, Ohkubo H, Hibi H, Ueda M. Mandibular lengthening by distraction osteogenesis using osseointegrated implants and an intraoral device: a preliminary report. ***J Oral Maxillofac Surg*** 1996;54:594-600.
22. Sawaki Y, Ohkubo H, Yamamoto H, Ueda M. Mandibular lengthening by intraoral distraction using osseointegrated implants. ***Int J Oral Maxiilofac Implants*** 1996;11:186-93.