Distraction osteogenesis: Our experience of six patients

Madhumati Singh, Anjankumar Shah, Beena Roopak, Vijay Anand S, Sathesh Kumar R., Amit B.

Department of Oral and Maxillofacial Surgery, Rajarajeshwari Dental College and Hospital, Bengaluru, Karnataka, India

Abstract

**Background:** Distraction osteogenesis (DO) is an established therapeutic technique for new bone formation during significant lengthening of the craniofacial skeleton without the need for bone grafting or orthognathic surgery. Hemifacial microsomia, mandibular retrognathia, posttraumatic or resection deformities of the mandible and multiple syndromes associated with maxillo-mandibular malformations have been successfully corrected using this technique. **Materials and Methods:** We report six cases who presented with a varied range of symptoms due to congenital and acquired deformities of the maxilla and mandible. DO was used for the correction of each of these deformities. Vertical alveolar DO was performed in two patients while two required mandibular distraction. Surgically assisted rapid maxillary expansion (SARME) was used in one patient. Both mandibular distraction and SARME was performed in the other patient. **Results:** In all cases, the planned lengthening was achieved without any immediate or delayed complications. **Conclusion:** Thus, DO consistently produce a measurable increase in the bone length with better functional and esthetic outcomes.

**Keywords:** Alveolar distraction, distraction osteogenesis, free fibula graft, platelet rich fibrin, surgically assisted rapid maxillary expansion

**Introduction**

Codivilla in 1905, first described distraction osteogenesis (DO) for lengthening of the femur. This technique was further developed and popularised by Ilizarov in 1988. In 1989, McCarthy was the first to clinically apply the technique of extra oral osteodistraction on four children with congenital craniofacial anomalies. In 1996, Chin and Toth reported the first application of vertical mandibular alveolar DO. In 1938, Brown first described surgically assisted rapid maxillary expansion (SARME), a form of DO, for palatal expansion.

DO is a biological process of new bone formation between the surfaces of bone segments gradually separated by the incremental traction. Application of the incremental traction to the reparative callus that joins the divided bone segments generates tension within the callus, which stimulates new bone formation parallel to the vector of distraction. These distraction forces also create tension in the surrounding soft tissues, including skin, muscle, nerves and blood vessels initiating a sequence of adaptive changes referred to as distraction histogenesis. These adaptive changes in the soft tissues allow larger skeletal movements and minimises the potential risk of relapse seen with acute advancement of osteotomized bone segments. Another advantage of mandibular distraction is that it can be performed in early childhood and infancy as against conventional orthognathic surgery which is preferred in non-growing patients. Complications like intraoperative nerve injury, post-operative displacement of bone segments due to inadequate bony contact and insufficient fixation, and partial or total relapse are avoided. Vertical mandibular alveolar DO is used for the augmentation of an affected area of the alveolar ridge, resulting in an increase in both alveolar bone height and labial sulcus depth. It is often used as an adjunctive procedure prior to implant placement and may be preferable to other methods, such as autogenous bone grafting. SARME results in a significant enlargement of the dental arch, the maxillary apical base and the palatal vault and is frequently used to correct skeletal maxillary transverse deficiency (MTD) in non-growing individuals.

**Subjects and Methods**

Six cases (4 males and 2 females) aged between 9 years and 36 years with varied maxillo-mandibular deformities were treated using DO. The details of patients and factors involved are enlisted in Table 1.

Three out of the six cases presented with mandibular hypoplasia. Patient 1 was diagnosed as a case of Treacher Collins Syndrome [Figure 1], while in the other two (patients 2 and 3), mandibular hypoplasia was non-syndromic. Patient 3 had...
**Table 1: Complete patient profile**

<table>
<thead>
<tr>
<th>Parameters</th>
<th>Patient 1</th>
<th>Patient 2</th>
<th>Patient 3</th>
<th>Patient 4</th>
<th>Patient 5</th>
<th>Patient 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age/sex</td>
<td>9 years/male</td>
<td>36 years/male</td>
<td>20 years/female</td>
<td>26 years/male</td>
<td>20 years/male</td>
<td>27 years/female</td>
</tr>
<tr>
<td>Complaints</td>
<td>Difficulty in mouth opening, and breathing, snoring, small chin</td>
<td>Severe sleep apnoea, limited mouth opening, difficulty in breathing, tracheostomy</td>
<td>Unesthetic appearance, difficulty in breathing, limited mouth opening, snoring</td>
<td>Nasal obstruction, mouth breathing and conductive hearing loss</td>
<td>Loss of teeth</td>
<td>Loss of teeth</td>
</tr>
<tr>
<td>Diagnosis</td>
<td>Mandibular hypoplasia associated with Treacher Collins syndrome</td>
<td>Non syndromic mandibular hypoplasia</td>
<td>Non syndromic mandibular hypoplasia</td>
<td>Maxillary transverse deficiency</td>
<td>Post traumatic Alveolar ridge defect of mandible</td>
<td>Post-surgical Alveolar ridge defect of mandible</td>
</tr>
<tr>
<td>Distraction site</td>
<td>Mandibular ramus</td>
<td>Mandibular ramus, midpalate</td>
<td>Mandibular body and ramus</td>
<td>Mid palate</td>
<td>Mandibular alveolar ridge</td>
<td>Mandibular alveolar ridge</td>
</tr>
<tr>
<td>Procedure done</td>
<td>Oblique ramus osteotomy followed by bilateral extra oral uni-directional distraction</td>
<td>Oblique ramus osteotomy followed by bilateral intra oral unidirectional distraction, SARME</td>
<td>Mandibular body and ramus osteotomy followed by bilateral extraoral bidirectional distraction</td>
<td>SARME</td>
<td>Vertical alveolar distraction</td>
<td>Vertical alveolar distraction</td>
</tr>
<tr>
<td>Latency period</td>
<td>7 days</td>
<td>7 days</td>
<td>7 days</td>
<td>7 days</td>
<td>5 days</td>
<td>5 days</td>
</tr>
<tr>
<td>Rate/rhythm</td>
<td>1 mm/day and 0.5 mm twice daily</td>
<td>1 mm/day and 0.5 mm twice daily</td>
<td>1 mm/day and 0.5 mm twice daily</td>
<td>1 mm/day and 0.5 mm twice daily</td>
<td>1 mm/day and 0.5 mm twice daily</td>
<td>1 mm/day and 0.5 mm twice daily</td>
</tr>
<tr>
<td>Distraction achieved</td>
<td>26 mm</td>
<td>26 mm (mandible), 13 mm (ramus), 12 mm (body)</td>
<td>26 mm (mandible), 13 mm (ramus), 20 mm (body)</td>
<td>12 mm</td>
<td>7 mm</td>
<td>8 mm</td>
</tr>
<tr>
<td>Further management</td>
<td>Auricular reconstruction, orthodontic treatment</td>
<td>Orthodontic treatment</td>
<td>Orthodontic treatment</td>
<td>Orthodontic treatment</td>
<td>Implant placement and prosthetic rehabilitation</td>
<td>Implant placement and prosthetic rehabilitation</td>
</tr>
</tbody>
</table>

SARME: Surgically assisted rapid maxillary expansion

Associated right condylar aplasia and left condylar hypoplasia. All of them presented with restricted mouth opening, snoring, breathing difficulty and unesthetic appearance. Patient 2, a case of mandibular hypoplasia with transverse maxillary deficiency, had a tracheostomy in situ as a part of treatment for severe sleep apnea. All of them were subjected to bilateral mandibular DO. Extra oral unidirectional distractor was used for patient 1, while intraoral unidirectional distractor was used in patient 2. Patient 2 was subjected to an additional LeFort I osteotomy, followed by Hyrax expander for palatal expansion to correct the narrow palatal arch [Figure 2]. Patient 3 was treated with an extra-oral bidirectional distractor [Figure 3]. Following a latency period of 1 week, activation was started at a rate of 1 mm/day and continued till the desired lengthening was achieved. This was followed by a consolidation period, which lasted for 8 weeks (patient 1 and 2) to 12 weeks (patient 3).

Patient 4, a case of MTD presented with nasal obstruction, mouth breathing and conductive hearing loss (due to recurrent otitis media). LeFort I osteotomy followed by hyrax expander for palatal expansion (SARME) was used to correct the narrow palatal arch [Figure 4].

Patients 5 and 6 were subjected to vertical intraoral alveolar DO for implant placement. Patient 5 had vertical alveolar bone loss in relation to teeth 31, 41, 42 and 43 regions secondary to trauma [Figure 5]. Patient 6 was a case of segmental mandibular resection for ossifying fibroma, resulting in a large alveolar defect in relation to teeth 31, 32, 33, 41, 42, 43, 44 and 45, treated with microvascular free fibular bone grafting a year back. In this patient, after performing an osteotomy on the graft, the distractor was activated for 2-3 mm and the gap was filled with bone marrow concentrate from the iliac crest enriched with platelet rich fibrin in order to improve the outcome of alveolar distraction [Figure 6]. Following a latency period of 5 days, activation was started at a rate of 1 mm/day and continued till the desired lengthening was achieved. This was followed by a consolidation period, which lasted for 8 weeks in both the patients (patients 5 and 6).

**Results**

In all six cases, the planned lengthening of bone was achieved without any major immediate or delayed complications. Mandibular distraction of 26 mm each was achieved in patients 1 and 2. In patient 3, ramus lengthening of 13 mm and body lengthening of 20 mm was achieved. Palatal expansion of 12 mm each was achieved in patients 2 and 4. Patients 5 and 6 attained a vertical alveolar distraction of 7 mm and 8 mm respectively. On follow-up, all patients showed a relapse ranging from 1 to 3 mm.
Post-surgical orthodontic correction was performed in patients 1, 2 and 3 while implant placement and prosthetic rehabilitation was considered for patient 5 and 6. Satisfactory improvement in aesthetics and functions was achieved in all patients. There was an improvement in mouth opening and resolution of breathing difficulties and snoring in patients 1, 2 and 3. Patient 2 was subsequently weaned off tracheostomy. Improvement in respiratory function and hearing occurred in patient 4. Patient 1 additionally required ear reconstruction and hearing aids as a part of further rehabilitation.

Discussion

Bilateral sagittal split osteotomy (BSSO) and DO are currently the most common methods used to surgically correct mandibular deformities.[5] DO allows major reshaping and sculpting of the facial bones i.e. changing the shape and form of the bones resulting in an improvement in the three-dimensional structural, functional, and aesthetic needs of the patient without complications that may be associated with BSSO.[2] It has also made an intervention possible in much younger patients.[1] On the other hand, BSSO is less predictable in the correction of severe mandibular deficiencies requiring lengthening of more than 8-10 mm.[5] The coupling of bone and soft tissue growth in

Figure 1: (a,b) Pre-operative frontal and lateral view of patient 1 with Treacher Collins syndrome, (c) panoramic radiograph with distractor, (d,e) post-operative views showing improvement in profile

Figure 2: (a,b) Pre-operative frontal and lateral view of patient 2 with tracheostomy in situ, (c) palatal distraction with hyrax expander, (d) patient with intraoral distractor, (e,f) post distraction lateral cephalogram and lateral view

DO results in a very low rate of relapse (7%) when compared to conventional surgical techniques in which relapse rate can be as high as 50%.[6] Hence, DO has gained a generalized acceptance over BSSO for the treatment of mandibular deformities.[2]

Unilateral distraction of the ramus, angle or posterior body of the mandible is indicated for the correction of hemifacial microsomia while bilateral advancement of the body is used in cases of severe retrognathia.[5] Depending on the desired direction of the distraction vector, a distraction device is fixed (to the bone, tooth or both) after performing the required osteotomies.[1] Controversy exists regarding the use of a corticotomy instead of osteotomy during DO. It is believed that preservation of intramedullary vessels during corticotomy is beneficial for bone regeneration and is an effective way to promote the maturity of the distracted callus.[7] However, it is better to perform an osteotomy in the mandible to allow better movement of the segments and increased control of the planned vector of elongation.

The first phase is the latency period during which the periosteal integrity is restored, and callus formation begins at the bony gap. This period usually varies from 5 to 7 days and is shorter in children due to the high rate of bone metabolism. The bone segments at either end of the gap are progressively distracted during the distraction period, wherein immature bone
is laid down within the gap. Distraction is performed at a rate of 1 mm/day with a frequency of 1-2 times daily. Faster rate of distraction is associated with fibrous non-union while a slower rate results in premature union.\textsuperscript{1} During the next phase known as a consolidation period which lasts for 6-8 weeks, the bone remolds into a mature state and the surrounding soft tissues adapt to their new positions. Successful DO depends on multiple factors, the most important being osteocyte viability and blood supply.\textsuperscript{1} Hence, thermal and mechanical bone injury at the distraction site is kept to a minimum.\textsuperscript{1}

Distraction devices are specially fabricated hardware used for the distraction process.\textsuperscript{3} External or extra-oral devices can be unidirectional, bidirectional or multidirectional thereby...
allowing manipulation of bone segments in multiple planes of space resulting in a greater flexibility and permitting corrections and improvisation during the process of distraction.\(^1,6\) In spite of ease of placement and removal of these devices, they are associated with a higher incidence of infections, displacement of the distractor and supporting pins and a potential risk of permanent facial scars.\(^6\) They are externally visible and cumbersome to use resulting in a negative psychosocial effect and requiring special care throughout treatment especially in children.\(^6\) These limitations paved the way for the development of intraoral devices.

The main advantage of the intraoral distractor is that it is socially convenient to the patient, without the discomfort associated with the extra oral devices.\(^6\) Higher quality distraction is possible with intraoral devices due to the proximity between bone and the frame, but they require a second major surgical procedure for removal.\(^1,6\) Intraoral devices have now evolved from unidirectional to bidirectional to multidirectional distraction.\(^9\)

Alveolar DO has been traditionally used as a definitive procedure for vertical augmentation of the alveolar ridge before dental implant placement (patient 5).\(^9\) It is also used as an adjunctive procedure to enhance the sites previously reconstructed with iliac bone grafts, radius, scapular and fibular free flaps (patient 6).\(^9\)

The free vascularized fibular flap was first used for the reconstruction of mandibular defects by Hidalgo in 1989.\(^11\) The fibular flap presents many advantages over other microvascular grafts, such as sufficient length of the bony segment (25-30 cm of dense cortical bone), good vascularisation, better quality of the bone, long vascular pedicle, and proper dimension for implant placement.\(^1,11,12\) However, since it does not offer sufficient bone height to restore the alveolar arch when reconstruction involves a dentate mandible, vertical alveolar distraction plays an adjuvant role.\(^11\) The distracted bone can receive dental implants as predictably as a native bone.\(^6\) Implant placement is recommended 10-16 weeks after the end of the activation period.\(^6\) Dual purpose devices, which are distractors and implants at the same time help to reduce the chair time and simplify the procedure.\(^6\)

Bone marrow concentrate enriched with autologous platelet-rich fibrin (PRF) was used in patient 6 to promote bone healing and regeneration. PRF is a biomaterial derived intra-operatively from the patient, that incorporates leukocytes, platelets, growth factors (platelet derived growth factor, transforming growth factor, vascular endothelial growth factor and insulin-like growth factor) and a wide range of glycoproteins in a dense fibrin matrix.\(^1,3,14\) It is now known that, bone formation may be improved by augmenting the distraction site with both osteoprogenitor cells such as bone marrow cells and osteoinductive growth factors obtained from PRF.\(^14\) The recombinant human bone morphogenetic proteins (rhBMP-2) such as BMP-2 and BMP-7 also have similar benefits.\(^1,3,14\)

SARME is a reliable method to expand the constricted maxilla, alleviate the narrow upper airway and improve the nasal dimensions along with airway resistance in patients with sleep apnoea due to severe craniomaxillomandibular deformities.\(^15,3\) Our patient was successfully weaned off tracheostomy following the above procedure. It also results in an improvement in nasal respiration and Eustachian tube functions thus reducing the incidence of middle ear infections, thereby improving the hearing (patient 4).

Recent developments continue to occur in the field of craniofacial DO. Biodegradable devices made of bioreabsorbable materials have been developed to abridge the need for a second operation for removal because they resorb, leaving no trace of ever having been inserted.\(^16\) Growth factors for promoting bone growth and calcium sulfate for accelerating bony consolidation can now be administered through a specially designed cannulated pin to improve the outcomes of distraction.\(^16\) Auto-distraction according to pre-programmed data is now possible with the use of microprocessors and miniature motorized distraction devices.\(^1\) Endoscopic techniques may be used to create osteotomies and to insert distraction devices that will convert the surgery for distraction into a minimally invasive surgery.\(^1\) Newer technologies, such as computer-assisted surgical planning, are now being applied to alveolar distraction.\(^10\)

Conclusion

DO has established itself as a reliable and evidence-based alternative in the treatment of bone discontinuity. It has been proven to be a safe and predictable procedure with a low incidence of major complications. We could successfully use DO for the correction of different maxillo-mandibular deformities in six patients resulting in good functional and aesthetic outcomes. Though not regarded as the “gold standard” method, it is still a very promising technique and has become widely accepted as the treatment of choice in lengthening of the mandible in both congenital and acquired defects. The indications for its use are increasing as also its popularity as an alternative to many conventional orthognathic surgical procedures.

References

5. Dolanmaž D, Karaman AI, Gurel HG, Kalayci A, Kucukkolbası H, Usumez S. Correction of mandibular retrognathia and