Case Report

DETERMINING THE IMPROVEMENT IN NASOPHARYNGEAL AIRWAY FOLLOWING THE RELEASE OF AN UNILATERAL TMJ ANKYLOSIS - A CASE REPORT

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ABSTRACT

TMJ Ankylosis is a debilitating disease with wide ranging functional and aesthetic manifestations. One of the primary concerns of the patient are the episodes of obstructive sleep apnea which are relived following arthroplasties in bilateral cases of TMJ Ankylosis due to increase in the nasopharyngeal airway. This phenomenon is also noted in unilateral cases but the possible mechanism and radiographic changes have not been found in literature. We present a case of a 7 year old female patient with TMJ ankylosis of the right side. Pre-operative photographs and a lateral cephalogram of the patient were taken. Saddle angle, depth angle, vertical angle, roof angle as well as the posterior height, total depth and length of the floor of the Nasopharynx were measured. The position of the hyoid bone was also measured by utilizing the values of H-Rgn, AA-H, C3-H and C3-Rgn. These measurements were compared with readings obtained on the post-operative lateral cephalogram and the differences were noted.

Keywords: *Ankylosis, Temporomandibular joint, obstructive sleep apnea syndrome.*

INTRODUCTION

Temporomandibular joint (TMJ) ankylosis is a pathological condition affecting one or both condyles resulting in progressive stiffening and immobilization of the joint[1]. It has varying prevalence throughout the world, with more cases reported in South East Asia as compared to Europe and North America. Worldwide studies on prevalence of this debilitating condition are virtually non-existent, although one community based study in India estimated the prevalence to be 0.46/1000 population of children[2]. The etiology of TMJ ankylosis has local and systemic contributing factors, trauma being responsible for 13-100% of cases reporting to all hospitals[3]. Systemic causes include otitis media, mastoiditis, tuberculosis, rheumatoid arthritis and fibrous dysplasia amongst many other causes[4,5,6,7].

The clinical picture of TMJ ankylosis shows a wide array of features that drastically impact the functional and aesthetic concerns of the patient and consequently lead to a poor quality of life[8]. This is compounded by delay in seeking treatment due to a misdiagnosis or lack of diagnosis by the general medical and dental practioners and lack of awareness amongst the general population itself[9,10,11]. Among unilateral cases of TMJ ankylosis, the patient usually presents as a case of hemifacial microsomia with mandibular retrognathia along with other classic signs of this disease. Obstructive sleep apnea syndrome (OSAS), though more commonly seen in bilateral TMJ ankylosis, is also a feature common in unilateral cases[12]. Studies have correlated OSAS with deficient oropharyngeal and hyopharyngeal airway spaces and accordingly construed soft tissue procedures such as uvulopalatopharynoplasty (UPPP) to help cope with this problem. The role of hard tissues in the development and consequently the relief of OSAS were not recognized until the 1980's when bimaxillary surgery was advocated to be safer and more successful than tracheostomies in treating OSAS[13,14]. This was correlated with bilateral TMJ ankylosis in the early 1990's and gap/interpositioning arthroplasties

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showed improvement in not just the mouth opening but also in a decrease in the episodes of sleep apnea. This clinical outcome was validated by post-operative lateral cephalograms which showed an increase in the nasopharyngeal airway and a more favorable position of the hyoid bone[15]. This effect has been noted in patients with unilateral TMJ ankylosis following the release of the ankylosis but has not been proven cephalometrically and the literature is silent on the mechanism involved.

We present a case of a 7 year old girl who showed discernible clinical and radiographic improvement in the nasopharyngeal airway following the release of the unilateral TMJ ankylosis. This case report hypothesizes that clinical and radiographic improvement would be seen on the basis of an "Arch" effect of the mandible upon release of the ankylotic mass which will relieve compression on the Nasopharynx on the affected side. This forward and anti-clockwise movement of the mandible (clockwise if the ankylosis is on the left side only) will also alleviate or greatly reduce the other associated sequale of unilateral temporomandibular joint ankylosis.

CASE REPORT

A 7 year old girl reported to the Department of Oral and Maxillofacial Surgery at Khyber College of Dentistry, Peshawar with the chief complaint of progressive limitation in mouth opening and lack of sleep at night due to recurrent episodes of breathlessness. The patient's attendant gave a positive history of trauma to the chin 1 year back for which she underwent no intervention. Clinical examination revealed a mouth opening of 10mm and discernible mandibular retrognathia. No movement was felt on palpation of the right condyle during opening of the mouth. Dental midlines were on and in concordance with the facial midline. A history of 4-5 sleep apnea episodes per night was also elicited. A scar was also visible on the lower chin. An orthopantomogram was ordered which showed diminished joint space on the right side and coronoid elongation. Antegonial notching was absent on the affected side. The patient was prescribed investigations for undergoing condylectomy with interpositioning arthroplasty under general anesthesia. A lateral cephalogram was ordered to assess the nasopharyngeal airway and hyoid triangle pre-operatively. The saddle angle, depth angle, vertical angle, roof angle as well as the posterior height, total depth and length of the floor of the nasopharynx were measured. The position of the hyoid bone was also measured by utilizing the H-Rgn, AA-H, C3-H and C3-Rgn parameters. The patient underwent the procedure and a 35mm mouth opening was achieved per operatively. The patient was discharged on the fifth post-operative day and recalled after 2 weeks for a fresh lateral cephalogram. The aforementioned parameters were measured again to check for any radiographic changes. A fresh history of the patient of the patient was also taken to check for improvement in occurrence of the sleep apnea episodes.

RESULTS

The improvement in the nasopharyngeal airway was analyzed through pre-op and post-op lateral cephalograms is shown in Table 1.

Table - 1: Comprision of pre-op and post-op cephalogram

	Post- op	Pre-op
Saddle angle	133°	130°
Angle of nasopharyngeal depth	58°	63°
Vertical angle of nasopharynx	50°	42°
Roof angle of nasopharynx	97°	97°
Posterior height of nasopharynx	38mm	40mm
Total depth of nasopharynx	21mm	39mm
Length of floor of nasopharynx	27mm	30mm

The pre-op distance between hyoidale (H) and retrognathion (Rgn) was found to be 21mm which increased by 7mm following the release of the ankylosis. The distance between the C3 vertebrae and hyoidale showed a decrease of 5mm, from 26mm (pre-op) to 21mm (post-op). The greatest change in length was noted in the C3 to Rgn parameter, showing an increase of 14mm in the post op lateral cephalogram as shown in Table 2. A point to note was that despite the release of the ankylosis, the post-op values remained quite shy of the normal readings seen while assessing the hyoid triangle (Table 3).

Table - 2: Pre-op and post-op position of hyoid bone

Position of Hyoid bone	Post-op	Pre-op
H-Rgn	28mm	21mm
AA-H	30mm	34mm
C3-H	21mm	26mm
C3-Rgn	42mm	28mm

Table - 3: Mean values of the hyoid triangle

Distance	Linear depth of nasopharynx
C3-Rgn	67.20mm, SD 6.6mm
C3-H	31.76mm, SD 2.9mm
H-Rgn	36.83mm, SD 5.8mm

DICUSSSION

Obstructive sleep apnea syndrome (OSAS) is a serious problem that affects multiple body systems and has many social ramifications associated with it. Apnea generally is defined as cessation of air flow at the level of nostrils and mouth lasting at least 10 seconds. Sleep apnea syndrome is diagnosed if more than 5 episodes of apnea is encountered per sleep hour[16].

A variety of factors that contribute towards OSAS have been put forward in literature. These include increased collapsibility of upper airway structures and reduction in size of airway. Along with that various alterations are there in craniofacial structures. Also surrounding soft tissue structure enlargement is often seen[17]. Day time symptoms are often silent in patients with OSAS, however such children are often under developed for their age. Also there is a poor nasal air way resulting in mouth breathing. During night time however the potential for obstruction increases. This is because the patient is supine, neck is not extended. Also secretion increases during sleep which contributes to increased apnea symptoms during night[18].

Micrognathia has been put forward as one of the causative factor for OSAS. Due to retrusion of mandible and tongue, there is decrease in size of airway. Thus OSAS is often a feature of unilateral and bilateral TMJ ankylosis.[12] The patient in current case report had unilateral TMJ ankylosis with retrognathia. Patient also had 4-5 episodes of apnea per night. This is in comparison with other studies. In a study by Kumar et al craniofacial deformities were reported to be the commonest cause of childhood OSAS[19].

Lateral cephalogram was used to evaluate dimensions of airway. Cephalometry is a useful tool as it demonstrates all the important parameters of airway obstruction. These include mandibular length, hyoid bone position and craniofacial extension. It is also considered a useful method of evaluating improvement in airway after surgical procedures. The role of cephalometry becomes more important in TMJ ankylosis where unaided visualization of the airway is not possible by using other means[20]. A variety of cephalometric perimeters are used to ascertain the pre-existing status of the upper airway. Zeitoun[15] utilized a variety of cephalometric landmarks in his study incorporating the distance of the posterior nasal spine, uvula and epiglottis to the posterior pharyngeal wall as well as the Frankfurt to mandibular plane angle (FMPA). His case series of 9 patients with bilateral TMJ ankylosis underwent a two stage treatment plan of mandibular advancement and ramus lengthening followed by ankylosis release 3 months later which was followed by post-operative radiographic assessment as well as correlation with arterial blood gases (ABG). Positive changes were noted in the distance between the posterior pharyngeal wall and the uvula as well as the epiglottis respectively. The FMPA showed a decrease after the 1st stage of treatment and increased after the release of the bilateral ankylosis. The ABG's showed no significant improvement in the post-operative period. No such correlation was done in the present case. Zeitoun[15] reported that all the patients except one showed complete remission of snoring and sleep apnea which is consistent with our study findings.

Other methods have been discussed in literature regarding measurement of upper airway in OSAS. These include physical examination, nasal examination including nasopharyngoscopy. Measures of upper airway collapsibility include pharyngeal critical pressure and negative expiratory pressure. Acoustic reflection, CT, and MRI are some other methods used for upper airway evaluation[21]. Due to a lack of patient's affordability; such investigations could not be carried out to obtain a 3D assessment of the improvement in the patient's airway

Craniofacial surgery has been advocated as a treatment option for relieving obstructive sleep apnea. Multiple options are available in this regard. These include tracheostomies, uvulopalatopharyngoplasties(UPPP) and osteotomies. Tracheostomy although plausible has a high mortality rate especially in children. Bimaxillary surgery has been suggested as the best alternative. Kumar et al[22] found that all the patients who had undergone surgical correction, improvement in airway was seen in all cases. Distraction osteogenesis has been recently described as a useful procedure for improving airway in cases of paedriatric OSAS. In a study by Bukhari[23], distraction osteogenesis was carried out on a total of 7 patients. All patients had retruded mandible, out of these 7 patients 5 had TMJ ankylosis.

Following distraction osteogenesis all these patients noticed relief of airway obstruction. Similarly in another study by Anantanarayanan[24] dimensions of airway improved to 31.33% with a 3.8% improvement in oxygen saturation levels. Similar results were obtained in the present case where the release of the ankylotic mass resulted in improvements of airway dimensions. According to a study by Gunaseelan[25], interpositional arthroplasty along with genial distraction was employed for a patient with OSAS associated with TMJ ankylosis. Improved in airway a dimension was seen following this treatment protocol.

Other studies have reported improvement in airway obstruction after mandibular advancement, thus increasing posterior facial height along with mandibular advancement causes widening in upper airway spaces. This brings the tongue forward and results in improvement in hyoid bone position[15]. This was noticed in the current study where condylectomy and interpositioning arthroplasty resulted in improvement of parameters such as angle of nasopharyngeal depth, total depth and length of floor of nasopharynx. This is significant since the majority of patients coming to our unit are non-affording, thus the option of condylectomy in such cases of unilateral ankylosis may prove to be of benefit in such cases where distraction osteogenesis cannot be done due to financial constraints.

CONCLUSION

The release of a unilateral TMJ ankylosis shows discernible clinical and radiological improvement in the nasopharyngeal airway in patient with obstructive sleep apnea syndrome.

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