

## Verrucous Carcinoma of Oral Cavity - A Case Report and the Review of Literature

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### ABSTRACT

Verrucous carcinoma (Ackerman tumor) is an uncommon variant of squamous cell carcinoma. It has some unique clinically and histological features different from squamous cell carcinoma. Here we report one such case of verrucous carcinoma and review etiology, pathogenesis and the management of this uncommon tumor.

**Key words:** Verrucous, carcinoma, Squamous cell carcinoma, Oral cavity, Human Papilloma Virus

### Introduction

A white lesion located in the oral cavity can pose problems for both the clinician and the pathologist. Verrucous carcinoma (VC) was first described by Lauren V Ackerman in 1948 as “a distinct variant of differentiated Squamous cell carcinoma (SCC) with low grade malignancy, slow growth and no or only low metastatic potential”. VC typically involves the oral cavity, larynx, genitalia and oesophagus[1]. VC of oral cavity is seen in patients who have a habit of use of snuff and chewing tobacco, called as “snuff dipper's cancer.” It manifests as a verrucous, exophytic or endophytic mass that typically develops at sites of chronic irritation and inflammation. It enlarges slowly but penetrates deeply into the skin, fascia and even bone. Various synonyms used to describe this tumor, are Ackerman's tumor, Buschke

Loewenstein tumor, florid oral papillomatosis, epithelioma cuniculatum, and carcinoma cuniculatum[2]. It usually appears as a painless, white, warty, exophytic plaque attached by a broad base resembling a cauliflower. It is seen more commonly in men than in women, in 6<sup>th</sup> or 7<sup>th</sup> decade of life. The most common sites of oral mucosal involvement include the buccal mucosa, followed by the mandibular alveolar crest, gingiva, and tongue [3]. The exact etiology of verrucous carcinoma is unknown, tobacco chewing and smoking are found to be the causative factors. Various treatment modalities include surgery, chemotherapy, radiation or combination of these and photodynamic therapy which has been recently reported [4]. Here we report one such case of verrucous carcinoma and review etiology, pathogenesis and the management of this uncommon tumor.

### Case Report

A 40 year old male came with a chief complaint of a large growth in posterior aspect of mouth since 5 years. The history of present illness revealed that the growth was initially small in size and had been slowly growing over the period of time. There was no

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pain or bleeding associated with the lesion. Past Medical history was non-significant. The patient gave a history of smoking cigarettes 3-4 packets/day since 15 years and alcohol consumption. Intra-oral examination revealed a mixed red and white well demarcated painless verruciform growth in left retro-molar region. The lesion appeared exophytic and cauliflower-shaped measuring 3.5cms X 3.5cms extending antero-posteriorly from lower third molar teeth to pterygo-madibular raphae. On palpation, the lesion proper was firm, leathery in consistency. Intra-oral examination reveals the presence of mixed red and white well demarcated painless verruciform growth in left retro-molar region. The lesion appeared exophytic and cauliflower-shaped measuring 3.5cms X 3.5cms extending antero-posteriorly from lower third molar teeth to pterygo-madibular raphae and superio-inferiorly from 1cm from upper buccal vestibule to lower buccal vestibule

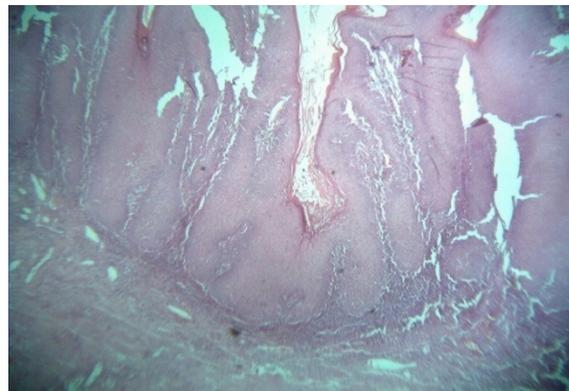
(Fig 1). On palpation, the lesion proper was firm, leathery in consistency. A provisional diagnosis of verrucous hyperplasia was arrived at and for confirmatory diagnosis a biopsy was performed. Local anaesthesia was give around the lesion, and the small portion of the lesion 1cm x 1cm (FIG 2) was incised for biopsy and sent for histopathological examination. The histopathological examination revealed epithelial proliferation with down growth of epithelium into connective tissue showing bulbous blunt rete pegs. In the epithelium, cleft like spaces lined by a thick layer of parakeratin extend from the surface deep into the lesion. Parakeratin plugging was noticed in the epithelium (Fig 3). Chronic inflammatory cells were present. Based on these findings a final diagnosis of VC was given. Complete excision of the lesion was done and follow-up after one year showed uneventful healing without any recurrence.



**Fig 1:** Presence of mixed red and white well demarcated painless Verruciform growth in left retro-molar region



**Fig 2:** Gross specimen



**Fig 3:** The histo-pathological examination revealed epithelial proliferation with down growth of epithelium into connective tissue showing bulbous blunt rete pegs

## Discussion

OSCC and VC are malignant tumours of the epithelial tissue origin. OSCC is the most common oral cancer and despite various attempts and approaches to intervene the disease, remains a serious problem of oral health worldwide. However, VC which is a highly differentiated variety of OSCC shows a low degree of malignancy. Oral cavity is the most common site of occurrence of VC, where it represents 2-12% of all oral cancers[1,5]VC may be seen at different sites including skin, paranasal sinus, bladder and anorectal region, male and female genitalia, sole of the foot, and ear. Schrader et al and Jordan reported VC as slow-growing, exophytic, well-demarcated hyperkeratotic lesions. These typically present as extensive, white, and warty lesions[6,7].The etiology of VC is not well-defined. Human papilloma virus (HPV) has been considered one of the causative factors. Smoking seems highly associated with the development of mucosal VC of the neck and head. Poor oral hygiene, presence of oral lichenoid, and leukoplakic lesions may act as predisposing factors[8].Shear and Pindborg reported that out of 28 patients with verrucous lesions, 24 (86%) used tobacco, and one was an areca quid chewer. Tobacco appears to be a major factor in causation of verrucous lesions[3].In our patient, cigarette smoking seemed the most causative factor among those mentioned aboveVC is found predominantly in men older than 55 years of age. In areas where women are frequent users of spit tobacco, however, elderly females may predominate. Hansen et al. found male and female ratio to be 4:1[9].The most common sites of oral mucosal involvement include the mandibular vestibule, the buccal mucosa, and the hard palate. Oral VC has a characteristic gross appearance. These lesions are almost always large, exophytic, soft, fungating, slow growing neoplasms with a pebbly mamillated surface. The lesion appears as a diffuse, well-demarcated, painless, thick plaque with papillary or verruciform surface projections resembling a cauliflower[1,3,5,7].The term “verrucous” was applied for lesions showing a keratotic exophytic surface composed of sharp or blunt epithelial projections with keratin-filled invaginations (plugging), but without obvious fibrovascular cores. The histological features of VC, for example, verrucous surface and “elephant feet-” like down growth seeming to compress the underlying connective tissue and typically showing

minimal or absent cytological atypia, are widely known. Distinction from classical SCC is a frequent problem also for clinicians because of the extensive nature of the lesion mimicking an invasive cancer[7-9]An important help could be offered by molecular approaches. VC shows the characteristics of cell kinetics that are similar to those of normal epithelium and not to conventional SCC. S-phase is confined to basal layer, unlike the invasive cancer. By flow cytometry, VC is a diploid lesion; on the contrary, the conventional SCC often shows aneuploidy and genomic instability The incidence varies from 4.5% to 9%, or even higher in some centres. In the oral cavity, VC constitutes 2 to 4.5 % of all forms of SCC[9,10].Rajendran, et al., recorded leukoplakia in association with OVC in 48% of their patients. The clinical association with leukoplakia and OVC is significant since untreated longstanding leukoplakia could progress to a verrucous cancer in time. Overexpression of the p53 (modulation of cell cycle control) oncogene is similar to that which has been observed in other head and neck cancers, and there is a suggestion that HPV and p53 may work through the same pathway[11].In our cases, the clinical diagnosis was verrucous hyperplasia, whereas final diagnosis was VC following the incisional biopsy. VC typically has a heavily keratinized, or parakeratinized, irregular clefted surface with parakeratin extending deeply into the clefts[12]. Our case presented histopathological findings similar to those mentioned above. The prognosis of VC is better than that of other kinds of life threatening malignant tumors. Various treatment modalities include, surgery, chemotherapy, radiation, or combination of these and photodynamic therapy have been recently reported. The use of buccal fat pad has increased in popularity because of its reliability, ease of harvest, and low complication rate. It has been used as a pedicle or free graft in reconstructing small to medium sized defects intraorally. Surgery is considered the primary mode of treatment for verrucous carcinoma. Irradiation alone or in combination with surgery is rarely performed. Combined therapy can be useful when the tumor extends to the retromolar area. Koch et al suggested that patients with oral cavity VC treated with surgery had better survival[11-13].In our case, we treated our patient with surgical excision.McClure et al reported that extensive lesions in the oral cavity may benefit from combined therapy.

When surgery is not indicated, other treatment modalities such as cytostatic drugs may be preferred;  $\alpha$ -interferon (IFN) seems to support the therapy by delaying the growth of the tumor but does not take the place of surgery alone[14]. The literature supports the concept that radiotherapy is contraindicated in the treatment of VC for the occurrence of radiation-induced anaplastic transformation. Ferlito and Recher reported that neck dissection is not indicated in laryngeal verrucous carcinoma because laryngeal VC has so far has never metastasized to the cervical lymph nodes or to other organs[15]. Verrucous hyperplasia and VC may not be distinguished clinically. It should be kept in mind that verrucous hyperplasia may transform into VC or squamous-cell SCC, acting as a potential precancerous lesion[1,3] Our case was VC. Thus, both clinicians and pathologists must be careful about warty and exophytic lesions in the oral cavity.

### Conclusion

OVC is a different clinicopathologic tumor distinguished from the usual OSCC because of its local invasiveness, non-metastasizing behaviour, and special clinical appearance. It is essential that the pathologist alerts the clinician to the progressive nature of the lesion and recommends complete excision or close follow-up. Further studies are still needed to compare the clinical behaviour of patients with VC with that reported in Western countries.

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