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### Verrucous carcinoma with oral sub-mucous fibrosis: An uncommon occurrence

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#### Abstract

Verrucous carcinoma is a warty variant of squamous cell carcinoma showing exophytic and fungating growth. Oral submucous fibrosis is a chronic, potentially malignant condition and shows a 4.5% malignant transformation rate to squamous cell carcinoma. Transformation of verrucous carcinoma from OSMF is a rare occurrence and needs to be documented for creating awareness amongst clinicians for its early surveillance.

The present report documents a case of verrucous carcinoma arising from OSMF in a 35 year old male, an uncommon combination.

**Keywords:** Verrucous carcinoma (VC), oral submucous fibrosis (OSMF), squamous cell carcinoma (SCC)

#### Introduction

Verrucous carcinoma (VC) is a clinically distinct subtype of squamous cell carcinoma (SCC) showing exophytic, fungating or cauliflower like growth. Chronic tobacco chewing has been attributed as the main cause.<sup>[1]</sup> The mean age of occurrence is usually 60-70 years with a male preponderance<sup>[2]</sup>.

Oral submucous fibrosis is a chronic condition, first described by Schwartz. It has also been defined as an insidious chronic disease affecting any part of the oral cavity and sometimes pharynx. Although occasionally preceded by and associated with vesicle formation, it is always associated with juxta-epithelial inflammatory reaction. Fibroblastic changes in the lamina propri accompany epithelial atrophy which leads to stiffness of the oral mucosa causing trismus and difficulty in eating<sup>[3]</sup>. Oral submucous fibrosis (OSMF), is a potentially malignant condition and shows a 4.5% malignant transformation rate to SCC<sup>[4]</sup>.

Transformation of verrucous carcinoma from OSMF is a rare occurrence and needs to be documented for creating awareness amongst clinicians for its early surveillance. Management of VC is usually surgical excision as these lesions are relatively radio-resistant<sup>[2]</sup>. The present report documents a case of verrucous carcinoma arising from OSMF in a 35 year old male.

#### Case Report

A 35-year old male presented with a chief complaint of difficulty in chewing with decreased mouth opening associated with a burning sensation for the past 3 years. The patient reported a habit of gutka and khaini chewing for the past 10 and 15 years with a frequency of 4-5 gutka per day and khaini 1-2 times a day respectively. Patient used to chew gutka and keep khaini in the right buccal vestibule for about 15 minutes followed by spitting. No associated history of alcohol consumption was reported by the patient.

Examination revealed marked reduction in inter-incisal mouth opening (<12 mm) with associated pain while opening(Fig1).Intraoral examination revealed the presence of an exophytic, cauliflower like proliferative growth on the right buccal mucosa(Fig.2). Size of the lesion was 50 X 30 mm extending 40mm posterior to the right commissure involving gingival and palatal mucosa opposite to 17, 18 extending up to pterygo-manibular raphae. Palpation confirmed inspection findings with regards to its shape, size and extent. Lesion was rough in texture, sessile, firm and tender. Patient reported presence of this growth for the past 1 year which initially though, was very small; has gradually progressed to the present extent. He also reported a gradual decrease in mouth opening with associated history of intermittent burning

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for the past 3 years and pain since 1 year while chewing especially hot/spicy food. Significant reduction in cheek flexibility and restricted tongue movement was present. Blanching was observed on both the sides of the buccal mucosa (Fig.3) involving labial mucosa, floor of oral cavity, ventral surface of tongue and soft palate. Complete examination of posterior part of oral cavity and pharynx could not be established due to restricted mouth opening. Diffuse fibrous bands were noted in anterior and posterior part of oral cavity associated with intense diffuse erythema bilaterally on buccal mucosa. Vertical fibrous bands were more appreciated in the posterior part of the oral cavity, opposite the molars. Lymph nodes were non-palpable and non-tender. It was surprising to note a verruciform growth associated with oral submucous fibrosis. As such occurrence is relatively rare; a tentative diagnosis of verrucous hyperplasia with oral submucous fibrosis was formulated. However, the suspicion of malignancy was not ignored as patient gave a strong history of smokeless tobacco habit associated with a pre-existing premalignant condition. OPG showed no signs of bony involvement of the lesion.

An incisional biopsy was performed and the histopathology revealed presence of hyperplastic squamous epithelium with excessive parakeratinization and bulbous rete pegs pushing into the connective tissue. Cleft like spaces were present showing parakeratin plugging (Fig.4). At higher magnification, hyperchromatic nuclei and numerous mitotic figures could be appreciated. (Fig.5) Overall features were suggestive of verrucous carcinoma.



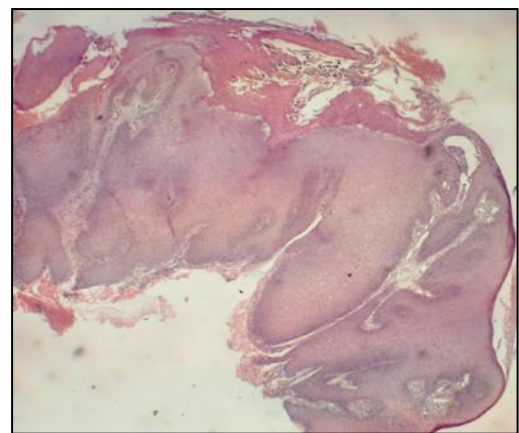
**Fig 1:** Showing reduced mouth opening (<12mm)



**Fig 2:** showing white fungating growth



**Fig 3:** showing diffuse erythema and blanching of right buccal mucosa.



**Fig 4:** Photomicrograph (4x) showing bulbous rete pegs, hyperparakeratinization and parakeratin plugging



**Fig 5:** Photomicrograph (40x) showing hyperchromatic nuclei and numerous mitotic figures.

**Discussion**

First described by Ackermann in 1948, verrucous carcinoma is a variant of squamous cell carcinoma and has multiple names such as Ackermann’s tumour, are Buschke-Loewenstein tumor, florid oral papillomatosis, epithelioma cuniculatum, and carcinoma cuniculatum [5]. VC is a relatively uncommon, locally aggressive, clinically exophytic, low-grade, slow-growing, well-differentiated squamous cell carcinoma with minimal metastatic potential. The prevalence of oral verrucous carcinoma has been accounted to be a mere 16.08% amongst cases of oral squamous cell carcinoma. [6] It has a male predisposition towards the sixth decade and is a slow growing locally invasive lesion, if left untreated. Distant

metastasis is uncommon [7]. A high percentage of reported cases revealed history of tobacco usage, use snuff or smoking among these subjects. Ill-fitting dentures have also been reported as one of etiology. Presentation of neoplasm is usually as an exophytic, papillary with a pebbly clefted surface. Nodal involvement if present is inflammatory [2].

In a cross sectional study by Punnya. V. Angadi *et al.* a total number of 205 known cases of OSMF were reviewed. The total numbers of cases associated with malignancy were 24 cases amounting to 11.75%. Within this, 9.75% were associated with squamous cell carcinoma and 1.95% with verrucous carcinoma. Out of the 24 patients, males amounted to 87% and 1.5% were females. Most patients in this group had the habit for more than 10 years. According to a national survey of head and neck verrucous carcinoma, survival rates in patients of VC treated by surgery alone or in combination with irradiation, survival rates significantly reduced from 86.7% for Stage I to 46.8% for Stage IV, emphasizing the need for early detection and treatment like in any other form of cancer [8].

Oral submucous fibrosis is a chronic, progressive, scarring precancerous condition of the oral cavity seen predominantly in the Indian subcontinent and South East Asia [9]. It is a chronic insidious disease and is well-recognized as a premalignant condition. A community-based epidemiological survey in three areas of India (north and south) recorded the following prevalence of OSMF: 0.36% in Ernakulam, Kerala, and 0.04% in Srikakulam district of Andhra Pradesh (both in South India), and 0.16% in Bhavnagar, Gujarat [10].

It occurs predominantly in the Indian subcontinent where the tobacco and betel nut habit is more prevalent. It has been regarded as a potentially malignant condition. The exact etiology of OSMF is unknown. The various hypotheses put forward so far suggest a multifactorial origin for this condition. Alongside the role of local irritants such as capsaicin, tobacco, areca nut, pungent and spicy foods, and alcohol, an underlying systemic predisposition is likely because of the geographical and ethnic distribution of OSMF. Among the systemic factors, the main ones incriminated are chronic iron and vitamin B-complex deficiency, anemia, and a genetic predisposition to the disease [11].

In a longitudinal study by Murti *et al.* sixty-six patients with oral submucous fibrosis were followed-up for a period of 17 yr (median observation 10 yr) in Ernakulam District, Kerala, India. Oral cancer developed in five (7.6%) patients. The malignant transformation rate in the same sample was 4.5% over a 15-yr observation period (median 8 yr). These findings impart a high degree of malignant potential to this condition [12].

Paymaster in as early as 1956 commented upon the precancerous nature of submucous fibrosis after observing a slow growing carcinoma in one third of his patients with the disease [13].

A study by Yusuf and Yong (2002) also does explain that the presence of lime within the betel quid/paan masala/gutkha mixture has been shown to release reactive oxygen species from extracts of betel nut could contribute to the cytogenic damage involved in the etiology of oral cancer [14].

All the reported cases in literature depict consumption of areca nut in some or the other form, pointing towards the fact that the primary lesion was OSMF which was succeeded by the formation of VC. In 3 cases gutka was consumed, one of its constituents being tobacco, which is the primary etiological factor for oral cancer, pointing towards the fact that VC must have arisen de novo and its occurrence with

OSMF is coexistential irrespective of the age of the patient [15].

Another study done in Taiwan by Shiao and Kwan (1979) on 35 cases of OSMF also demonstrated that betel nut chewing was highly correlated with the occurrence of the disease. They however pointed out that other factors like tobacco, alcohol and hot spicy foods were not etiologic agents. [16]

In the present case, subject was relatively young in 3<sup>rd</sup> decade of life with the habit of consuming khaini (tobacco quid) and gutka (a form of processed areca nut) for last 10 and 15 years respectively. It would not be prudent to generalize the findings with a single case report. However, authors' belief that the risk for verruciform like lesions increases with smokeless tobacco whereas risk for SCC increases with smoking among subjects with OSMF.

The literature still lacks sufficient documentation of OSMF associated with VC and no prospective studies exist that address this coexistence. Perhaps the epithelial atrophy common in this condition makes the epithelium more vulnerable to the action of the carcinogens from tobacco habits.

### Conclusion

Literature lacks studies relating to cases where OSMF has converted to verrucous carcinoma and there is dire need for the same to make the diagnoses less challenging and for a better understanding of the entity. A longitudinal, multidisciplinary approach combined with precise histopathological findings and a careful review of all available literature only can help the oral clinician identify this multifaceted entity facilitating an early treatment protocol for the afflicted patient.

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