Journal of Advanced Medical and Dental Sciences Research

@Society of Scientific Research and Studies ***NLM ID:*** *101716117*

Journal home page: www.jamdsr.com doi: 10.21276/jamdsr Indian Citation Index (ICI) Index Copernicus value = 100

(e) ISSN Online: 2321-9599; (p) ISSN Print: 2348-6805

Case Report

Triad of potentially malignant disorders- A Case Report and Review

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ABSTRACT:
Potentially malignant disorders in the oral cavity are in trend today with people having deleterious oral habits. Chronic irritation from smoking, betel nut chewing and alcoholism play synergistic effects. Lack of awareness about the signs and symptoms of Oral PMDs in general population and even healthcare providers is responsible for the diagnostic delay of these entities. This article reveals a rare case report of a patient having both premalignant lesion as well as premalignant condition giving a high risk of its transformation to malignancy.

Received: 16 October, 2022 Accepted: 19 November, 2022

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This article may be cited as: Bahal M, Saggar V, Bansal M, Sharma S, Bansal H, Kaur S. Triad of potentially malignant disorders- A Case Report and Review. J Adv Med Dent Scie Res 2022;10(12):67-71.

INTRODUCTION

As King James 1 of England commented "Smoking is a custom loathsome to the eye, hateful to the nose, harmful to the brain, dangerous to the lungs and in the black, stinking fume, thereof nearestresembling the horrible Strygian smoke of the pit that is bottomless.", smoking and tobacco consumption in today's world has proven to give birth to malignancies from tip to toe of the the body of the human. Oral cavity being the mirror of the whole body serves to issue warning signals to make theindividual understand the harms associated with the deleterious oral habits of smoking and tobacco consumption in the form of premalignant conditions and lesions. These lesions mark to warn the individual of the harms and the closeness to malignancies if the deleterious habits continue. Correct timing of the alarm bell to quit habits can save lives of millions. Presenting here with a case of the same in which the patient has triad of potentially malignant disorders. Conservative management and habit counselling has saved the lives of many from the serpent of oral malignancies.

CASE REPORT

A male patient of age 28 reported to the department of Oral And Maxillofacial Diagnosis and Radiology with a chief complaint of watery discharge from the right side of the cheek and difficulty in opening the mouth. On further consultation patient revealed that he was apparently normal then he noticed serous discharge from the right side of the cheek since one year, it healed one month back but has recurred and is non tender. Patient has difficulty in opening the mouth also and difficulty in chewing as well. Patient reveals is medical history saying one month back he visited the general physician who incised and drained the purulent discharge and was under medication since then.

Patient reveals the habit of smoking cigarette 3-4 times a day since the last 13 years. He consumes alcohol 1-2 times a week since the last 8 years.

On extraoralexamination, sinus opening was seen in the right cheek which drained purulent exudate, it was non tender on palpation, and was soft and swollen as well of size 0.7-1 cm in diameter approximately. Patient had inadequate mouth opening of 29 mm as well.

On intraoral examination patient had occlusal pit caries in relation to 18, generalized attrition and partially impacted tooth in relation to 48.

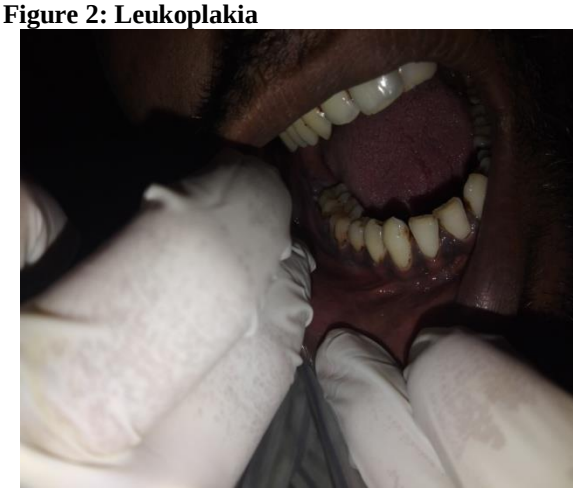
On examination of oral mucosa white plaque like deposits were found in the left and right buccal mucosa region extending superiorly to the occlusal table on the buccal mucosa and inferiorly in the mandibular vestibule, anteriorly in the vestibule and gingival sulcus region of 35 and 45, posteriorly till the

maxillary tuberosity on both sides. The lesion appeared to be rough and wrinkled showing characteristic cracked mud appearance. The lesion was raised, non tender, non scrapable suggestive of homogenous leukoplakia.[Figure 1 and 2]

Figure 1: Leukoplakia

White plaque like lesion visible in the buccal mucosa on the right side which is non scrapable and non tender with cracked mud like appearance.

Figure 2: Leukoplakia



White plaque like lesion visible in the attached gingiva and vestibule on the right side which is non scrapable and non tender with cracked mud like appearance.

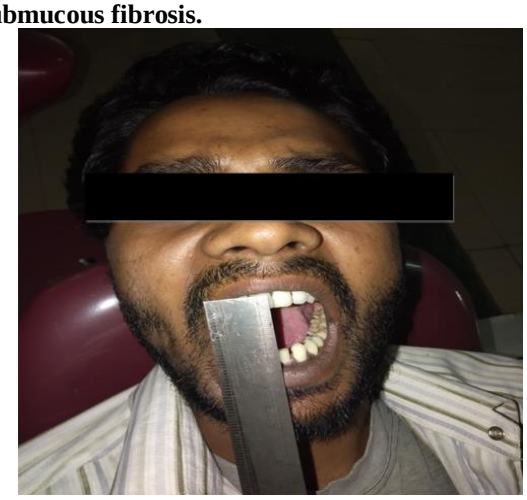
The differential diagnosis of frictional keratosis and plaque type of lichen planus was given which after histopathological examination was confirmed to be Homogenous leukoplakia.

Patient has reduced mouth opening of 25mm, diffuse blanching was seen in the right and the left buccal mucosal region. Uvula appeared shrunken and deviated to the left. On palpation white fibrous vertical bands were felt in the left and right buccal mucosal region extending from the corner of the mouth till the soft palate, uvula was shrunken showing hockey stick appearance, there was loss of flexibility of the oral mucosa with restricted tongue movement and Burning sensation on eating spicy as reported by te patient on a visual analogue scale of 9 on 10suggestive of oral submucous fibrosis grade 3. [Figure 3 and 4]

Figure 3: Oral Submucous Fibrosis

Diffuse blanching all over the buccal mucosa with stiffness of the buccal musculature, and ulcerations and burning sensation to the patient.

Figure 4: Reduced mouth opening due to oral submucous fibrosis.



Patient also had diffuse greyish black discoloration in his palate extending from the palatal gingival sulcus of maxillary anteriors till the posterior surface of the Hard palate which was non tender , non scrapable, flat suggestive of smokers Palate. [Figure 5]

Figure 5: Smokers palate

Grayish black discoloration in the hard palate.

Patient was given habit counselling and was sent for referral to the dermatologist, antioxidants medications were prescribed and the patient was called for follow up after 15 days, the patient is still on follow up.

CASE DISCUSSION

WHO has defined POTENTIALLY MALIGNANT DISORDERS as the risk of malignancy present in a lesion or condition either at the time of initial diagnosis, or at a future date.[2] WHO also classified the PMDs into A) PREMALIGNANT LESION , a benign lesion with morphologically altered tissue which has greater risk of transforming into malignancy, B) PREMALIGNANT CONDITION a disease or patients habit that does not necessarily alter the clinical appearance of local tissues but is associated with a greater than normal risk of precancerous lesion or cancer development in that tissue.[1]

LEUKOPLAKIA

Leukoplakia is a premalignant lesion most commonly affecting the oral cavity. It is one of the most common lesions found in the smokers mouth.The buccal mucosa (70%) is the most commonly affected site especially along the occlusal line , but it can occur in the whole of the buccal mucosa characterised by keratotic patches ,lesions can also be found on the floor of the mouth (42.9%), tongue (24.2%) and lip vermilion (24%).This term was first used by Schwimmer in 1877 , later WHO in 1980 defined Leukoplaka as "a white patch or plaque that cannot be characterized clinically or histologically as any other disease."[1,4]

In 2007, Warnakulasuriya et al proposed in a report that "Oral leukoplakia should be used to recognize white plaques of questionable risk having excluded known disorders that carry no risks for cancers".[4]

ETIOLOGY

Smoking, prolonged mechanical trauma, Epstein barr virus, candidiasis, HIV, HSV, HPV and reduced serum concentrations of vitamin A, marks to be most commonly associated to be the cause of leukoplakia.

CLINICAL MANIFESTATIONS

Leukoplakia has four stages which has different chances of malignant transformation.

Table 1: Stages of leukoplakia[5]

Early or Thin	0-2%
Homogenous or Thick	1% - 7%
Granular or verruciform	4% - 15%
Speckled or Erythroplakia	18% - 47%

HISTOPATHOLOGY

The classic features of dysplasia and potentially malignant disorders includes the presence of Irregular epithelial stratification, Loss of polarity of basal cells, Drop-shaped rete ridges, Increased number of mitotic figures, Abnormally superficial mitoses, Premature keratinisation in single cells (dyskeratosis), Keratin pearls within rete pegs, Nuclear pleomorphism: abnormal variation in nuclear shape, Cellular pleomorphism: abnormal variation in cell shape, Anisonucleosis: abnormal variation in nuclear size, Anisocytosis: abnormal variation in cell size, Increased nuclear size, Increased nuclear-cytoplasm ratio, Atypical mitotic figures and Increased number and size of nucleoli.[5,6]

MANAGEMENT

Consumption of carotenoids (β-carotene, lycopene); Vitamins A, C, and K; and fenretinide, bleomycin, and photodynamic therapy haveshown significant regression of the lesion.

Conventional surgical proceduresentail excision of the lesion. It can be accompanied with orwithout the placement of skin graft or any other dressingmaterial.

ORAL SUBMUCOUS FIBROSIS

Oral submucous fibrosis (OSMF) is a pre-malignant lesion of the buccal mucosa caused by chewingbetel quid. It results in the progressive inability to open the mouth.Patients with OSMF need treatment for trismus correction, reconstructive surgery for any simultaneous oral malignancy.[8, 10]Oral submucous fibrosis (OSF) is a premalignant condition mainly associated with thepractice of chewing betel quid containing areca nut, a habit common among South Asianpeople. It is characterized by inflammation, increased deposition of submucosal collagenand formation of fibrotic bands in the oral and paraoral tissues, which increasingly limited mouth opening.Ram Nathan has suggested that OSMF may be a mucosal change secondary to chronic iron deficiency calling it an Asian analogue of sidero-penicdysphagia. [15, 16]

Oral submucous fibrosis (OSMF) precancerous condition and is chronic, resistant disease characterized by juxta-epithelial inflammatory reaction and progressive fibrosis of the submucosal tissues. In 1966, Pindborg defined OSMF as "an insidious chronic disease affecting any part of the oral cavity and sometimes pharynx. It is associated with juxta-epithelial inflammatory reaction followed by fibroelastic changes in the lamina propria layer, along with epithelial atrophy which leads to rigidity of the oral mucosa proceeding to trismus and difficulty in mouth opening." Other terms used to describe this condition are juxta-epithelial fibrosis, idiopathic scleroderma of the mouth, idiopathic palatal fibrosis, submucous fibrosis of the palate and pillars, sclerosingstomatitis, and diffuse OSMF.[9]

It occurs at any age but most commonly seen in young and adults between 25 and 35 years (2^{nd}–4^{th} decade). Onset of this disease is insidious and is often 2–5 years of duration. It is commonly prevalent in Southeast Asia and Indian subcontinent. The prevalence rate of OSMF in India is about 0.2%–0.5%. This increased prevalence is due to increased use and popularity of commercially prepared areca nut and tobacco product - gutkha, pan masala, flavoredsupari, etc. The malignant transformation rate of OSMF was found to be 7.6%.[16, 17, 18]

CLINICAL FEATURES

Oral submucous fibrosis affects the oral tissue of the buccal mucosa, lips, soft palate, and occasionally the pharynx. The tissue is symmetrically affected and becomes progressively firm and pale. A common complaint is a progressive stiffness of the cheeks, which inhibits the ability to open the mouth. The oral mucosa appears pale and atrophic.[18]

HISTOPATHOLOGY

The earliest stage of the disease is characterized by chronic inflammation of the submucosal connective tissue. This stage is followed by a diffuse progressive fibrosis and atrophy of the overlying epithelium. The atrophic epithelium has a greater tendency to develop hyperkeratosis and epithelial dysplasia, which can progress to squamous cell carcinoma. For these reasons oral submucous fibrosis is considered a precancerous condition.[19]

TREATMENT

Oral submucous fibrosis is usually diagnosed when the disease is at an advanced stage and lesions are widespread. At this stage surgical treatment is usually not possible, but systemic and intralesional injections of corticosteroids have been used with some success.[20]

SMOKERS PALATE

Nicotine stomatitis was discovered in 1941. Nicotine stomatitis was found in pipe smokers where the mucous was not covered by the denture. In 1958, the first example of nicotine stomatitis was found on the hard palate. It appeared as circular reddish lesions on the orifice of the minor salivary glands. The prevalence of nicotine stomatitis is very high for pipe smokers. Irritants from smoke come in direct contact with the palate mucous, especially on 2/3 of the posterior hard palate.[21]

Nicotinic Stomatitis is been associated with pipe, cigarette, and cigar smoking, and, rarely, with chronic ingestion of high-temperature liquids. The mechanism of action is heat irritation from a tobacco product that acts as a local irritant, stimulating a reactive process. In patients who wear them, dentures often protect the palate from these irritants.[21]

ETIOLOGY

Nicotine stomatitis is lesions that form due to the physical irritation from smoke. Temperatures at the burning end of a tobacco cigarette reach 650°C (470°-812°C) and the core temperature of cigarettes can reach 824°-897°C. During smoke inhalation, the oral cavity can reach 190°C. The high temperature smoke coming in direct contact with the palate mucous causes irritation and inflammation at the orifice of the minor salivary glands on hard palate. Microscopy has explained the changes in cells around the orifice of the minor salivary glands. Squamous cells on the wall of the salivary gland duct undergo hyperplasia and parakeratosis occurs on the orifice mucous. [22, 23]

HISTOLOGY

Microscopy explained that the lumen orifice minor salivary glands were not closed. Infiltration of inflammatory cells from blood vessels into intracellular space and accumulation of mast cells occurs in the subepithelial tissue. A comparison of histological results between a healthy hard palate and a hard palate with nicotine stomatitis showed that only nicotine stomatitis underwent histological changes. Histological changes of hard palate tissue only occurred in 2/3 of the posterior hard palate. Nicotine stomatitis was not found on 1/3 of the anterior hard palate and soft palate. This is due to a low distribution of minor salivary glands on 1/3 of the anterior hard palate and soft palate.[24]

CLINICAL FEATURES

Nicotine stomatitis presents as a red circular lesion around the orifice of the minor salivary glands on hard palate mucous. In addition, it is found also characterized by the thickening of the epithelium due to extended irritation on the palate mucous.[24, 25]

TREATMENT

Treatment of nicotine stomatitis Nicotine stomatitis results in benign lesions and is reversible. Research by Walsh et al.4 explains that the hyperplasia around the orifice of the minor salivary glands will return to normal after two weeks without smoking.[25]

DECLARATION OF PATIENTS CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published but the anonymity is not guaranteed.

FINANCIAL SUPPORT AND SPONSORSHIP

Nil

CONFLICTS OF INTEREST

There are no conflicts of interest

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