Case Report

ORAL SUBMUCOUS FIBROSIS: A CASE REPORT

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Abstract

Oral submucous fibrosis (OSMF) is potentially malignant condition initially described in 1950s is commonly found in Indians and Asian population as the habit of betel chewing is common in this geographical area. It is a chronic progressive disorder with arecoline in areca nut as main etiological factor and presentation of clinical features depends upon the stage of disease. This report presents a clinical case of a 39 years old female patient presented with difficulty in mouth opening and burning sensation while eating which was diagnosed as Moderately Advanced Oral submucous fibrosis.

Keywords: Arecanut, Oral submucous fibrosis, Pathogenesis, Potentially malignant disorder.

INTRODUCTION

Oral submucous fibrosis is one of the chronic, insidious, incapacitating ailments encompassing oral mucosa, the oropharynx, and seldom involving, the larynx. It is reported primarily in the Indian population, having been long established in Indian literature since the time of inception of the Sushruta. It was defined by Pindborg and Sirsat an insidious, chronic disease that affects any part of the oral cavity and sometimes the pharynx. It may occasionally be preceded by and/or associated with the formation of vesicles. However, it is always definitely associated with a juxtaepithelial inflammatory reaction followed by a fibroelastic change in the lamina propria and epithelial atrophy that leads to stiffness of the oral mucosa and causes trismus and an inability to eat. The earliest description of this disease was given by Schwartz in 1952, who had coined the term atróphica idiopathic mucosa oris describing an oral fibrosing disease. Joshi et al had subsequently termed the condition as oral submucous fibrosis. Epidemiological data studies suggest that areca nut is the main aetiological factor for OSMF. Other etiological factors include chillies, tobacco, lime, nutritional deficiencies like iron, zinc,
immunological disorders, and collagen disorders.  
Arecoline has a definitive role in the pathogenesis of OSMF including fibroblastic proliferation and increased collagen formation (Figure 1). Of all other factors, Connective Tissue Growth Factor (CTGF) is chiefly associated with the onset and progression of fibrosis in many of the human tissues. Arecoline stimulated CTGF synthesis takes place through an interaction between ROS, JNK, NF-κappa B and p38 MAPK pathway. It also leads to an increase in the production of TIMP-1. The interaction between arecoline and keratinocytes has been reported to have induced the differentiation of myofibroblasts from the unspecific fibroblasts.  
Areca nut has also been found to contain tannin and copper. Tannin and copper have the ability to stabilize the collagen by cross-linking it. The collagen produced due to their combined action of tannin and copper is highly resistant to remodeling and phagocytosis.  

CASE REPORT
A 39 years old female came to the Department of Oral Pathology and Microbiology with the chief complaint of burning sensation in mouth on consuming spicy food since one year (Figure 2). She also reported difficulty in mouth opening. She had the habit of chewing supari, 2 packets/day since 6 years. On inspection, blanching was seen in the right and left buccal mucosa along with palate. Diffuse white lesion of size approximately 3.5 cm x 2 cm was present on the buccal mucosa extending from the retromolar area to the premolar area bilaterally (Figure 3). A reduced mouth opening was observed, with the interincisal width being 21 mm. The buccal mucosa was rubbery and inelastic. Vertical fibrous bands were present bilaterally on buccal mucosa posteriorly. On the basis of case history and oral examination a provisional diagnosis of oral submucous diagnosis was made put forth with differential diagnosis of Scleroderma, Leukoplakia, Oral lichen planus, and Iron deficiency Anemia. Blood investigations were done and all values were within normal limits. An incisional biopsy was performed under local anesthesia (Figure 4) and a soft tissue specimen measuring 0.8x 0.5 x 0.5 cm approximately and white in color was received for histopathological examination (Figure 5). Histopathological examination revealed parakeratinised stratified squamous epithelium showing atrophy along with loss of rete ridges. The underlying connective tissue stroma showed dense collagen fibers and many constricted blood vessels. Few chronic inflammatory cells predominantly being lymphocytes were present. Adipocytes and muscle tissue were also seen (Figure 6). Based on these histopathological findings, final diagnosis of moderately advanced Oral submucous fibrosis was made.

DISCUSSION
OSMF 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. In our case also patient had habit of chewing supari (arecanut). Under the influence of areca nut, fibroblasts differentiate into phenotypes that produce more collagen. The male-to-female ratio of OSMF varies by region, but females tend to predominate. As in our case also, a female patient was reported. Burning sensation and blanching of oral mucosa with reduced mouth opening is the first and foremost diagnostic features. Blanching is caused by impaired local vascular bed as a result of increased fibrosis and results in a marble-like appearance of oral mucosa. Our patient also reported with burning.
Figure 1: Pathogenesis of oral submucous fibrosis.

Figure 2: Extraoral photograph of the 39-years-old patient
Figure 3: Intraoral photographs showing blanching of left buccal mucosa and palate (a) and right buccal mucosa (b).

Figure 4: Photograph showing incisional biopsy w.r.t left buccal mucosa, Figure 5: Detailing of the specimen.

Figure 6: Histopathological sections showing parakeratinised stratified squamous epithelium with atrophy along with loss of rete ridges. Connective tissue stroma shows dense collagen fibers and many constricted blood vessels. Few chronic inflammatory cells predominantly being lymphocytes are present. Adipocytes and muscle tissue are also seen. (a)x40, (b)x100, (c)x400.
sensation in mouth difficulty in mouth opening. Clinical features like blanching of buccal mucosa and palate were present. Our case was diagnosed as moderately advanced OSMF as it shows similar histopathological features like thickened collagen bundles, less fibroblastic response, constricted blood vessels and presence of lymphocytes. 

CONCLUSION
This case report describes a 39 years old female patient with chronic habit of supari chewing who was diagnosed with moderately advanced OSMF. Considering the malignant potential of the disease, early diagnosis and proper management is essential in reducing the mortality of oral cancer.

REFERENCES
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