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Case Report

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VERRUCOUS CARCINOMA IN AN UNCOMMON LOCATION: A CASE REPORT

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ABSTRACT:

Verrucous carcinoma is a rare and distinct clinicopathologic variant of well differentiated squamous cell carcinoma. Its correct clinical and histopathological examination is very important in diagnosis and treatment planning. A case report of histologically proven oral verrucous carcinoma on a rare location tongue is hereby presented associated with history of tobacco chewing.

Key words: Carcinoma, Oral Cavity, Tongue, Verrucous

INTRODUCTION

Verrucous carcinoma (VC) is an unusual but distinct variety of well differentiated squamous cell carcinoma first outlined as a clinico-pathologic entity by Ackerman in 1948.[1]It is also called as Ackerman's tumour, Buschke-Loewenstein tumor, Florid oral papillomatosis and Epithelioma cuniculatum. [1,2] Primarily being a squamous mucosal lesion, this lesion may also be found on cutaneous surfaces. Whether the carcinoma occur in the upper aero digestive tract, genitalia or on extremities, they are the same neoplasm with slow growing, locally invasive and non-metastasizing behaviour. [3] In upper aero digestive tract the predominant risk is seen in the oral cavity and larynx.[3] In the oral cavity, verrucous carcinoma constitutes 2 to 4.5 % of all forms of squamous cell carcimonas. [4] The precise aetiology is not known, although factors such as, smokeless tobacco, betel quid chewing, poor oral hygiene and human papilloma virus have been associated.[5]

The term "Verrucous" is given because of its fine, finger like surface projections.[6] The difficulty in the clinical or histopathological diagnosis of verrucous carcinoma has been attributed to the slow growth and its benign histologic features. The dorsum of the tongue is an extremely unusual location for VC, as most cases intraorally are noted on the gingiva and the buccal mucosa, followed by the mandibular vestibule, hard palate, and areas of chronic tobacco placement.[7] We hereby present a case with oral verrucous carcinoma on the dorsum of the tongue which is an extremely rare location for this lesion. A through literature search was done and the reported cases with verrucous carcinoma on the tongue are listed in Table 1.

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CASE DETAIL:

A 56 years old male presented to our department with a complaint of growth on the tongue since 3 months which was associated with mild pain and gradually increasing in size. Growth was associated with burning sensation in the mouth which got aggravated by hot and spicy food. There was no history of trauma, oral bleeding but patient reported of dysphagia and speech problems. No paresthesia, anesthesia was reported. No associated systemic symptoms like fever, weakness, malaise. Tongue movements were normal. Patient is hypertensive and was on medication for the same. Patient was chewing pan, tobacco and areca nut 20 per day since 25 years. On intraoral inspection of the lesion single ulceroproliferative, exophytic sessile lesion with cauliflower-like appearance was seen on the anterior and left lateral border of the tongue which was approximately 2cm x 2 cm in size (Figure 1). Presence of extensive white areas on the entire surface of tongue, floor of mouth, gingiva on the right aspect of teeth, buccal mucosa extending till pterygomandibular raphe region (Figure 2). A provisional diagnosis of malignancy in respect to tongue & homogenous leukoplakia in respect to gingiva, tongue, floor of mouth, buccal mucosa was given. On performing toluidine blue staining the lesion showed positive uptake of the dye (Figure 3). Patient was advised biopsy of the lesion. The biopsy was inconclusive as sample was inadequate. A repeat biopsy revealed hyperplastic parakeratinized stratified squamous epithelium showing both exophytic and endophytic growth. Parakeratin plugging was seen extending into the epithelium. The rete pegs are bulbous with the basal layer of epithelium showing minimal atypia in the form of basilar hyperplasia with intact basement membrane. The underlying connective tissue showed presence of chronic inflammatory cells chiefly lymphocytes with presence of dilated vascular spaces engorged with RBCs, thus a final diagnosis of verrucous carcinoma was rendered.

Reported cases of verrucous carcinoma on tongue

Name of the author	No of cases	Article title
Lai-Kuan Zhu et al[13]	2	A clinicopathological study on verrucous hyperplasia and Verrucous carcinoma of the oral mucosa
Vilela FA et al[14]	1	Verrucous carcinoma of the tongue
Anjana sadasivan et al[5]	5	Verrucous lesion of oral cavity treated with surgery: analysis of clinicopathologic features and outcome.
Junu Ojha et al[7]	1	White lesion on the dorsum of tongue
Michiro Kawakami et al[9]	2	Verrucous Carcinoma of the Tongue: Report of two cases

Kuldeep Singh et al[4]	1	Verrucous Carcinoma (Ackerman's Tumour) of Mobile Tongue
D.O Awange et al[15]	1	Oral verrucous carcinoma: report of two cases and review of literature
Tadashi Terada[16]	3	Verrucous Carcinoma of the Oral Cavity: A Histopathologic Study of 10 Japanese Cases
Luciane H. Azevedo et al[8]	2	Treatment of Oral Verrucous Carcinoma With Carbon Dioxide Laser

Table 1: Showing the reported cases of verrucous carcinoma on the tongue

DISCUSSION:

Verrucous Carcinoma is recognised as a nonaggressive variant of well differentiated squamous cell carcinoma. It has an exophytic, cauliflower-like appearance and occurs predominantly in the elderly above 55 years with male predilection.[8] The aetiology is indeterminate, but consumption of tobacco in the form of chewing, snuffing, or heavy smoking is considered the most important factor. According to few studies human papillomavirus (HPV), mainly genotypes 2, 6, 11, 16, and 18, could be another possible causative factor.[7]Our case also had histories of tobacco chewing. Regional lymph node metastases are exceedingly rare, and distant metastases have not been reported. [9]

VC is primarily an oral lesion; however, it has also been reported in other sites, such as the nasal cavity, oesophagus, larynx, and genitals.[7] This case report is unusual since a rare location has been involved. The dorsum of the tongue is an extremely rare location for VC.[7] Its clinical association with leukoplakia is significant, and the evidence indicates that untreated leukoplakia may develop into a verrucous carcinoma in time.[10]The clinical presentation varies from an initial superficial, well-defined thick, white lesion which later progresses as more diffuse with irregular borders, with exophytic or plaque like growth, and with papillary or verruciform surface projections as seen in our case. The intensity of the white colour depends on the amount of keratin production. The lesion may also present as pink or erythematous.VC may also invade the bone, cartilage, muscles, and the salivary glands if not treated.[7]Verrucous carcinoma has a heavily keratinized, or parakeratinized, irregular clefted surface with parakeratin extending deeply into the clefts. The prickle cell layers show bulbous hyperplasia. The tumour has a well-defined lower border and basal lamina. Atypia is minimal, and there is usually a subepithelial inflammatory infiltrate.[10] Our case presented with similar histopathological findings. For an accurate diagnosis, the biopsy specimen must contain the full thickness of the epithelial component along with the underlying connective tissue.[11] The surgeon must take a specimen with full thickness of the tumour to make a correct diagnosis. However, it is still sometimes difficult to provide adequate specimens from regular biopsies of large lesions thus, VC is often under

diagnosed. When VC was highly suggested by clinical appearance, patients should undergo deep incisional biopsies under general anaesthesia.

The preferred treatment of VC is complete surgical excision without radical neck dissection. Surgical resection may be required in advanced cases with extensive involvement. Radiation and chemotherapy may be used as adjuncts. According to some authors, radiotherapy is usually not indicated because it carries a risk of development of a poorly differentiated carcinoma within the VC. Chemotherapy is reserved for inoperable cases but is not considered as an effective treatment, as it reduces the size of the lesion only temporarily. Few studies in the literature also support alternative treatments, such as combination therapy of radiation and chemotherapy or radiation therapy and surgery.[12] This patient was advised with surgical excision but due to financial constraints they opted out for treatment. Other therapeutic options for treatment are electrosurgery, cryosurgery and lasers.[8] VC has an excellent prognosis because of its low-grade nature and high level of differentiation. More than 90% of patients are disease-free after 5 years.[12]

CONCLUSION:

VC of the oral cavity is a different clinicopathologic tumour distinguished from squamous cell carcinoma because of its local invasiveness, nonmetastasizing behaviour, and can be a difficult diagnosis because of its diverse appearance. The dorsum of the tongue is an infrequent site for its existence. Adequate sampling is required to avoid histopathological misdiagnosis. Thus both clinicians and pathologists must be careful about warty and exophytic lesions in the oral cavity.

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FIGURES:

Figure 1: Clinical picture of exophytic growth on the tongue

Figure 2: Clinical picture showing the presence of extensive white areas on the surface of tongue, gingiva, buccal mucosa extending till pterygomandibular raphe region

Figure 3: Toluidine blue staining of the growth showing positive uptake of the dye

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