Mandibular distraction osteogenesis for obstructive sleep apnoea secondary to TMJ ankylosis

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Abstract: Temporomandibular joint (TMJ) ankylosis in a paediatric population is uncommon and a challenging problem to treat. From a technical perspective, ankylosis is difficult to treat as the surgeon should consider the potential effects of time and growth on the outcome. The cognitive and emotional development of the patient and the role of the parents are other factors that can affect the management and treatment results in children. Therefore, surgical planning, post-operative management and comprehensive multidisciplinary approach are needed to ensure an ideal outcome. We present a case of a four years old girl presented with mandible hypoplasia in anterior-posterior direction secondary to left TMJ ankylosis. Her condition is further complicated with presence of severe trismus, adenoid hyperplasia grade III and moderate obstructive sleep apnoea. She underwent two stages of surgical intervention which were bilateral mandibular distraction osteogenesis via submandibular approach followed with left condylectomy and gap arthroplasty with interpositional myofascial temporalis flap 4 months later. Mouth opening successfully improved from an initial interincisal opening of 20mm to 28mm post-surgery. Her obstructive sleep apnoea was also resolved. In this case report, we highlight the role of stereolithography model in surgical planning and a multidisciplinary approach for optimum surgical correction of this maxillofacial deformity.

Key words: Distraction osteogenesis, Temporomandibular joint, Obstructive sleep apnoea.

Introduction

Temporomandibular joint (TMJ) ankylosis in a paediatric population is uncommon and a challenging problem to treat. Ankylosis is defined as the immobility and consolidation of a joint due to disease, injury or surgical procedure. TMJ ankylosis can be classified by location (intra-articular or extra-articular), type of tissue involved (eg, bone, fibrous, or fibro-osseous) and extent of fusion (complete or incomplete). The presence of TMJ ankylosis in childhood may cause the development of mandible hypoplasia or mandible asymmetry. This is due to the condyle being the growth center and if severe injury occurs to the condyle, it may result in ankylosis which will cause interference with the development of the bone. Facial asymmetry will increase progressively as maturity is reached. When the mandibular hypoplasia is severe, patients may develop feeding difficulty and respiratory obstruction.

Obstructive Sleep Apnoea (OSA) is a possible outcome in patients with severe mandibular hypoplasia. OSA is a disorder and clinically defined by the occurrence of daytime sleepiness, loud snoring, witnessed breathing interruptions, or awakenings due to gasping or choking in the presence of at least 5 obstructive respiratory events (apneas, hypopneas or respiratory effort related arousals) per hour of sleep. Complications of OSA include failure to thrive, pulmonary hypertension, cor pulmonale, and sudden death.

Distraction osteogenesis is the regeneration of bone between two vascularised bone surfaces that are separated by osteotomy and gradual traction using a mechanical device. The application of distraction osteogenesis to the maxillofacial region was first reported in 1972 by Snyder et al. (1973). The distraction devices can be divided into extraoral and intraoral devices. The extraoral distraction devices are use in management of severe micrognathia patients. It must be noted that patients are susceptible to obvious
facial scarring and increased risk of injury to the facial nerve branches post operatively. Intra-oral devices have been developed to prevent these complications. The advantages of intraoral distractors are as follows: no external scars, less soft tissue trauma, near total concealment of the device and improved superior psychological tolerance. However, in infants and young children, there is not sufficient space subperiosteally to accommodate the whole intraoral distractor.

We present a case of a four years old girl who presented with mandible hypoplasia in anterior-posterior direction complicated by the presence of severe trismus, adenoid hypoplasia grade III and moderate obstructive sleep apnoea secondary to left TMJ ankylosis.

Case Report

A 4 years old girl presented to Oral & Maxillofacial Clinic, University Kebangsaan Malaysia, Medical Centre, Kuala Lumpur with facial asymmetry and limited mouth opening. Her mother noted this problem since she was two years old following attempts for solid food feeding. She suffered from severe Neonatal jaundice within 24 hours of life secondary to ABO incompatibility. She then had infection at the left preauricular region on day two of life and aspiration cytology was performed by the otorhinolaryngology surgeon. She presented with snoring during sleeping since infancy and it progressively worsened since a year ago. There was no history of trauma to the mandible. She is the youngest of three siblings and none of her siblings have a similar deformity.

Upon examination, she presented with mandible hypoplasia in anterior-posterior direction and a severely retruded soft tissue pogonion with deep labiomental fold. There was also severe trismus with only 2mm of vertical mouth opening. She also could not perform any protrusion and lateral excursion movements of the lower jaw. Intraorally, she presented with class II incisor and molar relationship with an overbite of 3mm and an overjet of 10.5mm. The chin midline was deviated 6mm to the left when compared with the facial midline.

CT scan showed a deformed left mandibular condyle with the articular surface flattened and irregular. CT scan also showed loss of normal concavity of the articulating surface of the left mandibular fossa. The left TMJ space was markedly reduced. The right TMJ was normal. It was concluded that she had mandibular hypoplasia in anterior-posterior axis secondary to left TMJ ankylosis.

She was referred to a pediatric respiratory specialist and a Polysomnography study was performed. She was found to have moderate obstructive sleep apnoea (OSA) with AHI 8.9. This resulted in her needing to use the Continuous Positive Airway Pressure (CPAP) machine. ENT assessment later also found her to have a grade III Adenoid hypertrophy.

CT scan was repeated one month prior to surgery and a stereolithographic (STL) model was constructed. The roles of STL model in this case were for bending the distractor foot-plates, planning the distraction vector and fabricating the surgical guide. The STL model was also necessary to assist the osteotomy of the mandible and to optimize the surgical time (Figure 1).

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Figure 1. Intraoral mandibular distractors. (A) Perioperative on Sterolithography model. (B) Intraoperative image shown position of the intraoral mandibular distractor as planned. (C) Sterolithography model shown ostotomies and the distractor foot-plates planning preoperative. (D) Positioning fabricated surgical guide intraoperative.
She was planned to undergo two stages of surgical intervention. In the first stage surgery, bilateral mandibular distraction osteogenesis was performed to resolve her OSA. The mandibular distraction vector was anteriorly to achieve advancement of mandible. Distraction was start post op day three for 3 times a day at a rate of 1mm per day. Distraction on right side was performed for 8 days and left side 13 days. The differential distraction was intentional to achieve symmetry of the chin. She was recovered well postoperatively (Figure 2).

CT scan was repeated after 3 month postoperatively prior to second stage surgery. The aims of the CT scan were to assess the bony consolidation at the distraction sites and to assess proximity of internal carotid artery to the ankylosis site. CT scan revealed good consolidation of bone at the distraction site bilaterally and the left carotid canal is located approximately 6 - 7mm medial to medial margin of the ankylosed joint. Most inferiorly, the cervical internal carotid artery is located 9mm posteromedial to the medial margin of the ankylosed joint.

Four months after the distraction surgery, she underwent the second stage surgery for removal of the mandibular distractor followed with left condylectomy, bilateral coronoidectomy and gap arthroplasty with interpositional myofacial temporalis flap. This aims of this surgery are to treat the trismus and prevent recurrence of ankylosis. Nasal endoscopy and adenoidectomy was performed by the Otorhinolaryngology surgeon in same setting. Intraoperative findings noted good bone consolidation at distraction gap bilaterally, bony and fibrous union ankylosis of left TMJ and 80% enlarged adenoid.

Figure 2. Clinical images of patient (A) Preoperatively, (B) Post op 2 weeks 1st surgery with intraoral mandibular distractors, (C) Post 6 months 2nd surgery.
Mouth opening was successfully improved from an initial interincisal opening of 2mm to 28mm post-surgery. Lateral skull (cephalometric) radiograph postoperative revealed pharyngeal posterior airway space more pattern compared with preoperative view (Figure 3). Her OSA also improved from moderate to mild OSA which removed the need for CPAP overnight.

Discussion

Ankylosis is most commonly associated with trauma (31–98%), local or systemic infection (10–49%), or systemic diseases\(^5\). Unilateral TMJ ankylosis usually presented with limitation of mouth opening, facial asymmetry and upper airway obstruction (night snoring or OSA)\(^6\). Ankylosis will occur, as a result of destruction of the growth center and limited mobility of the mandible, gross changes in mandibular shape and size, as well as the surrounding functional matrix (muscles, ligaments, and airway)\(^7\).

The traditional way to treat such patients has always been to deal first with the ankylosed joint and allow the patient to open the mouth, then to treat any facial deformity later on\(^8–20\). This case report highlighted the management of left TMJ ankylosis in a paediatric patient with 2 stage surgery by performing mandibular distraction osteogenesis followed with releasing the TMJ ankylosis later. This was decided based on previous experience of the authors in distracting non-ankyloitic mandible where when distracting in anterior-posterior direction, both the proximal and distal segment were pushed away. When the proximal segment (containing condyle) was distracted posteriorly, TMJ posterior dislocation and external auditory meatus compression could happen. Thus in an ankyloitic TMJ, taking the advantage of an immobile condyle would allow only anterior direction distraction of the distal segment, while the proximal segment remain firm in place.

Mandibular distraction osteogenesis was reported in unilateral asymmetry cases would improve facial asymmetry and retrognathia (50.1%), correct the slanted lip commissure (24.7%), and improve or level the mandibular occlusal plane (11.1%)\(^21\). For bilateral mandibular distraction, its effective preventing tracheostomies of neonates or infants with respiratory distress (91.3%), relieving symptoms of OSA in adults (100%) and children (97%)\(^21\). Additionally, a study performed by Xiaofeng et al. in 2008\(^22\), revealed all their patients have a better profile and the narrow upper airway was relieved with their AHI improved from 69.6 ± 27.3 preoperatively to 3.3 ± 2.5 postoperatively distraction osteogenesis for the patients of OSA with craniomaxillomandibular deformities.

The STL model was constructed in this case prior for the surgery. STL model play an important role in surgical planning of the craniomaxillofacial deformity cases. The interesting about this STL model are the surgeon can handle a practical model with three dimensional analysis of the deformity and planning of correction\(^23\). Preoperatively, STL model helped the surgeon to assess the bone quality and plan their osteotomies site, bending the plates and screws and to fabricate surgical guide. More importantly, in distraction osteogenesis, STL model allows the planning and visualization of the distraction vector thus allowing assessment of the final desired outcome of the distracted bone. Therefore, in directly it may reduce or optimized the operating time and achieved more accurate surgical outcome\(^23\).

Conclusion

Mandibular distraction osteogenesis was effective in treating mandibular deformity and relieving symptoms of OSA. The stereolithography model played an important role in surgical planning and multidiscipline approach was crucial for optimum surgical correction of this maxillofacial deformity.

References


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