Management of Obstructive Sleep Apnea and Non Apneic Snoring with Maxillo-Mandibular Distraction Osteogenesis

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Abstract
Background: Recently Distraction Osteogenesis (DO) has been used successfully for advancing maxilla and mandible to treat sleep disordered breathing like Obstructive sleep apnea and non apneic snoring. Prime advantage of DO is slow stretching of soft tissues and bone formation allowing greater advancement and enhancement of airway volume.

Patients and Methods: 10 cases with sleep disordered breathing due to maxillo-mandibular discrepancy of varying age groups were treated by various DO techniques.. All the cases showed gross improvement not only in facial and dental aesthetics but also respiratory symptoms. A mean increase of 5.14 mm and 1.85mm with respect to posterior airway space and retro palatal space was observed in mandibular corpus lengthening cases. All the cases showed improvement subjectively and objectively.

Conclusion: DO has been found to be valuable and gives us the ability to both prevent and correct the development of sleep disordered breathing.

Introduction
Sleep disordered breathing affects all age groups and the most common of them are non apneic snoring and obstructive sleep apnea. It is the most common yet often under appreciated conditions in Cranio-facial deformities. Maxillo-Mandibular distraction osteogenesis is one of the most contemporary and effective surgical treatment options [1] [2] [3].

OSA is a potential life threatening disorder caused by partial or complete obstruction of upper airway. If the obstruction sites are retroglossal or retro palatal maxillo-mandibular advancement would prove beneficial. Certain craniofacial abnormalities are susceptible to OSA and non apneic snoring. These include hypoplastic mandible, sagittal maxillary deficiency, narrow maxillary arch, long face syndrome, inferiorly and posteriorly positioned hyoid bone. Distraction Osteogenesis (DO) can be performed in all age groups and is a valuable tool that gives us the ability to both prevent and correct the development of sleep disordered breathing [1] [2] [3].

Distraction Osteogenesis is a biologic process of new bone formation between vascularised margins of bone segments gradually separated by incremental traction [4]. The basic concept of the procedure is induction of new bone formation along the vector of pull obviating the need for a bone graft. Finally, the increase in resulting bone stock will provide more reconstructive options in the future for traditional orthognathic surgical techniques. The traction force generates tension in the callus that connects the bone segments and this in turn stimulates bone formation. Distraction force also creates tension in the soft tissues including blood vessels, ligaments, cartilage, muscles, nerves which initiate a
sequence of changes termed as distraction histogenesis. Prime advantage of DO is slow stretching of soft tissues and bone formation allowing greater advancement than standard techniques. Advancing the maxilla and mandible in this manner will allow the enhancement of the upper airway volume gradually. This is similar to the titration accomplished when using adjustable mandibular advancement devices.

A study was carried out at our centre to assess the effectiveness of DO in OSA and non apneic snoring. The objectives were to compare results objectively and subjectively before and after the procedure.

**Patients and Methods**

All patients with severe maxillo-mandibular discrepancy with OSA or non apneic snoring undergoing DO for bilateral corpus lengthening, mid face advancement, transverse maxillary expansion during the last 39 months were considered as prospective study candidates (n=10). Clinical examination, PA and lateral cephalometric studies were performed according to established protocol for all the patients. Polysomnography (PSG) was done in cases with severe mandibular hypoplasia and respiratory symptoms. Three dimensional CT scan was also done in cases operated for TMJ ankylosis and syndromic cases. Comprehensive treatment plan based on the data base addressing facial aesthetics, occlusion and respiratory symptoms were formulated.

The treatment protocol followed in 8 cases included orthodontic preparation, distraction surgery, orthodontic therapy during distraction and consolidation, and post consolidation orthodontic treatment for settling the occlusion. In two young children with Treacher-Collins syndrome, DO was carried out without orthodontic intervention. The distraction surgery in all the cases was performed under GA.

The facts taken into consideration for building up the data base, both pre and post therapy, included age, sex, diagnosis, type of distraction and distractor device used, degree of lengthening, improvement on Apnea Hypopnea Index (AHI), Pre and Post treatment Posterior airway space (PAS) - Retro palatal space (RPS) are summarized in table 1.

<table>
<thead>
<tr>
<th>Case NO</th>
<th>AGE</th>
<th>SEX</th>
<th>DIAGNOSIS</th>
<th>Type of distraction osteogenesis</th>
<th>Type of distractor used</th>
<th>Clinical outcome</th>
<th>Maximal mouth opening (mm)</th>
<th>Pre and treatment PAS RPS (mm)</th>
<th>Retropalatal airway space changes</th>
<th>AHInpred Post/Post Resp Symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>15</td>
<td>F</td>
<td>OSA</td>
<td>Corpus lengthening</td>
<td>IO-medical body distractor</td>
<td>Good improvement in profile; chin projection</td>
<td>60/90</td>
<td>0/12</td>
<td>57</td>
<td>58.6/14.1</td>
</tr>
<tr>
<td>2</td>
<td>23</td>
<td>F</td>
<td>OSA</td>
<td>Corpus lengthening</td>
<td>IO-medical body distractor</td>
<td>Satisfactory improvement in profile; chin projection with upper airway volume</td>
<td>52/70</td>
<td>0/11</td>
<td>6/8</td>
<td>48.2/0.6</td>
</tr>
<tr>
<td>3</td>
<td>22</td>
<td>M</td>
<td>OSA</td>
<td>Corpus lengthening</td>
<td>IO-medical body distractor</td>
<td>Excellent improvement in profile; chin projection and occlusion</td>
<td>49/70</td>
<td>4/11</td>
<td>6/8</td>
<td>60.8/4</td>
</tr>
<tr>
<td>4</td>
<td>03</td>
<td>M</td>
<td>Treacher-Collins syndrome</td>
<td>Zurich paediatric distractor</td>
<td>Excellent improvement in profile; chin projection</td>
<td>33/48</td>
<td>3/10</td>
<td>4/6</td>
<td></td>
<td>Dreamtanned</td>
</tr>
<tr>
<td>5</td>
<td>18</td>
<td>M</td>
<td>Class II Div 2 infraorbital with intractable snoring</td>
<td>Corpus lengthening</td>
<td>IO-medical body distractor</td>
<td>Excellent improvement in profile; chin projection</td>
<td>67/74</td>
<td>0/11</td>
<td>7/8</td>
<td></td>
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<tr>
<td>6</td>
<td>14</td>
<td>F</td>
<td>Maxillary hypoplasia</td>
<td>Air face advancement</td>
<td>Rigid external distractor</td>
<td>Excellent improvement in profile; Good final occlusion</td>
<td>82/58</td>
<td>1/11</td>
<td>7/7</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>22</td>
<td>M</td>
<td>OSA</td>
<td>Mandibular lengthening</td>
<td>IO-medical body distractor</td>
<td>Excellent improvement in profile; Good final occlusion</td>
<td>55/69</td>
<td>5/12</td>
<td>6/8</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>18</td>
<td>M</td>
<td>Severe maxillary mandibular deficiency with snoring</td>
<td>Surgically assisted surgery</td>
<td>Hyrax appliance</td>
<td>Excellent improvement in facial aesthetics, occlusion</td>
<td>22/54</td>
<td>11/11</td>
<td>6/8</td>
<td></td>
</tr>
<tr>
<td>9</td>
<td>16</td>
<td>F</td>
<td>Malocclusion (maxillary deficiency) snoring and breathing</td>
<td>Corpus lengthening</td>
<td>IO-medical body distractor</td>
<td>Excellent improvement in profile; chin projection, occlusion</td>
<td>72/79</td>
<td>8/12</td>
<td>7/8</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>6</td>
<td>M</td>
<td>Treacher-Collins syndrome</td>
<td>Zurich paediatric distractor</td>
<td>Excellent improvement in profile; chin projection, occlusion</td>
<td>43/97</td>
<td>5/9</td>
<td>5/7</td>
<td></td>
<td>Excellent improvement objectively subjective</td>
</tr>
</tbody>
</table>
Pre and post cephalometric data with respect to PAS, RPS, and mandibular length was compiled as per established protocol and was considered for this study [5].

**Results**

The patient data and results are summarized in Table 1. All the cases in our series showed improvement in facial aesthetics, occlusion and respiratory symptoms. A mean increase of 5.14 mm and 1.85 mm with respect to PAS and RPS respectively was observed in cases with bilateral corpus lengthening. In one case of mid face advancement the RPS increased by 5 mm and snoring completely ceased. In the case with transverse maxillary deficiency, surgically assisted rapid maxillary expansion resulted in 12 mm increase in inter molar distance, severe crowding could be relieved and patient reported complete cessation of snoring. One case of severe OSA due to severe mandibular hypoplasia and was CPAP dependent was weaned of CPAP following bilateral corpus lengthening with DO [6]. A three year old child tracheostomy dependent since birth was decannulated one month following DO (Fig 1, 2, 3).

**Discussion**

One of the most important indications for maxillo-mandibular distraction osteogenesis is severe OSA as a result of mandibular hypoplasia or maxillary deficiency. This is because DO addresses the main obstructive sites i.e., PAS and RPS increase in the 3-dimensions. Rachmiel and his co-workers analyzed bilateral mandibular distraction with compromised airway by 3D CT scan and concluded that DO of hypoplastic mandible, volume of hypoplastic mandible and upper airway increases eliminating the symptoms of OSA and preventing tracheostomy [7]. Although traditional orthognathic surgery are effective in treating mild and moderate OSA, the results are disappointing in severe OSA and when BMI > 28 and in syndromic cases. This is because a traditional osteotomy has a physiological limit of approximately 10 mm of advancement which may be insufficient to open the upper airway for effective relief of obstruction in the obese patient or in those with severe disease. In the present series, we achieved a mean...
advancement of 14 mm and in one of the severe OSA case we achieved 21 mm (Figs 4,5,6). This is indeed the reason for significant improvement in respiratory symptoms. We have reported statistically highly significant (p<0.001) increase in total anterior face height and corpus length and excellent soft tissue response to large mandibular advancement with intra oral bilateral mandibular distraction osteogenesis [8] [9]. The cases considered in this study also consisted of cases of mandibular deficiency without respiratory symptoms [8]. However we are of the view that all mandibular deficiency cases are potential OSA and advancing the mandible vide DO would not only improve masticatory function and facial aesthetics but also prevent OSA in the future.

In our series we followed the same protocol as recommended in the literature [10] [11] [12]. But we often used functional appliances like mandibular advancement device prior to and after corpus lengthening [8]. In cases of non apneic snoring associated with mandibular deficiency, use of mandibular advancement appliance during pre distraction orthodontic stage for neuromuscular adaptation showed reduced or ceased snoring. This was an indication to the treating team that surgically advancing the mandible would eliminate snoring. Pre-distraction orthodontics involved placing the dentition ideal to the skeletal bases using fixed orthodontic appliances and adjuncts. The highlights of the procedure involves performing osteotomy and placement of distracters (Figs 7,8). In case of mandibular corpus lengthening, osteotomy cuts were placed distal to the existing last molar. Lefort I level osteotomy was done for maxillary advancement and the same surgery coupled with mid palatal split was done for surgically assisted rapid maxillary expansion (SARME). Latency period of 5 days was followed; rate of DO was 1 mm/day and rhythm of DO was 0.5 mm/12 hr/day. Interarch elastics were used during DO to mould the regenerate [13]. Respiratory symptoms gradually improved as the distraction progressed and all the cases showed gross improvement in the respiratory symptoms following completion of distraction. In a recently published article, Baur and Helman are of the opinion that the amount of DO can be individualized for each patient by performing PSG after 15-18 mm of advancement. If the PSG demonstrates a normal AHI, DO can be stopped to allow for consolidation. If AHI is elevated DO continues for 25 mm [14].

Fig 4: Pre treatment Lateral Cephalogram of 21 year old male with severe mandibular hypoplasia secondary to growth disturbance following TMJ ankylosis and severe OSA. Note the gross reduction in PAS.

Fig 5: Ortho pantomo gram of the same patient at the end of distraction.

Fig 6: Post treatment lateral cephalogram showing gross improvement in facial profile, mandibular size and PAS.
Maxillo-mandibular DO anteriorly positions the maxillary and mandibular framework and their attending muscular attachments. In addition to addressing the obstructive sites namely, retroglossal and retro palatal regions, provides additional tension for genioglossus muscle and increases the available room in the floor of the mouth for the tongue and also improves tongue posture. Literature recommends maxillo-mandibular advancement as the most effective treatment of OSA other than tracheostomy and the success rate approach 100% [15] [16]. This procedure has been reported to have high degree of effectiveness in patients who are not successful with first phase surgery and who undergo this procedure even though they do not have maxillo-mandibular deficiency [17].

Studies with mandibular advancement devices has shown that there is increase in the size of pharyngeal airway at both post palatal and post lingual airway dimensions although greater increase is at the former [18]. We have observed the same in corpus lengthening cases.

Oral appliances have been found to be the most acceptable for non apneic snoring, mild to moderate OSA [19] [20]. But in the present series non apneic snoring was associated with severe maxillo-mandibular discrepancy and the patients had sought consultation primarily for the latter. In addition to gross improvement in facial profile and dental aesthetics, snoring completely ceased in these cases (Table1) [8] [9].

Breathing difficulties during sleep may occur or exacerbate following repair particularly pharyngoplasty and palate repair in cleft palate children [21]. In the present series, one case with severe maxillary deficiency secondary to operated cleft lip-palate and associated snoring was treated comprehensively with orthodontics, alveolar grafting and mid face advancement using rigid external distracter. Her RPS was found grossly inadequate (1mm) which increased to 6 mm following mid face distraction and snoring completely ceased and there was no deterioration in speech either (Figs 9 & 10). Recently, the Pillar procedure has been successfully used for snoring and OSA. In this procedure polyester inserts were implanted into patient’s soft palate to cause stiffening effect and thereby allow air to flow more freely through the patient’s airway [22]. This procedure is likely to gain more acceptances in non cleft cases in the future.

Cistulli P has reported that OSA patients who have narrow maxillae and under gone palatal expansion either...
with orthodontic appliance or by surgery show significant improvement in the number of respiratory events that occur following expansion [23]. We have observed the same with complete cessation of snoring in one of our case who under went surgically assisted rapid maxillary expansion for severe constriction of maxilla and intractable snoring (Fig 11,12). In this case the PAS and RPS were within normal limits. There is a reduction in nasal resistance and expansion at anterior nares following rapid maxillary expansion, therefore improvement in breathing [24]. A recent study to compare the effects of rapid maxillary expansion and surgically assisted rapid maxillary expansion on nasal volume using acoustic rhinometric methods concluded significant increase in nasal volume and decrease nasal resistance in two groups [25].

The application of a continuous stretching force on the two bone segments through the use of a device triggers the conditions for growth. The undifferentiated cells in the bone marrow evolve into osteoblasts and begin formation of interlaced bone tissue orientated into parallel lines. Muscle and soft tissue mass increase via a process referred to as distraction histiogenesis. Clinically, this offers a distinct advantage as several craniofacial anomalies have soft tissue hypoplasia, in addition to deficient bony structures. This is especially effective in the stretch of the supra hyoid group of musculature along with histogenesis in and around the tonsillar fossa with resultant effect in the palatoglossus and palatopharyngeous musculature resulting in an increase in the posterior pharyngeal airway space facilitating smooth and unlaboured breathing activity. Neurovascular elements contained within distracted bony segments also are stimulated to regenerate. In all our cases we found healthy new bone in the distraction zone both radiographically and clinically during the removal of the distractors. Osteocyte viability is essential to provide an adequate source of osteoblastic activity at the distraction site, hence careful surgical technique to minimize thermal or mechanical bone injury must be ensured. Similarly, an adequate blood supply to the distraction site & an intact periosteum and endostem are critical to osteogenesis. In our surgical protocol, the bone cuts were restricted to osteotomy aimed at preserving the endosteal tissues thus facilitating an optimal bone healing [26].

OSA is common in a number of abnormalities associated with various congenital syndromes that involve mandibular hypoplasia (e.g. Pierre –Robin, Treacher Collin and Goldenhars syndrome) [27][28]. Steinbacher and coworkers recently studied mandibular advancement by DO for tracheostomy dependent children with severe micrognathia. The results of this preliminary study indicate that mandibular advancement by DO is a
potentially viable treatment option for tracheostomy dependent children with upper airway obstruction secondary to micrognathia. We accomplished the same in one case (Table 1).

Distraction due to its versatility has established itself as a procedure of choice in a multitude of specific conditions. Cases treated with this technique show better stability with good quality of bone with long-term stability of the desired results.

**Conclusion**

DO for treatment of non apneic snoring and OSA caused by mandibular or maxillary deficiency is dependent on many factors including age of the patient. Unlike orthognathic surgery, DO can be applied in all age groups. The results were found to be stable and the respiratory symptoms in all the cases improved. It addresses the obstruction sites namely the retroglossal and retro palatal regions comprehensively and increases upper airway dimensions providing a 100% cure. The technique is indeed valuable and gives us the ability to both prevent and correct the development of sleep disordered breathing.

**References**

22. Friedman M. Palatal stiffening as a CPAP alternative. Sleep review; Journal for sleep specialists. Jun 2007. (sleepeditor@ascendmedia.com).


