

Oral Submucous Fibrosis: A Dual Case Study

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Abstracts: Oral submucous fibrosis (OSMF) is a potentially debilitating condition primarily associated with betel nut and tobacco use and drinking alcohol. This report presents two cases of OSMF, highlighting the importance of early diagnosis, appropriate management, and follow-up to prevent progression and complications. The discussion underscores the significance of patient education and cessation of harmful habits in halting the disease's advancement and the potential for malignant transformation.

Keywords: Premalignant Lesion, Oral Submucous Fibrosis, Oral Mucosa, White Lesion, Precancerous Disorder.

1. BACKGROUND

Oral submucous fibrosis (OSMF) is a complex and debilitating condition that primarily affects the oral cavity's mucosal lining [1]. It is characterized by the progressive accumulation of fibrous tissue beneath the oral mucosa, leading to a range of clinical symptoms and functional impairments. OSMF is most prevalent in regions where the consumption of betel quid, a mixture containing areca nut, slaked lime and tobacco is common [1][2]. It primarily affects adults in South and Southeast Asia, particularly in countries like India, Sri Lanka, Taiwan, Bangladesh and Pakistan. It is more common in males but is increasingly affecting women as well with the peak age of onset typically falling between 20 and 40 years [2].

The etiology of OSMF is intricate and multifactorial, driven by several contributing factors. Chief among these is the habitual consumption of areca nut, frequently in the form of betel quid. Areca nut contains alkaloids, such as arecoline, known to induce fibrosis and inflammation within the oral mucosa [3]. Furthermore, the use of tobacco products, either by chewing or smoking, significantly amplifies the risk of OSMF. The compounds present in tobacco, particularly in smokeless forms, synergize with areca nut to potentiate the fibrotic effects. Genetic factors have also been explored as potential contributors to susceptibility, although their specific role remains a subject of ongoing research. Additionally, nutritional deficiencies, environmental and cultural practices all contribute to OSMF's development. Understanding these multifactorial causes is crucial for effective prevention and treatment [2].

The clinical presentation of OSMF encompasses a spectrum of symptoms and signs, the severity of which can vary among affected individuals. A central feature is the reduction in mouth opening, a phenomenon referred to as trismus, owing to the formation of fibrous bands within the submucosal tissues. Patients often report a burning sensation in the oral cavity, particularly during the consumption of spicy or hot foods. Altered taste perception is another common complaint, where individuals may experience a loss or alteration in their ability to taste different flavors. Clinically, the oral mucosa takes on a pale or blanched appearance due to fibrosis, coupled with the presence of palpable fibrous bands [2].

Diagnosing Oral Submucous Fibrosis (OSMF) is a comprehensive process that combines clinical assessment and when necessary confirmatory tests. The initial step involves a thorough clinical evaluation conducted by a healthcare professional. This assessment is instrumental in identifying hallmark signs such as limited mouth opening, palpable fibrous bands, and mucosal changes, all of which are indicative of OSMF. Equally significant in the diagnostic process is the patient's history of areca nut, betel quid, and tobacco product usage, as this information plays a pivotal role in establishing a potential link to OSMF. [2][4]

In certain cases, particularly when confirming the diagnosis and assessing the extent of fibrosis is essential, a histopathological examination through biopsy of affected oral tissues may be recommended. Biopsy entails the collection of a small tissue sample that is subsequently examined under a microscope. Histopathological findings typically reveal substantial fibrosis, increased collagen deposition, inflammatory infiltrates, and epithelial changes. These microscopic changes align with the diagnosis of OSMF and serve to differentiate it from other oral conditions [2][4].

Grading systems are frequently employed to gauge the severity of OSMF. One of the most commonly used grading systems is Khanna and Andrade's grading system, which categorizes OSMF into four distinct grades based on clinical features and mouth-opening measurements. This grading system aids clinicians in classifying patients and devising appropriate management strategies [4].

Grade 1 OSMF encompasses less than one-third of the oral cavity, characterized by mild blanching, burning sensation, recurrent ulceration, stomatitis, and mouth dryness. Patients in this grade can achieve a mouth opening of up to 35 mm. Grade 2 OSMF extends to one-third to two-thirds of the oral cavity, presenting with blanching of oral mucosa, a mottled and marble-like appearance, palpable fibrotic bands, and involvement of the soft palate and premolar area. Mouth opening typically ranges from 25 to 35 mm, accompanied by a 33% reduction in cheek flexibility. Grade 3 OSMF involves greater than two-thirds of the oral cavity, exhibiting severe blanching, broad thick fibrous palpable bands at cheeks and lips, rigid mucosa, depopulated tongue, restricted tongue movement, shrunken bud-like uvula, floor of the mouth involvement, and lymphadenopathy. Mouth opening in this grade varies from 15 to 25 mm, with a 66% reduction in cheek flexibility. Grade 4 OSMF represents advanced stages, marked by leukoplakia changes, erythroplakia, ulcerating lesions, and suspicious malignant transformations. Mouth opening is severely limited, often falling below 15 mm or entirely absent [4].

Timely diagnosis and intervention during the initial stages of OSMF are paramount to prevent progression to more severe stages and the associated risk of malignant transformation. A comprehensive understanding of the various types, stages, and grading systems of OSMF is crucial for healthcare professionals to provide appropriate care, including patient education, cessation support, and tailored treatment strategies, ensuring effective management of this complex condition [4].

Managing OSMF entails a comprehensive approach aimed at arresting disease progression, alleviating symptoms, and mitigating the risk of malignant transformation. Crucial elements of management encompass immediate cessation of areca nut, betel quid, and tobacco product usage, though this often presents a formidable challenge due to the addictive nature of these substances. Patients may require counselling and robust support mechanisms to achieve successful cessation. Additionally, maintaining good oral hygiene is essential to prevent secondary infections and complications. Medications, such as corticosteroids and antioxidants, may be prescribed to manage symptoms and reduce inflammation. Physical therapy, in the form of physiotherapy, can assist in enhancing mouth opening and jaw mobility. Long-term follow-up is imperative to monitor disease progression and evaluate the potential for malignant transformation [2].

2. CASE PRESENTATION

Case 1: A 25-year-old male, with a history of chewing betel quid and tobacco for 5 years, sought medical attention due to progressive mouth-opening difficulty (limitation and pain), burning sensations in the oral buccal mucosa, and altered taste perception. During clinical examination, notable findings include :fair oral hygiene with green teeth staining, patient was CL II div II, a limited mouth opening, with an interincisal distance measuring 19 mm as shown in figure 1 (A)(B). Palpable fibrous bands were evident within the oral cavity, accompanied by the whiteness of the oral mucosa(patchy white lesion) as shown in figure 1 (C). These clinical manifestations collectively led to a provisional diagnosis of Oral Submucous Fibrosis (OSMF). This case underscores the importance of early diagnosis and intervention in OSMF patients.

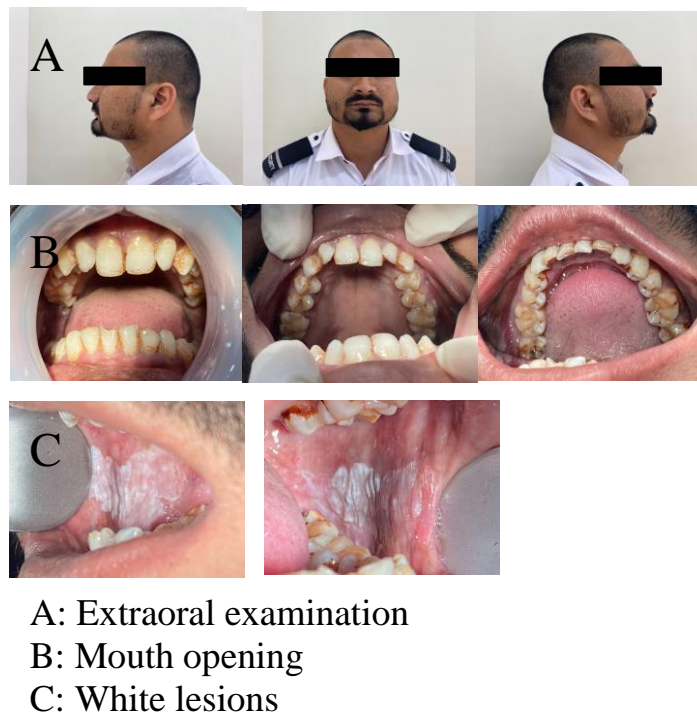


Figure 1. Clinical Assessment of case 1 with Oral Submucous Fibrosis (OSMF)

Case 2: a 45-year-old male presented for a routine check-up, reporting no specific complaints. Notably, he had successfully quit smoking, a habit that he had maintained for 15 years, approximately one year ago. However, during the clinical examination, palpable fibrous bands and mixed red and white lesions of the oral mucosa were observed as shown in figure 2. In addition to that, a limitation in mouth opening of 31mm was measured. A provisional diagnosis of OSMF was established based on these clinical findings along with his history.



Figure 2. Clinical Assessment of case 2 with Oral Submucous Fibrosis (OSMF)

3. INVESTIGATION

Both patients underwent a comprehensive clinical examination. Lymph node evaluation revealed non-significant changes. Assessment of the lesion, observation and palpation of the buccal mucosa revealed stiffness due to the presence of a fibrous band with limitation in the mouth opening. Histopathological examination of a biopsy specimen from the affected buccal mucosa revealed atrophic oral epithelium with excessive bony keratinization overlying the mucosa showing a thickened fibrocollagenous band. Prominent kerato-hyalinized granules were seen in figure 3 and some melanin pigmentation was also seen in the basal epithelial layer. There were no signs of atypical epithelial changes. Additionally, cone-beam computed tomography (CBCT) scans were performed to assess the extent of

fibrosis and evaluate any bony involvement in case 1 there was no bony involvement.

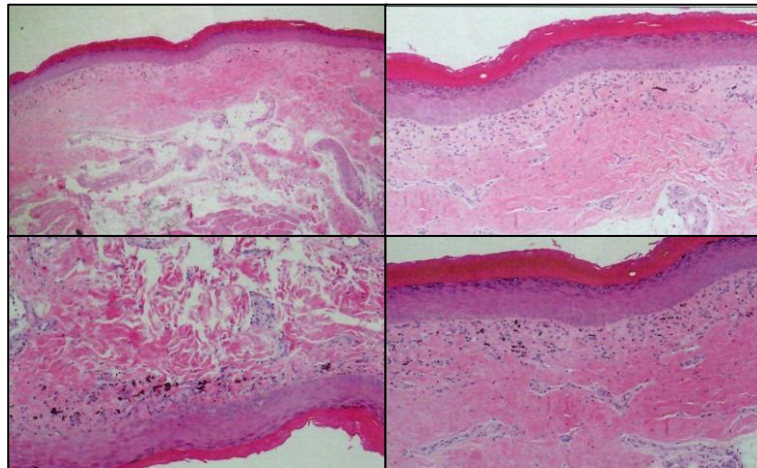


Figure 3. Histopathological Examination of Atrophic Oral Epithelium in OSMF

4. DIAGNOSIS

Case 1: Upon evaluating the clinical signs and symptoms presented by the patient, including difficulty in mouth opening, the initial differential diagnosis considered localized bulk lichen planus, squamous cell carcinoma and Oral Submucous Fibrosis (OSMF). However, after obtaining a comprehensive history revealing the patient's habit of chewing betel quid and tobacco for 5 years, coupled with the clinical findings of limited mouth opening, palpable fibrous bands and whitening of the oral mucosa, OSMF was confirmed as the provisional diagnosis. The history of betel quid and tobacco consumption aligns with the known risk factors for OSMF and histopathology confirmed the presence of OSMF.

Case 2: In this case, the differential diagnosis encompassed conditions such as lichen planus, pemphigus vulgaris, and leukoplakia due to the clinical presentation of palpable fibrous bands and blanching of the oral mucosa. However, the comprehensive evaluation, including clinical features, and consideration of the patient's history of tobacco consumption (15 years of smoking), collectively supported the diagnosis of Oral Submucous Fibrosis (OSMF). The association between OSMF and tobacco use, particularly in long-term smokers, played a pivotal role in confirming the provisional diagnosis.

These differential diagnoses underscore the importance of thorough clinical assessments and obtaining a detailed patient history to accurately diagnose and differentiate conditions with overlapping clinical presentations. In both cases, the final diagnosis of OSMF was reached based on a combination of clinical findings and relevant patient history.

5. TREATMENT

Patients' education about the danger of smoking and smokeless tobacco and chewing betel nut that long time use will result in a pre-malignant lesion which consequently can change to squamous cell carcinoma if the patient continues on his bad habit. Additionally, the patient was instructed to avoid spicy, warm and sour food and drinks, avoid tooth brushing and use alcohol-free mouthwash. Furthermore, to improve the mouth opening, the patients were instructed to use the wooden sticks (tongue blades) to be inserted between the incisal edges of the central incisors, starting with a comfortable height that does not exert excessive pressure on the temporomandibular joint and does not produce pain. This practice was repeated two times a day and gradually increased the number of wooden sticks whenever he was comfortable doing so. The patients were treated with dexamethasone injection 1CC mixed with Lidocaine with epinephrine 1/100000 to be injected at several points on each side of the lesion. An application of topical anesthesia gel was applied to the lesion a few minutes before injection. This treatment was repeated weekly

for 3 weeks with a remarkable improvement in the signs and symptoms in addition to the mouth opening.

6. OUTCOME AND FOLLOW UP

Throughout the course of treatment, the patient's progress and response to therapy have been closely monitored. During the initial visit of case 1, the patient presented with a limited mouth opening of 19 mm in figure 1 (B). As part of the prescribed exercise regimen, the patient began using 14 wooden sticks twice a day to aid in improving mouth opening. Additionally, during this visit, the patient received dexamethasone injections, each consisting of 1cc mixed with Lidocaine with epinephrine 1/100,000, administered at several points on each side of the lesion. At the second visit, there was a notable improvement, with the mouth opening measuring 23 mm (figure 4,A). The patient reported that using 15 wooden sticks was comfortable while using 16 wooden sticks caused some discomfort and mild pain. This signified an initial response to the treatment, a gradual expansion of jaw mobility, and a noticeable reduction in the lesion, with continued dexamethasone injections. The third visit demonstrated continued progress, with the mouth opening measuring 26 mm (figure 4,B). Inserting 16 wooden sticks had become challenging but was not painful, indicating ongoing improvement in jaw mobility and lesion reduction. Upon the fourth visit, the patient continued to exhibit positive developments. The mouth opening measured 27 mm (figure 4,C), and inserting 16 wooden sticks was now comfortable for him. Improvement in the lesion was evident in each visit, with a noticeable reduction in size and severity. On the fifth visit, the patient's mouth opening further improved, measuring 28 mm (figure 4,D). While the patient was able to insert 17 wooden sticks, it was noted that when the sticks were removed, some degree of discomfort and pain was experienced. This suggested ongoing progress but indicated that residual fibrosis and sensitivity persisted, emphasizing the importance of continued therapy.

These follow-up assessments underscore the effectiveness of the prescribed treatment plan in gradually improving mouth opening, reducing fibrosis, and addressing discomfort. Importantly, there was consistent improvement in the lesion in each visit, with a noticeable reduction in its size and severity. The patient's commitment to the recommended exercise regimen and continued dexamethasone injections have yielded tangible results. Regular follow-up visits will remain crucial in tracking the patient's journey toward enhanced oral function and overall well-being, with a focus on managing Oral Submucous Fibrosis (OSMF).

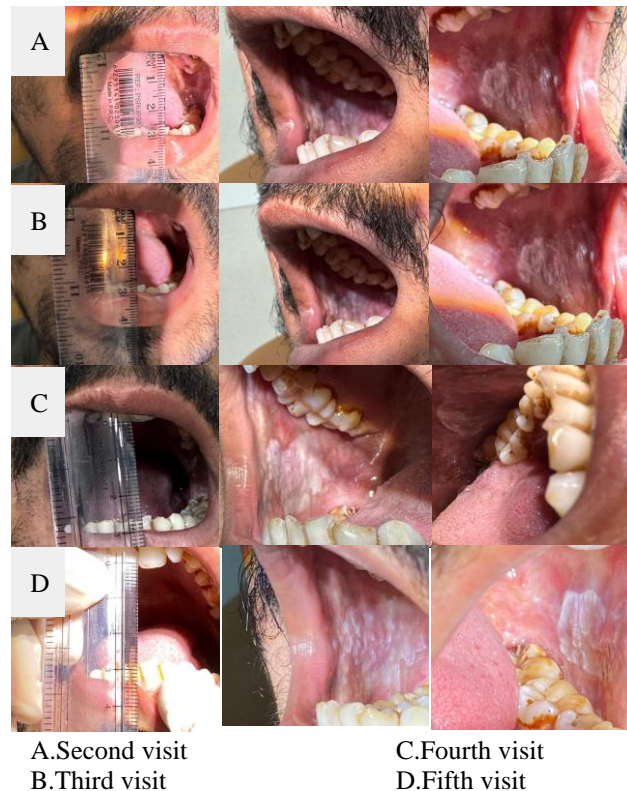


Figure 4. Management Progress in Case 1 with Ongoing Treatment

The initial visit of case 2 was presented with no pain or discomfort however stiffness was noted during examination of the patient. Upon examination of the patient, he presented with a mouth opening of 31mm (figure 5). On the second visit, the patient was given his first dexamethasone injections of 1cc combined with Lidocaine and epinephrine 1/100,000 were given to the patient at multiple points in both left and right cheeks. When the patient came for the third appointment no improvement had been noted. The patient mentioned that he had not been following the post-treatment instructions which could have led to the lack of progress in his case. He was then injected again with the same dose of dexamethasone at his next visit. After this the patient was not been able to come back to the clinic for a follow-up appointment, however, it can be concluded that due to the patient's carelessness, no progress was found. As mentioned before, follow-up appointments are crucial in order to track patient progress and monitor the case to prevent further deterioration of the disease.



Figure 5. Mouth limitation of case 2

7. DISCUSSION

The Oral submucous fibrosis (OSMF) is known to be an inflammatory disease that is characterized by the distinct widespread fibrosis of the submucosal oral soft tissues, which causes the oral mucosa to become noticeably hard, impairing one's ability to open the mouth and causes trouble projecting their tongue [5]. In the above cases, both patients had suffered from limited mouth opening, mostly case 1 which was recorded to be 19mm, as it was the patient's chief complaint. The mucosa becomes stiff and non-elastic as the condition worsens, severely limiting the patient's ability to open their mouth. Clinically, individuals have a blanching of the oral mucosa that is dull and marble-like in appearance. Stomatitis symptoms include erythematous mucosa, vesicles, mucosal ulcers, blotchy melanotic mucosal pigmentation, and mucosal petechiae may be visible in the early stages [5]. Vertical and round fibrous bands may be palpable in the buccal mucosa and in the pericommissural region as the condition worsens. The bands running in the blanched mucosa may give the tissue a mottled, marble-like appearance.[5]. These characteristics combined with both patients' long-term history of smoking can reach a conclusion to confirm the provisional diagnosis of OSMF. The oral mucosa of patients with OSMF feels burning, which is frequently made worse by spicy meals. Initially, increased salivation is seen, but as the condition worsens, salivary flow is reduced, leading to dry mouth and intolerance to spicy food, particularly chili [5]. The patient in case 1 had noticed his condition worsening with the consumption of spicy food in addition to an altered taste perception, in comparison to the patient in case 2 who had nothing to report in that regard. Both patients share the habit of smoking, case 1 has smoked tobacco and chewed pan for 5 years, but he stopped 3 years ago from his first visit. Patient 2 was a smoker for 15 years but stopped a year from when he came into the clinic. This is one of the important factors for the causation of the disease according to the literature. Coming to the histopathology of OSMF, the biopsy sample from the first case afflicted buccal mucosa exhibited atrophic oral epithelium with extensive bone keratinization overlaying the mucosa and a thicker fibro-collagenous band. There were prominent kerato-hyalinized granules and moderate melanin pigmentation in the basal epithelial layer which indicates OSMF. Case 1 made substantial progress in mouth opening since he kept all of his appointments and followed all of the post-operative instructions. The instructions included mouth-opening exercises with wooden tongue blades, avoiding spicy, warm, and sour foods, and total cessation of smoking. Both the dexamethasone injections along with the home aftercare worked hand in hand and produced improvement throughout the sessions when compared to the first visit. There is a significant variation in prognosis between cases 1 and 2. Case 2 did not follow the instructions or the appointments provided, resulting in a lack of progress in mouth opening when comparing the two cases. Following up with allocated appointments and abiding by post-operative instructions aids in reaching a better prognosis over time.

CONCLUSIONS

These two cases of oral submucous fibrosis (OSMF) underscore the importance of early diagnosis, appropriate management, and follow-up. OSMF is primarily linked to betel nut, tobacco, and alcohol use. Timely diagnosis and intervention, including patient education and cessation of harmful habits, are crucial to prevent progression and

potential malignancy. Treatment involves medication, oral hygiene, and physical therapy. Case 1 showed significant improvement due to adherence to post-treatment instructions, while Case 2's non-compliance hindered progress. These cases emphasize that early diagnosis and comprehensive care are essential for OSMF management, leading to improved oral function and overall well-being. Moreover, these cases highlight the significance of regular dental check-ups and comprehensive oral health assessments to detect and manage OSMF, even in patients with no overt symptoms.

CONFLICT OF INTEREST

The author declares no conflict of interest.

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