CASE REPORT

Oral submucous fibrosis: a premalignant condition in a 14-year-old Indian girl

Anshula Deshpande, Shital Kiran, Steffi Dhillon, Rachappa Mallikarjuna

Department of Pedodontics and Preventive Dentistry, K M Shah Dental College and Hospital, Sumandeep Vidyapeeth, Vadodara, Gujarat, India

Correspondence to
Professor Anshula Deshpande, dranshula@gmail.com

SUMMARY

A 14-year-old Indian girl presented with difficulty in mouth opening and burning sensation while eating. On examination, blanching of the oral mucosa with diffuse white pigmented lesion of size 3.5 to 2 cm along with melanotic pigmentation was seen on the left buccal mucosa posteriorly. The patient was diagnosed with oral submucous fibrosis. A comprehensive treatment plan was made based on conservative management that included motivation and intense counselling of the patient and her parents so that she quits the habit of chewing areca nut and tobacco, along with systemic treatment of vitamin B complex supplements, antioxidants, multivitamins and oral physiotherapy. We present this case to highlight the difficulties faced by the clinical practitioners in providing treatment because of the taboos and myths associated with surgical treatment modality in rural population as well as to emphasise the menace of increasing consumption and availability of tobacco and areca nut to children.

BACKGROUND

Oral submucous fibrosis (OSMF) is a chronic condition characterised by progressive stiffening of the oral mucosa with the resultant inability to open the mouth.[1] It is a premalignant condition of the oral cavity with the major aetiological factor being the consumption of areca nut. Stage III OSMF is seen in a young girl aged 14 years that depicts the increasing prevalence of chewing habit of areca nut and tobacco among school-going children in India. The ignorance and lack of awareness regarding the malignant potential of the areca nut chewing among the Indian population are evident. The early involvement is seen because the young mucosa is more vulnerable and responds immediately to insult. This case highlights the threat of ready availability of sweetened and flavoured areca nut in the market to the young children at low cost, resulting in a life-threatening premalignant condition.

CASE PRESENTATION

A 14-year-old girl reported to the department of paedodontics and preventive dentistry for routine dental checkup in the camp organised by the hospital with difficulty in opening the mouth and burning sensation while eating spicy food with no other dermatological or systemic problem (figure 1). She had the habit of chewing flavoured areca nut and scented tobacco, 2 packets/day since 1 year. History of present illness revealed that she is chewing it, as that helps in reducing her appetite.

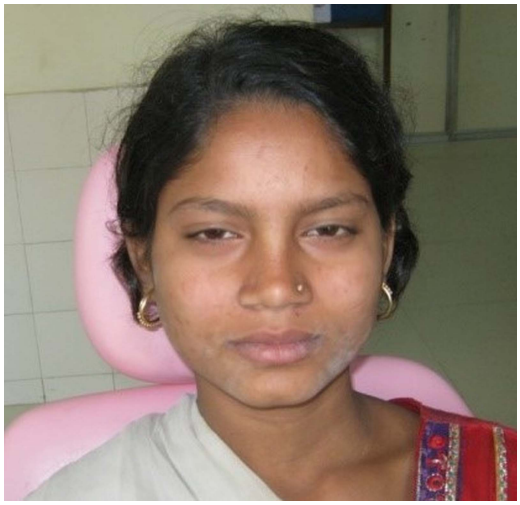

Figure 1 Extraoral photograph of the 14-year-old girl.

She further added that she works as a maid at various houses so after consuming the combination of flavoured areca nut and scented tobacco, she does not feel tired and hungry while working. While recording the details of her chewing habit, she revealed that she mixes the separately available packets of flavoured areca nut and scented tobacco and keeps the mixture in the left buccal vestibule in the second molar region. On general physical examination, pallor was observed in the lower sclera, tongue, oral mucous membrane and palmar creases depicting anaemia.

On oral examination, generalised dental stains were present. On inspection, blanching was seen in the right and left buccal mucosa along with palate (figures 2–4). The oral mucosa was white and pale. Diffuse white lesion of size approximately 3.5–2 cm

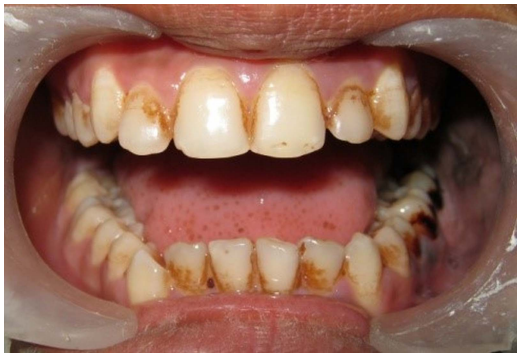

Figure 2 Generalised dental stains can be seen on labial surfaces.

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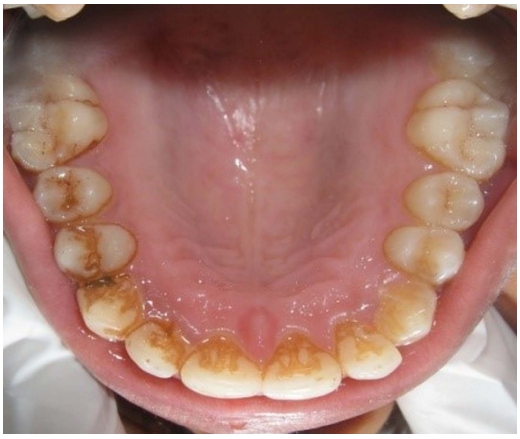

Figure 3 Generalised dental stains on palatal and occlusal surfaces of maxilla.

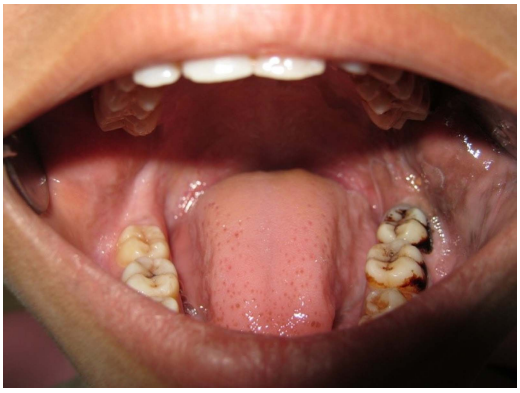

Figure 5 Blanching of both sides of mucosa is seen. Diffuse white lesion and the melanotic pigmentation are visible on the left side of buccal mucosa where the patient places the areca nut mixed with tobacco quid.

along with multiple diffuse areas of melanotic pigmentation were present on the buccal mucosa extending from the retromolar area to the premolar area on the left side (figure 5). A reduced mouth opening was observed, with the interincisal width being 30 mm. On palpation, all the findings were confirmed. The buccal mucosa was rubbery and inelastic, though fibrous bands were not present. The diffuse white lesion on palpation was found to be flat, suggestive of developing oral leukoplakia. Root stumps of 85 were present. Angle's class I molar relationship was recorded for the right and left sides. Considering the combination of clinical findings and habit of areca nut chewing along with tobacco, a provisional diagnosis of stage III OSMF was given by two expert clinicians, independently.

INVESTIGATIONS

A biopsy confirming the histological features of the disease and complete blood count to diagnose anaemia should be performed, but the patient's parents did not give the consent for it as they considered it to be inauspicious and a taboo.

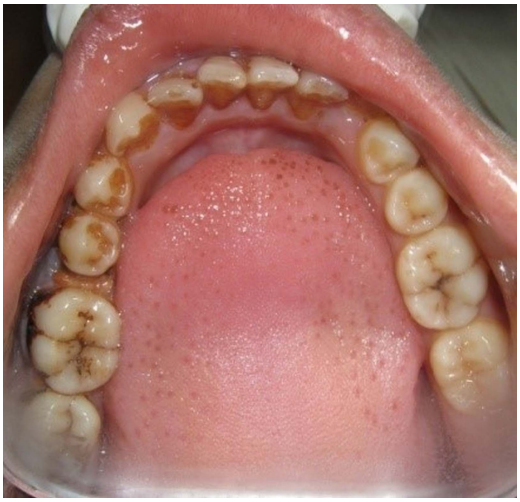

Figure 4 Generalised dental stains on lingual and occlusal surfaces of mandible.

DIFFERENTIAL DIAGNOSIS

Looking at the clinical signs and symptoms of the disease such as difficulty in mouth opening, the diagnosis that can be thought of are localised scleroderma and OSMF. But after knowing the history of having the habit of chewing areca nut along with tobacco and looking at the clinical signs and symptoms, OSMF was confirmed as provisional diagnosis.

TREATMENT

The three main treatment modalities available to treat the patients of OSMF are medical therapy, surgical treatment and oral physiotherapy. Considering the age of the patient and adhering up to her parents' belief of not doing any surgical intervention, a comprehensive treatment plan was made, based on the concept of conservative management for oral submucous fibrosis.

The first step was motivating and counselling the patient and her parents so that she quits her habit. During the intense counselling session with the patient and her parents, carcinogenic potential of chewing tobacco and areca nut was explained. The threat of conversion of the premalignant condition into malignancy was explained to them and the importance of immediate quitting of the habit was emphasised.

Vitamin B complex capsules 500 mg once a day that contains pantothenic acid 50 mg, thiamine 50 mg, riboflavin 50 mg, niacin 50 mg, pyridoxine 50 mg, choline 21 mg, inosital 50 mg, cobalamin 50 µg, biotin 50 µg, folic acid 400 µg and antioxidant capsules once a day containing vitamin A 2000 µg, vitamin C 100 mg, vitamin E 8 mg and bioflavinoids 500 mg were initially prescribed for 1 month to boost up the nutritional status of the patient. Along with that iron tablets 100 mg/day were prescribed to treat the patient who was also suffering from anaemia.

Along with it, diet counselling was given so that her dietary requirements are met. She was advised to take five meals a day that contain fruits, vegetables and grains. Consumption of fresh green vegetables such as spinach, beans, peanuts, egg yolk, wheat bread, milk and fruits, particularly citrus fruits were advised as these foods would provide the daily requirement of the body of various micronutrients and vitamins.

Oral physiotherapy that included ballooning exercises were taught to the patient. Instructions were given to blow the

balloon 15–20 times a day for 1 month. Hence, the customised comprehensive treatment plan for this patient included only medical treatment and oral physiotherapy. Ultrasonic scaling was performed to remove stains, plaque and calculus and it would further help in evaluating the continuation of habit during consequent visits. Oral hygiene instructions were given to the patient that included brushing twice a day using modified bass technique. Extraction of root stumps of 85 was performed under local anaesthesia with epinephrine.

OUTCOME AND FOLLOW-UP

The patient turned up for follow-up after 1 month and increase of 1 mm in mouth opening was noticed, on account of the tissue remodelling achieved by combination of medical therapy and oral physiotherapy that included vitamin B complex capsules, antioxidants and iron supplements (figures 5 and 6). Reduction of burning sensation while eating was also observed; though clinically no changes were observed in the status of the diffuse leucoplakic lesion on the left buccal mucosa. The patient has been kept on further follow-up visit at 3-month interval and was advised to continue the same treatment regime until advised for any change during the follow-up.

DISCUSSION

OSMF is a chronic, insidious disease that is associated with significant functional morbidity and an increased risk for malignancy.[2] Various factors have been thought as causative agents for OSMF. Some of the factors implicated in the aetiology of this disease include areca nut chewing, ingestion of chillies, genetic processes, immunological process and nutritional deficiencies.[3] High copper content of areca nut upregulate lysyl oxidase activity which results in fibrosis.[4] The major areca nut alkaloids are arecoline, arecaidine, arecolidina, guyacoline and guacine. Arecoline, the most abundant alkaloid, might have cytotoxic effects on cells and is also demonstrated to promote collagen synthesis.[5] A long-term population-based cohort study demonstrated that this condition occurred only among those who chewed areca nut in one form or the other.[6] Here, in the present case, the patient had the habit of areca nut chewing along with scented tobacco since 1 year.

It is found that areca nut suppresses the appetite and has a psycho-stimulating effect.[7] The parents usually do not want to intervene as they are aware of the appetite-suppressing potential of areca nut. Since she belonged to a lower socioeconomic rural population and frequently skipped meals, she consumed 2 packets/day. She was a sufferer of child labour and hence could pay for the smokeless tobacco that is readily available in the Indian market. The content of each packet was betel nuts, catechu, lime, cardamom, menthol, natural artificial flavours, mixed spices and added flavour along with separately available packet of scented tobacco. The diffuse white pigmented lesion suggestive of oral leukoplakia was seen in the left buccal vestibule extending from the retromolar area to the premolar area, where she placed the quid. The aetiological description identifies two categories of leukoplakia: those of unknown aetiology and those associated with, or results from, the use of tobacco.[8]

Figure 6 Photograph showing the increased mouth opening after 1 month.

It has been reported earlier that patients with submucous fibrosis had oral cancer as an associated finding in 5.2% and leukoplakia in 26%.[9] The present case falls under stage III classification of oral submucous fibrosis in which leukoplakia is observed, which is termed as sequelae of OSMF according to Pindborg.[10] The classification is divided into three stages based on the progression of clinical features of the disease. The clinical signs and symptoms of the disease 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. The fibrosis in the soft tissue leads to trismus, difficulty in eating and even dysphagia. The diagnosis of the disease can be made based on clinical signs and symptoms. Out of the above clinical features, burning sensation on eating spicy foods, paleness of the buccal mucosa and palate, leukoplakia, stiff and inelastic buccal mucosa and trismus were present in this patient. The peak incidence of this condition is in the 35–54-year age group.[11] But, in the present case, the age of patient is just 14 years that highlights the need for establishment of a preventive and intervention programme in the school for the malicious habit, so that young children can be benefited. Shirzaii[12] also reported a case of OSMF in a 15-year-old boy: the first case report in Iran and Shah *et al*[13] reported a case of OSMF in an 11-year-old Bangladeshi girl living in the UK. Hayes[14] reported OSMF in an Indian girl aged 4 years.

OSMF is a potentially malignant condition.[11] The malignant transformation rate is found to be 4.5–7.6%.[15] The possible precancerous nature of OSMF was first described by Paymaster, who observed the occurrence of squamous cell carcinoma in one-third of his patients with the disease.[16] This accounts for the immediate attention to make a comprehensive treatment plan to cure the disease. In the present case after motivating and counselling the patient to quit the habit, a combination of medical therapy that included vitamin B complex capsules, antioxidants and iron supplements along with oral physiotherapy was advocated, which helped in tissue remodelling to increase mouth opening.

The main problem in treating such a case of OSMF is that the parent does not understand the ill effects and malignant potential of areca nut and tobacco. The overall consumption rate of areca nut and tobacco in India is very high leading to increased prevalence of development of such a premalignant condition in children and adults. Hence, the emphasis on generation of awareness regarding the morbidity of the disease is to be addressed among the general population. Because of the taboos and myths regarding the disease and its cure; efficiently delivering the treatment to the diseased becomes difficult as the cooperation desired from the patient's part is not available. There is a dire need for establishment of a preventive and intervention programme for the malicious habit, the first step being so that young children can be benefited.

Learning points

- Oral submucous fibrosis is one of the most prevalent premalignant conditions in the Indian subcontinent.
- Common signs and symptoms of the disease are difficulty in opening mouth, burning sensation on eating, particularly spicy food, and problems in hearing and speaking.
- The various treatment options are medical, surgical and oral physiotherapy; for the individual patient, customised treatment plan is to be made based on the extent of the disease.
- This case highlights the association of child labour and development of such addicting habits because of the easy availability of these psycho-simulating products at low price.
- In India, lack of awareness and taboos associated with the surgical and interventional treatment modalities are prevalent, urging a serious need for establishment of a preventive and intervention programme for the malicious habit, so that young children can be benefited.

Contributors AD and RM were involved in clinical examination and diagnosis of the patient. AD and SK were involved in diagnosis and preparing a treatment plan. SD was involved in execution of treatment plan, providing motivation, counselling and medication to the patient and her parents. All authors were involved in preparation of the manuscript.

Competing interests None.

Patient consent Obtained.

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