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Case Report
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Ankylosis of Temporomandibular Joint: Surgical Management

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Abstract

A child with unilateral temporomandibular joint ankylosis, micrognathia and obstructive sleep is presented. The safe management demands a multidisciplinary approach to select the best treatment option and sequence. The main aim of treatment is to achieve the highest Maximal Incisal Opening possible and immediate relieve of the narrow posterior airway space through release of the ankylotic bone mass. Satisfactory mouth opening was achieved in the immediate postoperative period but post-operative Apnea-Hypopnea Index does not return to normal level without additional mandibular advancement technique to further correct the retrognathia. Intensive jaw exercise is mandatory to achieve and maintain an acceptable Maximal Incisal Opening.

Introduction

Ankylosis of Temporomandibular Joint (TMJ) is a condition in which there is fibrous, osseous or fibro-osseous fusion between the mandibular condyle and the roof of mandibular fossa in the temporal bone, resulting in loss of normal rotational and translational movement. In children, ankylosed TMJ may affect both the structure and function of facial skeleton depending on its onset and magnitude. Underdeveloped mandible with asymmetrical face, limited chewing ability and oral hygiene care, narrowed airway and speech alterations are among its observed effects. Management of TMJ ankylosis in children differs from adult as it involves growth period and developing dentition.

Case Presentation

A 12-year old boy was referred to our hospital in November 2017 for inability to open his mouth. It was noticed during dental check-up by our School Dental Health Services. According to his mother, her son's condition was actually noticed since early childhood. However, they did not seek any medical consultation at that time because it was not associated with any pain. Loss of jaw function only allowed this young boy to eat soft diet and the food were inserted through natural front teeth space. He was born full term through normal delivery and has achieved normal growth milestones. He is the last of ten siblings and all other siblings are healthy. During sleep, his mother admitted that he snores heavily, but claimed to be active during school day. He has never been admitted into hospital for any ailments nor having any history of trauma or chronic infection to the ear, face and jaw.

The patient has a relatively small stature weighing 20 kg and a height of 132 cm. There is a slight asymmetry of the neck-shoulder border but the chest and back looks normal. There is no deformity in other parts of the body. Craniofacial assessment revealed a normal head shape, normal and symmetrical eyes and ears position. He has a short asymmetrical lower face; with a retruded "Bird's chin" (Figure 1). The facial nerve is intact bilaterally. However the left lower lip is devoid of pin prick and pressure sensation when compared to the normal right side. The patient is unable to open his mouth and palpation of the pre-auricular region revealed no movement of the left condyle and very slight rotational movement of the right condyle. The patient himself confirmed that he is able to move the right Temporo-Mandibular (TMJ) Joint slightly but the left TMJ has always been stiff and fixed. There was no fullness around the left zygomatic arch and pre-auricular region, showing normal skin without scar or ear tags and a normal ear anatomy. Intraorally, he has permanent dentition stage, a shifted lower dental midline and severely proclined upper incisors. Dental intercuspation showed zero Maximal Incisal Opening (MIO) making it impossible to examine the tongue.

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Table 1: TMJ Ankylosis in Young Patients. Treatment sequence, options and their challenges.

Recommended Procedure	Advantages	Challenges
Release of ankylosis followed by gap arthroplasty only [8].	Address the zero mouth opening in a single surgery, very quick and spontaneous results. Immediate improvement in nutrition and body weight. Allow opportunity for pre surgical orthodontics to manage complex malocclusion and plan an ideal Orthognathic Surgery later when facial growth is completed (classical treatment).	i) Mandible is still growing, and might necessitate second surgery for occlusal correction. ii) Pain, crepitus, degenerative changes and limited movements following gap arthroplasty. iii) Possibility of relapse following gap arthroplasty. iv)Facial deformity is still not corrected
Release of ankylosis followed by interpositional arthroplasty [8].	Address the zero mouth opening in a single surgery, very quick and spontaneous results. Immediate improvement in nutrition and body weight. Allow opportunity for pre surgical orthodontics to manage complex malocclusion and plan an ideal Orthognathic Surgery later when facial growth is completed (classical treatment).	i) Temporal hollowing may occur due to rotation of temporalis muscle into the surgical gap site. ii)Facial deformity is still not corrected
Distraction osteogenesis of mandible in the first phase and followed by release of ankylosis (with gap or interpositional arthroplasty) in the second phase operation [21,22].	Address the severe obstructive sleep apnea problem first by distracting the mandible forward. Able to achieve better facial profile. Patient and family are motivated to do further surgery based on improvement in facial appearance.	i) Not easy to achieve the ideal vector of movement for the mandible due to complex malocclusion. ii) High risk of airway occlusion if infection and edema occur during the distraction procedure because the TMJ is still ankylosed. Thus may need prolong tracheostomy for safe distraction procedure.
Release of ankylosis (with gap or interpositional arthroplasty) followed immediately by distraction osteogenesis [3,18,19].	Correction of mouth opening and advancing the mandible in one single operation. Able to control and decide the amount of mandibular advancement to enlarge the posterior airway to correct obstructive sleep apnea. Improve feeding immediately.	i) Unable to perform immediate intensive mouth opening exercise due to distraction osteogenesis procedure. ii) Early jaw exercise may hinder the new callus formation at the healing osteotomy sites.
Release of ankylosis and arthroplasty with costochondral graft reconstruction [18,23]; followed by conventional orthodontics	A functional condyle with good growth potential of the graft particularly in the pediatric age group. Re-establish joint function and harmonious jaw function. May commence pre-surgical orthodontics. Might be able to catch-up with normal skeletal profile and achieve good functional and esthetic results and avoid orthognathic surgery	i) Donor site morbidity. ii) Growth of graft is unpredictable, may overgrow and lead to mandibular prognathism, later needing orthognathic surgery for mandibular set-back. iii) Unpredictable costochondral graft growth may interfere with presurgical orthodontics outcome.

Figure 1: Left TMJ ankylosis. Micrognathia with chin point deviated to the left (A) and retrognathic mandible with loss of normal mandibular-neck angle (B).

Figure 2: Pre-operative CT Scan images in coronal (A), sagittal (B) and axial (C) view shows a thin gap between the deformed bifid left condyle and skull base. The ankylotic bone mass seems to extend medio-laterally (A) as well as anteriorly in relation to the glenoid fossa (B).

Nasal endoscopy using a flexible scope revealed hypertrophic inferior turbinate in right nasal cavity while it was not possible to pass the scope in the narrower left nasal cavity. The adenoids were hypertrophied filling the posterior choanae. The epiglottis and both vocal cords were normal. 3D Computed tomography scan of the craniofacial region revealed a large bony mass that forms a fusion between the left mandibular fossa of the temporal bone and abnormally bifid-shape condylar head region of the left mandible. Both coronoid processes appeared elongated superiorly, and the right TMJ complex appeared normal (Figure 2). A Stereolithographic (STL) skull model was constructed for assessment and surgical planning (Figure 3). The patient was referred to Respiratory Clinic for polysomnography. Apnea-Hypopnea Index (AHI) score obtained was 37.5 indicating a severe obstructive sleep apnea condition.

A diagnosis of left TMJ ankylosis associated with severe obstructive sleep apnea was established. Following a multidisciplinary discussion and treatment work-up, surgery to release the ankylosed TMJ was performed in January 2018. The aim was to achieve a Maximal Incisal Opening (MIO) of about 30 mm and correction of obstructive sleep apnea. Fluticasone nasal spray and oral antihistamine was prescribed and Continuous Positive Airway Pressure (CPAP) commenced prior to the surgery.

The surgical procedure began with fibreoptic-assisted nasotracheal intubation, followed by an elective tracheostomy. The ankylosed left TMJ area was accessed through a left pre-auricular incision which was extended hemi-coronal into the scalp and the surgical flap was reflected anteriorly exposing the left condylar region. Upon approaching the left condylar region, a large hard and avascular was seen occupying the whole TMJ area leaving no clue of the original anatomy of the site. Dissection was done all along the zygomatic arch and once the extent of the bony overgrowth was confirmed, bony cut was performed from the sigmoid notch anterior superiorly and carried postero-inferiorly beneath the zygomatic arch towards the

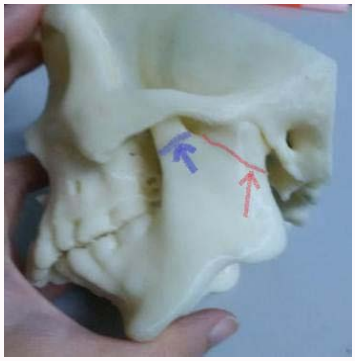

Figure 3: Stereolithography model of the skull, printed to aid surgical planning. Arrows on the model indicate the planned surgery, excision of the ankylotic bone mass (red arrow) and coronoidectomy (blue arrow).

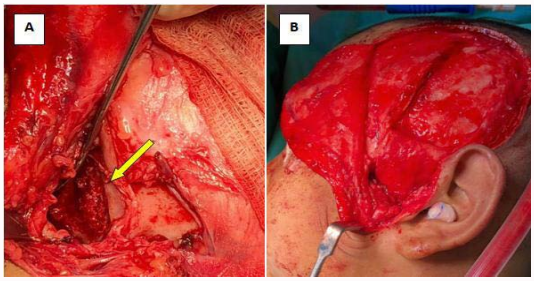

Figure 4: Intra-operative surgical view following excision of the ankylotic bone mass leaving a one cm gap defect beneath the zygomatic arch, as indicated by arrow (A) and downward rotation of the anterior part of temporalis muscle flap sutured into the defect (B).

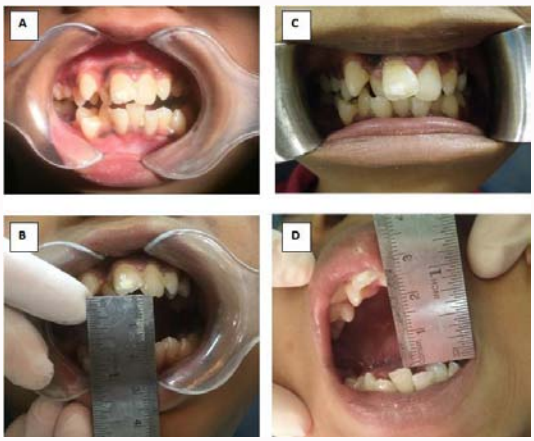

Figure 5: Intra-oral views of occlusion and maximal mouth opening achieved at different time points. Post-operative day-3 showing an unstable occlusion (A), and his maximal incisal opening of 15 mm (B). At six months post-operative, his occlusion is stabilized (C) and maximal incisal opening is 28 mm.

posterior border of the ramus (Figure 4A). This maneuver released the ankylotic bone mass from temporal bone and some degree of mouth opening was immediately achieved.

Bilateral coronoidectomy was performed allowing further intra-operative MIO up to 32 mm anterior part of the temporal is muscle was then raised and rotated inferiorly to fill in the surgical gap defect (Figure 4B). Wound closure done in layers with a drain left for three days. His post-operative period was uneventful and the tracheostomy tube was kept for one week. Jaw exercise commenced early on third

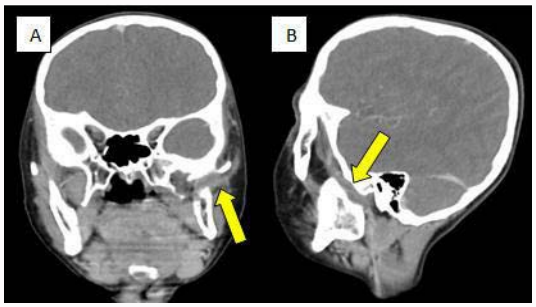

Figure 6: CT scan ImageIN coronal (A) and sagittal (B) view at six months post-operative showing a gap remained with interpositional temporalis flap *in-situ* (arrow), with no evidence of calcifications that may suggest re-ankylosis.

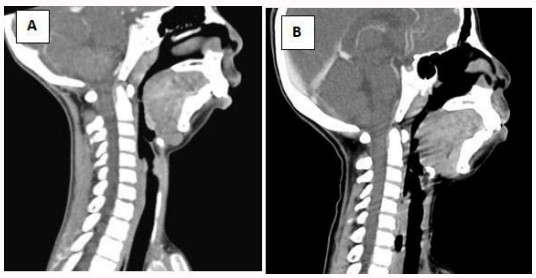

Figure 7: Sagittal view of CT scan. Preoperative view showing retrognathic mandible associated with a narrow retro-glossal airway space (A) and post-operative view showing an anterior displacement of the mandible producing a wider retro-glossal airway space (B).

post-operative day. All were well except for a slight paresis of the left zygomatic branch of facial nerve and an initial early post-operative unstable occlusion presenting with a left unilateral open bite (Figure 5a). Patient was discharged after ten days and encouraged to perform intensive jaw exercise to maintain and further improve mouth opening.

At 6 months post-operative review, patient had recovered well and weighing 25 kg. Paresis of left facial nerve had resolved completely and MIO has increased to 28 mm with ability to perform lateral excursive movements of the mandible. Mother claimed that his night time snoring has reduced significantly and he seems to be more active as compared to pre-operative stage. A repeat post-operative polysomnography was performed and his Apnea-Hypopnea Index has improved to 10.3 per hour. Post-operative CT scan at six months did not reveal any evidence of re-ankylosis (Figure 6). Both patient and his mother are very happy with the mouth opening and a stable functional occlusion (Figure 5c).

Discussion

Etiology of TMJ ankylosis in children may rarely be congenital [1] but more commonly acquired [2] following trauma to the TMJ and infection such as ear infection and odontogenic cause [3]. In this case, although the mother claimed complete trismus to have taken place since early childhood days, we believe that ankylosis activity may have actually started around the age of 7-9 years old. His facial asymmetry is mainly limited to the lower face and there were no retained deciduous molars, followed by eruption of their permanent successors. Should ankylosis occur at earlier age, they might be retained as the patient is unable to remove them when they were loose in the mouth. The ankylosis activity continued gradually over the time with excessive bone formation and mineralization which gave

rise to our findings of the broad size of ankylotic mass and elongated ipsilateral coronoid process. As there was no history of pain or ear symptoms, infection may not be the cause and trauma to the left TMJ could have been the most probable reason. An absence of sensation in the left lower lip further confirmed this etiology which could be due to inferior alveolar nerve injury during the traumatic episode in the left mandible.

Intubation and safe access to the airway in TMJ ankylosis cases is extremely challenging. Blind nasal intubation and retrograde intubation are alternative interventions [4]. Although a wake nasal intubation is a safe alternative, its application in children is challenging [5]. Elective tracheostomy seems to be the safest and most preferable airway management choice in children with TMJ ankylosis. The fibreoptic assisted intubation was successfully done in this case followed by a tracheostomy for continuous delivery of general anesthesia and a secured and protected airway in the postoperative period. Indeed the diagnostic preoperative nasoendoscopy done in this case was crucial to detect presence of soft tissue obstruction or any other anomaly in the upper pharyngeal airway thus avoiding "mishaps and surprises" during anesthesia.

Surgical procedures advocated in managing ankylosis in children include release of ankylosis with gap arthroplasty; interpositional arthroplasty, joint reconstruction, and distraction osteogenesis and orthognathic surgery. The sequence of these procedures are debatable and generally depends on age and growth maturity, extend of facial deformity, general health and self motivation (Table 1).

Gap arthroplasty is the oldest technique employed in the surgical management of TMJ ankylosis [6] and offers a simple operation with low surgical cost. Despite these advantages, we did not advocate this treatment option as it carries the disadvantage of chronic pain, crepitus, and occasional degenerative changes and associated with limited movements that may lead to reankylosis. Studies revealed that there was higher percentage of recurrences among patient treated with gap arthroplasty alone, as compared to interpositional arthroplasty using either remainder of the disc, temporalis flap or costochondral graft [6,7]. In terms of maximal mouth opening (MIO) achieved, Junli Ma *et al.* found that the difference between gap arthroplasty and interpositional arthroplasty technique was insignificant, giving only a 1.96 mm difference [8]. Another study by Danda *et al.* Also concluded that there is no significant difference in diet scores, weight of the patients as well as maximal mouth opening achieved [9].

In this case, interpositional arthroplasty using temporalis muscle and fascia was performed in supporting the idea that presence of vital soft tissue in the surgical defect may prevent from re-ankylosis [10]. Temporalis muscle was taken due to its close proximity to the surgical site, easy usage, rich blood supply and minimal donor-site morbidity. The interpositional material creates a barrier between the upper ramus and the skull base, thus preventing fibrous adhesions across the gap. However, in long term, the fascia may undergo contracture and resorption and finally end up as scar tissue. Besides autogenous grafts, other alloplastic materials such as acrylic [11] and methyl methacrylate [12] have been used as interpositional materials with varying results.

A bilateral coronoidectomy was performed intra-operative which allowed further improvement in the MIO up to 32 mm. Both coronoid processes were elongated superiorly. Long standing TMJ ankylosis contributes to coronoid hyperplasia and elongated coronoid process may result in limited mouth opening [13]. The restricted mouth opening reduces temporalis muscle activity, contributing to formation of fibrosis of temporalis muscle tendon. Hyperactivity of the suprahyoid muscles when attempting to open the mouth induces development of coronoid hyperplasia through the mechanism similar to distraction osteogenesis [14].

Distraction osteogenesis may be applied to the retruded ankylosed mandible to correct secondary deformities and widen thyropharyngeal airway by mobilizing the genioglossus muscle and the whole tongue forward. In turn, the symptoms of snoring and obstructive sleep apnea May improve. Zhang *et al.* advocated performing distraction osteogenesis of the mandible as the initial surgery [15], followed by arthroplasty or TMJ reconstruction. Since TMJ ankylosis frequently leads to more complex facial deformity which are not amenable by simple distraction procedure, some patients require orthognathic surgery to improve occlusion and facial profile along with or following arthroplasty or TMJ reconstruction. The patient in this case study was only 12 years old and a decision was made to defer orthognathic surgery till the facial growth is complete. This will also give more time for the orthodontist to begin presurgical orthodontics that will allow a more favourable occlusion during orthognathic operation. Performing distraction osteogenesis before releasing ankylosis will enable advancement of the mandible but without achieving favourable dental occlusion.

Temporomandibular joint reconstruction is not commonly performed following release of ankylosis. It can be performed either in the same operation session or in a later reconstruction session. This procedure is more commonly done in children using costochondral grafts to take advantage of the potential growth cartilage, thus allowing more natural forward growth of the mandible and possibly simplifying orthodontics and avoiding future orthognathic surgery.

Regardless of which surgical technique is selected, the most important post-operative care is intensive mouth opening physiotherapy which must be diligently followed by the patient himself with the aim of avoiding recurrence and further improving the MIO. Although patients are commonly blamed for lack of compliance in aggressive jaw exercise, one of the possible outcomes in this TMJ surgery is re-ankylosis. It generally begins with a gradual decrease in MIO distance followed by loosing range of mandibular motion, most commonly within the first year after surgery. However, the onset varies from 1 month to as long as 13 years post-operatively [16]. The major cause of re-ankylosis is thought to be inadequate resection of ankylotic part and poor compliance of the patient with postoperative mouth-opening physiotherapy. Other factors include wound infection, and a foreign body reaction caused by interpositional materials [17]. Long-term follow-up is the norm.

The success of this case was clearly demonstrated with a maintained MIO of 28 mm, enabling normal feeding and an abrupt increase in body weight from 20 kg to 25 kg within six months. Snoring during sleep was eliminated and the Apnea-Hypopnea Index has improved from a preoperative score of 37.5 to 10.3 and CT scan image at six months post-operative does not show any foci of calcification in the left TMJ site (Figure 6). Although resection of the ankylosed left TMJ has tremendously improved the mouth opening and at the same time able to produce a slight anterior displacement of the mandible, thus widening the posterior pharyngeal airway at the retro-glossal level, however the improvement in the AHI is still unsatisfactory (Figure 7). At this stage, a mandibular advancement appliance for sleep disordered breathing, which is able to protract the mandible forward,

may be recommended. This appliance may stimulate the growth of the right condyle as well widening the posterior airway and further improve the Apnea-Hypopnea Index.

Conclusion

Safe and effective management of a child presenting with a triad of TMJ ankylosis, micrognathia and obstructive sleep apnea requires a multidisciplinary approach. The treatment sequence is tailored to individual needs depending on the age at diagnosis and expertise available. The main aim of treatment is to achieve the highest MIO possible and immediate relieve of the narrow posterior airway space through release of the ankylosis. Intensive mouth opening physiotherapy is mandatory to achieve and maintain an acceptable MIO.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the parents have given their consent for their son's images and other clinical information to be reported in the journal. The parents understand that the patient's name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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