



RESEARCH ARTICLE

VERRUCOUS CARCINOMA OF TONGUE HAILING TOGETHER WITH HPV 16 - A REPORT OF A RARE CASE

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ABSTRACT

Verrucous carcinoma (VC) was first described as a distinct clinico-pathological entity by Ackerman in 1948. The name of this variety of Squamous cell carcinoma makes it distinctive. Although this carcinoma has been reported in extra-oral sites, it is a rare intra-oral entity, commonly found in oral mucosal sites. The common sites of occurrence of VC are buccal mucosa> gingiva> alveolar ridge> palate and floor of mouth and very rarely on tongue. Here we report a rare case of a large Verrucous carcinomatous lesion on left lateral anterior region of tongue in 78 year old female associated with human papilloma virus 16 without any history of tobacco related habits.

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INTRODUCTION

Verrucous carcinoma is a rare tumor, first described by Lauren V. Ackerman in 1948 (Ackerman, 1948). In 75% of cases it is found in the oral cavity, this tumor is predominantly seen in males with the peak occurrence in the sixth decade, and smoking is the main recognized etiopathological factor. Additionally human papilloma virus (HPV) type 16 was identified as a causative agent in 1% of patients (Ackerman, 1948). It is considered as a special form (garden) variety of squamous cell carcinoma with a characteristic verrucous presentation. Within the oral cavity it is localized most commonly in the hard palate and floor of the mouth. Localization within the tongue is very rare and has specific clinical and histological features. The tumor has slow growth, locally invasive in nature and has no metastatic potential (Alkan et al., 2014). This tumor is characterized with a deep penetrating type of growth. Histologically it creates thick verrucous and blunt wide stromal indentation, with the basement membrane preserved.

The tumor consists of a well-differentiated stratified epithelium with no characteristics of atypical cell. Mitoses is very rare and usually found only at the basement layer. Externally tumor surface is usually covered with a thick layer of keratin and is diagnosed in only 1 to 3 people of every 1000,000 persons each year (Bouquot, 1998). Etiopathogenesis of oral verrucous carcinoma is strongly associated with the chronic use of tobacco related habit that is pan chewing, smoking and snuff dipping. Clinically it produces a thick white plaque resembling an exophytic cauliflowerlike growth. The purpose of this article is to discuss this carcinomatous lesion which was present on the left side of tongue in association with HPV 16.

Case Report

A 78 year old female patient reported to the Department of Oral and Maxillofacial Surgery, D Y Patil University School of Dentistry, Nerul, Navi Mumbai with chief complaint of growth (swelling) on the tongue since last 5 years. Patient noticed a small innocuous growth (swelling) on the tongue five year back which had gradually increased to the present size; patient also started having pain and discomfort in the lesion since last one month.

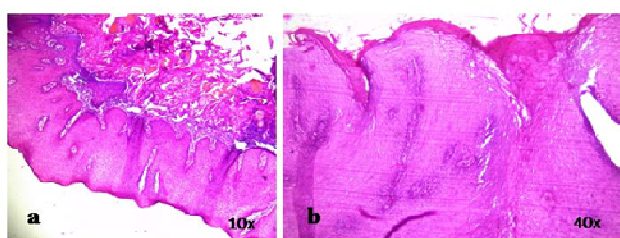
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She however did not give any history of tobacco related habit and neither suffering from any systemic ailment, patient also had no history of any chronic trauma to the tongue and no other relevant family history. On intra-oral clinical examination an exophytic papillomatous cauliflower like fungating growth on left half of the tongue, was seen extending from the left lateral border to the tip of the tongue (Fig1a). Size of the lesion was approximately 3x2cm in anteroposterior and mesiodistal dimension.



**Fig. 1. a. Intraoral clinical photograph
b. Immediate post operative Co2 laser excision of the lesion**

The rest of the tongue was depapillated, on palpation the lesion showed no evidence of bleeding and induration. No evidence of palpable lymphnode in submandibular and cervical region. Based on history and clinical finding a provisional diagnosis of verrucous hyperplasia or verrucous carcinoma was given. The patient was taken for incisional biopsy which was performed under local anaesthesia. Histopathologic evaluation of the given specimen revealed hyperplastic bulbous squamous epithelial rete ridges almost at the same level throughout the specimen. epithelium showed parakeratin clefting hyperkeratosis and koilocytes, however there was no evidence of dysplasia. Underlying connective tissue was fibrocellular with mild to moderate inflammatory cell response with close approximation to the epithelium. Based on these findings a histopathological diagnosis of verrucous carcinoma was derived.



**Fig.2a. H&E stained photomicrograph showing bulbous squamous epithelium and parakeratin clefting
b. H&E stained photomicrograph showing parakeratin clefting and koilocytes**

The patient was further scheduled for wide surgical excision of the lesion. The surgery was carried out using CO2 laser knife under general anesthesia (Fig 1b).

The histopathological examination of the excised specimen confirmed our previous histopathological diagnosis (Fig 2a, b). This specimen was then subjected to Immunohistochemical staining using HPV16 antibody, in which the epithelial cells showed a positive staining for HPV16 (Fig 3a, b). Thus the final diagnosis of verrucous carcinoma associated with HPV 16 was reached.

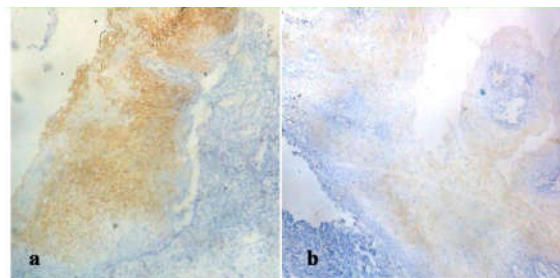


Fig 3a & b. Photomicrograph showing Positive immunohistochemical staining for HPV 16 antibody

DISCUSSION

Verrucous carcinoma is a distinct variant of well differentiated squamous cell carcinoma with low grade malignancy, slow growth and no metastatic potential. It occurs in elderly patient around 60 to 70 years of age and is a common finding in males. These lesions are found to be occurring more often on the skin, penis, vulva and uterine cervix, and rarely in the oral cavity.² Its occurrence in the oral cavity is commonly associated with habits of smokeless Tobacco, betel nut chewing or poor oral hygiene. Hence also termed as Snuff dippers' cancer (Koch *et al.*, 2001; Mc Guirt, 1983). However certain authors have reported the occurrence of VC in non tobacco habitués. On further investigations some of these patients were found to be HPV positive, hence relating its co-existence. The likelihood of detecting HPV in VC was found to be 29.5% (Malik *et al.*, 2014). VC in the oral cavity is commonly found on buccal mucosa, gingiva, alveolar ridge, palate and floor of the oral cavity, its occurrence on the tongue is a rarity as in our case (McCoy, 1981). Oral Vc can be associated with HPV 16 or 18. In the present case we found the stain positive for HPV 16. The patient was further treated with surgical excision which is considered as a primary treatment protocol according to the literature (Lubbe *et al.*, 1996).

Conclusion

Verrucoid lesions in the oral cavity, whether diagnosed as verrucous hyperplasia or verrucous leukoplakia, should be thoroughly investigated. Occurrence of verrucoid lesions in non-tobacco habitués patient should further be investigated for viral etiology and treated accordingly

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