Case Report

Indian Journal of Dental Education
Volume 6 Number 4, October - December 2013

Oral Submucous Fibrosis Transforming into Carcinoma: A Case Report

Sushruth Nayak*, Sandhya Shrivastava, Prachi Nayak***, Rolly gupta****, Aparajita Beera*****

Abstract

Oral submucous fibrosis is a chronic fibrotic disease of the oral cavity and oropharynx associated with the chewing of the areca quid. This condition occurs predominantly among Indians and to a much lesser extent in other Asian population. The condition affects large population of the Indian sub continent. Interestingly the prevalence and the incidence rates are high in South India where the incidence of oral cancer is also very high. Betel quid (BQ) chewing is a popular oral habit with potential links to the occurrence of oral cancer. A malignant transformation rate of 11.7% is reported with Oral submucous fibrosis (OSMF) which was seen predominantly in males (87%). Most cases with malignant transformation in OSMF had occurred gradually over a long period of time. Here we are presenting a rare case of OSMF which developed carcinoma within a short span of 4 months, thus giving evidence for the aggressive potential of OSMF. In spite of extensive studies, the pathogenesis and other aspects of oral submucous fibrosis remains unclear till today.

Keywords: Oral submucous fibrosis; Areca nut; Carcinoma.

Introduction

Sushruta, the father of ancient Indian Medicine described a condition termed "Vidari" under mouth and throat diseases, characterised by progressive narrowing of mouth and burning sensation, particularly on eating spicy food and depigmentation of oral mucosa.[1] All these are the characteristic features of oral submucous fibrosis. "Atrophia idiopathica (trophica) mucosae" was the first name given to oral submucous fibrosis by Schwartz in 1952.[1,2] Lal in 1953, named the condition as "diffuse oral submucous fibrosis". [3] From that time it has remained as an enigma in spite of innumerable studies done over the past five decades.

Author's Affilation: *Asso. Professor, **Professor & Head, ***Reader, ****Senior Lecturer, *****Post Graduate student, Department of Oral and Maxillofacial Pathology & Microbiology, Chhattisgarh Dental College and Research Institute, Rajnandgaon 491441, Chhattisgarh, India.

Reprints Requests: Dr. Sushruth Nayak, MDS, Asso. Professor, Department of Oral and Maxillofacial Pathology & Microbiology, Chhattisgarh Dental College and Research Institute, Rajnandgaon-491441, Chhattisgarh, India.

E-mail: sushruthnayak@yahoo.com

The major presenting complaint is a progressive inability to open the mouth due to accumulation of inelastic fibrous bands in the juxta epithelial region of the oral mucosa. This severity impairs eating and oral hygiene care. Nasopharyngeal involvement may cause palato-pharyngeal incompetence and deafness and fibrosis within the oesophagus and may present as dysphagia. The epithelium overlying the fibrous condensation become atrophic in 90% of cases and is the site of malignant transformation in 5-10% of cases. Histologically, this condition bears resemblance to localized scleroderma in which epithelial atrophy and dermal fibrosis are associated with a chronic inflammatory infiltrate.[4]

The buccal-mucosa, soft palate and the retro-molar area are commonly affected sites. The mucosa in these regions develops a blotchy marble like pallor and progressive stiffness of sub-epithelial tissue. When tongue is involved it becomes rather immobile, frequently diminished in size and often devoid of papillae. Fully developed cases show sub mucous fibrotic bands that are palpable on the buccal mucosa, soft palate and labial mucosa.[5]

The possible precancerous nature of Oral submucous fibrosis (OSMF) was first

mentioned by Paymaster in the year 1956. A malignant transformation rate of 11.7% is reported with OSMF which was seen predominantly in males (87%).[6] The malignant transformation rate of OSMF has been reported to be around 7.6% over a 17-year period.[7] We are presenting a rare case of OSMF which developed carcinoma within a short span of 4 months, thus giving evidence for the aggressive potential of OSMF.

Case Report

A 45 year old female patient reported to the department with the chief complaint of inability to eat and decreased mouth opening since 3 to 4 months. Mouth opening was initially 3 fingers wide which later reduced to 1 finger wide. Patient also had the complaint of a painful ulcer on the right buccal mucosa since 1 month for which she took medication. Medication did not reduce the pain nor heal the ulcer. She had the habit of gutkha chewing since 10 years about 3 to 4 packets per day.

Figure 1: Palpable Bands with Ulceration

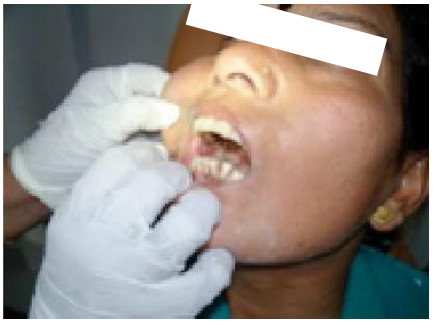


Figure 2: Reduced Mouth Opening

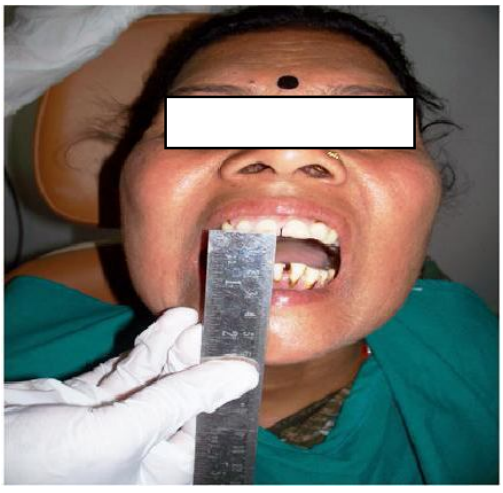


Figure 3: Grossed Specimen

Intraoral examination of the lesion proper revealed blanching of the right and left buccal mucosa with palpable vertical bands and an ulcerated lesion with yellowish slough on it in the right buccal mucosa (Figure 1). The ulcer was located in the retromolar region with regular borders and fixed to the underlying tissue. The Submandibular lymph nodes were normal on palpation. Mouth opening was reduced with interincisal distance being 1.4 cm (Figure 2).

All the hematological examinations were under normal range except for Hb% which was on a lower level (7.2 gm%). Incisional biopsy was performed and sent for histopathological examination. On macroscopic examination the lesion measured

Figure 4: Atrophic Epithelium Under 40x

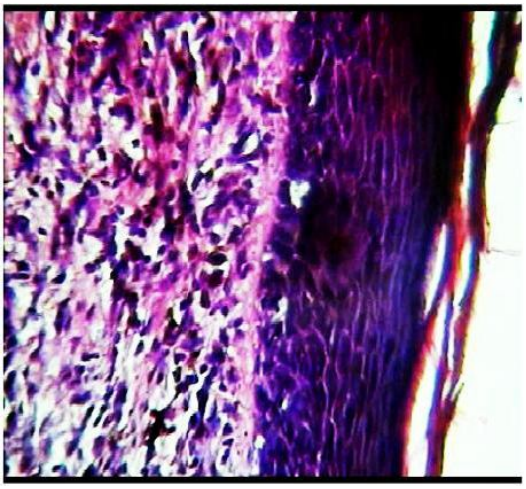
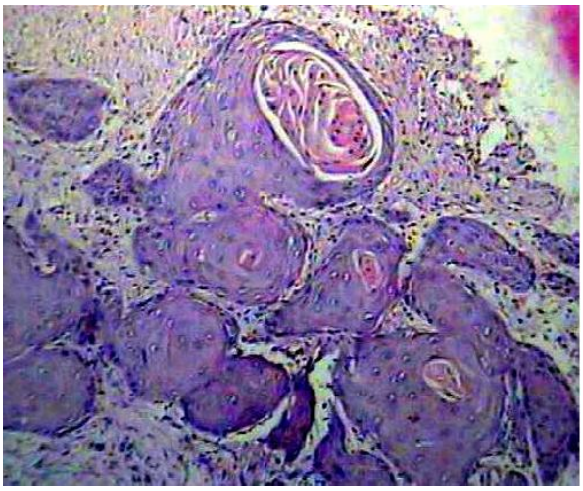


Figure 5: Epithelial Islands Within the Connective Tissue Under 10x



about 0.6 x 0.5 cm, creamish brown in color, soft to firm in consistency with regular margins (Figure 3). The H&E stained section revealed the presence of parakeratinised stratified squamous epithelium overlying the connective tissue component. The epithelium was atrophic without any rete ridge formation (Figure 4). The underlying connective tissue was composed of strands and islands of epithelial cells showing dysplastic features like keratin pearl formation, cellular and nuclear pleomorphism and increase number of nucleoli (Figure 5). Based on the histopathological findings the diagnosis of Oral submucous fibrosis transforming into well differentiated squamous cell carcinoma was made.

Discussion

Oral sub-mucous fibrosis is a chronic condition of the oral mucosa, first described among the five East African women of Indian origin under the term, atrophia idiopathica (tropica) mucosae oris. Since then this condition has also been described as, idiopathic scleroderma of the mouth, idiopathic palatal fibrosis and sclerosing stomatitis. In India this condition was first described as diffuse oral sub-mucous fibrosis and as sub-mucous fibrosis of the palate and pillars. Oral submucous fibrosis occurs predominantly among Indians and to a much lesser extent in other Asiatic people.[3]

It is a well-recognized potentially malignant condition of the oral cavity. Besides being regarded as a precancerous condition, it is a seriously debilitating and progressive disease. [8] Once the disease has initiated it is not amenable to reversal at any stage of the disease process even after cessation of the habit. It causes significant morbidity (in terms of loss of mouth function as tissues become rigid and mouth opening becomes difficult) and mortality (when transformation into squamous cell carcinoma occurs).[9]

Previous studies on the pathogenesis of oral submucous fibrosis have suggested that the occurrence may be due to;

- Clonal selection of fibroblasts with a high amount of collagen production during the long-term exposure to areca quid ingredients.
- Stimulation of fibroblast proliferation and collagen synthesis by arecanut alkaloids.
- By fibrogenic cytokines secreted by activated macrophages and T-lymphocytes in oral submucous fibrosis tissues.
- By decreased secretion of collagenase.
- Deficiency in collagen phagocytosis by oral submucous fibrosis fibroblasts.
- By production of collagen with a more stable structure (collagen type-I trimer) by oral submucous fibrosis fibroblasts.
- By stabilization of collagen structure by (+) catechin and tannins from the arecanut.
- By an increase in collagen cross linkage as caused by upregulation of lysyl oxidase by oral submucous fibrosis fibroblasts.

Genetic susceptibility may also be associated with oral submucous fibrosis because raised frequencies of HLA-A10, B_7 and DR_3 are found in oral submucous fibrosis patients compared to normal subjects. Further HLA typing done

by the use of the polymerase chain reaction (PCR) also demonstrates significantly increased frequencies of HLA-A24, DRB1-11 and DRB3—0202/3 antigens in oral submucous fibrosis patients.[10]

Diagnosis of OSF 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.[11] The potentially malignant nature of this condition has been well documented. A malignant transformation rate of 7.6% over a period of 10 years was described in an Indian cohort and the relative risk for malignant transformation may be as high as 397.3.[12] A malignant transformation rate of 11.7% is reported with OSF which was seen predominantly in males (87%).[6]

The present case is a rare one since the OSMF developed into carcinoma within a short span of 4 months, thus giving evidence for the aggressive potential of OSMF.

Conclusion

It is obvious from the present case that oral sub mucous fibrosis should be regarded as a premalignant condition that predisposes to the development of oral cancer. There is more need for public awareness about the potential complications of betel quid & tobacco chewing and transformation risk into carcinoma.

References

1. Gupta SC, Yadav YC. "Misi" an etiologic factor in oral submucous fibrosis. *Indian Journal of Oto Laryngology.* 1978; 30(1): 5-6.
2. Mohd Akbar. Oral submucous fibrosis – A clinical study. *JIDA.* 1976; 48(9): 365-73.
3. Daftary DK, Murthi PR, Bhonsle RB, Gupta PC, Mehta FS, Pindborg JJ. Oral precancerous lesions and conditions of tropical interest. In: Prabhu SR, Wilson DF, Daftary DK, Johnson NW, editors. Oral diseases in the tropics. Oxford: Oxford University Press; 1992, 417 – 22.
4. Haque MF, Meghji S, Khatib U, Harris M. Oral submucous fibrosis patients have altered levels of cytokine production. *J Oral Pathol Med.* 2000; 29: 123-28.
5. Neville BW, Damm DD, Allen CM, Bouquot JE. Epithelial pathology. Oral and maxillofacial pathology. 2nd ed. Philadelphia: Elsevier; 2002, 350.
6. Punnya V Angadi KP, Rekha. Oral submucous fibrosis: a clinicopathologic review of 205 cases in Indians. *Oral Maxillofac Surg.* 2011; 15: 1–9.
7. Gupta PC, Bhonsle RB, Murti PR, Daftary DK, Mehta FS, Pindborg JJ. An epidemiologic assessment of cancer risk in precancerous lesions in India with special reference to nodular leukoplakia. *Cancer.* 1989; 63: 2247–52.
8. Nair U, Bartsch H, Nair J. Alert for an epidemic of oral cancer due to use of the betel quid substitutes gutkha and pan masala: a review of agents and causative mechanisms. *Mutagenesis.* 2004; 19(4): 251–62.
9. Ajit A, Miriam PR, Lewei Z, Sumanth KN. Oral Submucous Fibrosis, a Clinically Benign but potentially Malignant Disease: Report of 3 Cases and Review of the Literature. *JCDA.* 2008; 74(8): 735-40.
10. Rajendran R. Benign and malignant tumors of the oral cavity. In: Rajendran R, Shivapathasundaram, editors. SHAFER'S Textbook of ORAL PATHOLOGY. 5th ed. New Delhi: Elsevier; 2006, 136-39.
11. Hayes PA. Oral submucous fibrosis in a 4-year-old girl. *Oral Surg Oral Med Oral Pathol.* 1985; 59: 475-8.
12. Gupta PC, Nandakumar A. Oral cancer scene in India. *Oral Dis.* 1999; 5: 1-2.