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Case Study

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MALIGNANT TRANSFORMATION OF ORAL SUBMUCOUS FIBROSIS: A CASE REPORT

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ABSTRACT
Oral submucous fibrosis(OSMF) is a high risk premalignant condition, commonly seen among south east Asian population and characterized by changes in the connective tissue fibers of the lamina propria and deeper parts leading to stiffness of the mucosa, restricted mouth opening & mortality (when transformation into squamous cell carcinoma). OSMF is an acquired disease usually associated with the habit of areca nut chewing. Here, we are presenting a case of oral submucous fibrosis showing malignant potential and development of oral squamous cell carcinoma.

KEYWORD: Oral submucous fibrosis, Premalignant condition, Malignant Transformation.

I. INTRODUCTION

Oral submucous fibrosis is a potentially malignant condition of the oral cavity, characterized by a juxta-epithelial inflammatory reaction followed by a fibroblastic changes in the lamina propria and associated epithelial atrophy. The disease affects oral cavity as well as upper third of the esophagus.[1]

OSMF has 7-30% malignant tranformation rate to the oral squamous cell carcinoma.[2]

In 1956, Paymaster first mentioned the precancerous nature of submucous fibrosis & described the occurrence of slow growing squamous cell carcinoma in one third of the cases with submucous fibrosis.[3]

The present paper describe a case of OSMF, turning into malignancy in a 43 years old male with clinical, histopathological features and review the literature on OSMF and increase awareness of this condition.

II. CASE REPORT

A male aged 43 years old reported to the outpatient Department of Oral Medicine and Radiology with chief complaint of restricted mouthopening and burning sensation in the mouth over the past 2 years. The patient also complaints of growth in his inner left cheek region since past 4 months.

Patient was apparently asymptomatic 2 years back, later he experienced progressive difficulty in mouth opening and burning sensation when consuming spicy food, thereafter patient visited to dentist and he had begun treatment with intralesional steroids injections 1 year earlier on diagnosis of OSMF.

Patient gave history of removal of his lower left molar (38), 7 months back.

After extraction, 3 month back he noticed growth in his lower left buccal mucosa, initially growth was smaller and increased gradually to the present size with a throbbing type of pain present since past 7 days.

As mentioned above past dental history revealed intralesional steroid injections 1 year back and extraction of lower left third molar, 7 months back. No significant past medical & family history.

Habit history: Patient has habit of pan chewing (arecanut and tobacco in the form of jarda) since 20 years, 3 to 4 times in a day. He kept the pan quid in left buccal vestibule of mouth for 10-15 minutes then he spit it out.

Clinical Findings

General examination revealed patient has moderately built (figure:1) with steady gait and normal vital signs. There were no signs of pallor in the conjunctiva, cyanosis and icterus in the sclera was noted. Skin was normal in appearance and nails shows no sign of koilonychias, beading, clubbing, cyanosis or pallor.

Extraoral examination revealed a single ovoid lymphnode palpable in the left submandibular region,

measuring approximately 2x3 cm, which was firm in consistency, tender and was freely mobile in all planes.

Intraoral examination revealed pale, blanched right & left buccal mucosa & soft palate. The mouthopening was restricted with interincisal distance being approximately 29mm, along with restricted protrusive tongue movements noted. Uvula appearance was deformed and shrunken. When the patient was asked to blow out air with closed lips, the usual puffed cheek appearance was not seen, suggesting loss of cheek elasticity.

In addition, intraorally a ulcero-proliferative growth (figure: 2) seen on the posterior aspect of left buccal mucosa involving gingiva, alveolus from 36 to 38 tooth region with irregular in shape, size approximately measured 3x5 cm, extending superiorly 2 cm below the upper buccal vestibule, inferiorly until retromandibular pad area, anteriorly 4.5 cm away from the corner of mouth & posteriorly until the pterygomandibular raphe region.

The surface of the growth appeared rough, ulcerated and was covered by white necrotic slough and over the surface indentations caused by the cusp of corresponding teeth (26, 27) noted. The surface of the mucosa adjacent to the growth appeared erythematous and the teeth 37 associated with growth was mobile.

On palpation buccal mucosa was tough & leathery with palpable 2-3 fibrotic bands running vertically and extending to the retromolar region. The growth was non-tender, indurated, fixed to the underlying structure, firm in consistency and no bleeding on mild provocation was evident.

On correlating the chief complaint, history of habit and clinical examination, a provisional diagnosis of Oral submucous fibrosis along with malignant proliferative growth on left buccal mucosa was made and TNM staging was T3N2aM0 (Stage- III).

Investigations

Orthopantomogram (OPG) was taken for evaluation of any bony involvement, which revealed (figure:3) destruction of the interdental bone with furcation involvement and widening of periodontal ligament space of mesial root associated with tooth 37, missing teeth 38 seen to be noted and generalized interdental boneloss present.

Figure: 1 Extraoral profile of the patient.

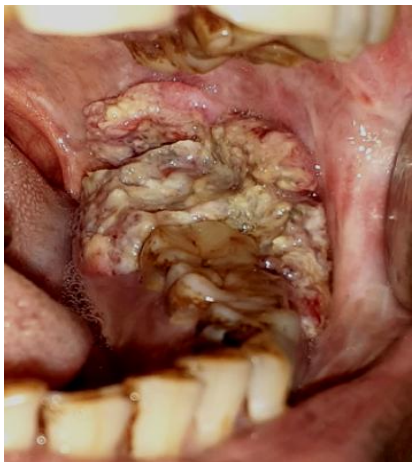

Figure 2: Intraoral view of the patient with ulcero- proliferative growth seen on the lower left buccal vestibule.

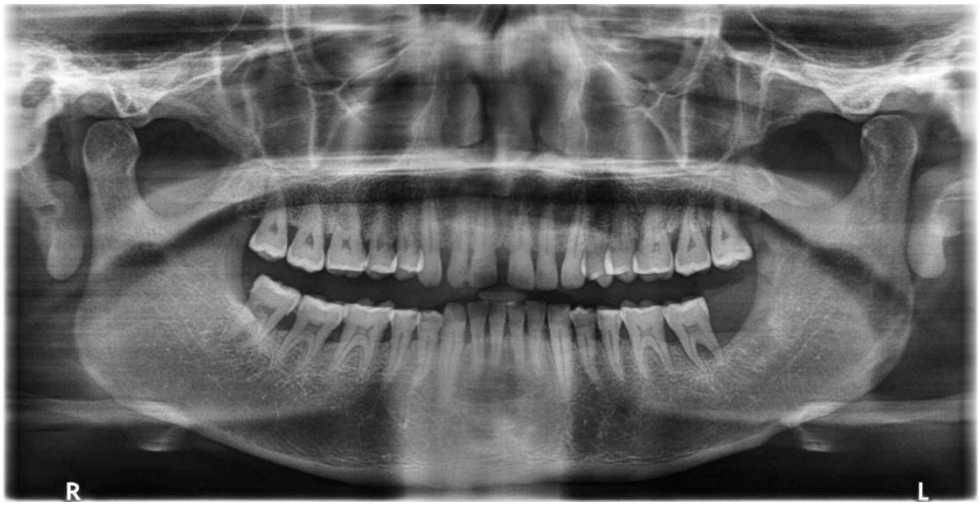

Figure: 3 Orthopantomogram (OPG), revealed destruction of the interdental bone with furcation involvement and widening of periodontal ligament space of mesial root associated with tooth 37, missing teeth 38, and generalized interdental boneloss noted. Noted.

Histopathological Findings

To confirm the diagnosis incisional biopsy was performed and which revealed (figure:3) overlying epithelium is thick atropic parakeratinized stratified squamous in nature with evidence of ulceration can be seen. The basement membrane is breached. The underlying connective tissue shows infiltration by malignant epithelial cell in the form of cords and islands. Malignant epithelial cells show features of cellular atypia like nuclear and cellular pleomorphism, prominent nucleoli loss of cohesion, increased and abnormal mitosis. Rest of connective tissue shows dense chronic inflammatory cell infiltrate, minor salivary gland acini.

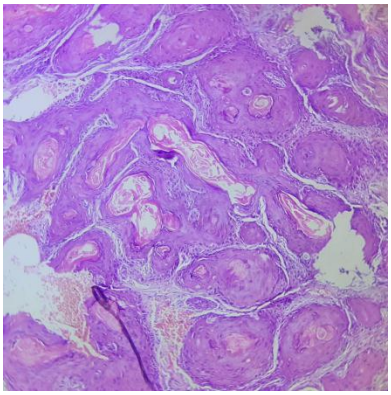

Figure: 4 Histopathological picture.

On the basis of clinical, radiographical and histopathological findings the present case was diagnosed as oral submucous fibrosis obscuring with well differentiated buccal squamous cell carcinoma. The patient was refered to department of oral and maxillofacial surgery and advised to undergo a surgical procedure involving excision of the lesion with a wide clearance, hemimandibulectomy and radical neck dissection.

III. DISCUSSION

Oral submucous fibrosis is an insidious chronic disease affecting any part of oral cavity and sometimes pharynx although occasionally preceded by and /or associated with juxta epithelial inflammatory reaction followed by a fibroelastic change of the lamina propria with epithelial atrophy leading to stiffness of the oral mucosa and causing trismus and inability to eat.[4] In 1952, Schwartz first describe a fibrosing condition in a series of Indian women living in East Africa and coined the term 'Atropica IdiopathicaTropica Mucosae Oris'.[3]

In 1953. Joshi described Oral submucous fibrosis is an oral condition and causes changes similar to those of systemic sclerosis but limited to oral tissue.[1]

Oral submucous fibrosis cases mainly reported in India, and also occurred in Srilanka, Malaysia, Nepal, South Vietnum and Thailand.[5] In India, higher prevalence rate (0.2 to 0.5%) reported from the southern states.[1]

Many etiological factors trigger the occurence of OSMF. The most common etiology behind the development of OSMF is areca nut chewing. As reported in our case patient has habit of chonic pan chewing along with areca nut and tobacco in the form of jarda.

Tobacco containing areca nut in the market has been associated with increase in the frequency of OSMF.[6] The amount, frequency and duration of chewing areca nut betel quid are clearly related to the development of OSMF.[7] The direct contact of the quid mixture with oral tissue causes continous irritation by various components, including biologically active alkaloids (arecoline, arecaidine, arecolidine, guavacoline, guavacine) and copper.[1] Other factors include genetic basis, nutritional deficiencies, autoimmunity, salivary constituents, etc.[1]

OSMF affects both males and females. Male –female ratio was found to be 2.3:1.[1] The most common site of

OSMF is buccal mucosa followed by soft palate, uvula, lips, tongue and floor of mouth. The signs and symptoms of OSMF are due to inflammation and fibrosis. The most common initial symptoms of OSMF is burning sensation of oral mucosa while chewing spicy food, followed by dry mouth, blanching of the oral mucosa and ulceration.[1]

Blanching of the mucosa is the most common earliest sign and caused by impairment of local vascularity because of increasing fibrosis and acquires marble- like appearance. [1] Blanching may be localized, diffuse or reticular. As the disease progresses (advance stages of OSMF), the most prominent feature is a fibrous band located beneath an atropic epithelium.[8] Fibrous bands leads to loss of resilience, which interfers with speech, tongue mobility and restricting mouth opening.[8]

Due to development of fibrosis lip becomes thick, rubbery, difficult to evert them and the opening of mouth is altered to an elliptical shape. Fibrosis causes cheeks thick and rigid, and usual puffed –out appearance of the cheeks is missing during blows a whistle or tries to inflate a balloon. [1] In the tongue, initial changes is depapillation at the tip, lateral margins may occur and fibrosis may lead to restriction in protrusion of tongue movements.[1]

In some cases of OSMF, fibrosis extend posteriorly involve to soft palate and uvula. In advance cases uvula becomes 'bud-like' or 'hockey stick' appearance. Gingiva is less commonly affected, when involve it becomes fibrotic, blanched.[1]

Potentially malignant nature of OSMF has been well documented in the literature. Oral submucous fibrosis has 7-30% malignant tranformation rate to oral squamous cell carcinoma.[2]

A malignant transformation rate of 7.6% over a period of 10 years was mentioned in an Indian cohort and the relative risk for malignant transformation may be as high as 397.3.[9]

In many observational studies revealed in OSMF subjects there is a habit of areca chewing compared to that of general population.[3]

OSMF does not regress spontaneously after cessation of habit. Once the disease is present, it either persists or become more severe with involvement of other areas of oral mucosa.[3]

According to Thomas et. al., tobacco chewing was the most important risk factor for development of premalignant lesions and major etiological factor for cancer in the Indian population.[1] Oral cancer may develop in person who consume areca nut but do not consume tobacco in any form.[10]

Auto oxidation of areca nut poly phenols in the saliva produced reactive oxygen species which are crucial in the initiation and promotion of oral cancer.[1]

The two important histological changes observed in patients with iron deficiency anaemia are; i) epithelial atrophy, ii) lack of proper maturation of epithelium.[11] Cytochrome oxidase and Fe containing enzymes are responsible for normal maturation of epithelium. In iron deficiency anaemia, low level of these enzymes are noted, thus epithelial atrophy and lack of maturation of epithelium occurs.[1]

Also iron deficiency anaemia result in improper vascular channel formation and concomitant decreased vascularity, which causes easy percolation of esters of arecoline. These esters induces fibrogenesis and resultant fibrous tissue deposition which manifest as fibrotic bands. Iron deficiency together with other initiating factors may contribute to this pathological conditions. It is well documented that post cricoids carcinoma, esophageal carcinoma and oral carcinomas shows an association with iron deficiency anaemias.[1]

A plethora of therapies may be used to manage OSMF, like restriction of habit/ behavioral therapy, medicinal therapy as supportive treatment, antioxidants, intralesional steroids and surgical interventions.[1]

In our case report patient had oral submucous fibrosis since 2 years. For OSMF he received intralesional steroid injections therapy 1 year back and experienced slight decrease in mucosal stiffness and burning sensation in his mouth. Post treatment patient was non compliant for his addiction habit (arecanut and tobaco) and he developed ulcero-proliferative growth in the left buccal mucosa involving gingiva, alveolus from 36 to 38 tooth region since 4 months. For confirmatory diagnosis incisional biopsy performed, which revealed well differentiated squamous cell carcinoma, developed from premalignant condition (OSMF).

CONCLUSION

In the present reported case patient had history of chonic pan chewing along with areca nut and tobacco (jarda), that contains harmful ingredients which had led to the development of potentially malignant condition (OSMF) which turn into malignancy. Oral cancers causes significant mortality and morbidity. It has a good prognosis when detected at an early stage, but almost two-third of oral cancer patients are diagnosed at a late stage, which leading to extensive treatment and low survival rates. Therefore, oral health care professionals should have a basic awareness of such type of potentially malignant conditions and oral cancers.

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