ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)

IJDSIR : Dental Publication Service

Available Online at: www.ijdsir.com

Volume – 3, Issue – 5, October - 2020, Page No. : 45 - 52

Oral Submucous Fibrosis a Disease with Malignant Potential in Left Retromolar Region – A Case Report

[1]Dr. Sowmya Hemantha Kumar, [2]Dr. Arathy Lankupalli .S, [3]Dr. K. Saraswathi Gopal

Meenakshi Ammal Dental College and Hospital, Chennai, Tamil Nadu – 600095

Corresponding Author: Dr. Sowmya Hemantha Kumar, Meenakshi Ammal Dental College and Hospital, Chennai, Tamil Nadu – 600095

Citation of this Article: Dr. Sowmya Hemantha Kumar, Dr. Arathy Lankupalli .S, Dr. K. Saraswathi Gopal , " Oral Submucous Fibrosis a Disease with Malignant Potential in Left Retromolar Region – A Case Report", IJDSIR- October - 2020, Vol. – 3, Issue - 5, P. No. 45 – 52.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Oral submucous fibrosis (OSMF) is a higher risk precancerous condition characterized by changes in the connective tissue fibers of the lamina propria and deeper parts leading to stiffness of the mucosa and restricted mouth opening. Patients have marked difficulties in chewing, swallowing and speaking. It largely occurs in Indians and other population of the Indian subcontinent with certain oral habits. In patients with submucous fibrosis, the oral epithelium becomes atrophic and thereby becomes more unguarded to carcinogens. It is currently accepted that chewing areca is the main etiological factor for developing OSF. The atrophic epithelium shows first an intercellular edema and later epithelial atypia associated with moderate epithelial hyperplasia. From then on, carcinoma may develop any time. It is suggested that submucous fibrosis should be regarded as a condition that causes predisposition to the development of oral cancer. Here we are presenting a case of oral submucous fibrosis showing malignant potential and development of oral squamous cell carcinoma.

Keywords: Oral squamous cell carcinoma, Oral sub mucous fibrosis, Areca Nut, Malignant potential

Introduction

Cancer is one of the main causes of death in all communities, its relative position ranges with age and sex. Worldwide, oral cancer is the sixth most common cause of cancer related- death, although many people are unknowing of its existence [1]. In the International classification of diseases (ninth revision- WHO), oral cancer is classified under the rubrics 140 (lip), 141 (tongue), 143 (gingiva), 144 (floor of the mouth), and 145 (other parts of the mouth) [2]. Of all the histologic variants of oral cancer, oral squamous cell carcinoma is the fifth most common cancer globally [3]. The high occurence of oral carcinomas in India can be presented in part by the habit of tobacco and betel quid chewing [4]. OSCC is a also major source of morbidity and mortality in

the Indian subcontinent.Oral precancer is characterized by WHO (World Health Organization) into 'precancerous lesions' (e.g. Leukoplakia, Erythroplakia) and 'precancerous conditions' (Eg. Oral sub mucous fibrosis, Lichen Planus). It has been catalogued in literature and regularly observed in clinical practice that many cases of OSCC are associated with or preceded by precancerous lesions and conditions for a varying length of time[5]. They distribute the same etiologic factors with oral cancer, particularly the use of tobacco, and manifest the same site and habitrelationships. These precancerous lesions and conditions can consequently serve as a paragon for investigating the chemopreventive approach for controlling oral cancer. The identification and management of precancers, hence, comprise a vital oral cancer control measure[6,7].Oral sub mucous fibrosis (OSMF) may be defined as an insidious, chronic disease affecting any part of the oral cavity and sometimes pharynx. Although occasionally preceded by and/or associated with vesicle formation, it is always associated with a juxta—epithelial inflammatory reaction followed by fibro elastic change of the lamina propria, with epithelial atrophy leading to stiffness of oral mucosa and causing trismus and inability to eat. Overall, estimates of OSMF exhibit a confinement to Indians and Southeast Asians, with global frequency rate in India to be about 0.2–0.5 % and prevalence by gender varying from 0.2 to 23% in males and 1.2–4.57 % in females [8]. It has been recommended that ingestion of chillies, genetic susceptibility, nutritional deficiencies, altered salivary constituents, autoimmunity and collagen disorders may be involved in the pathogenesis of this condition. The state is well recognized for its notable malignant potential, the incidence of which varies from one region to another, as revealed by a number of studies [9]. Pindborg [10] in 1980, from a sequence of epidemiological surveys conducted in India and South Africa found malignant change in 3 to 6 % of patients having OSMF. In 1985, Murti et al. [11] assessed malignant transformation rate of 7.6 % in a 10-year mean observation period.In perspective of increasing number of patients of OSCC who have related OSMF in the clinical practice, the incidence of OSCC associated with OSMF appear to be much higher than that reported in the literature.

Case Report

A 54year old male patient (fig:1) came to the op with a chief c/o of pain in left lower back tooth region for past 10 days. History revealed that, the Pain is intermittent, radiating in nature. Aggravates upon consuming hot and spicy food.On personal history, patient gives a h/o chewing smokeless tobacco since 15 years, 4-5 packs/day,stacks in his left and right buccal vestibule for 10-15 mins and spits it out and alcohol consumption since 15 years, daily.On clinical examination, Intraorally, presence of an ulceroproliferative growth evident in the left retromolar region (fig:2) , roughly measures 4*2.5cm in its greatest dimension. Surface appears to be inflamed and erythematous. Extends, anteriorly, distal aspect of 35; posteriorly, retromolar region; superiorly, 1.5cm away from the upper buccal vestibule; inferiorly, extends into lower buccal and lingual vestibule; along with blanching evident in left and right buccal mucosa(fig:3), perioral region (fig:4,5), hard and soft palate, floor of the mouth and shrunken uvula (fig:6,7). On palpation, the inspectory findings such as site, size and extent are confirmed. Surface is rough ,erythematous with indurated base. Firm in consistency. Non-tender and bleeding is present. Presence of multiple vertical fibrotic bandsevident in left and right buccal mucosa, perioral region extends from upper to lower buccal vestibule.Maximum mouth opening : 25mm (fig:8); Tongue Protusion : 6mm (fig:9). On Lymph Node Examination, presence of palpable two

lymph nodes in left submandibular region. Correlating the history and clinical findings, a provisional diagnosis of Grade V Oral Submucous Fibrosis leading to non healing ulceroproliferative growth in left retromolar region suggestive of malignancy and TNM Classification: T4aN2M0.IOPA, revealed generalized horizontal pattern of bone loss extending approx 3mm below CEJ of all teeth evident (fig:10).OPG, revealed no of teeth present: 30, missing 28,38 and presence of generalized mild horizontal pattern of bone loss extending approx 3mm below CEJ of all teeth evident (fig:11).On incisional biopsy, H & E section (fig:12) shows connective tissue with numerous malignant epithelial islands exhibiting features of pleomorphism, hyperchromatism, increased nuclear cytoplasmic ratio, individual cell keratinization along with keratin pearl formation. Chronic inflammatory cell infiltrate. Suggestive of Well Differentiated Squamous Cell Carcinoma. On CT Neck- Plain and Contrast, evidence of an ill defined, irregular soft tissue density lesion showing mild heterogenous post contrast enhancement is noted in the left buccal space, extending to retromolar space with mild erosion of adjacent mandible. The lesion measures 3.8*2.5*1.7 cms. Multiple level 1A, bilateral level 1B and level II lymphadenopathy (fig:13).Patient was scheduled for surgical excision. Under GA, patient in supine position with neck flexed and head turned to right. Modified schobinger incision was made and subplatysmal flap was elevated. Neck dissection continued to remove fibro fatty tissue at level I –V by sacrificing sternocleidomastoid and preserving internal jugular vein, spinal accessory nerve and hypoglossal nerve. Segmental mandibuloectomy with wide local excision of ulcero-proliferative growth involving left retromolar trigone and alveolar margin of mandible and maxilla. Pectoralis major myocutaneous flap along with the pedicle harvested and rotated to cover the mandibular defect area

On Gross specimen (fig:14), Labelled as left segmental mandibulectomy with upper alveolar mucosal margins:

- Received a specimen of left mandibulectomy measuring 10*6*4cms with six teeth
- A ulceroproliferative growth measuring 8*3*2.5cms is present in alveolar border reaching upto retromolar trigone

On Histopathological report, H & E section (fig:15) shows connective tissue with numerous malignant epithelial islands exhibiting features of pleomorphism, hyperchromatism, increased nuclear cytoplasmic ratio, individual cell keratinization along with keratin pearl formation.

Chronic inflammatory cell infiltrate. Suggestive of Well Differentiated Squamous Cell Carcinoma.

On Final Diagnosis, Well Differentiated Squamous Cell Carcinoma $T_{4a}N_1M_0$ and Pathology Staging: $PT_3N_0M_0$ was given.

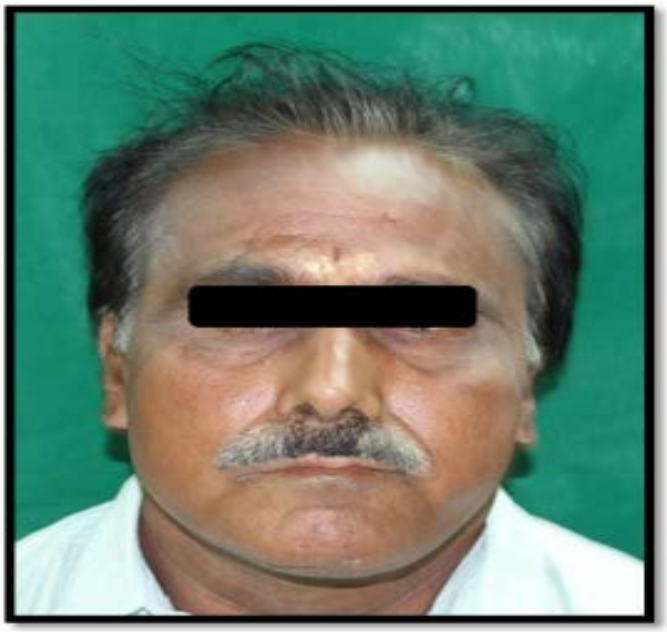

Fig. 1: Patient Profile

Intra – Oral Examination

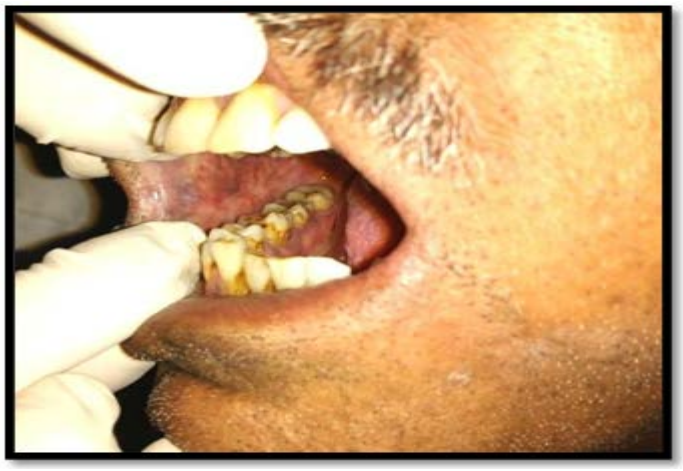

Fig. 2: right buccal mucosa

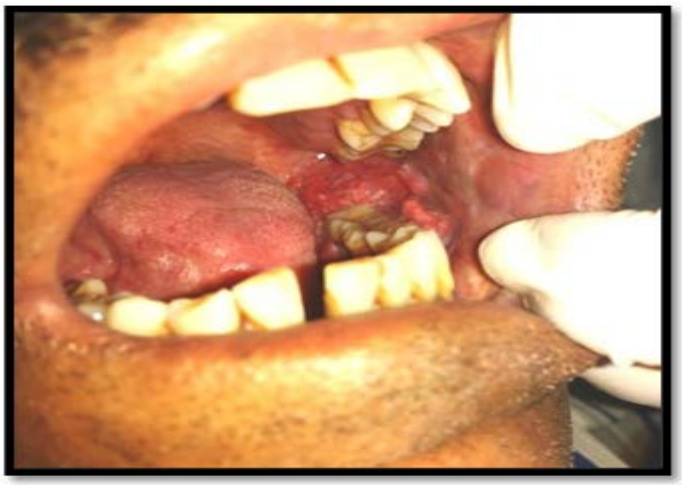

Fig. 3: Left Buccal Mucosa

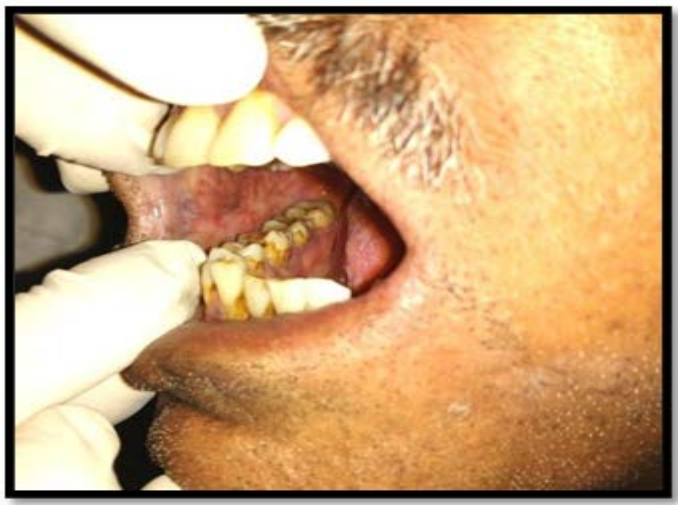

Fig. 4: UPPER LABIAL MUCOSA

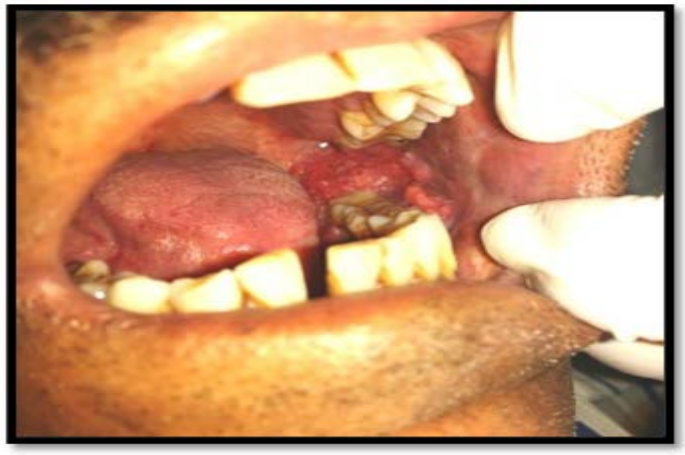

Fig. 5: Lower Labial Mucosa

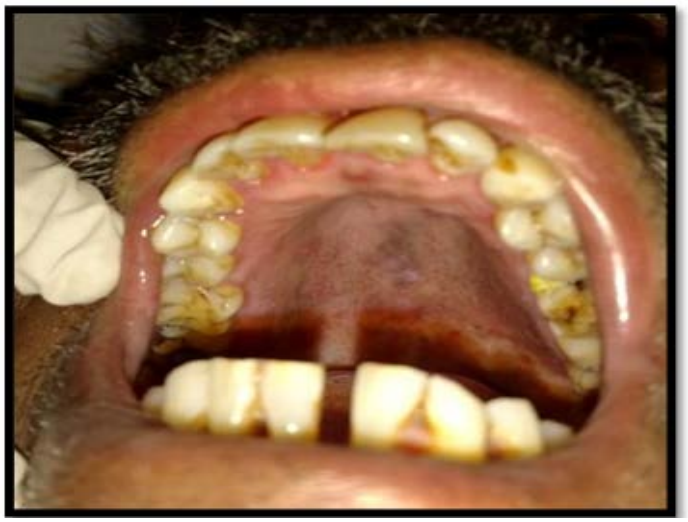

Fig. 6: Hard Palate

Fig. 7: Soft Palate & Uvula

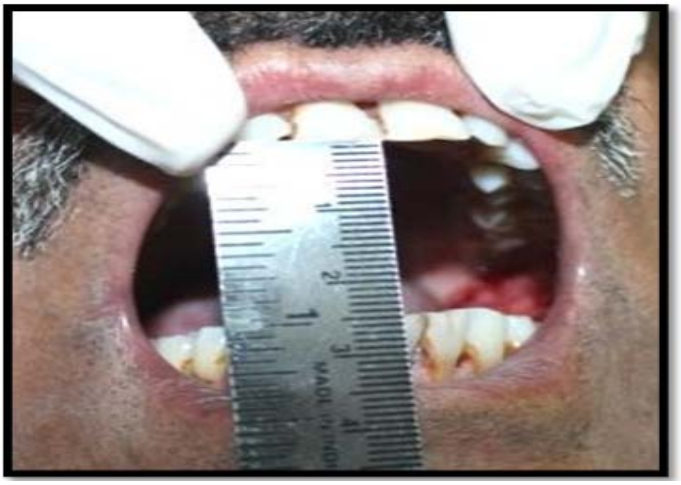

Fig. 8: Inter-Incisal Distance

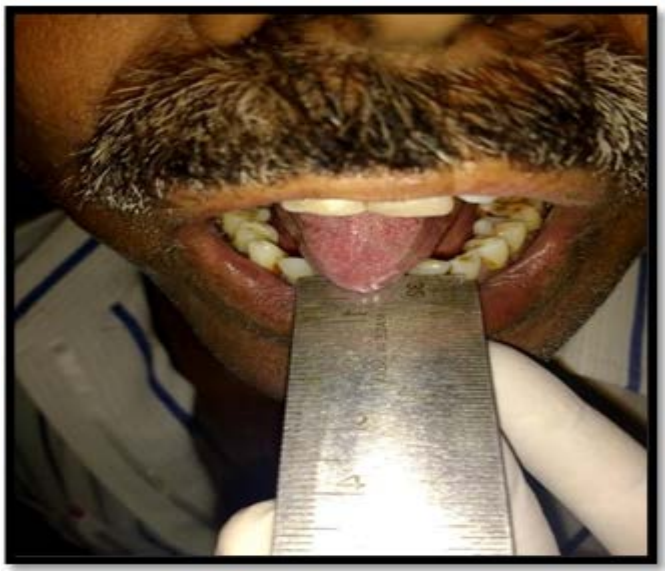

Fig. 9: Tongue Protusion

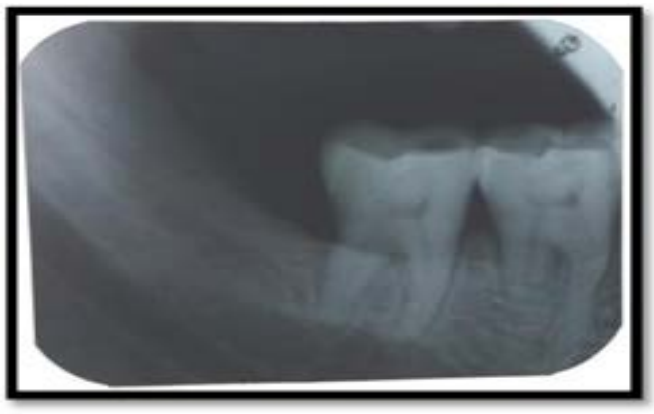

Fig. 10: intraoral periapical radiograph

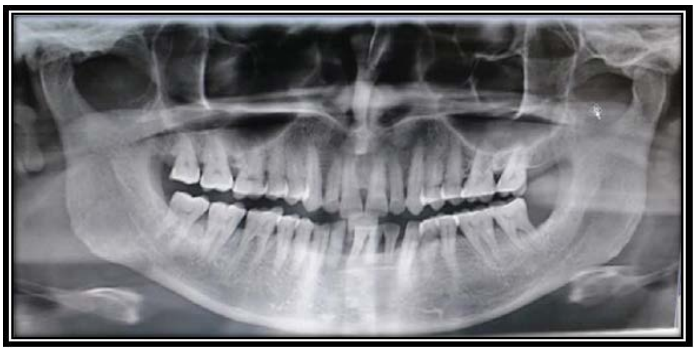

Fig. 11: Orthopantomogram

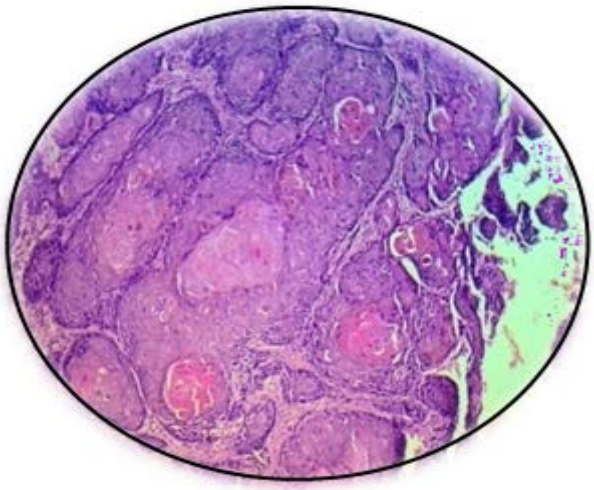

Fig. 12: Histopathological Picture – 10x

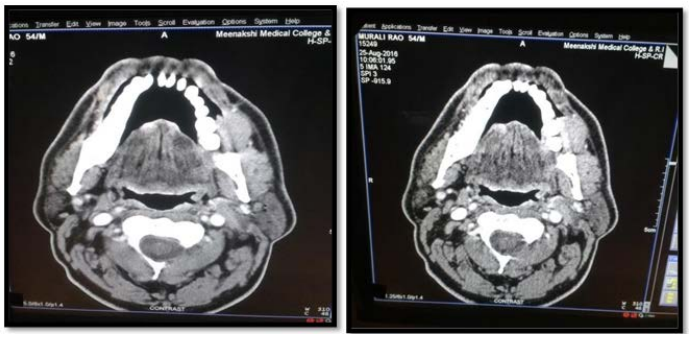

Fig. 13: CT Scan Images

Fig. 14: Excised Gross Specimen

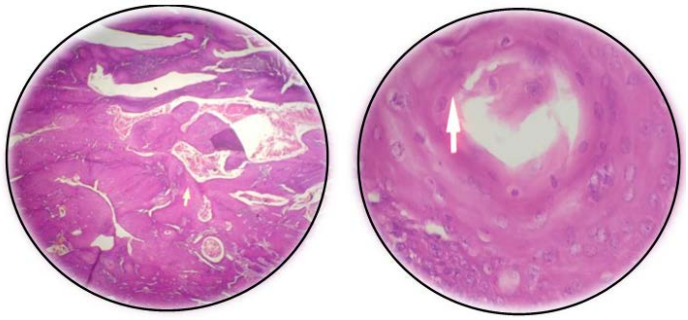

Fig. 15: Histopathological Picture – 10x & 40x

Discussion

OSMF, a potentially premalignant condition of the oral cavity and oropharynx has been a substance of thorough research since the last 50 years. Initially literature emphasises on the studies done by Joshi, Sirsat and Khanolkar, ABN Rao and other workers, related to etiology, clinical features, histopathological features and malignant potential of the disease [12]. Oral Submucous fibrosis is a "insidious chronic disease affecting any part of the oral cavity and sometimes the pharynx, occasionally preceded by vesicle formation, always associated with juxtaepithelial inflammatory reaction followed by a fibroelastic change of lamina propria with epithelial atrophy leading to stiffness of the oral mucosa, trismus and inability to eat"[13]. Prevalence rate is 0.2 to 0.5% with higher percentage reported from the southern states in India[14].Unlike other precancerous lesions, OSMF is insidious in origin and does not regress, either spontaneously or with cessation of habits[15]. The condition may endure neither stationary or become severe, leaving an individual handicapped both physically and psychologically[16].A diverse of factors trigger the disease process. Factors include areca nut chewing, genetic basis, nutritional deficiencies, auto immunity, and saliva.OSMF and cancerOSMF is considered as a premalignant condition. A malignant transformation rate of 7.6% over a span of 10 years was described in an Indian cohort and the relative risk for malignant transformation may be high [17].Thomas et al from South India proposed tobacco chewing was the most principal risk factor for multiple oral premalignant lesions and may be a major etiological factor for cancers on the oral epithelium in the Indian population [18]. Reactive oxygen species(ROS) generated during the auto oxidation of areca nut poly phenols in the saliva of commercial areca nut chewers are pivotal in the initiation and promotion of oral cancer[19]. Nitrosation of areca nut alkaloids also give rise to areca nut specific nitrosamines that have been revealed to be mutagenic, genotoxic and are competent of inducing tumours in experimental animals.Epithelial atrophy and lack of proper maturation of epithelium are the two key histological changes marked in the oral mucosa of patients with iron deficiency anaemia 39. Further Fe deficiency results in improper vascular channel formation and concomitant decreased vascularity, therefore creating easy percolation of esters of arecoline. These esters restoring fibrogenesis and the resultant fibrous tissue deposition potentially will manifest as fibrotic bands. Cytochrome oxidase, an Fe containing enzyme is reasonable for the normal maturation of epithelium. In Fe deficiency anaemia, low levels of this enzyme are discern, thus epithelial atrophy and lack of maturation takes place. It is thoroughly documented that Post cricoids carcinoma, esophageal carcinoma and oral carcinomas exhibit an increased inherent relation with Fe deficiency anaemias. Malignant transformation in OSMF could be elucidated in a analogous manner[20].

Conclusion

Oral health is an inherent component of general health and well being. Regardless of great improvements in the oral health status, the overload and impact of oral diseases related to the consumption of carcinogenic products like tobacco, betel nut quid are still high, particularly in South Central Asian countries. Areca nut

quid being consumed in diverse forms plays a major role in the etiology of Oral submucous fibrosis, a precancerous condition.An altered oral mucosa appears in OSMF, following extended period of chronic deficiency of iron and folic acid. Iron is crucial for maintaining the integrity of epithelia and normal functioning of enzymes. Iron deficiency jointly with other initiating factors may contribute to this pre-existing pathological response of the lamina propria; leading to a defective inflammatory reparative response, culminating in fibrotic healing.An abundance of interventions may be used to manage Oral submucous fibrosis: Discontinuation of habit, Oral physiotherapy, antioxidants, intralesional steroids, and surgical interventions.Inculcating the patients about the harmful effects of tobacco consumption, should hence be the primary objective in dental health camps.It is accordingly empirical to high point the malignant potential of OSMF and take all the inevitable steps towards prevention of the disease through public awareness rather than waiting for pointless effort to treat the same ineffectively. The inscription ‘‘prevention is better than cure’’ becomes more pertinent for such complicated clinical premalignant conditions.

References

1. Mehta FS, Hammer JE (1993) Tobacco related oral mucosal lesions and conditions in India, III edn. Basic Dental Research Unit, TIFR Publication, Mumbai
2. Shah JP, Johnson NW, Batsakis JG (2003) Oral cancer. Martin Dunitz, Taylor and Francis Group, London
3. Kramer IR, Lucas RB, Pindborg JJ, Sobin LH (1978) World Health Organization Collaborating Center for Oral Precancerous Lesions. Definition of leukoplakia and related lesions: an aid to studies on oral precancer. Oral Surg Oral Med Oral Pathol 46:518–539
4. Sankaranarayanan R (1990) Oral cancer in India: An epidemiologic and clinical review. Oral Surg Oral Med Oral Path 69(3):325–330
5. Subapriya R, Thangavelu A, Mathavan B, Ramachandran CR, Nagini S (2007) Assessment of risk factors for oral squamous cell carcinoma in Chidambaram, Southern India: a case- control study. Eur J Cancer Prev 16(3):251–256
6. Pindborg J, Sirsat S (1966) Oral submucous fibrosis. Oral Surg Oral Med Oral Pathol 22(6):764
7. RajendranR(1994) Oral submucous fibrosis:Etiology, pathogenesis, and future research. BullWorld Health Organ 72(6):985–996
8. Pindborg JJ, Zachariah J (1965) Frequency of oral sub-mucous fibrosis among 100 South Indians with oral cancer. Bull WHO 32:750–753
9. Paymaster JC (1956) Cancer of the buccal mucosa—clinical study of 650 cases in Indian patients. Cancer 9(3):431–435
10. Phatak A (1979) Fibrin producing factor in Oral Sub-Mucous Fibrosis. Indian J Otolaryngol Head Neck Surg 31(4):103–104
11. Murti PR, Bhonsle RB, Gupta PC , Daftari DK, Pindborg JJ, Mehta FS. Etiology of oral submucous fibrosis with special reference to role of arecanut chewing. J Oral Pathol Med 1995; 24: 145-52.
12. Ramanathan K. OSMF-An alternative hypothesis as to its causes. Med J. Malaysia 1981; 36: 243-5.
13. Canniff JP, Harvey W, Harris M. Oral Submucous Fibrosis; Its pathogenesis and management. British Dent J1986;160: 429-34.
14. Shafer WG, Hine MK, Levi MB: A text book of Oral Pathology. 4th edition, Philadelphia, Saunders.

15. Prabhu SR, Walson DF, Daftari DK and Jonson NW: Oral Diseases in the tropics, 1993: Oxford university press.
16. Paymaster JC. Cancer of the buccal mucosa; A clinical 650 cases in Indian patients. Cancer 1956; 9: 431-5.
17. Rao ABN: Idiopathic Palatal fibrosis. Br. J. Surgery. 1962:50:23-25.
18. Hamner JE, Looney PD, Chusad TN; Submucous fibrosis. J Oral Surg Oral Med Oral Pathol 1974; 37: 412-21.
19. Nishant Raj Chourasia • Rajiv M. Borle • Ankita Vastani, Concomitant Association of Oral Submucous Fibrosis and Oral Squamous Cell Carcinoma and Incidence of Malignant Transformation of Oral Submucous Fibrosis in a Population of Central India: A Retrospective Study, J. Maxillofac. Oral Surg. (Oct–Dec 2015) 14(4):902–906
20. Shamimul Hasana, Osama Sherwania, Sameer Ahmeda, Mohd Abbas Khana, Oral submucous fibrosis turning into malignancy- A case report and review of literature, Journal of Orofacial Sciences, 10 Oct, 2011