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Oral Submucous Fibrosis: Understanding the Disease through a Case Series

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ABSTRACT

'Vidari' is a crippling fibrotic disorder affecting mouth commonly seen in India and South East Asia. It's always optimum and highly desirable to beat oral submucous fibrosis(OSMF) before it becomes oral cancer. OSMF is a huge dilemma for clinicians worldwide specifically in south east Asia due to elusive pathogenesis. The main characterstic of OSMF is the build up of fibrous tissue in the sub mucosal layer of mouth, which can lead to a loss of flexibility and movement in mouth and can cause difficulty in opening the mouth and speaking, as well as eating and swallowing.We are presenting a case series of OSMF highlighting the clinically observed features. These features might not be seen together.

KEYWORDS: Areca nut, OSMF, clinical features, tobacco, treatment plan, grading.

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INTRODUCTION

The oral cavity is considered to be a mirror of the body's health. Oral cavity speculates the symptoms of various systemic disturbances. "idiopathic scleroderma of the mouth, juxtaepithelial fibrosis, idiopathic palatal fibrosis, diffuse oral submucous fibrosis, and sclerosing stomatitis" and a plethora of other names have been used to describe this disabling condition.(7)T. Sheikh was a pioneer when he studied this condition in India on 5 women from Kenya. It was S.G Joshi who coined the term Oral submucous fibrosis. Born in Copenhagen, Denmark 1921, J.J Pindborg had a keen interest for the topic and left his own town to live in India and research for the same. Jack Wild once said," until I was diagnosed with oral cancer I had never heard of it." As per WHO statistics the global incidence of cancers of the lip and oral cavity is estimated to be 377,713 new cases and 177,757 deaths in 2020.(6) A major cause of mortality in head and neck cancer is because of the presentation of the disease in very late untreatable stages. As per NCRP 2020 data, 75% of head and neck cancers are presented in the late stage and only 25% of patients report in early stages. (4)

OSMF is considered a premalignant condition and malignant transformation is seen in approximately 7.6% cases. (4,5) It is only blatant to look for the etiological factors and ways to prevent this menacing condition. This article focuses on various clinical features and treatment plans seen in OSMF.

CASE SERIES

CASE 1

A 62-year-old patient reported to the department of OMDR with the chief complaint of loose teeth and wanted to get them treated.

Past medical and dental history were not contributory. Patient brushes his teeth once daily with a toothbrush and toothpaste.

Patient gives a history of supari chewing for 30 years.

The patient also gives a history of bidi and cigarette smoking from 30 years and had no intent of leaving it whatsoever.

On extraoral examination, the patient had a reduced mouth opening of 22 mm.

The patient reported a dull pain in the ears during mastication.

On intraoral examination, diffuse blanching in a lace-like pattern was seen in the buccal mucosa bilaterally from the maxillary vestibule superiorly to the mandibular vestibule inferiorly from the angle of the mouth anteriorly till the retromolar pad region posteriorly occupying the soft palate.

On palpation, the buccal mucosa was stiff with decreased flexibility of orofacial musculature.

Vertical fibrotic bands were palpable bilaterally around the pterygomandibular raphe. Uvula was deviated to the left side and shrunken.Tongue movement was restricted with difficult protrusion and blanching on the palate was visible.

Thick white, keratotic, non-scrapable, wrinkled patch seen wrt left lip commissure showing classic cracked mud appearance s/o Homogenous leukoplakia.

The case was diagnosed as Grade 5b OSMF.

CASE 2

A 42 year old patient reported to the department of OMDR with the chief complaint of pain in the lower right region of the jaw for 15 days and burning sensation on eating spicy and hot food for a year now. The pain was dull, intermittent and radiating.

As further history was recorded it was highlighted that the patient had undergone a cyst removal surgery 4 years back.

Patient also admitted that he had been chewing supari for 6 years.

On extraoral examination, the mouth opening was severely reduced to 12mm. Bleeding was seen at angle of mouth bilaterally s/o angular cheilitis.

On intraoral examination, very poor oral hygiene with grade 3 calculus and grade 2 mobility in lower anteriors was noticed. opaque, diffuse blanching was seen on the buccal mucosa bilaterally with intervening normal mucosa giving it a lace like appearance. Erythematous patches and petechiae were quite visible on the buccal mucosa. Buccal mucosa was inelastic, on palpation, rough vertical fibrous bands were felt from 33 to 38 and 43 to 48 tooth regions. Tongue movement was severely reduced, the patient could barely protrude his tongue till incisal edges of his lower anteriors.Diffuse blanching on palate was observed. Due to reduced mouth openeing, uvula could not be appreciated.

The case was diagnosed with Grade 4a OSMF.

CASE 3

A 45 year old patient came to the department of oral medicine and radiology with a chief complaint of reduced mouth opening and pain on opening mouth. The pain was dull, intermittent, non radiating. Past medical and dental history were recorded but were not contributory.

Patient had a history of occasional alcohol consumption. Patient also gave a history of paan/ supari consumption 5-6 times a day for 20 years.

On extraoral examination, mouth opening was reduced to 28 mm.

On intraoral examination, white, keratotic fibrotic bands are seen in the mandibular raphe region on right and left sides of the mouth in 38 and 48 region making the buccal mucosa rigid and limiting the mouth opening. Patient also gave a history of burning sensation on eating hot and spicy food. Patient had a large tongue and had limited tongue movement. Protrusion of the tongue was seen when the patient opened his mouth. Patient had very poor oral hygiene with grade 2 calculus and grade 2 stains.

The case was diagnosed with Grade 2 OSMF.

CASE 4

A 30 year old patient came to the department of oral medicine and radiology with a chief complaint of pain in the lower left back region of the jaw for 15 days. The pain was dull, intermittent, non radiating, aggravates on mastication and relieves on its own. Past medical and dental history were recorded but were not contributory.

Patient was hesitant to admit to any tobacco abuse.

On extraoral examination, non tender firm on palpation with no elevated temperature, non fluctuant swelling extending from inferior border of mandible to zygomatic process of maxilla involving the body of mandible was observed.

On intraoral examination, reduced mouth opening to 26mm was observed along with the presence of white keratotic,firm, inelastic bands in the buccal mucosa bilaterally extending from 48-44 region and 38-34 region. Patient had no burning sensation whatsoever.

Patient had a carious 46, on radiographic examination, a dento-alveolar abscess was diagnosed. Patient also exhibited bad odour and poor oral hygiene with grade 3 stains. Patient had gingival recession and attrition in lower anteriors. Tongue protrusion was normal.

The case was diagnosed with Grade 2 OSMF.

CASE 5

A 51 year old male patient reported to the department of OMDR with the chief complaint of mass in the oral cavity on the right side for 2 months. Patient reports that he started having pain in the lower right posterior region, visited the dentist who said the tooth was mobile and the tooth shed subsequently.

Patient is asthmatic since childhood and takes nebulizer during season change.

Patient gave a history of beedi smoking and supari chewing 4 times a day for 30 years.

On intraoral examination, a hard proliferative overgrowth present in the right mandibular vestibule extending from 44 to 47 region causing obliteration. On palpation the mass was irregular, firm to hard in texture extending from the retromolar pad area to the occlusal table level. No bleeding on slight digital palpation s/o squamous cell carcinoma of buccal mucosa.

Submandibular lymph nodes of the right side were tender and firm on palpation.

Patient had severely reduced mouth opening of 15 mm with firm, inelastic buccal mucosa. On palpation, vertical fibrous bands were felt in the molar region. In the soft palate, the fibrous bands were seen to radiate from the

pterygomandibular raphe in a scar-like appearance. Uvula was deviated and shrunken.

The case was diagnosed Grade 3 OSMF & squamous cell carcinoma of buccal mucosa.

CASE 6

A 28 year old patient reported to the department of OMDR with the chief complaint of pain in the lower left back tooth region and was unable to open his mouth comfortably for 2 years now.

Patient gives a history of tobacco chewing for 5 years and is off the habit for 5months.

Case was diagnosed as Grade 3 OSMF.

Patient looked lean and has lost 5 kgs in a span of 1 year.

On extraoral examination, the patient had a reduced mouth opening of 18mm.

On further intraoral examination, blanched buccal mucosa was firmly attached to the underlying tissues. Palatal blanching was evident. On palpation, firm vertical fibrous bands were felt around the premolar region. Tongue protrusion was limited. Patient was asked to blow out cheeks and whistle, he was unable to blow his cheeks for more than 10 seconds and was completely unable to whistle.

DISCUSSION

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 juxta epithelial inflammatory reaction followed by fibroelastic changes of lamina propria, with epithelial atrophy leading to stiffness of oral mucosa and causing trismus and inability to eat. - PINDBORG. (2)

The prevalence of OSMF in India has been estimated to range from 0.2-2.3% in males and 1.2-4.6% in females . Interestingly, the ratio of women with OSMF is higher than women with head and neck cancer, as compared to men. The most common site of OSMF is the buccal mucosa and retro molar region.(6)Although the etiopathogenesis of this disease is multifactorial, areca nut-chewing in any formulation is considered the main causative agent. Contributory risk factors suggested include chewing of smokeless tobacco, high intake of chilies, toxic levels of copper in foodstuffs and masticatories, vitamin deficiencies, and malnutrition resulting in low levels of serum proteins, anaemia and genetic predisposition.

STAGES OF ORAL SUBMUCOSA FIBROSIS (5)

Stage	Functional staging	Clinical staging
Stage 1	Interincisal openings of 35 mm and above	• Burning sensation in the mouth, • Acute ulceration and recurrent stomatitis • No associated mouth opening limitation
Stage 2	Interincisal openings of 26-35 mm	• Mottled and marble-like buccal mucosa • Dense, pale, depigmented fibrosed areas alternated with pink normal mucosa • Occasional red erythroplakic patches • Widespread sheets of fibrosis
Stage 3	Interincisal opening of 15-25 mm	• Pale buccal mucosa firmly attached to the underlying tissues • Palpable vertical fibrous bands in the premolar area • Unable to blow out cheeks and whistle • In the soft palate, the fibrous bands were seen to radiate from the pterygomandibular raphe or the anterior faucial pillar in a scar-like appearance • The lips may be affected with atrophy of the vermilion border
Stage 4 (4a and 4b)	Interincisal opening of 15 mm and below	• Thickened, shortened, and firm fauces, with the tonsils compressed between the fibrosed pillars • Small, shrunken, fibrous bud uvula • Narrowed isthmus, presence of circular band around entire lip and mouth • Restricted tongue movement, diffuse papillary atrophy • Atrophy of the vermilion border. • Premalignant and malignant changes

TREATMENT OPTIONS

Long term follow up and habit cessation are the primary and most important treatment steps for patients in early stages of OSMF.(6) Habit cessation through proper habit counselling is the first and foremost treatment plan of Oral Submucous Fibrosis, since a lot of emphasis is laid on the areca nut chewing habit in contribution to OSMF. The patient needs to be educated patiently for the same. Physiotherapy is seen as an adjuvant treatment plan as it is more compliant and economical for the patient.

Special 26 treatment plans have been discussed for OSMF. Intralesional steroid injection is one of the modalities for the symptomatic relief of burning and may also be combined with hyaluronidase and placental extract intralesional injections to improve mouth opening. (1,2)

IFN gamma plays a significant role in the treatment of OSMF for the reason that it has an immuno-regulatory effect. IFN-gamma is a known antifibrotic cytokine, effect of which was considered on collagen synthesis by arecoline-stimulated OSMF fibroblast.(4)Honey possesses strong bactericidal and bacteriostatic property. It is mainly effective against gram-positive bacteria such as A pyogenes, S. mutans, S.aureus etc. Honey has shown strong antimicrobial activity because of its low water content and low pH. Other than these, honey contains glucose oxidase which produces hydrogen peroxide which has a strong bactericidal action. (3,5)

Enzymes such as collagenase, hyaluronidase and chymotrypsin are being used for the treatment of OSMF. Hyaluronidase by breaking down hyaluronic acid (the ground substance in connective tissue) lowers the viscosity of intercellular cement substance. (4)

CONFLICT OF INTEREST- Nil

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