Oral Submucous Fibrosis In A Young Boy

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Citation

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Abstract

Shushrutha in ancient medicine described a condition, “Vidari” under mouth and throat diseases. He noticed a progressive narrowing of mouth, depigmentation of oral mucosa & pain on taking food. Schwart in 1952, described five Indian women from East Africa with “Atrophia idiopathica (tropica) mucosae oris”. In 1953, SG Joshi coined the term Oral submucous Fibrosis for a similar condition. The disease predominantly affects people of South-East Asian origin.[1] We describe a case of a 9 year old child with this condition.

INTRODUCTION

Shushrutha in ancient medicine described a condition, “Vidari” under mouth and throat diseases. He noticed a progressive narrowing of mouth, depigmentation of oral mucosa & pain on taking food. Schwart in 1952, described five Indian women from East Africa with “Atrophia idiopathica (tropica) mucosae oris”. In 1953, SG Joshi coined the term Oral submucous Fibrosis for a similar condition. The disease predominantly affects people of South-East Asian origin.[1]

Pindborg JJ & Sirsat SM in 1966, defined Oral Submucous fibrosis (OSF) as “an insidious chronic disease affecting any part of the oral cavity & sometimes the pharynx. Although occasionally preceded by &/or associated with vesicle formation, it is always associated with juxta-epithelial inflammatory reaction followed by a fibro-elastic change of the lamina propria with epithelial atrophy leading to stiffness of mucosa causing trismus & inability to eat”.[2] OSMF is a premalignant condition which is defined by World Health Organization as “a generalized pathological state of the oral mucosa associated with a significantly increased risk of cancer”. The possible precancerous nature of OSF was first described by Paymaster, who observed the onset of slowly growing squamous cell carcinomas in one-third of such patients.[3]

CASE REPORT

A 9 year old male patient reported to the Department of Oral Medicine & Radiology I.T.S C.D.S.R. with the chief complaint of restricted mouth opening (2.5cm, Figure1) and burning sensation on taking hot and spicy food. The patient had the habit of chewing tobacco in a smokeless form, 1-2 packets since last 2-3 years. He also had the habit of keeping quid in the buccal sulcus, 2-3 times per day. Intra-oral examination revealed bilateral fibrous bands on the buccal mucosa on palpation. Blanching of mucosa (Figure 2 & 3) was also evident.

Figure 1

Figure 1: Showing restricted mouth opening

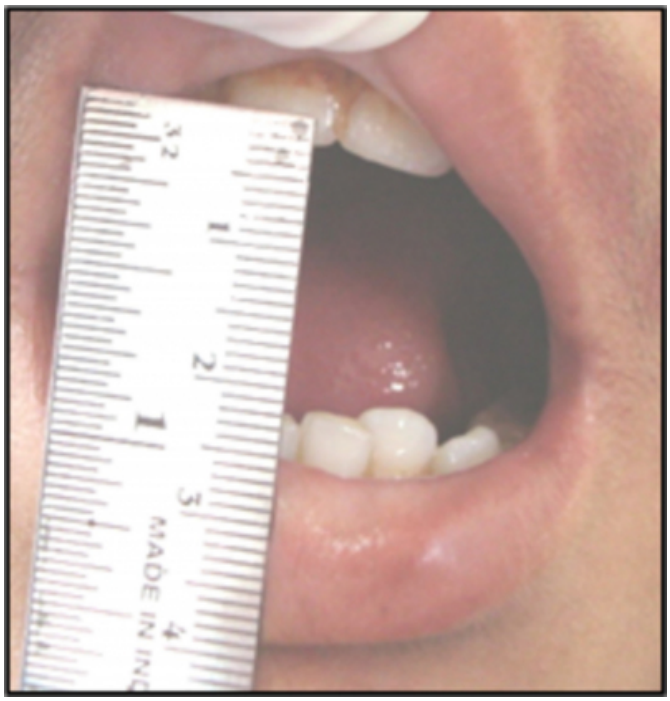


Figure 2

Figure 2: Blanching of labial mucosa

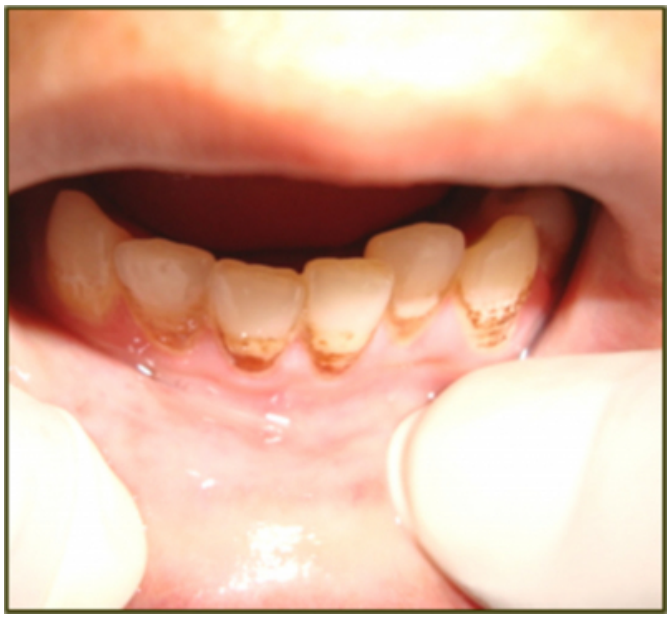
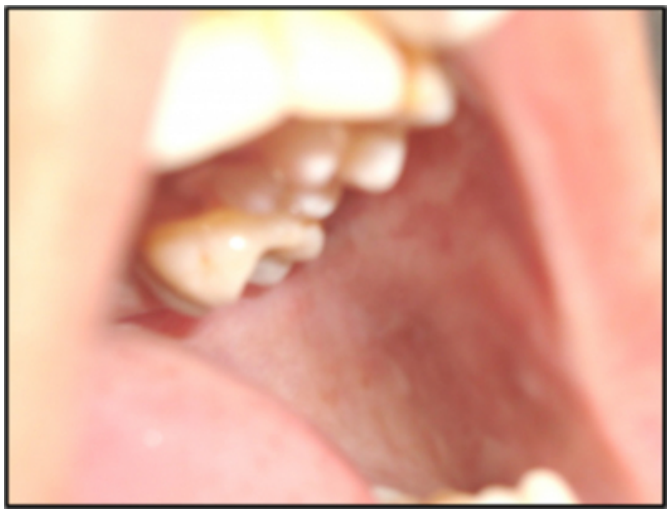


Figure 3

Figure 3: Blanching on the buccal mucosa



A provisional diagnosis of oral submucous fibrosis was made. An incisional biopsy was taken from the same region.

Incisional biopsy from the buccal mucosa revealed an atrophic keratinized stratified squamous epithelium. Hyalinization of the connective tissue and occasional chronic inflammatory cell infiltration is evident in the subepithelial region. Perivascular chronic inflammatory cell infiltrate was also seen (Figure 4). Dense bundles of collagen was seen extending deep into muscle fibres (Figure 5). A diagnosis of oral submucous fibrosis-Moderately advanced stage was made.

Figure 4

Figure 4: Photomicrograph showing atrophic keratinized stratified squamous epithelium. Subepithelial Hyalinization is evident(X10, H&E). Perivascular chronic inflammatory cell infiltrate is also seen (Inset. X40 H&E).

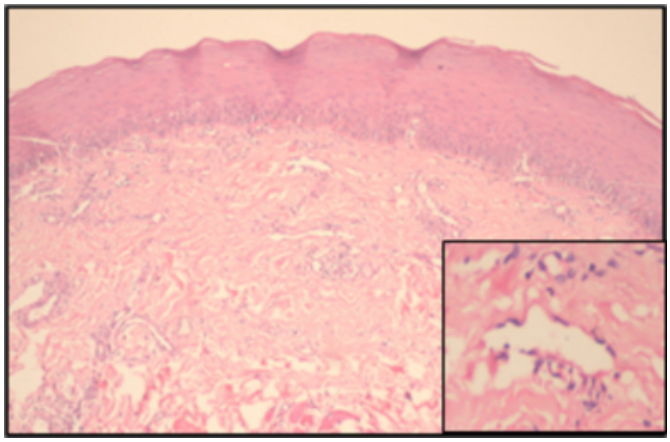
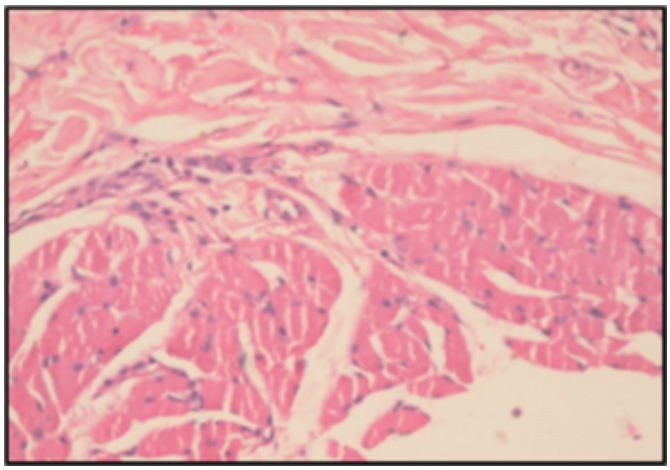


Figure 5

Figure 5: Photomicrograph showing dense collagen bundles extending into and between the muscle fibres (X40, H&E).



DISCUSSION

Hypersensitivity to chillies, betel nuts and nutritional deficiencies have all been suggested at various times to account for the pathogenesis of Oral Submucous Fibrosis. The pathogenesis of the disease is believed to be multifactorial.[4] A combination of factors such as genetic alterations, role of infectious agents such as Candida and viruses, carcinogenic agents associated with arecanut and tobacco which are known to have genotoxic effects can best explain the pathogenesis of this unique condition. Additionally, the pre-conditioning of the oral mucosa by various deficiency states make it more vulnerable to the effects of carcinogens in the pathogenesis of this disease. Clinically, the condition commonly affects individuals

between 20 -40 years. In India, general female preponderance is observed. This may be attributed to deficiency state of iron and B complex deficiency.[5]

The onset of the disease is insidious over 2 to 5 year period. Oral submucous fibrosis is a well recognized condition in adults, although there are few reported cases of the condition in children. Yusuf & Yong described the occurrence of the disease in a 12 year old child.[6] Johnson NW et al, in a case-control study showed the prevalence of the disease in a substantial number of young men.[7]

The present case also highlights the occurrence of OSF in a young boy of 9 years. The etiology for such case appears to be genetic susceptibility. No precise genetic basis for OSF have been identified However, the incidence of malignant transformation in patients with OSF ranges from 3-19.1%[6] indicating that genetic abnormalities such as altered/amplified oncogene expression can be implicated in the pathogenesis of OSF. Genetic predisposition can further render the oral mucosa more susceptible to chronic inflammatory changes on exposure to carcinogens derived from betel quid components including tobacco. Another etiologic factor is autoimmunity. Many connective disorders, including rheumatoid arthritis and systemic lupus erythematous have been associated with unique HLA-DR antigens, a similar association has been sought for OSF. Elevated levels of immunoglobulins G and A have been found in serum of patients with OSF. Similar finding is seen in scleroderma, a disease resembling OSF histologically.[8]

CONCLUSION

Thus from the data currently available on OSF, the role of genetic abnormalities still needs to be defined for OSF, particularly affecting younger individuals. In near future, research into genetic, viral and immunologic aspect should provide an excellent model for studying genetic-environmental-immunologic-nutritional interactions in disease pathogenesis. Finally, to prevent occurrence of the disease in younger individuals there is a need for improved primary prevention in different community groups as well as targeted initiatives for groups or individuals at risk.

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