

# Management of Severe Mandibular Retrognathia in the Adult Patient Using Distraction Osteogenesis

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Distraction osteogenesis is a powerful technique for creating new bone during significant lengthening of the mandible without the need for bone grafting and associated donor site morbidity. The majority of clinical applications of mandibular lengthening with distraction osteogenesis techniques reported in the literature are in growing patients.<sup>1-8</sup> For the purposes of this article, an adult patient is defined as a patient who has completed skeletal growth. This includes female patients from the approximate age of 15 years and older and male patients from the approximate age of 17 years.<sup>9</sup>

Distraction osteogenesis techniques were initially applied to adult patients predominantly with tumor-related segmental defects of the mandible. Reconstruction of these segmental defects were undertaken using transport distraction osteogenesis techniques with good success.<sup>10,11</sup> However, distraction osteogenesis applications continue to expand and adult patients with severe mandibular retrognathia have undergone mandibular lengthening with distraction osteogenesis.<sup>4,12,13</sup>

## Anatomic Considerations With Severe Mandibular Retrognathia

Severe mandibular retrognathia can be classified as congenital or acquired. Congenital abnormalities that are associated with severe mandibular retrognathia or micrognathia include craniofacial syndromes such as hemifacial microsomia, Pierre-Robin syndrome, Treacher-Collins syndrome, and Nager syndrome. Adult patients with craniofacial syndromes may have

undergone previous surgery at an earlier age, but unfavorable postsurgical growth or skeletal relapse may have occurred. These patients may have undergone previous autologous bone grafting and reconstruction with costochondral or iliac bone grafts in efforts to treat their severe growth deficiencies.<sup>14</sup> This often results in significant residual deformity in adulthood, with unusual bony anatomy. Traditional surgical approaches in these previously reconstructed patients are difficult at best. Distraction osteogenesis plays a significant role in the treatment of these adult patients who have undergone previous surgery.

Severe mandibular retrognathia also can develop following maxillofacial trauma and mandibular fractures, which may have occurred in an adult or as a child. Condylar fractures occurring at an early age can result in subsequent bony and/or fibrous temporomandibular joint ankylosis and/or deficient mandibular growth. Temporomandibular joint ankylosis in a growing child would typically be treated surgically in an effort to maintain jaw function and improve growth. These patients may present in adulthood with severe residual mandibular retrognathia requiring further surgical intervention. Frequently, rudimentary bony anatomy is encountered, which makes traditional osteotomies difficult to perform, and bone grafting may be required. Immigration patterns throughout the world are changing dramatically, and adult patients who have emigrated from a country where surgical treatment for severe mandibular retrognathia in a growing child was not available may present for treatment in North America.

In adults with severe mandibular retrognathia, compromise or camouflage treatment is frequently used to reduce the distance of mandibular advancement. Typically, extraction of maxillary first premolars occurs, with orthodontic retraction of the maxillary incisors. This may adversely effect the nasolabial angle and upper lip support and ultimately limit the amount of mandibular advancement and the degree of profile improvement. Even with advancement genioplasty, these patients may still appear retrognathic at the completion of treatment.

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Patients who previously underwent orthognathic surgery with complications such as skeletal relapse can present with challenging bony anatomy. This may be due to a previous unfavorable bilateral sagittal split ramus osteotomy (BSSO), infection, or possible idiopathic condylar resorption. Secondary osteotomies are difficult to perform and may require external approaches and bone grafting. In such cases, distraction osteogenesis for mandibular lengthening offers a simple solution without the need for bone grafting or vascular compromise.

Adult patients with complications from previous mandibular tumor resection and reconstruction can also present with acquired severe mandibular retrognathia. Bone graft infection, loss of the graft, and a residual mandibular continuity defect with retrognathia can occur. Radiation therapy can be a contributing factor to a hypovascular scarred tissue bed, which presents further challenges in restoration of mandibular form and function.

Severe mandibular retrognathia also can be a contributing factor in obstructive sleep apnea. The classic picture of daytime hypersomnolence, disrupted sleep patterns, and subsequent cardiovascular and neurologic sequelae can be significant. Surgical advancement of the mandible and/or maxilla in adult patients with obstructive sleep apnea has resulted in significant improvements in their Respiratory Disturbance Index (RDI), often eliminating the need for nasal continuous positive airway pressure.<sup>15,16</sup> The limiting factor with traditional surgical mandibular advancement is the distance of advancement and the potential for relapse. Distraction osteogenesis provides an opportunity to provide greater lengthening of the mandible, with potentially greater stability and less relapse compared to conventional surgery.<sup>17</sup> These techniques can be applied to the mandible as well as the maxilla to provide significant resolution or correction of obstructive sleep apnea. The gradual lengthening of the mandible can be titrated by polysomnography to the desired amount for effective change.

Many adult patients with severe mandibular retrognathia present with dentoalveolar crowding and narrow, constricted mandibular arches. Traditionally, this has been treated by dental extractions for arch alignment and coordination, in preparation for surgical movement of the mandible. This may cause occlusal difficulties, particularly if a transverse skeletal deficiency of the mandible is present. Buccal-lingual transverse discrepancies in occlusion may then be present after mandibular advancement. Mandibular widening by distraction osteogenesis following a midline symphyseal osteotomy creates increased width, arch length, and available space for correction of dentoalveolar crowding.<sup>18</sup> Simultaneous mandibular widening and advancement can be undertaken for

correction of the 3-dimensional occlusal and skeletal problems associated with the severely retrognathic mandible.

### **Biologic and Technical Considerations in Mandibular Lengthening by Distraction Osteogenesis**

Integral to the overall success of mandibular lengthening with distraction osteogenesis in adult patients is an understanding of the biologic basis of the technique, with proper preoperative planning and vector selection. Distraction osteogenesis involves an osteotomy, a latency period, a distraction device activation period, a bony consolidation period, and a bony remodeling period.<sup>19-21</sup> The distraction osteogenesis intraoral devices are contoured and the osteotomies are partially completed. The distraction osteogenesis devices are then stabilized with bicortical or monocortical screws, the osteotomies are completed, device activation is undertaken to ensure movement of the bony segments, and then the device is backed down to the zero position.

Typically, a linear osteotomy is created through the mandible with burs or saws, except in the location of the inferior alveolar neurovascular bundle. The osteotomy is completed with osteotomes, creating a fracture. Corticotomy and other osteotomies have also been used in mandibular distraction osteogenesis.<sup>22,23</sup>

The latency period allows resolution of inflammation secondary to the osteotomy and surgical placement of the device. It also allows initial organization of the hematoma and induction of pleuripotential mesenchymal cells and endosseous and periosteal cells into fibroblasts and osteoblasts. During this time, type 1 collagen is laid down and osteoid production occurs. The latency period ranges from 0 to 10 days, although the most common latency period is 5 days, and is applicable in adults.

The distraction process, or callus manipulation, occurs at a rate ranging from 0.5 mm to 2 mm a day. The rate will depend on the age of the patient and the type of osteotomy. The gold standard for clinical distraction osteogenesis is 1 mm a day, divided into 2 or 4 activations per day. The distance of distraction is determined by the amount of skeletal and occlusal change desired. Transoral activation arms are typically removed under local anesthetic and/or sedation at the completion of the distraction.

The consolidation period in adults should be a minimum of 3 months and can extend up to 6 months as needed. Consolidation time is related to the magnitude of the distraction distance and the age of the patient. Adequate stability of the bony segments is important during distraction and the consolidation

period to allow an optimum regenerate to occur. Timing of distraction device removal can be related to the quantity of distraction regenerate, as documented by radiographs, ultrasound, or a computed tomography (CT) scan. Distraction device removal can be carried out on an outpatient basis under sedation or general anesthesia, particularly if additional ancillary orthognathic or surgical procedures are being performed. Further bony remodeling of the distraction regenerate under function will occur up to 1 year or longer after the period of distraction. The regenerate bone will ultimately be virtually indistinguishable from the adjacent bone and will withstand normal physiologic loading.

Preoperative 3-dimensional vector selection is important to achieve a predictable aesthetic and functional outcome following mandibular lengthening with distraction osteogenesis in adults. Diagnostic aids in selecting the distraction vector include clinical evaluation; panoramic, lateral, and posteroanterior cephalometric radiographs; and a CT scan with 3-dimensional reconstruction. Stereolithographic models are also useful for selection of osteotomy location, preoperative distraction device contouring, and vector selection when unusual anatomy is present. Model surgery using occlusal casts mounted on a semiadjustable articulator, in conjunction with cephalometric analysis and lateral and posteroanterior cephalometric prediction tracings, may further help to define the vector and distance of distraction. Video imaging linked to cephalometric prediction tracings may allow complete visualization of the treatment goals. In bilateral mandibular distraction osteogenesis, the vector is typically parallel to the occlusal plane and close to the sagittal plane. Variations will occur in vector selection in asymmetric mandibles. Multidirectional distraction devices provide the opportunity to adjust the vector of distraction during the distraction process, as needed.

### **Advantages of Distraction Osteogenesis Over Traditional Surgical Techniques**

The BSSO is the workhorse osteotomy of traditional mandibular advancement surgery. Advancements greater than 10 mm require a long sagittal split, which can be technically difficult to achieve. Alignment of the proximal and distal segments also can be difficult due to lateral flaring of the proximal segment. This may result in torquing of the mandibular condyle laterally,<sup>24,25</sup> resulting in spatial changes with respect to the articular disc. This can contribute to greater internal derangement, particularly if rigid fixation is used. Application of rigid internal fixation becomes more complex with larger advancements. Bicortical

screws and/or bone plates may not provide adequate osseous stability or may be difficult to apply. Various techniques have been described to overcome these difficulties, including bone grafting, intentional lingual cortex fracture, and lengthy periods of maxillo-mandibular fixation.

Many severe mandibular retrognathic mandibles have some component of asymmetry. Asymmetric advancement of the mandible increases proximal segment flare, particularly with large advancements, which may intensify the temporomandibular joint changes.

Lengthening of the severely retrognathic mandible with distraction osteogenesis overcomes many of the problems of the BSSO. Frequently, a linear osteotomy is used and vector selection and lengthening are accomplished by the application of an intraoral distraction device. A linear or beveled osteotomy avoids flaring of the proximal segment compared with the BSSO. No bone grafting is required, and significant lengthening of the mandible, up to 10 to 20 mm or greater, can be obtained. New bone is created during distraction through a cascade of cellular and molecular events. This membranous bone, parallel to the direction of distraction, matures into bone similar to the adjacent bone (cortical bone surrounded by intramedullary cancellous bone) during the consolidation period.<sup>21,26</sup>

When a parasagittal vector is used for significant mandibular lengthening, there is less torque on the temporomandibular joint. Additionally, the temporomandibular joints are gradually loaded during the activation of 1 to 2 mm a day. Proximal segment rotation in an anterosuperior direction after a BSSO can result in changes in masticatory muscle orientation and positional changes in the mandibular condyle. In mandibular distraction osteogenesis, the devices are contoured and placed prior to osteotomy and the proximal and distal segments are maintained in their original position postosteotomy. This has a decreased impact on the temporomandibular joint compared with traditional osteotomies.

Mandibular advancements greater than 10 mm are difficult to achieve with the BSSO, because of resistance of the soft tissues and problems with maintaining adequate bony contact for osseous healing. Stretching of the periosteum, muscle, and fascia, a great distance requires extensive stripping of these tissues (possible suprahyoid myotomy) off of the bone, which may decrease vascularity to the segments.

Mandibular lengthening with distraction osteogenesis offers many advantages over conventional orthognathic surgical techniques. Slow lengthening of the osseous segments results in soft tissue histogenesis, a combination of gradual stretching of the soft tissues

and cellular proliferation. An increased number of myocytes, along with adaptations in sarcomere length, have been reported in muscle in response to distraction osteogenesis.<sup>27,28</sup> These favorable adaptive changes maintain the soft tissue attachments to the bone and hence there is a greater blood supply to the distraction site and mandible than with conventional osteotomies. They also allow greater mandibular lengthening, with minimal or no relapse.

### Distraction Osteogenesis and Relapse

An average of 2 mm,<sup>29</sup> and up to 30% of sagittal relapse, have been reported following mandibular advancement using BSSO and wire fixation.<sup>28</sup> The greater the acute lengthening, the greater is the propensity toward relapse. Advancements of greater than 10mm are more prone to skeletal relapse, which can occur at the osteotomy site or at the mandibular condyle.<sup>30</sup>

Rare occurrences of relapse have been reported with mandibular lengthening by distraction osteogenesis. This is attributable to use of external devices and allowing inadequate time for consolidation of the distraction regenerate, or to device loosening.<sup>1</sup> Appropriate distraction device stability and an adequate time of bony consolidation are important for a successful distraction regenerate to develop.

Relapse after 10 to 20 mm mandibular lengthening with distraction osteogenesis is minimal or nonexistent if an adequate distraction protocol and bony consolidation periods are used.<sup>31,32</sup> A consolidation period of at least 3 months is indicated for significant mandibular lengthening, with the exact time based on radiographic visualization of cortical bone in the distraction regenerate. Buried intraoral distraction devices offer greater patient acceptance and more adequate time for bony consolidation than external distraction devices.

The advantages of distraction osteogenesis for large mandibular advancements become clear when one compares a 20-mm mandibular advancement using distraction osteogenesis with an inverted L osteotomy in which an interpositional bone graft (donor site) is needed, and there is difficulty in stabilizing the proximal and distal segments and bone graft adequately. Consider the acute tension and stretching of soft tissue with this magnitude of advancement, which would frequently require an external approach. Will there be condylar resorption, bone graft remodeling, and skeletal relapse over time? If infection were to occur, there could be significant or total loss of the bone graft and disastrous consequences.

Distraction osteogenesis at 1 mm/d for 20 mm will achieve superior results, with favorable soft tissue adaptation, stable distraction bone stock, and less

acute loading of the temporomandibular joints. There is no donor site morbidity, and an intraoral approach would avoid the significant skin scars from external approaches. Although infections have occasionally been reported with mandibular distraction osteogenesis, it appears to have a minimal effect on the distraction regenerate.<sup>33</sup> This is likely due to the angiogenesis that occurs during the distraction process. A new blood supply, along with new, viable bone, is created during the distraction process, which appears to be relatively infection-resistant.

### Temporomandibular Joint Considerations

Distraction osteogenesis for mandibular lengthening is indicated for adult patients with internal derangements, presurgical or postsurgical condylar resorption, and degenerative joint disease. Distraction osteogenesis should be considered in these patients even if only moderate lengthening of the mandible is anticipated.

It is documented that acute loading of the temporomandibular joint from conventional mandibular osteotomies and lengthening can exacerbate temporomandibular joint problems.<sup>34,35</sup> The amount of stripping of periosteum and musculature off the proximal segment may decrease its vascularity and result in further condylar remodeling and/or resorption.

Patients with idiopathic condylar resorption after mandibular osteotomy are frequently treated with camouflage maxillary surgery in an attempt to mask the mandibular retrognathism. Distraction osteogenesis allows treatment of significant mandibular retrognathism secondary to idiopathic condylar resorption. There is need for less surgical access to create a linear body-ramus osteotomy and apply intraoral distraction devices, with potentially less disturbance to soft tissue vascularity, than with the usual mandibular osteotomy. Distraction osteogenesis causes gradual loading of the problematic temporomandibular joints rather than the acute joint loading associated with acute mandibular advancement.

Adult patients with juvenile rheumatoid arthritis, may have had previous surgery, or present de novo. The unusual condylar resorption, short ramus height, and severe micrognathia present difficult problems for performing typical mandibular advancement surgery. Surgical modalities that are frequently used include an inverted L or C osteotomy and simultaneous bone grafting from an external approach. Alternatively, costochondral grafts or replacement with alloplastic temporomandibular joints have been reported.<sup>36</sup> Distraction osteogenesis can be used to create increased ramus height and mandibular the body length without the need for bone grafting. Greater

distances can be achieved in lengthening of the mandible, with secondary favorable soft tissue adaptation.

There is minimal evidence of significant remodeling of the temporomandibular joint secondary to mandibular lengthening with distraction osteogenesis documented in animal studies.<sup>37</sup> Use of bidirectional and tridirectional distraction devices offers improved vector control during mandibular distraction, and appears to cause less remodeling of the temporomandibular joints than unidirectional devices.<sup>38</sup>

### Neurosensory Changes Associated With Mandibular Distraction Osteogenesis

Various neurosensory outcomes have been reported in humans and animals in response to mandibular distraction osteogenesis. Careful surgical technique during the osteotomy and distraction device placement are important to avoid injury to the inferior alveolar nerve. With the development of buried intraoral devices, permanent injury to the facial nerve has virtually been eliminated.

Clinical reports on sensory changes in the inferior alveolar nerve vary from none<sup>39,40</sup> to neurosensory deficits ranging from 25% to 50% of patients undergoing mandibular distraction osteogenesis.<sup>41,42</sup> Action potentials measured during 10 mm of distraction osteogenesis of the mandible in dogs revealed minimal deleterious effects on the inferior alveolar nerve.<sup>43</sup> There were no significant differences in jaw jerk voltage of the mental nerve between control and unilateral mandibular distraction osteogenesis sites. The authors concluded that these were only mild inferior alveolar nerve injuries secondary to slow traction.<sup>44</sup> Bilateral mandibular distraction osteogenesis at a rate of 1 mm/d in goats also appeared to be tolerable and safe for the inferior alveolar nerve. However, distraction rates of 2 mm/d may result in significant nerve degeneration.<sup>45</sup>

During the BSSO for mandibular advancement, damage to the inferior alveolar nerve can occur during the osteotomies, while splitting the mandible or from medial retraction. The inferior alveolar nerve may be manipulated significantly, particularly if it is partially embedded in the proximal segment and requires decortication and freeing. Application of rigid fixation with cortical screws and/or miniplates may directly injure the inferior alveolar or can create nerve compression injury.<sup>46</sup>

Various authors have reported statistically significant neurosensory injury from the BSSO, depending on the age of the patient, magnitude of advancement of the mandible, and degree of nerve manipulation.<sup>47</sup> Other authors have reported permanent changes in inferior alveolar nerve sensation ranging from 15% to

87% at 1 year after osteotomy.<sup>48,49</sup> Additionally, increased inferior alveolar nerve damage from the screws used for rigid internal fixation, as well as permanently altered lingual nerve sensation has been reported at 1 year after sagittal split osteotomy.<sup>50</sup>

It appears, after a review of the literature, that temporary and permanent neurosensory changes can occur both with mandibular distraction osteogenesis and with conventional osteotomies for lengthening of the mandible. There is a wide range of reported clinical incidence of permanent altered sensation, which may be related to the surgical technique. However, it would appear that distraction osteogenesis techniques for significant mandibular lengthening of 10 mm to 20 mm may result in less neurosensory changes than the conventional BSSO or inverted L osteotomy.

### Summary

Mandibular lengthening with distraction osteogenesis in adult patients with severe mandibular retrognathia is a significant alternative to traditional surgical techniques. Multidirectional buried intraoral distraction devices have overcome some of the obstacles of earlier external distraction devices and produce good vector control, occlusion, and aesthetic results. The occlusal outcomes are aided by concomitant orthodontic therapy. Distraction osteogenesis techniques do require additional surgical training, a thorough understanding of the biologic process, and careful preoperative planning. It is technique sensitive and the surgical skills and experience of the surgeon reduce the complication rate and optimize treatment outcomes. Refined distraction osteogenesis techniques for the treatment of severe mandibular retrognathia in adults provides the opportunity for more favorable treatment outcomes compared to traditional surgical procedures, with less morbidity and minimal complications.

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