

Case Report

Bilateral fibro-osseous TMJ ankylosis in Marie-Strumpell disease: a case report

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ABSTRACT

Reporting a case of bilateral fibro-osseous TMJ ankylosis in Marie-Strumpell disease. Managed with interposition gap arthroplasty with temporalis myofascial flap. Marie-Strumpell disease or ankylosing spondylitis is a chronic inflammatory disorder affecting predominantly the axial skeleton although peripheral joint involvement may be a significant feature. Temporomandibular joint involvement in ankylosing spondylitis varies from 4% to 35%. A 53-year-old male patient reported to our department with a chief complaint of difficulty in opening his mouth and difficulty in eating food. The case was diagnosed as a case of bilateral fibro-osseous TMJ ankylosis associated with ankylosing spondylitis. The individual was managed with aggressive resection of ankylosed segments and inter-position of the temporalis myofascial flap bilaterally with early mobilization and aggressive physiotherapy. Aggressive resection of the ankylosed mass with inter-positional gap arthroplasty with early mobilization and aggressive physiotherapy is a highly effective and safe surgical management option for the management of TMJ ankylosis and with acceptable immediate and long-term outcome with minimum re-ankylosis and have produced satisfactory mandibular movements.

Keywords: Ankylosis, Temporomandibular joint, Marie-Strumpell disease, Temporalis myofascial flap

INTRODUCTION

Mandibular hypomobility results from a variety of disorders affecting temporomandibular joint (TMJ) and surrounding structures. It is classified according to the location (intra- or extra articular), type of tissue (fibrous, bony, or fibro-osseous), and extent of fusion (complete, incomplete).¹

In ankylosis, the normal architecture of the joint is altered. It is a severe disability that results in restricted mouth opening, difficulty in mastication, poor oral hygiene, rampant caries, and disturbances of facial and mandibular growth, speech, yawning, and nutrition, and overall health and quality of life of the individual. There may be associated restricted airway problems (obstructive sleep

apnea-hypopnea syndrome). When this disease occurs in a growing child, it mostly affects mandibular development, leading to complex and extensive dentofacial deformities with associated malocclusion.^{2,3}

Ankylosis may occur as a result of trauma (13-100%), local or systemic infections (0-53%), or systemic diseases like psoriasis, ankylosing spondylitis or Marie-Strumpell disease, rheumatoid arthritis.⁴

Ankylosing spondylitis (AS) also known as Bechterew's disease or Marie-Strumpell disease is a chronic inflammatory disorder affecting predominantly the axial skeleton although peripheral joint involvement may be a significant feature.⁵ The frequency of TMJ involvement in patients with ankylosing spondylitis (AS) has varied from

4% to 35%. It is more common in men and produces generalized stiffness in involved joints.⁶

Early diagnosis and aggressive surgical treatment are crucial if the worst sequelae of this condition are to be avoided. Our paper reports a case of bilateral fibrous-osseous ankylosis of the TMJ associated with Ankylosing spondylitis.

CASE REPORT

A 53-year-old male patient reported to our department with a chief complaint of difficulty in opening his mouth and difficulty in eating food for two years with a consistent decrease in mouth opening (Figure 1).

The individual was a known case of Ankylosing spondylitis for 11 years, for which he was being managed at the internal medicine department with NSAIDs. Physical examination revealed besides a weakly frail body, there was a limitation in the mobility of the spine with thoracolumbar kyphosis and no neurological deficit. He was unable to look up and unable to straighten himself while walking. On extra-oral examination, no facial asymmetry was noticed, and no scar or injury marks were seen. On palpation, no TMJ movements were appreciated while opening the mouth and there was no tenderness in the TMJ region. Intraoral examination revealed poor oral hygiene with inter-incisal mouth opening restricted to 8 mm. There were multiple grossly decayed teeth 18, 17, 48, 38, and 37 with generalized attrition and gingival recession. On the basis of clinical examination, the patient was diagnosed as a bilateral temporo-mandibular ankylosis related to ankylosing spondylitis with a differential diagnosis of trismus due to long-standing infection related to 38, 48. Radiographic examinations comprised of orthopantomogram and computed tomography (Figure 2 and 3).

An OPG revealed moderate anti-gonial notching with obliteration of right and left TMJ space, CT scan showed a coronal image with severe cortical irregularities in the mandibular condyle and temporal fossa, with no condylar translation at full mouth opening it also revealed a lack of structural organization in both TMJ. Based on clinical as well as radiographic findings, a diagnosis of bilateral fibrous-osseous TMJ ankylosis secondary to Ankylosing spondylitis was confirmed.

Initially, patient was managed conservatively with non-invasive, non-surgical physiotherapy with ice cream sticks for one month but with no positive results. Subsequently, the patient was advised for surgical intervention. The surgical procedure was explained to the patient and his relatives and a written consent of the patient was obtained. Surgical technique- the procedure was performed under general anesthesia, fiber-optic nasal intubation was done by a pediatric anesthesiologist, since the individual was a case of ankylosing spondylitis much manipulation of the neck was not be possible. Both the

TMJ was approached through Akayat and Bramley approach (Figure 4).⁷

The periosteum over the arch was incised horizontally and the joint entered through a T- shaped incision over the lateral capsule. After exposure, fibrous and osseous adhesions were identified and aggressive excision of 1.5 cm of condylar stumps was carried out, with special emphasis on the medial aspect to ensure complete resection of the ankylotic mass (Figure 5).

Precautionary, bilateral coronoidectomy and stripping of temporalis, medial pterygoid and masseter were performed through intra-oral incision after achieving adequate inter-incisal mouth opening. Inter-positing of finger-shaped temporalis myofascial flap bilaterally, by rotating over the arch into the joint was done as described by Pogrel and Kaban (Figure 6).⁸

The flap was sutured medially, anteriorly, and posteriorly with 4-0 proline sutures. Vacuum drains were placed on both sides, subsequently, wounds were closed in layers, and pressure with a crepe bandage was applied. Physiotherapy was commenced on the first postoperative day. No weakness with respect to the facial nerve was noted during the postoperative period.

Post-operatively injectable antibiotics (injection augmentin 1.2 g and injection metronidazole 400 mg) and injection diclofenac sod 75 mg IM were prescribed for 5 days followed by tablet combiflam (paracetamol and ibuprofen) SOS. Maxillo-mandibular fixation was maintained using guiding elastics and arch bars for six weeks. The patient was advised aggressive physiotherapy which consisted of active hinge opening, lateral excursions, and manual finger stretching in front of mirror four to five times a day for 20 min.

The individual was followed up for one year and an inter-incisal mouth opening of approximately 40 mm was maintained (Figure 7).



Figure 1: Severely restricted mouth opening.

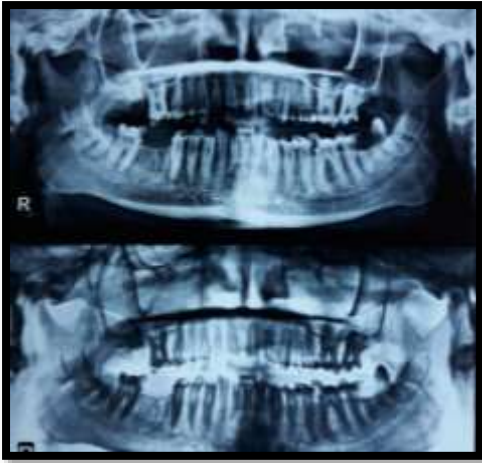


Figure 2: OPG showing obliteration of TMJ spaces bilaterally.



Figure 5: Bilateral removal of ankylotic mass.



Figure 3: CT Revealing the cortical irregularities and alteration of morphology- bilateral TMJ.



Figure 6: Placement of temporalis myofascial flap.



Figure 4: Surgical exposure of TMJ using alkayat and Bramley incision.



Figure 7: Adequate post-operative mouth opening.

DISCUSSION

TMJ involvement secondary to Ankylosing spondylitis has varied from 4% to 35%. Many theories have been postulated for the involvement of TMJ in Ankylosing spondylitis.

Davidson et al reported that the restricted mouth opening in patients with ankylosing spondylitis may be due to the proximity of the chin to the neck.⁵ Other studies reported that difficulty in opening the mouth may be due to the flattening and erosions of the mandibular condyle.⁷ Wenghoefer et al found that the limited jaw mobility in such patients might also be caused by an elongation of the mandibular coronoid process.⁸ Helenius et al reported that patients experience pain, stiffness, headache, and restricted movements in TMJ.⁹ Wenneberg and Kopp et al in 1982 reported that 13% of patients has restricted mouth opening, and 31% had tenderness of the TMJs compared with the control group (with 4% having restricted mouth opening and 1% having pre-auricular tenderness).¹⁰

Ramos-Remus et al found that ankylosing spondylitis had more variability in TMJ mobility than controls and showed an increased frequency of condylar erosions, flattening, sclerosis, and temporal flattening.¹¹ Resnick (1974) reported that 32% of his sample of 25 consecutive patients with long-standing ankylosing spondylitis had tomographic TMJ abnormalities.¹² The most common feature was joint space narrowing, followed by erosions, reduced mobility, osteophyte formation, excessive sclerosis, and extensive erosion.

TMJ ankylosis is a serious potential danger that can lead to airway embarrassment in the event of life-threatening odontogenic infections. Hence, prompt treatment should be initiated as soon as the condition is diagnosed, with the main goal of restoration of normal motion and functional harmonious jaw movements.¹³ There are three basic surgical goals in the treatment of TMJ ankylosis. These are to achieve functional joint movement, to prevent re-ankylosis, and to achieve normal growth and development in the case of children.

Topazian's et al review of gap arthroplasty without interposition reported a recurrence rate as high as 53%.¹⁴ To reduce the chance of re-ankylosis, some authors suggest radical condyle and neck removal as well as coronoidectomy.¹⁵ The main disadvantage of this is the loss of vertical ramal height. Re-ankylosis is less likely if material is interposed between the divided bone ends.¹⁶ Autogenic tissues have gained popularity over allogenic materials because the latter tend to initiate foreign body reactions.¹⁷ Interposition arthroplasty using locally available vascularised temporalis myofascial flap has been found to be a reliable method to prevent re-ankylosis with acceptable immediate and long-term outcomes especially in adults and costochondral grafts with fascia interposition in children.^{17,18} It obviates the

disadvantages of alloplastic materials as well as non-vascularised autogenous tissues.

In our case, we followed the surgical protocol developed by Kaban and Bouchard with few modifications for the treatment of Bilateral TMJ ankylosis with ankylosing spondylitis: (1) aggressive resection of the ankylosis segment, (2) bilateral coronoidectomy, (3) lining of the joint with temporalis fascia, (4) IMF with arch bars and guiding elastics for six weeks for achieving functional occlusion, and (5) early mobilization and aggressive physiotherapy which included active hinge opening, lateral excursions, and manual finger stretching in-front of mirror four to five times a day for 20 min.¹³

The biological and physiological basis for increasing the range of motion using dynamic exercises is well documented in literature. The potential benefits of TMJ opening and closing exercises are improved muscle vascularity, increased muscle mass and protein metabolism, decreased muscle fatigue and increased strength, reversal of the atrophic and degenerative changes within the joints, and restoration of the normal internal fibrous structure anatomy.¹⁹

CONCLUSION

Ankylosis of the TMJ is a seriously debilitating condition, which prevents normal feeding habits, impairs speech and causes facial deformity, and affects social life, but with proper diagnosis and early surgical intervention, with good patient compliance and with an intensive follow-up, the excellent results can be achieved.

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REFERENCES

1. Rowe NL. Ankylosis of the temporomandibular joint. J R Coll Surg Edinb. 1982;27(2):67-79.
2. Zhu S, Wang D, Yin Q, Hu J. Treatment guidelines for temporomandibular joint ankylosis with secondary dentofacial deformities in adults. J Craniomaxillofac Surg. 2013;41(7):e117-27.
3. Gundlach KK. Ankylosis of the temporomandibular joint. J Craniomaxillofac Surg. 2010;38(2):122-30.
4. Kaban L, Pogrel MA, Perrott DH. Complications in oral and maxillofacial surgery. 1st ed. Philadelphia: WB Saunders; 1996.
5. Davidson C, Wojtulewski JA, Bacon PA, Winstock D. Temporo-mandibular joint disease in ankylosing spondylitis. Ann Rheum Dis. 1975;34(1):87-91.
6. Arora P, Amarnath J, Ravindra SV, Rallan M. Temporomandibular joint involvement in ankylosing spondylitis. BMJ Case Rep. 2013;2013:bcr2013009386.
7. Locher MC, Felder M, Sailer HF. Involvement of the temporomandibular joints in ankylosing spondylitis

- (Bechterew's disease). *J Craniomaxillofac Surg*. 1996;24(4):205-13.
8. Wenghoefer M, Martini M, Allam JP, Novak N, Reich R, Bergé SJ. Hyperplasia of the coronoid process in patients with ankylosing spondylitis (Bechterew disease). *J Craniofac Surg*. 2008;19(4):1114-8.
9. Helenius LM, Hallikainen D, Helenius I, Meurman JH, Könönen M, Leirisalo-Repo M, et al. Clinical and radiographic findings of the temporomandibular joint in patients with various rheumatic diseases. A case-control study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2005;99(4):455-63.
10. Wenneberg B, Kopp S. Subjective symptoms from the stomatognathic system in ankylosing spondylitis. *Acta Odontol Scand*. 1982;40(4):215-22.
11. Remus C, Major P, Gomez-Vargas A, Petrikowski G, Hernandez-Chavez A, Gonzalez-Marin E, et al. Temporomandibular joint osseous morphology in a consecutive sample of ankylosing spondylitis patients. *Ann Rheum Dis*. 1997;56(2):103-7.
12. Resnick D. Temporomandibular joint involvement in ankylosing spondylitis. Comparison with rheumatoid arthritis and psoriasis. *Radiology*. 1974;112(3):587-91.
13. Kaban LB, Perrott DH, Fisher K. A protocol for management of temporomandibular joint ankylosis. *J Oral Maxillofac Surg*. 1990;48(11):1145-51.
14. Topazian RG. Etiology of ankylosis of temporomandibular joint: analysis of 44 cases. *J Oral Surg Anesth Hosp Dent Serv*. 1964;22:227-33.
15. Rajgopal A, Banerji PK, Batura V, Sural A. Temporomandibular ankylosis. A report of 15 cases. *J Maxillofac Surg*. 1983;11(1):37-41.
16. Barry M. The TMJ. Carthy M, eds. *Plastic surgery*. WB Saunder's: 1990; 1475-1513.
17. Ahmad QG, Siddiqui RA, Khan AH, Sharma SC. Interposition arthroplasty in temporomandibular joint ankylosis. *Indian J Otolaryngol Head Neck Surg*. 2004;56(1):5-8.
18. Abbas I, Jamil M, Jehanzeb M, Ghous SM. Temporomandibular joint ankylosis: experience with interpositional gap arthroplasty at Ayub Medical College Abbottabad. *J Ayub Med Coll Abbottabad*. 2005;17(4):67-9.
19. Chun-Li L, Yu-Chan K, Lun-Jou L. Design, Manufacture and clinical evaluation of a new TMJ exerciser. *Biomed Eng Appl Basis Comm*. 2005;17:135-40.

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