



## CASE SERIES

## Oral Submucous Fibrosis - Case Series

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## ABSTRACT

Oralsubmucous fibrosis is regarded as a collagen metabolic disorder with an overall increased collagen production and decreased collagen degradation resulting in increased collagen deposition in the oral tissues, and fibrosis due to alkaloid exposure. We report three case reports of oral submucous fibrosis. Clinical findings and histopathologic findings were consistent with the diagnosis of stage 2 oral submucous fibrosis. The patient was educated on the effects of areca nut and chances of malignant transformation.

**Keywords:** Malignant Transformation; Oral Submucous Fibrosis; Pathogenesis; Management

## 1 INTRODUCTION

OSMF is a chronic insidious disease characterised by progressive submucosal fibrosis of the oral cavity and the oropharynx<sup>(1)</sup>. In 1966 Pindborg defined OSMF as “An insidious chronic disease affecting any part of the oral cavity and sometimes pharynx”<sup>(2)</sup>.

The disease manifests as blanching of the mucosa, burning sensation of the oral mucosa and sometimes with appearance of vesicles in the mucosa. A distinguishing feature of the condition is the appearance of fibrous bands in the lips, cheeks, and soft palate that causes restricted mouth opening. OSMF is histopathologically characterized by fibrosis that affects the oral cavity pharynx, and also the upper part of the oesophagus

The aetiology of the disease is multifactorial and various factors have been reported to cause OSMF that includes betel quid chewing, smokeless tobacco, local irritants like chillies, nutritional deficiency.

The primary cause of OSMF is now known to be the areca nut alkaloid arecoline. Arecoline seems to have a role in the pathogenesis of OSMF by promoting fibroblast growth and

elevating collagen synthesis<sup>(1)</sup>.

OSMF is one of the oral potentially malignant disorder with the highest rate of malignant transformation. Paymaster in 1956, first described the malignant potential of OSMF and had reported that an estimated malignant transformation rate of 7-13%<sup>(3,4)</sup>.

Various treatment modalities have been used for treating OSMF with varied outcomes. Management of OSMF currently is focussed on habit cessation, alleviating symptoms of burning sensation and restricted mouth opening. Various agents that have been used to treat and manage OSMF with that includes iron and multivitamin supplements, as well as a variety of drugs such as collagenase, pentoxifylline, lycopene, chymotrypsin, human placenta extracts, intraleisional injection of steroids, and hyaluronidase<sup>(5)</sup>.

## 2 CASE SERIES

## 2.1 Case 1

A 47year-old female patient came to the department with complaint of burning sensation on having spicy food

since two years. Visual analogue score was 6 for burning sensation. patient gives history of eating arecanut 2-3times per day since two years. On intra-oral examination, pale, blanched appearance of right and left buccal mucosa extending anterior-posteriorly from the retro-commissure area up to the retro -molar area and superior-inferiorly extending from approximately 5 mm above and below the line of occlusion. Marble stone appearance of mucosa with generalized tobacco stains on teeth and hard palate was also observed. Vertical bands were palpable in the buccal mucosa with respect to molar region on right side and the retro-molar area. perioral bands were palpable with respect to the mandibular labial mucosa. Uvula was shrunken and tongue movements was normal. Mouth opening was 35mm (Figures 1, 2 and 3).



**Fig. 1: Intra-oral picture of Blanching of the Buccal Mucosa**



**Fig. 2: Intra-oral picture of Blanching of the Palatal Mucosa**

Based on the clinical findings a provisional diagnosis of STAGE II Oral Sub mucous Fibrosis (classification according to Khanna JN et al<sup>(6)</sup>) was made.

Patient was referred to oral and maxillofacial surgery for further treatment. Incisional biopsy was done and was send for histopathological evaluation. On the basis of Histopathological findings the final diagnosis was Oral Sub mucous Fibrosis with mild dysplasia.



**Fig. 3: Incisional Biopsy specimen**

### 2.1.1. Treatment

Patent counselling was done and was advised to quit the habit and the capsule of SM fibro was prescribed once daily for 3 months. Treatment given-intralesional injection Hyaluronidase twice a month.

The patient was on follow up for 3 months. The visual analogue score for burning sensation was reduced to 2 and there was resolution of the lesion and reduction in palpable bands.

### 2.2 Case 2

A 40 year old male patient came to the department with a chief complaint of reduced mouth opening since 6 months. The patient stated that his mouth opening has gradually reduced over the last six months. The patient stated that he developed a burning sensation in the mouth on consuming spicy and coarse food in the past three months. Visual analogue score was 4 for burning sensation. The patient gives history of gutkha chewing with slaked lime since 6 years. On oral examination multiple white keratotic patch was present on the commissure of the lip of left buccal mucosa measuring approximately 2x2cm in size with generalized melanosis surrounding mucosa appeared normal. Single fibrous Vertical bands were palpable in the buccal mucosa with respect to left posterior molar region . Uvula was shrunken. Tongue movements was normal (Figures 4, 5 and 6).

Based on the clinical findings a provisional diagnosis of STAGE II Oral Sub mucous Fibrosis with leukoplakia of left buccal mucosa (classification according to Khanna JN et al<sup>(6)</sup>). Incisional biopsy was taken. On the basis of histopathological findings the final diagnosis was oral submucous fibrosis with mild dysplasia.

### 2.2.1. Treatment

Patent counselling was done and was advised to quit the habit and the capsule of SM fibro was prescribed once daily for 3 months. Treatment given-intralesional injection



Fig. 4: Biopsy site at right buccal mucosa



Fig. 5: Marking of biopsy site on right buccal mucosa



Fig. 6: Incisional Biopsy specimen

tobacco stains on teeth and hard palate. Vertical bands were palpable in the right and left buccal mucosa with respect to left and right posterior molar region and the retro-molar area. Perioral band were palpable with respect to the maxillary and mandibular labial mucosa. Uvula was shrunken tongue movements was restricted. Based on the clinical findings a provisional diagnosis of STAGE II Oral Sub mucous Fibrosis was made. Incisional biopsy was taken. On the basis of Histopathological findings the final diagnosis was Oral Sub mucous Fibrosis with mild dysplasia (Figures 7, 8 and 9 )



Fig. 7: Biopsy site at right buccal mucosa



Fig. 8: Marking of biopsy site on right buccal mucosa

Hyaluronidase twice a month

The patient was on follow up for 3 months. The vas score for burning sensation was reduced to 1 and there was resolution of the lesion and reduction in palpable band

### 2.3 Case 3

A 47year-old male patient brought to the department with complaint of burning sensation in mouth on having spicy food since two years. There was a history of eating arecanut 4-6times per day for the last one year. On Intra Oral examination, pale, blanched appearance of floor of the mouth , right and left buccal mucosa extending from the retrocommisural area up to the retro molar area anterior posteriorly and superior inferiorly from approximately 5 mm above and below the line of occlusion. Marble stone appearance of mucosa was also observed with generalized



Fig. 9: Incisional Biopsy specimen

### 2.3.1. Treatment

Patent counselling was done and was advised to quit the habit and the capsule of SM fibro was prescribed once daily for 3 months. Treatment given-intralesional injection Hyaluronidase twice a month

The patient was on follow up for 3 months. The vas score for burning sensation was reduced to 1 and there was resolution of the lesion and reduction in palpable bands

## 3 DISCUSSION

Oral submucous fibrosis is regarded as a collagen metabolic disorder with an overall increased collagen production and decreased collagen degradation resulting in increased collagen deposition in the oral tissues, and fibrosis due to alkaloid exposure<sup>(7,8)</sup>. Oral Sub mucous Fibrosis was first described by SCHWARTZ in 1952 among five East African women of Indian origin<sup>(8,9)</sup>. Oral submucous fibrosis is seen predominantly in Asians who chew betel nut quid or its variants such as gutkha (mitha pan), kiwam, zarda and pan masala. The easy availability of packaged dried product such as gutkha, is increasing the incidence of this condition, as it consists of a greater dry weight of areca nut quid along with other OSMF producing ingredients than the traditional quid rolled in betel leaf (paan). Only betel nut chewing does not cause OSMF, contribution of other ingredients like slaked lime is more important in the causation of OSMF<sup>(10,11)</sup>. Areca nut chewing, smoking tobacco, smokeless tobacco and hypersensitivity to chillies are the causative agents in genetically predisposed patients. OSMF mostly affects the buccal mucosa, lips, retro molar areas, soft palate. It is a chronic condition characterized by mucosal rigidity of varying intensity due to fibro-elastic transformation of the juxta-epithelial layer<sup>(4,5)</sup>. This leads to restricted mouth opening when the tongue is involved, its protrusion may be impaired. Early lesion present as a blanching of buccal mucosa, imparting a marble-like appearance but later lesions reveal palpable fibrous bands rendering the mucosa pale, rigid and stiff. This leads to progressive inability to open the mouth, burning sensation, pain and dysphagia. Most affected individuals report a burning sensation of the oral mucosa aggravated by spicy food. The presence of palpable fibrous bands is a diagnostic criterion for this condition. Iron and vitamin B12 deficiency has been associated particularly in conjunction with other factors<sup>(7,12)</sup>. Oral submucous fibrosis can be seen at any age, but is rare in young children. The predominant age group affected is 20-40 years. Compared to traditional betel nut quid, gutkha chewing tends to begin at a younger age and has a shorter time to the development of disease, so cases of OSMF have begun to appear in younger age group. OSMF is a potentially malignant condition<sup>(13,14)</sup>. The malignant transformation rate is found to be 4.5–7.6%<sup>(7,12)</sup>. The possible precancerous nature of OSMF was first described by Paymaster, who observed the occurrence of squamous cell carcinoma in one-

third of his patients with the disease .

In his study From a total of 3627 intraoral malignant tumours, 650 cases of histologically proven buccal mucosal cancer was observed between the years of 1941 and 1947 in Indian population. According to his study intraoral and pharyngeal neoplasms together account for 45 per cent of malignant transformation<sup>(4,5)</sup>.

The management of OSMF is aimed at habit cessation, providing symptomatic relief, improvement in mouth opening and prevention of malignant transformation. Abundant published research is available in the medical literature pertaining to application of various medications like corticosteroids, anti-oxidants, pentoxifylline, hyaluronidase, placentrix and adjuvants that include curcumin, aloe vera, sesame oil in the management of OSMF<sup>(15-17)</sup> .

However the results have not been convincing and there is lack of robust scientific evidence. This has led researchers to explore newer modalities in the management of OSMF of which, Lasers has been largely utilized in the management of OSMF particularly fibrotomy, but have yielded controversial results.

### 3.1 Malignant potential of OSMF

In 1956, Paymaster first estimated the OSMF's malignant potential rate to be 7–13%. A recent study predicted that 2-8% of cases with OSMF that progress to squamous cell carcinoma (SCC) are malignant. In India and other south-east Asian nations, the frequency of OSMF varies from 0.2-1.2%. With production and import totalling 622,260 and 80,419 tonnes, respectively, in the fiscal year 2013–2014, India is both the greatest consumer and producer of areca nuts. In India, where betel nut, betel leaf, and tobacco use are more common, mouth cancer is more likely to occur, is more prevalent, and is associated with higher morbidity and mortality<sup>(3,15)</sup> .

Primary prevention has been proved to be achievable and effective in decreasing the incidence of OSMF, thus efforts must be undertaken in that regard<sup>(18)</sup>.

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