A Case Report of a Patient Suffering From Oral Submucous Fibrosis

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Abstract
A case of oral submucous fibrosis occurring in a 51 year old man is presented. This paper reviews the aetiology, clinical presentation and treatment modalities of oral submucous fibrosis. This case highlights the link between oral submucous fibrosis and the regular use of areca-nut (pan) and the newer trans-cultural oral tobacco products. The case report underlines the danger that human face with products which are clearly targeted at them by the tobacco industry.

Keywords: Oral submucous fibrosis, irritants, genetic, auto-antibodies

1. Introduction
Oral mucosal lesions associated with betel quid, areca nut and tobacco chewing habits, were reviewed at a consensus workshop in Kuala Lumpur, Malaysia in 1994\(^1\). Criteria and guidelines were proposed to define, describe and identify lesions such as submucous fibrosis (OSMF) which had been described three decades earlier by Pindborg and Sirsat\(^2\) as a chronic insidious disease affecting any part of the oral cavity and may extend to the pharynx and the oesophagus, and may be preceded or associated with vesicle formation. It is always associated with juxta-epithelial inflammation and followed by fibro-elastic change of the lamina propria with epithelial atrophy leading to stiffness of the oral mucosa and causing trismus. OSMF is seen most frequently in communities resident in the Indian sub-continent and has a reported incidence of between 0.2 – 1.2% of the urban population attending dental clinics\(^3\). The condition predominantly affects women with a female: male ratio of 3:1 and characteristically first presents in adulthood between the ages of 45 – 54 years\(^4\). Cases of OSMF have also been reported in individuals of South Asian origin living outside the sub-continent but it is extremely rare in White populations\(^4\). The present report describes a case of OSMF presenting in a 51 year old male of India.

2. Case Report
A 51 year old male patient came with a chief complain of reoccurrence of ulcers in oral cavity. In addition, the patient suffered discomfort and a burning sensation affecting the oral mucosa, particularly when eating spicy foods (Fig. 1).
Medical history indicated previous deficiency of both iron and vitamin B12. The iron and vitamin B\textsubscript{12} deficiencies were fully investigated. There was no abnormality on barium meal and follow-through. The patient was receiving regular iron supplements and vitamin B\textsubscript{12} injections. Previous special investigations for the possible presence of Crohn's disease were negative. The patient confirmed that there was a long history of tobacco chewing among both parents: the mother is a non-smoker but consumes chewing tobacco 2 - 3 times per day; the father both smokes and chews tobacco. The patient only occasionally chews betel-quid but regularly chews supari (a sweetened form of areca-nut). This has occurred since the age of 30 years. The patient is aware of the new tobacco products (Gutkha) but did not use these on a regular basis. The parents and all the other family members were unaware of the harms of supari or chewing tobacco products, believing them to be a natural product which aid digestion. Intra-oral examination revealed a number of ulcers that were characteristic of recurrent minor aphthous stomatitis. Oral hygiene was poor and the dentition required some restoration. No other soft tissue or mucosal abnormalities were detected at the first examination. Management involved reassurance and advice on cessation of areca-nut chewing, and chlorhexidine mouth rinse. Further examination revealed that the buccal mucosa was pale and fibrous bands were palpable within the buccal mucosa. Mouth opening was reduced to 25 mm. A clinical diagnosis of OSMF was made and the patient was once again strongly advised to stop the consumption of areca-nut. Prednisolone as 5 mg dispersible tablets was prescribed, to be used as a mouthwash three times daily when ulcers occurred. The oral discomfort had led to poor oral hygiene and there was evidence of pericoronitis associated with a lower second molar. The patient was prescribed metronidazole tablets 200 mg three times daily for 5 days and instructed to use chlorhexidine 0.2% as a mouthwash twice daily.

3. Discussion

The aetiology of OSMF remains uncertain and at the present time there is evidence suggesting a combination of factors is likely to be involved. The main proposed factors include the following:

Areca-nut is traditionally chewed throughout India as pan supari\textsuperscript{5}. The mixture is held adjacent to the buccal mucosa and slowly chewed over a long period of time. Tissue culture studies using human fibroblasts by Harvey\textsuperscript{6}, suggests that areca-nut alkaloids, particularly acroleine and arecaidine, were involved in causing OSMF. Furthermore it was demonstrated that extracts of areca-nut stimulated collagen synthesis by 170\% over the control studies. Many other experimental studies have also shown a strong correlation between OSMF and areca-nut chewing. Iron and vitamin B\textsubscript{12} deficiency has been implicated particularly in conjunction with other factors. In 1919 Paterson and Brown-Kelly independently described the condition of chronic dysphagia and mucosal atrophy in women who had chronic anaemia – this was later termed sidero-penic anaemia or Brown-Kelly-Paterson Syndrome and has the potential for cancerous change in the oropharynx\textsuperscript{7}.

An increased frequency of HLA-10, DR-3 and DR-7 has been noted in patients with OSMF. There would appear to be a predisposition in females with a ratio of women to men of 3:1. Studies have shown raised levels of a number of immunoglobulins including IgA, IgE and IgD. Auto-antibodies to gastric, parietal and thyroid microsomal and antinuclear antibodies have also been found in 65\% of patients with OSMF. Diagnosis of OSMF is usually based on the clinical signs and symptoms, which include oral ulceration, burning sensation (particularly with spicy foods), paleness of the oral mucosa,
and occasional leukoplakia. The most characteristic feature is the marked vertical fibrous ridge formation within the cheeks, and board like stiffness of the buccal mucosa. The fibrosis in the soft tissue leads to trismus, difficulty in eating, and even dysphagia. Biopsy of the tissue is rarely performed due to the observation that such investigation results in further fibrous scar formation and worsening of the symptoms. At the present time there is no cure for OSMF and management consists of elimination of the ingestion of implicated irritants. Successful prevention in the early stages of the condition has been shown to produce improvement in symptoms.

The close association between areca-nut chewing and the development of OSMF with a risk of oral cancer has been demonstrated in a number of studies. A case control study showed that the condition only occurred among those who chewed areca-nut in one form or another. However, this observation is still not fully conclusive and other factors such as genetic and nutritional influences also have a role to play. As there is no cure for the condition education and prevention seem the only way to reduce the risk of OSMF in the South Asian community. The association of the condition with the development of oral cancer highlights the importance of education to limit OSMF. 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 OSMF. Subsequent studies have reported that the incidence of carcinoma varies in OSMF from 2–30%. More recently the chewing of gutkha; a sweetened mixture of tobacco and betel-nut, has increased in this country and its use is thought to be commercially aimed at children. An active preventative approach is required to hopefully limit the potential for the development of oral malignancy.

4. Conclusion

OSMF described in the present case was difficult to manage in part caused by the age and poor oral hygiene maintenance. In addition he was found to be a poor attender and this made monitoring and managing the earliest stages of the condition difficult. It seems inevitable that OSMF is likely to worsen since he would not appear to be able to stop the ingestion of areca-nut. If the condition does worsen, he may in the long term need surgical intervention with grafting and there is always the possibility of malignant change and therefore close monitoring of his oral mucosa is essential.

References: