Management of obstructive sleep apnea syndrome secondary to temporomandibular joint ankylosis by mandibular elongation using distraction osteogenesis

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Abstract

Obstructive sleep apnea syndrome (OSAS) is associated with repetitive nocturnal upper airway obstruction leading to daytime sleepiness, cardiovascular derangements, and can be a debilitating, even life-threatening condition. The most favorable treatment for patients with OSAS is multidisciplinary care by a team that represents various dental and medical disciplines. Prescribed therapies might include weight loss, behavior modification, oral appliances, soft tissue surgery, skeletal surgery, or some combination of approaches. Osteogenesis by mandibular distraction has proved effective in children in the treatment of obstructive apnea syndrome associated with congenital malformations. In the adult, the possibility of using distraction osteogenesis in the management of OSAS remains to be defined. We report a case of an adult patient treated for OSAS secondary to temporomandibular joint ankylosis by mandibular distraction followed by interpositional arthroplasty.

Keywords: Distraction osteogenesis, obstructive sleep apnea syndrome, temporomandibular joint ankylosis

Introduction

Obstructive sleep apnea syndrome (OSAS) is associated with repetitive nocturnal upper airway obstruction leading to daytime sleepiness and cardiovascular derangements. The surgical management of OSAS, including the use of laser-assisted uvulopalatoplasty (LAUP), uvulopalatopharyngoplasty (UPPP), genioglossus advancement, hyoid suspension, tongue base reduction, and maxillomandibular advancement (MMA) are primarily designed to enlarge airway dimensions and decrease airway collapsibility. Of the available surgical options, MMA has been shown to be highly effective in the improvement of upper airway dimensions. Distraction osteogenesis (DO) in the maxillofacial region was first investigated by Snyder et al. in the canine mandible. Karp et al. showed that bone formation during DO in the maxillofacial region is similar to that in long bones, which is predominately by intramembranous ossification. Since that time, McCarthy et al. reported on mandibular lengthening in pediatric patients with hemifacial microsomia by DO; this technique has gained increased popularity in oral and maxillofacial surgery, including the management of OSAS. We report a case of an adult patient treated for OSAS secondary to temporomandibular joint (TMJ) ankylosis by mandibular elongation using distraction osteogenesis followed by interpositional arthroplasty.

Case Report

A 23-year-old male patient referred to Department of Oral & Maxillofacial Surgery with a chief complaint of inability to open the mouth and facial asymmetry which on further evaluation led to the history of snoring, excessive daytime sleepiness, and tongue falling back during sleep. No complications had been reported at birth, and there was no subsequent history of trauma to the facial skeleton. Both the patient and his parents were aware of this, but delayed evaluation because of economic circumstances. The initial clinical examination revealed an obviously hypoplastic mandible with chin receded on the affected side and a class II dental relationship. Radiographic investigations included orthopantomogram, lateral skull, and CT scan. These images confirmed bony ankylosis of the left TMJ with bilateral elongation of the coronoid. He was further evaluated with overnight polysomnography and was diagnosed with OSAS secondary to TMJ ankylosis.

The following staged surgical treatment was planned which
included mandibular elongation using DO followed by release of TMJ ankylosis through interpositional arthroplasty. General anesthesia was secured through tracheostomy due to difficult intubation conditions. Using a vestibular approach, vertical osteotomy was then made posterior to the second molars. External pin distractors were placed parallel to the occlusal plane, and the pins were inserted percutaneously [Figures 6a and b. The distractors were fixed to the bone, tested by activating them 3 mm, and then returned to a neutral position [Figure 7]. Distraction was begun 3 days postoperatively at 1 mm/day (one 0.5-mm turn twice daily). Although initially painful, it was well tolerated by the patient. Distraction was continued to 12 mm, and marked clinical improvement was evident at this stage. The patient was followed-up for 3 months without any untoward incident. Follow-up radiographs obtained at this time showed ossification of the distraction callus. After 3 months, the distraction appliances were removed under local anesthesia and second surgery for release of TMJ ankylosis was planned.

General anesthesia with nasotracheal intubation and total muscle relaxation was administered. A preauricular incision with a temporal extension was made [Figure 8]. The temporalis muscle was lifted from the infratemporal fossa toward the anterior at the pericranial level, while the zygomatic root was uncovered. The ankylosed TMJ was palpable and an incision was made directly onto the bone, exposing the ankylosed TMJ. Excision of the fibrous tissue and ankylosic bony mass was carried out using drill and saw. The TMJ was lined with a temporalis muscle and fascia flap rotated over the arch into the joint. The flap was sutured medially, anteriorly, and posteriorly with 4-0 Vicryl. Postoperative pain medications and vigorous postoperative physiotherapy
for the first 2 weeks and once every 2 days for the following 2 weeks. There was a considerable increase in the mouth opening and change in the lateral and frontal profile of the patient [Figures 9-11] and decreased snoring and tongue falling back during sleep. Postoperative orthopantomogram and lateral skull radiographs revealed ossification of the callus and marked improvement in the lateral profile [Figures 12 and 13]. Conventional orthodontic therapy was advised to obtain well-coordinated and well-aligned dental arches. Presently, the patient is undergoing fixed orthodontic treatment and he is under regular follow up.

Discussion

TMJ ankylosis is a very distressing structural condition that denies the victim the benefit of a normal diet and opportunities in careers that require normal speech ability. It also causes severe facial disfigurement that aggravates psychological stress. TMJ ankylosis during early childhood may lead to disturbances in growth, or cause asymmetry
and serious difficulties in eating and breathing during sleep.[8] This ailment is caused by various factors including trauma, systemic, and local inflammatory conditions, as well as neoplasm in the TMJ area, and can only be relieved by a direct surgical procedure.[8] Management of TMJ ankylosis is mainly through surgical intervention. It is necessary to use an interpositional material to prevent TMJ re-ankylosis after arthroplasty, and this particular aspect of the treatment has been the subject of numerous discussions. A variety of interposition materials have been used, including temporalis muscle and fascia, dermis, auricular cartilage, fascia lata, fat, lyodura, silastic, silicone, and various metals. The most commonly used interpositional material is temporalis muscle flap fascia flap.[8]

Improvement of the airway dimensions by skeletal advancement remains the most effective surgical therapy for OSAS.[9-12] Because the extent of airway improvement is correlated with the amount of skeletal advancement, large skeletal movement with rigid skeletal fixation and autogenous bone grafting are usually performed with improved outcome.[13] DO offers several advantages over the conventional techniques by eliminating the need of bone grafting and involving less surgical dissection because the lengthening is the result of natural bone healing in a gap created by a simple osteotomy. The incremental skeletal movement allows accommodation of the soft tissues, thus enabling large skeletal movement that cannot be achieved by conventional techniques. The improved soft tissue accommodation also improves the stability of the new skeletal position. Despite the advantages of DO, several significant disadvantages exist. Although less surgical dissection is necessary for DO, the procedure is highly technique sensitive, especially in achieving the proper alignment of the distraction devices.[14]

Mandibular elongation by progressive distraction is gradual and thus better tolerated by the patient, both functionally and morphologically. The procedure is less time consuming, and placement of the distractors causes no particular problem apart from the care required in achieving the proper vectors of distraction. These vectors must lie in the occlusal plane to prevent a gap being produced during elongation. Also, they should not be convergent, to avoid any strain on the
temporomandibular joints. Mandibular distraction has been used more frequently in the child and, in particular, it has enabled treatment of the malformations responsible for OSAS.\cite{15,16} However, few results are available on the application of this technique in the adult with OSAS. The development of miniaturized intraoral distractors and reduction in their cost should result in more widespread use of the procedure. Moreover, techniques for maxillary distraction are constantly evolving and the surgical techniques are becoming simpler; certain authors consider that mandibular distractors can be placed under local anesthesia.\cite{15} These developments will certainly make surgical management of patients with severe OSAS easier. This case report shows that DO may be applicable in selected adult patients for skeletal advancement in the treatment of OSAS. Further investigations are necessary to determine the potential of this technique.

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