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Management Of Bilateral Temporomandibular Joint Ankylosis With Micrognathia And Obstructive Sleep Apnea Syndrome Using Interpositional Arthroplasty And Distraction Osteogenesis – A Case Report

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Abstract

Temporomandibular joint (TMJ) Ankylosis posses many problems like inability to open the mouth, poor oral hygiene, difficulty in eating proper food, unaesthetic appearance due to growth retardation and in severe cases difficulty in breathing specially during night depending on the time of occurance of injury. If the TMJ Ankylosis occurs the early stage of life it affects both the form and function of the patient. One of the most common treatments used to treat temporomandibular joint (TMJ) ankylosis is interpositional arthroplasty (IPA). When patients come with severe mandibular dysplasia either orthognathic surgery or distraction osteogenesis (DO) can be considered. Simultaneous IPA with DO has been used by many surgeons correct TMJ ankylosis associated with facial asymmetry/micrognathia as as it enables them to simultaneously reconstruct the neocondyle and correct facial malformations eliminating the need for second surgery. Here we have narrated a case of an young male patient suffering from bilateral TMJ Ankylosis with micrognathia treated by simultaneous IPA and DO and regular follow up was done.

Keywords: Interpositional Arthroplasty(IPA), Obstructive Sleep Apnea(OSA), Apneac-Hypoapneac Index(AHI)

INTRODUCTION

Micrognathia is caused by bilateral temporomandibular joint (TMJ) ankylosis in a developing kid. Micrognathia can cause airway blockage. The obstructive sleep apnea and hypopnea syndrome (OSAHS),

induced by TMJ ankylosis and mandibular micrognathia, can have a major influence on patients' mental well-being. Tracheostomy and tracheal intubation would be required in severe cases.[1-3]
TMJ ankylosis causes condyle degradation as well as mandibular body and ramus deficits over time. Micrognathia may commonly be treated by autoplastic bone transplantation[4,5] or distraction osteogenesis, whereas trismus can be treated with gap arthroplasty, condylectomy, or TMJ reconstruction[6]. Distraction osteogenesis is a prominent treatment for mandibular lengthening that works well for patients with a mixed dentition. Snyder et al. began utilising it in the head and neck in 1973, after orthopaedic surgeons had widely utilised it to stretch long bones[7]. Distraction osteogenesis has been used to achieve mandibular broadening, alveolar height augmentation, bifocal distraction for mandibular defect rectification, and craniofacial lengthening.
Although simultaneous facial asymmetry treatment has been recorded, gap arthroplasty, or joint repair, is often done first, followed by correction of secondary mandibular and maxillary growth abnormalities[8]. The first report of simultaneous gap arthroplasty and DO for the treatment of micrognathia in TMJ ankylosis was in 1999[9]. Simultaneous repair removes the need for a second operation and is more cost effective.
We report a case of a young patient with significant bony temporomandibular joint (TMD) ankylosis, distraction osteogenesis was employed to extend the mandible in conjunction with a condylectomy and interpositional arthroplasty.

CASE REPORT

A 17-year-old male patient presented to the Department of Oral and Maxillofacial Surgery with the chief complaint of gradual decrease in mouth opening since birth and facial asymmetry, which on further examination revealed a history of snoring, excessive daytime sleepiness, and tongue falling back during sleep (Fig. 1a & 1b). There had been no problems reported at delivery, and there had been no later history of damage to the face bones. There is a possibility of forceps delivery during birth that the parents are unaware of. Both the patient and his parents were aware of the issue, but postponed evaluation due to financial constraints.

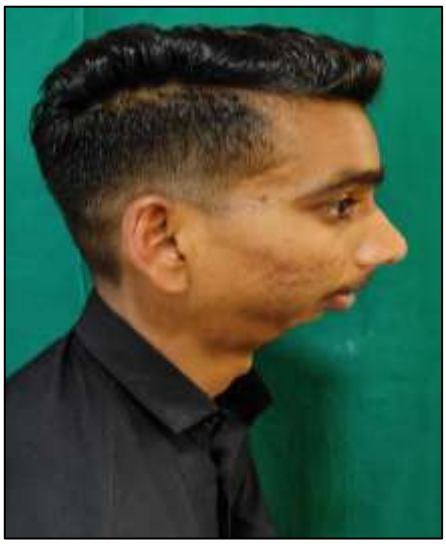

Fig. 1a

Fig. 1b

The intraoral health was poor, with several teeth suffering from caries and residual roots and no condylar motions in the TMJ. There was a distinct convex facial profile and significant mandibular retrognathism, creating the impression of a "bird face." There was also an increased naso-labial angle and no cervicomental angle. Furthermore, the patient's inability to lie flat on the bed resulted in considerable snoring, as well as trouble breathing.
Radiological testing revealed the condyle abnormalities, and the jaw looked to be undeveloped below and forward. OPG revealed a pronounced gonial angle and obliteration of the joint space between the glenoid fossa and the condyle, as well as bilaterally enlarged coronoid processes. According to cephalometric analysis, an angle reflecting the maxillary protrusion formed by the sella turcica point, nasion point, and upper alveolar point (SNA) is 79.5 degrees, and an angle reflecting the mandibular protrusion formed by the sella turcica

point, nasion point, and inferior alveolar point (SNB) is 60.1 degrees. This was due to severe skeletal class II (Fig 2a & 2b).

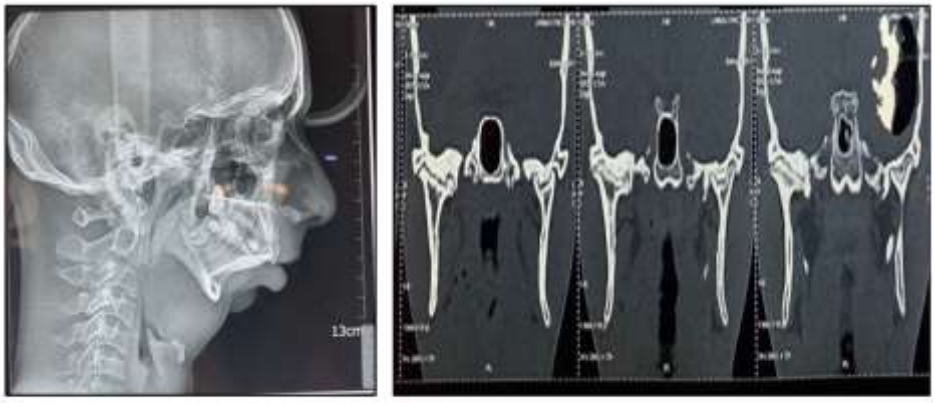

Fig. 2a Fig. 2b

Fig 2a showing Lateral Cephalogram and Fig 2b showing CT image

Further examination revealed that the patient also had typical signs and symptoms of obstructive sleep apnea (OSA). The patient complained of excessive daytime tiredness, snoring, breathing difficulties owing to tongue fallback, and dry mouth. Polysomnographic testing verified all of these symptoms. The results revealed an apnea-hypoapnea index (AHI) of 55.4, indicating severe OSA. While the patient was awake, the average oxygen saturation was 95%, but while sleeping, it plummeted to 67%.

Based on the findings, the diagnosis was a triad of bilateral TMJ ankylosis, microganthia, and OSA. We intended to treat bilateral TMJ ankylosis with interpositional arthroplasty with temporomyofascial flap and distraction osteogenesis with intra-oral distractors for mandibular lengthening at the same time.

Awake fibreoptic intubation was performed while under general anaesthetic. The Alkayat bramley incision was noted, and the dissection was done layer by layer. Ankylotic mass of about 15 x 20 mm was found and removed. A coronoidectomy was also carried out. Following that, a temporomyofascial flap was extracted and inserted into the space produced. The operation was repeated on the opposite side. The mouth openness attained after ankylotic mass removal and coronoidectomy was 40mm. Osteotomy cuts were created bilaterally distal to the distal most tooth for the implantation of intra-oral distractors. Screws were used to fix an intra-oral unidirectional distraction device so that the vectors of distraction were parallel to the lower border of the jaw. Following suction drain placement, the preauricular incision was closed in two layers with 3-0 vicryl suture and the skin was closed with 4-0 ethilon suture. To prevent post-operative pain, a tracheostomy was done. Hemostasis was verified and accomplished from all locations, and general anaesthesia was reversed..

After a three-day delay, the distractor was activated at a pace of one millimetre each day, twice a day, for 0.5 millimetres each time. The anticipated amount of distraction was 23mm. To avoid a recurrence, the patient was kept on a regular follow-up schedule and exercises were conducted on a regular basis (Fig 4a & 4b).

Fig. 3

Fig 3 showing 1 month follow up lateral profile

Fig. 4a

Fig. 4b

Fig 3a showing 3rd day follow up OPG and Fig 3b showing 10th day follow up OPG

DISCUSSION

TMJ ankylosis happens when the mandibular condyle and the glenoid fossa of the temporal bone merge. It hinders a child's ability to grow their facial skeleton, which results in a retrognathic mandible, facial asymmetry, a deviated chin, midline shift, occlusal cant, tooth crowding, and unerupted teeth. Reduced mouth opening also contributes to periodontal disease, dental caries, and poor oral health. In addition to having a narrower airway due to retrognathic chin and jaw, some of these patients may also suffer sleep apnea[10].

Only direct surgical intervention can treat this disorder, which is caused by a variety of reasons including trauma, systemic, local, and inflammatory disorders, as well as tumours in the TMJ region. Surgery is the primary method of treatment for TMJ ankylosis. It is crucial to use an interpositional drug after arthroplasty to stop TMJ re-ankylosis; this specific treatment element has been the subject of several discussions. As interposition materials, the temporalis muscle and fascia, dermis, auricular cartilage, fascia lata, fat, lyodura, silicone, and other metals have all been used. The interpositional material that is employed most frequently is the temporalis muscle flap fascia flap[11].

For surgeons, treating TMJ ankylosis with secondary abnormalities continues to be a serious clinical problem. The literature has provided descriptions of numerous surgical methods that can be used to treat this issue. In the majority of situations, combining these strategies will produce satisfactory results. The order in which distraction osteogenesis and arthroplasty should be carried out to treat TMJ ankylosis with subsequent abnormalities, however, is still a matter of debate.

In 2004, Krishna Rao et al. and Sudhir kumar et al. evaluated the potential of distraction osteogenesis for concurrent mandibular deformity repair. Six kids with mandibular deformity and temporo-mandibular joint ankylosis were included in this investigation. In this investigation, uniaxial double pin distractors with Schanz pins were employed. The patients underwent retromolar mandibular osteotomy with distractor insertion and gap arthroplasty simultaneously. On the fifth day after surgery, distraction began. On the first post-operative day, the patients were given dynamic exercises for the temporo-mandibular joint. On follow-up, every patient had a good mouth opening. All of these patients had their mandibular abnormality cosmetically corrected well. The patients were put on orthodontic therapy as a result of some degree of malocclusion that developed during the course of the treatment[12].

In 2007, R. Gunaseelan, et al. and P. Anantanarayanan et al. presented a case of obstructive sleep apnoea syndrome secondary to TMJ ankylosis that was corrected by a new technique employing simultaneous genial distraction along with interposition arthroplasty. A four-month postoperative evaluation of the distracted segment revealed good stability while retaining the prior occlusion[13].

Feiyun et al. and wei et al. in 2010 conducted a study on 16 patients with bilateral TMJ ankylosis and micrognathia. The 16 patients' three-dimensional craniomaxillofacial models were created utilising fast prototyping and computed tomography. On the models, simulation surgery and individual internal DO were carried out. The procedure involved simultaneous DO of the mandibular body and transport DO for arthroplasty of the temporomandibular joint. The diversion began the seventh day following surgery. The rate of diversion

was 0.8 mm per day. After surgery, the patients started opening their mouths actively. In addition to curing the obstructive sleep apnea and hypopnea syndrome, all patients had their micrognathias fixed. Preoperatively, the average mouth openness was 4.6 mm, and it grew to 33.5 mm after surgery. The sella-nasion-supramental angle's average range increased from 68.7° preoperatively to 77.6° after surgery. In the distraction spaces, bone growth was seen. A follow-up duration of 29.7 months was used (range 6 to 52). Throughout the follow-up period, no patient experienced any sequelae or recurrence of micrognathia, temporomandibular joint ankylosis, or either condition[14].

A case report presented in the year 2016 by Sharma A. et al. and Paeng J. et al. where Gap arthroplasty and concurrent intraoral distraction were intended to alleviate the condition. TMJ ankylosis and related asymmetry of the face. The ramus-condyle segment was successfully prolonged, and the mouth opening range was greatly increased, following gap arthroplasty and 23 mm of distraction. For four months after distraction, the resulting interocclusal space was steadily maintained with an occlusal splint. After prosthetic treatment, satisfactory occlusion was eventually attained. Osseous contouring and augmentation surgery was used to address the remaining mandibular asymmetry. Twenty-four months following the last treatment, the mouth-opening range remained at 35 mm[15].

Zhang. W et al. and Zhang T et al. did a study on 40 patients. Their ages ranged from nine to fifty-three (mean age 24.5 years). Thirty of these individuals had bilateral TMJ ankylosis, while ten had unilateral TMJ ankylosis. Additionally, 27 patients had the obstructive sleep apnea-hypopnea syndrome (OSAHS). The initial procedure for all patients was distraction osteogenesis, which was followed by arthroplasty or TMJ repair. In addition to or following arthroplasty or TMJ reconstruction, some patients received orthognathic surgery to enhance their occlusion and facial structure. Improvements in maximal interincisal opening (MIO), appearance, and respiratory function were used to assess the treatment benefits. After the course of treatment was complete, all patients displayed improvements in MIO and appearance, and the snoring symptom vanished. The airway's width was greatly expanded. Four patients relapsed during a follow-up period that spanned from 6 to 85 months (mean 28.3 months). This study suggests that, especially for patients with OSAHS, treating TMJ ankylosis with secondary deformities by distraction osteogenesis as the initial surgery and arthroplasty or TMJ reconstruction as the second-stage treatment may achieve favourable results; however, some patients may need orthognathic surgery[16].

Sadakah A. et al. in 2006 included 9 patients in his study. This study's objective was to determine whether transoral bimaxillary distraction osteogenesis was feasible before applying intraoral mandibular distraction to release temporomandibular joint (TMJ) ankylosis. Along with a mandibular osteotomy on the afflicted side, a bilateral Le Fort I osteotomy was carried out (s). To preserve the preoperative dental occlusion, an intraoral distractor (or distractors) was placed in the lower jaw, followed by an intermaxillary fixation (IMF). After a latency period of 5-7 days, the distractor was actuated twice daily by 0.5 mm. A consolidation period of 6 to 8 weeks came next. Following a periauricular incision to remove the TMJ ankylosis, a gap arthroplasty was completed, and mandibular movement was restored once the IMF and distractor were taken out. Clinically and radiologically, the best outcomes were obtained with the fewest relapses and comorbidities. It is possible and perhaps preferable to distract a malformed mandible and maxilla intraorally before relieving TMJ ankylosis[17].

Li J. et al. in 2012 performed a study wherein twelve patients with bilateral TMJ ankylosis and micrognathia (ages 17 to 27) received arthroplasty as the first surgical surgery, followed by orthodontic care and the second surgical technique, osteodistraction and advancement genioplasty, to address mandibular micrognathia. Mouth opening, radiography, medical photography, and respiratory function were used to assess the clinical outcomes. A minimum of 8 months and a maximum of 36 months were spent following up with the patients. All of the patients' TMJ ankylosis was successfully relieved, increasing the average mouth openness from 3.3 mm preoperatively to 35.8 mm postoperatively. Interestingly, the micrognathia was fixed, and the obstructive sleep apnea and hypopnea condition was treated. Orthodontic treatment led to a satisfactory occlusion[18].

In 2019 Hakim-Hassan et al. conducted a retrospective study on 20 patients with mandibular hypoplasia secondary to TMJ ankylosis. They were managed by two-stage surgical strategy was used to treat them, with MDO coming after gap arthroplasty in the first stage. The integrity of the skeletal system and soft tissues, as well as the preservation of the MIO, were examined in the patients. Four time points were used to evaluate lateral cephalograms: prior to distraction (T1), following a consolidation period with or without genioplasty (T2), one year after consolidation (T3), and at the longest follow-up (T4) (T4). Statistical comparisons were made between the skeletal and soft-tissue alterations at various time points in each group. Gap arthroplasty without a costochondral graft was used to treat all ankylosed joints with the exception of three. Preoperatively, the MIO was 8.2 2.1 mm; postoperatively, it was 40.2 1.7 mm. Following the consolidation phase, MIO fell to 23 6.5 mm. After the distractions were removed, the patients were told to resume active physiotherapy in order to regain the pre-distraction MIO, which was sustained during the short-term follow-up. There was an average

follow-up of 8.5 + 1.5 years. At the conclusion of the follow-up, ankylosis had returned in two patients. After MDO with or without genioplasty, cephalometric examination showed significant improvements in the hard- and soft-tissue structures. All groups saw a number of severe long-term relapses, but none of them returned to their preoperative levels[19].

Obstructive sleep apnea syndrome (OSAS) is characterized by persistent nocturnal upper airway obstruction, which compromises cardiovascular function and makes people feel exhausted during the day. Surgical techniques for OSAS include laser-assisted uvulopalatoplasty (LAUP), uvulopalatopharyngoplasty (UPPP), genioglossus advancement, hyoid suspension, tongue base reduction, and maxillomandibular advancement in order to expand airway dimensions and minimise airway collapsibility (MMA). One of the best surgical methods for expanding upper airway dimensions has been identified as MMA[20].

To relieve the restriction in mouth opening and craniofacial deformity simultaneously, some surgeons have suggested doing interpositional arthroplasty and distraction osteogenesis at the same time. Three incidences of distraction osteogenesis and temporalis flap application occurring simultaneously were reported by Dean, Alamillos, Piero, and colleagues[9]. This would save the need for a second surgery and lessen the financial burden on the patient[21]. However, this method may have the following drawbacks: (1) the effect of distraction osteogenesis may be unsatisfactory due to the unstable condyle; (2) there may be conflict between physical activity and distraction; (3) a second surgery may still be necessary to remove the distraction devices; and (4) a single procedure won't completely resolve all of the issues faced by patients with OSAS[22]. Other surgeons have claimed that distraction osteogenesis should be done in the second stage after arthroplasty in the first to address the issue of limited mouth opening. In the initial phase, this procedure could restore the mandible's movement to guarantee proper food intake, assisting in preparing the patient for the ensuing, more involved procedures. In order to more precisely correct the deformity, surgeons could also modify the surgical plan based on the patient's specific circumstances. However, additional surgery might still be required, and addressing the deformity later on might make things more difficult[23].

Distraction osteogenesis is the first course of treatment advised by the current authors to restore the length and position of the mandible, followed by arthroplasty or TMJ reconstruction to restore the mouth opening. The two treatments mentioned above are preferable to this therapy sequence for the reasons listed below. First off, people with OSAS can safely follow this treatment plan. Before releasing the TMJ ankylosis in these individuals, distraction osteogenesis should be carried out so that the surgeons can simultaneously cure the mandibular bone deficit and expand the airway volume, making the subsequent surgery safer. If TMJ ankylosis was treated early, the shorter mandibular ramus and postoperative oedema would cause the already compromised airway space to worsen. This could result in severe apnoea and a rapid drop in blood oxygen saturation, necessitating an urgent tracheotomy in the immediate aftermath of surgery[24].

Along with lengthening the mandible, simultaneous interpositional arthroplasty with DO for TMJ ankylosis corrects severe facial asymmetry, occlusal cant, midline shift, and creates room for yet-to-erupt teeth. In our perspective, this approach for TMJ ankylosis helps shorten the duration of treatment, eliminates the need for a second operation (if they were performed as a two-stage procedure), and prevents the need for a second tracheostomy if fibreoptic intubation could not be performed. It lowers the price by reducing the number of procedures. The benefits of single stage simultaneous interpositional arthroplasty with DO are numerous, and neither orthognathic surgery nor costochondral graft can match or compete with its capacity to treat micrognathia brought on by TMJ ankylosis.

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