Oral Submucous Fibrosis in a 26-Year-Old Sri Lankan Man Living in South Korea

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INTRODUCTION

Oral submucous fibrosis (OSF) is a chronic insidious scarring disease affecting the oral cavity, and sometimes the pharynx and upper digestive tract. Although occasionally preceded by, or associated with, vesicle formation, it is always associated with a juxtaepithelial inflammatory reaction followed by fibroelastic change of the lamina propria, with epithelial atrophy leading to stiffness of the oral mucosa.\(^1\) The clinical manifestations include blanching and leathery texture of oral mucosa, depapillation and reduced movement of tongue, restricted mouth opening and dry mouth.\(^2\)

Geographically, OSF has a specific distribution and predominantly affects people of South Asia and Southeast Asia–India, Bangladesh, Sri Lanka, Pakistan, Taiwan, southern parts of China, etc.\(^3\) Previous report estimated that globally, about 2.5 million people suffer from OSF, but Indian studies in 2002 have reported that over 5 million people are affected in India alone (0.5% of the Indian population).\(^4\) OSF is also reported among Asian migrant communities living in the North America, parts of Europe, and in South Africa.\(^5\) With an increased immigration from the Indian subcontinent, Korean dental practitioners seem more likely to encounter this disease in the near future.

The present report describes a case of OSF presenting in a 26-year-old Sri Lankan man who is resident in South Korea.

CASE REPORT

A 26-year-old Sri Lankan man presented to the Department of Oral Medicine, Kyungpook National University Dental Hospital with a complaint of trismus and intraoral stiffness. Mouth opening limitation occurred 1-month ago without apparent initiating event. He noted bilateral tightening and pain on the buccal mucosa when opening the mouth. He visited a local medical clinic and was prescribed muscle relaxant before visiting Kyungpook National University Dental Hospital. However, his symptoms were not improved. The patient has no history of significant systemic disease except diverticulitis, trauma, and...
temporomandibular joint sound. He reported a past habit of chewing areca nut 2-3 times a day for 5-6 years while he was living in Sri Lanka.

Clinical examination revealed maximum unassisted mouth opening of 25-mm with bilateral buccal mucosal pain. No other abnormal extraoral findings were noted. Left and right buccal mucosa showed evidence of marble-like appearance and leathery texture (Fig. 1). Maxillary and mandibular vestibules were tight, and fibrous bands were palpable on the lower labial mucosa. There were no pathologic findings including infection, fracture or any other abnormalities on panoramic radiograph (Fig. 2).

A diagnosis of OSF was clinically made based on anamnesis and clinical examination. As the disease is known as a potentially malignant disorder of the oral cavity, the patient was referred to the department of oral and maxillofacial surgery for further evaluation. Facial computed tomography with enhancement demonstrated no remarkable pathologic findings. It was considered unnecessary to perform a biopsy, which would result in further fibrous scar formation and aggravation of the symptoms. Triamcinolone acetonide 5 mg was injected into each buccal mucosa once a month or every two months for 9 months. Following the conservative management, the patient reported remarkable improvement of discomfort, and maximum unassisted mouth opening was increased to 34-mm. He is being followed up without any alteration in the lesion.

DISCUSSION

OSF is a premalignant condition prevalent in South Asian populations.\(^2-5\) It was known that Schwartz first described with the term ‘atrophia idiopathica mucosae oris’ in 1952.\(^3,5\) Subsequently, Joshi coined the term ‘submucous fibrosis’. The etiology of the disease was thought to be multifactorial, and various etiological factors, including capsaicin in chilies, iron and/or vitamin B complex deficiency, genetic predisposition and autoantibodies, have been implicated.\(^5\) However, a number of studies have provided overwhelming evidence that the strongest risk factor for OSF is areca nut, the endosperm of the fruit of the Areca catechu palm tree.\(^2-6\) The nut is a masticatory substance used predominantly by peoples of South and Southeast Asia. Furthermore, it was reported that the development of the disease was directly proportional to the frequency and duration of chewing the nut.\(^5\) OSF is considered a collagen metabolic disorder with

![](https://example.com/image1.png) Intraoral photographs of the buccal mucosa.

![](https://example.com/image2.png) Panoramic radiograph.
histological changes in the extracellular matrix of the lamina propria and connective tissue of the oral mucosa.\textsuperscript{3-5} Areca nut contains alkaloids, flavonoids and copper, all of which are thought to interfere with homeostasis of the extracellular matrix.\textsuperscript{6} Alkaloids, including arecoline, arecaidine, guvacine, and guvacoline, are involved in stimulating fibroblasts to produce excessive collagen. Flavonoids are also known to inhibit collagenase, thereby decreasing collagen degradation. It was demonstrated that the high concentration of copper in areca nut enhanced the activity of lysyl oxidase, an enzyme essential to the final cross-linking of collagen fibers. Areca nut also elicits localized mucosal inflammation through a recruitment of activated T cells and macrophages, subsequently leading to an increase in cytokines and tumor growth factor beta (TGF-beta). TGF-beta is found to play a major role in fibrosis by increasing collagen production and inhibiting collagen degradation.

Clinical manifestations depend on the stage of the disease. The most common initial symptoms and signs include a burning sensation or intolerance to spicy food, blanched oral mucosa, ulceration and dry mouth.\textsuperscript{7} Blanching of the oral mucosa is caused by impaired local vascularity due to increasing fibrosis and usually involves buccal mucosa, labial mucosa, soft palate and anterior faucial pillars. In some cases, blanching may be accompanied by small vesicles that rupture to produce ulcers. In the advanced stage of the disease, the fibrosis leads to restriction in mouth opening and causes difficulty in mastication, speech, swallowing and maintaining oral hygiene.\textsuperscript{7-8} Tongue involvement usually presents with the reduced movement and depapillation.\textsuperscript{9} Loss of hearing may occur secondary to blockage of Eustachian tubes.\textsuperscript{2,4,5,8} Fibrosis can extend into the esophagus, possibly leading to the reduced esophageal mobility and subsequent dysphagia.\textsuperscript{2,4,5,8}

Biopsy is rarely performed on OSF patients due to the observation that such procedure leads to further fibrous scar formation and exacerbation of the symptoms. Isaac et al.\textsuperscript{9} evaluated the microscopic features of OSF and found that OSF with nonspecific chronic inflammation and fibrosis was the most common histological findings, followed by OSF with atrophy of the epithelium and dense fibrosis in the submucosa, OSF with lichenoid reaction, OSF with ulceration, OSF with pseudoepitheliomatous hyperplasia and OSF with dysplasia.

Various classification and staging systems for OSF have been proposed. Pindborg and Sirsat\textsuperscript{10} developed the histopathological classification system in 1966. The clinical classification was proposed based on symptoms by Wahi et al.\textsuperscript{11} Khanna and Andrade\textsuperscript{12} combined histopathological and clinical features of the disease to aid in treatment planning. However, no classification system has been universally accepted. In this case, OSF was classified as grade 2 according to the grading system presented by Kerr et al.\textsuperscript{13}

OSF is associated with an increased risk of oral malignancy. Previous studies reported that malignant transformation rate varies from 7% to 30%.\textsuperscript{4} Although the underlying mechanism of this association remains to be elucidated, it may be related to the fact that an atrophic mucosa may be more vulnerable to carcinogen penetration. Furthermore, OSF may be associated with oral leukoplakia and other premalignant lesions, such as erythroplakia and lichen planus.\textsuperscript{13} It was reported that leukoplakia with OSF had the increased risk of oral cancer. A number of studies were conducted to identify potential predictive oral cancer markers for OSF patients. Teh et al.\textsuperscript{13} investigated the existence of genomic instability regarding the carcinogenesis in OSF and found that 23 hot spot loss of heterozygosity loci were associated with malignant transformation.

Currently, there is no cure for OSF which is considered irreversible once formed. Treatment is based on the severity of the disease. Cessation of areca nut chewing before development of trismus often might resolve OSF.\textsuperscript{5} However, once trismus has developed, treatment aims to restore range of jaw movement, stop the progression of the disease and minimize the risk of malignant transformation. Medical treatment includes intralesional injections of steroids, collagenase, hyaluronidase, interferon gamma, and placental extracts.\textsuperscript{4,5,8} Steroid is thought to decrease inflammation and collagen formation. Collagenase is hoped to facilitate the breakdown of fibrotic areas. It is thought that hyaluronidase, interferon gamma and placental extracts alter collagen synthesis. Other treatment modalities include use of micronutrients and minerals, carbon dioxide laser, pentoxifylline, lycopene, immunized milk, and turmeric.\textsuperscript{5,8} Surgical interventions are required in patients with severe trismus and include excision of fibrous bands, coroniodectomy,
myotomy and soft tissue reconstruction with split thickness skin graft or allograft. It should be noted that aggressive physical therapy following surgery is essential to prevent recurrence of trismus.

OSF is a potentially precancerous condition that causes significant functional morbidity if not properly treated. Dental practitioners in South Korea should be reminded of this disease as it seems to be increasing with population migration.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES