Verrucous Carcinoma of the Buccal Mucosa – A Case Report

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Abstract
Oral verrucous carcinoma (OVC) is a rare but well differentiated form of oral squamous cell carcinoma mainly seen in sixth or seventh decade of life with male predominance. Smokeless tobacco is found to be the most etiological factor in development of verrucous carcinoma although other potentially malignant lesions may act as a predisposing factor. As there are very few cases of VC and OSMF reported in the literature we present a rare case of OVC in association with oral submucous fibrosis of an elderly patient with tobacco chewing habits.

Keywords: Exophytic growth, Oral cancer, Oral submucous fibrosis, Tobacco chewing, Verrucous carcinoma, Verrucous hyperplasia.

Introduction
Cancer is an important public health problem in many parts of the world. According to the International Agency for Research on Cancer of the World Health Organization (IARC–WHO), cancer rates are expected to increase at an alarming rate: from 10 million new cases globally in 2000 to 15 million in 2020.1 Oral cancer is among the 10 most common cancers worldwide, and is especially seen in disadvantaged elderly males. Most cancers of the oral cavity are oral squamous cell carcinomas (OSCC), and tobacco, alcohol and betel use are the main risk factors.2

Oral Verrucous Carcinoma (OVC), is a variant of Squamous Cell carcinoma (SCC), was first described by Lauren V Ackermann in 1948 so it was known as “Verrucous Carcinoma of Ackermann” or Ackermann’s Tumor” other names used in literature are Buschke-Loewenstein tumor, florid oral papillomatosis, epitheliomacuniculatum, and carcinoma cuniculatum.3

The term VC (Verrucous carcinoma) refers to those exophytic mucosal or cutaneous squamous tumors that are heaped above the epithelial surface with a papillary micronodular surface and pushing margins. VC representing 2–12% of all oral cancers is mainly found in elderly men and closely aligned with the use of tobacco.4 The most common site of occurrence is oral cavity, other sites being larynx, pyriform sinus, esophagus, nasal cavity and paranasal sinuses, external auditory meatus, lacrimal duct, skin, scrotum, penis, vulva, vagina, uterine cervix, perineum, and the leg.3

This heavily keratinized, well differentiated variant of squamous cell carcinoma shows warty-like aspects and lacks conventional cytologic findings of atypia, exhibiting only locally invasiveness and no metastatic potential.5 Lymph node involvement and distant metastasis are rare in VC. It occurs more frequently in males over the sixth decade. Histopathologic and clinical diagnosis of oral VC may be difficult, so close cooperation between pathologist and surgeon is required in order to identify the nature of the lesion.6

We report a case of Verrucous carcinoma in the oral cavity in a 52 year old male patient with tobacco chewing habits.

Case Report
A 52 year old patient reported to the department of Oral medicine and Radiology with a chief complaint of oral discomfort. The patient elicited positive history of tobacco and areca nut mixed with slaked lime chewing 6 times per day for last 20 years and used to keep it in the left buccal vestibule. 2 years ago he experienced burning sensation on consumption of hot and spicy food and difficulty in mouth opening. 6 months ago he experienced a growth on the left buccal mucosa. The growth was initially small in size and increased gradually over time, with lateral and vertical expansion till the present size.

On clinical examination a white exophytic growth with pebbly surface and finger like projections was seen on the left buccal mucosa (Figure 1) at the line of occlusion and about 1 cm posterior from the left commissural area surrounded by a thick smooth homogenous plaque extending posteriorly till the ramus region and anteriorly till the labial vestibule. The lesion was irregular in shape of approximately 2x3 cm in size. An extensive ulceration was observed with irregular...
borders, 2 cm in size adjacent to 37 and 38 surrounded by erythematous atrophic area. Both the lesions were tender on palpation, firm in consistency and indurated. Patients mouth opening was 21 mm. Blanching with vertical fibrotic bands were palpable on both right and left buccal mucosae posteriorly (Figure 2).

A clinical diagnosis of Verrucous carcinoma of left buccal mucosa and OSMF was made. Single left submandibular lymph node was enlarged of approximately 1.5 x 1 cm in size, ovoid in shape, tender on palpation, mobile and firm in consistency.

Patient was sent for blood investigations and the report revealed Haemoglobin level of 12 gm%, Raised ESR of 42 mm. An incisional biopsy was performed under local anesthesia and two specimen from anterior (specimen 1) and posterior (Specimen 2) parts of the left buccal mucosa were sent for a histopathological examination.

Microscopic examination showed Specimen 1 - parakeratinized stratified squamous epithelium showing epithelial hyperplasia with normal cell maturation pattern, rete ridges were wide and elongated with parakeratin plugging. Specimen 2 – parakeratinized stratified squamous epithelium which is dysplastic with tumor cells arranged in form of sheets invading underlying fibrocellular connective tissue.

Discussion
Due to the mechanism by which oral mucosa undergoes transformation in OSCC, i.e. chronic exposure to carcinogens in the form of tobacco, betel nut, with alcohol as a cocarcinogen, there are a wide variety of molecular alterations that have been associated with carcinogenesis. In addition to dysregulation of oncogenes and tumour suppressors, cytogenetic changes, epigenetic changes and mitochondrial mutations have been implicated in development of OSCC. These alterations are reflected in varying degrees in potentially malignant disorders.7

The etiology of verrucous carcinoma is not well defined. Persons who use smokeless tobacco are at a higher risk of developing verrucous carcinoma than non-tobacco users. Human papilloma virus has been identified in the cells of this tumor but is still undetermined.8 VC, is a less common tumour, represents 4.5-9% of oral squamous-cell carcinomas. The most common sites of occurrence within the oral cavity are the buccal mucosa, gingiva and the alveolar ridge; other sites of involvement include the palate, floor of the mouth and lip.9 VC in the oral cavity is characterized by a cauliflower-like exophytic growth with a cleft, pale, warty, fungating, locally aggressive, ulcerated tumor attached by a broad base, is well circumscribed and it is clearly demarcated from the adjacent mucosa with a pebbly mamillated surface.10

Oral submucous fibrosis (OSF) is a disease mainly associated with the chewing of areca nut, an ingredient of betel quid, and is prevalent in South Asian populations. It causes significant morbidity (in terms of loss of mouth function as tissues become rigid and
morbidity and mortality (when transformation into squamous cell carcinoma occurs). The direct contact of the quid mixture with oral tissues results in their continuous irritation by various components, including biologically active alkaloids (arecoline, arecaidine, arecodoline, guvacoline, guvacine, flavonoids (tannins and catechins) and copper. There are very few cases of VC with OSMF reported in literature. It has been proved that 20% of the cases of VCs have small foci of well-differentiated SCCs within them and such tumors should be correctly recognized. A correct biopsy with sufficient depth can lead to the precise differentiation between the comparatively better VCs and those with frank malignant foci. In our present case we have seen development of OSCC within Verrucous carcinoma.

A distinction should be made between verrucous hyperplasia and verrucous carcinoma. Verrucous hyperplasia was described by Shear and Pindborg in 1980. It is more superficial and does not extend deeper than the surrounding normal epithelium. It shows dysplasia and can later develop into verrucous carcinoma or squamous cell carcinoma. Verrucous carcinoma, on the other hand, extends more deeply, pulling the adjacent normal epithelium at its margin. Verrucous carcinoma can develop de nova or from pre-existing leukoplakia and in older individuals. OSMF, a premalignant condition caused by chronic betel nut chewing, can also lead to the development of verrucous carcinoma irrespective of the age of the patient.

Conclusion

It is well established that Verrucous carcinoma can arise from potentially malignant disorders and it is expected that early screening and diagnosis of these disorders can help in improving the overall health quality of the patient and prevent mortality. As VCs in oral cavity are a distinct clinical entity with varied histopathology so it becomes mandatory for the oral physicians to detect and identify its course of development. We report a case of Verrucous carcinoma in association with OSMF which is a rare occurrence.

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References