

# Unusually rare occurrence and recurrence of temporomandibular joint ankylosis and oral submucous fibrosis simultaneously in a patient: A surgeon's dilemma

### ABSTRACT

We here present a rare case of simultaneous occurrence and recurrence of TMJ ankylosis and submucous fibrosis in the same patient. Patient presented with limited mouth opening at both occasions. However, the diagnosis of submucous fibrosis was overlooked at the first presentation. The patient reported with recurrence of both the conditions after a period of 15 years. We presume that noncompliance of jaw exercises aggravated both the conditions as one led to the other. Since fibrosis sets in earlier than bony fusion, we theorized that the patient had acquired OSMF before TMJ ankylosis recurrence. The case was managed with release of fibrous bands intraorally followed by release of TMJ ankylosis and interposition with dermis fat graft. Physiotherapy is extremely essential when either pathology is treated in patients, especially when both are present in a single patient.

**Keywords:** Oral submucous fibrosis, temporomandibular joint ankylosis, TMJ ankylosis, Recurrence, Submucous fibrosis

### INTRODUCTION

Difficulty in opening the mouth or complete inability to do so can occur due to a number of reasons. Perhaps, we are here the first to describe a case report of bilateral temporomandibular joint (TMJ) ankylosis and oral submucous fibrosis (OSMF) occurrence in a single patient who was once treated for the same dual problem and again reported to us with recurrence of both the conditions.

It is said that "If you diagnose two rare conditions in the same patient, you are rarely correct." In this case, TMJ ankylosis was released the first time, but we failed to achieve any mouth opening, following which, fibrous bands were palpated intraoperatively and thus released, to achieve a full mouth opening.

The same patient reported again after 15 years with recurrence of both the conditions, and there was a quandary as to which condition led to recurrence of the other.

### CASE REPORT

A 40-year-old female came in the year 2000 with the chief complaint of reduced mouth opening for 2 years. She had a history of trauma due to fall at a young age.

Computed tomography scan had revealed bony bridging between the zygomatic arch and the condyles bilaterally,

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thus a diagnosis of bilateral TMJ ankylosis was made (Sawhney's type 3) [Figure 1].<sup>[1]</sup>

Ankylosis was released bilaterally, however, when the jaw stretcher was applied, no mouth opening was achieved, further examination revealed bilateral intraoral fibrous bands suggestive of OSMF, Type 4 (as per the Khanna and Andrade's classification).<sup>[2]</sup> Releasing incisions were taken intraorally to release the fibrotic bands. Mouth opening was achieved. Palatal island flaps were designed bilaterally by making an incision, 3–4 mm away from the gingival margin to midpalatal midline, and were raised posteriorly, based on the greater palatine artery. The flaps were rotated buccally, posterior to the second molar, and sutured to cover the raw areas. A specimen of the fibrotic tissue was sent for histopathological examination.

The biopsy showed severely atrophied epithelium, dense fibrosis and hyalinization of the connective tissue, and degenerating

muscle fibers in the subepithelial connective tissue thus confirming the diagnosis of submucous fibrosis [Figure 2].

The patient again reported after 15 years in 2016 with recurrence of both the conditions, the patient had failed to comply with jaw exercises following the first surgery [Figure 3]. This time, the fibrotic bands were released first. Fibrin glue was applied to cover the raw areas [Figure 4]. Dermis-fat interposition arthroplasty was done. Postoperatively, the patient is on a regular follow-up and has a stable mouth opening.

## DISCUSSION

Over the past 150 years, much has been said and done as far as TMJ ankylosis is concerned. Ironically, its cure still remains a conundrum. Similarly, OSMF, another disabling disease is characterized by stiffness of the oral mucosa, trismus, and frequent recurrent stomatitis and inability to eat.<sup>[3]</sup> The incidence of two different diseases occurring in the same patient is an exception rather than the rule.

To establish the etiopathogenesis as to what came first is challenging. In this case, the patient came to us with the recurrence

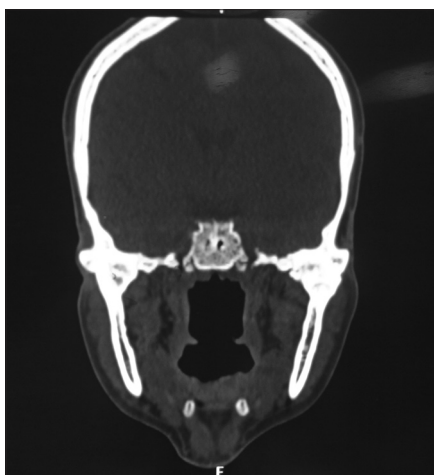


Figure 1: Computed tomography scan showing bilateral temporomandibular joint ankylosis

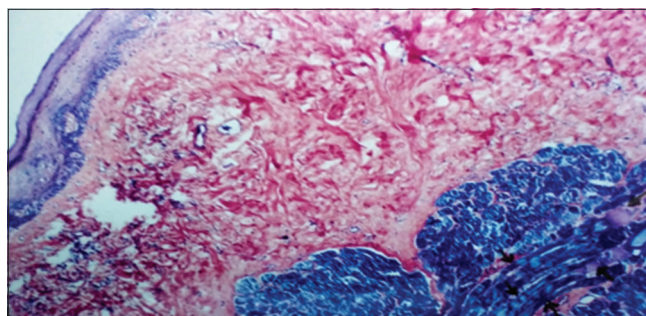


Figure 2: Histopathological photomicrograph showing severely atrophied epithelium, dense fibrosis, and hyalinization of the connective tissue



Figure 3: Postoperative showing satisfactory mouth opening



Figure 4: Fibrin glue applied to the raw areas

of both the entities, bilateral TMJ ankylosis and OSMF after 15 years. There was obvious frustrating fallout Intraoperatively in the first surgery, when we were unable to establish a mouth opening due to submucous fibrosis. Postoperatively, she reported being habituated to areca nut chewing, the role of which is well established in submucous fibrosis.<sup>[2]</sup>

At the first presentation, it is difficult to decipher the sequence of events, as the patient had a history of both trauma and areca nut chewing. The aspect of this case that is worth pondering is what recurred first and did one entity lead to the recurrence of the later. However, our assumption is that noncompliance of jaw exercises aggravated both the pathologies. Since fibrosis sets in earlier than bony fusion, we theorized that the patient had acquired OSMF before TMJ ankylosis recurrence.

Physiotherapy is extremely essential when either pathology is treated in patients, especially when both are present in a single patient. To avoid relapses, wide resection of the ankylotic mass is pivotal to establish the free movement of the joint.<sup>[4]</sup>

Although there is no interpositional graft material that fulfills all conditions, the purpose of using dermis-fat interposition was multifold. There is availability of ample interpositional material, can be passively placed, dermis acts like a scaffold to hold the graft.<sup>[5]</sup>

Treatment of submucous fibrosis is a standard involving surgical excision of fibrous bands with bilateral coronoidectomy. The highlights of using palatal flaps at the first surgery were considered to be low percentage of fibrosis, the donor area being in close proximity, and of a similar texture and color, low donor-site morbidity, and no secondary surgery for detachment of pedicle. Furthermore, because of the use of a mucoperiosteal flap pedicled to the greater palatine artery, the chances of shrinkage, sloughing, and contracture were considered to be minimal. However, due to the advent of various biological membranes and fibrin glue, palatal flaps have fallen into disfavor and are no longer used in our practice. We used fibrin glue for coverage of the raw surface, it is easy to apply and shows excellent healing. Over the years, fibrin glue has been used in various surgeries as a biological dressing.<sup>[6]</sup> Having both tissue adhesive and hemostatic properties, application of fibrin glue in these raw areas helps control intraoperative bleeding, accelerates postoperative healing, and reduced pain and discomfort, thus facilitating physiotherapy. The tissue adhesive property of fibrin glue was studied in 1940 by Young and Medawar.<sup>[7]</sup> Several homologous commercial preparations of fibrin glue are available such as Tisseel™, that was used in our case.

The ultimate treatment goal of restoring normal mouth opening and function poses a significant challenge to

surgeons due to lack of patient compliance and a higher recurrence rate in both the entities, that is, ankylosis and OSMF. Postoperative physiotherapy is truly the mainstay to prevent both re-ankylosis and submucous fibrosis.

## CONCLUSION

Recurrence of both the conditions in the same patient highlights the importance of establishing etiology and cause of recurrence in such cases. Although the case was unique, whether there was any correlation between the two conditions still remains a dilemma. Nevertheless, our goal was to prevent recurrence using dermis fat as interposition and regular jaw exercises.

Through this article, we also highlight the importance of complete intraoral checkup to rule out any possibility of OSMF. We also add that for every OSMF patient, an orthopantomogram is a must to rule out any TMJ ankylosis.

## Declaration of patient consent

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

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## Conflicts of interest

There are no conflicts of interest.

## REFERENCES

1. Su-Gwan K. Treatment of temporomandibular joint ankylosis with temporalis muscle and fascia flap. *Int J Oral Maxillofac Surg* 2001;30:189-93.
2. Khanna JN, Andrade NN. Oral submucous fibrosis: A new concept in surgical management. Report of 100 cases. *Int J Oral Maxillofac Surg* 1995;24:433-9.
3. Pindborg JJ, Sirsat SM. Oral submucous fibrosis. *Oral Surg Oral Med Oral Pathol* 1966;22:764-79.
4. Al-Moraissi EA, El-Sharkawy TM, Mounair RM, El-Ghareeb TI. A systematic review and meta-analysis of the clinical outcomes for various surgical modalities in the management of temporomandibular joint ankylosis. *Int J Oral Maxillofac Surg* 2015;44:470-82.
5. Dimitroulis G. The interpositional dermis-fat graft in the management of temporomandibular joint ankylosis. *Int J Oral Maxillofac Surg* 2004;33:755-60.
6. Rajurkar SG, Kazi N, Sharma A, Deshpande M, Meshram D, Ingole S. Fibrin glue-a sealant for oral and maxillofacial surgery. *Int J Curr Adv Res* 2017;6:6014-6.
7. Young JZ, Medawar PB. Fibrin suture of peripheral nerves. *Lancet* 1940;275:126.