

Verrucous Carcinoma (Ackerman's Tumor) of the Tongue – Case Report

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Abstract: *Verrucous carcinoma (VC) is a relatively rare well-differentiated variant of squamous cell carcinoma with a specific clinical appearance and histopathologic features. It can grow large in size and could be locally destructive with low rate of metastasis. We present a case of 78-year old female with VC of the tongue. Clinical and histopathologic findings are discussed as well as the treatment plan*

Keywords: verrucous carcinoma, squamous cell carcinoma, oral cavity, tongue

1. Introduction

Verrucous carcinoma (VC) is a rare variant of squamous cell carcinoma with a specific biology, clinical appearance and potentially deceptive histopathologic appearance. VC was reported for first time in 1941 by Friedell and Rosenthal and the comprehensive description of the entity is given in 1948 by Ackerman [1]. It is also called Ackerman's tumor.

The mucous membranes of head and neck region are sites of predilection for VC, with the larynx and oral cavity especially at risk [2],[5]. It accounts for nearly 5% of all squamous cell carcinomas in the oral cavity. According to Ackerman the tumor can be found on the buccal mucosa in 60% of cases, in 30% on the alveolar process and gingiva, and in 10% on the mouth floor [1]. Other known sites of occurrence are the upper aerodigestive tract (verrucous carcinoma), on cutaneous surface, genitalia (condyloma acuminatum) and on the extremities (carcinoma cuniculatum) [2]. The tumor is predominantly seen in men (women:men – 1:3) over fifth and sixth decade.

The etiopathogenesis of VC in the oral cavity remains unclear. However it is thought to be developed from a benign precursors, or to be associated with poor oral hygiene, tobacco use (inhaled along with smokeless tobacco), alcohol abuse, and some studies report the role of human papilloma virus in VC appearance and growth [[8],[9],[17]].

Macroscopically Ackerman's tumor appears to be an exophytic grey to red lesion with papillomatous, rough surface. The histologic appearance of VC is of well-differentiated hyperplastic squamous epithelium. The finger-like projections are covered by ortho- and/or parakeratinized surface layer [1]. Furthermore, regional lymph node metastasis is rare and distant metastases have not been observed. Locally destructive margins, described as "pushing border" at interface with the connective tissue could be seen, as well as a thick zone of non-keratinizing cells without mitotic activity (lacking the S-phase cells) [10]. VC is described as a benign variant of squamous cell carcinoma [4], however it can grow very large in size and is

usually associated with strong inflammatory reaction in the underlying stroma.

To date, a limited number of studies have been conducted focusing on the clinicopathologic features, patterns of treatment and outcome of the head and neck VC. The purpose of this article is to present a rare case of VC in the oral cavity; to discuss the clinical presentation, differential diagnosis and treatment in the context of current scientific literature.

2. Case Report

A case of 78-year-old woman is presented with an exophytic growth on the tongue, painful on swallowing, for a period of 3 months. An exophytic formation was found during the clinical examination on the right side of the tongue, with rough, papillary surface; red color with greyish white regions, well-defined margins and no infiltrative induration in the adjacent tissues (fig. 1). The growth was 2x1 cm in its greatest dimensions, T1N0M0 stage (no regional or distant metastasis were found on the clinical examination or CT scan), dense with mild pain on palpation. There is no history of trauma or bleeding.

The patient underwent macroexcision taking 5-8 mm oncological margin for histological verification of the diagnosis. The pathohistological examination revealed the features of VC (fig. 2 A, B). The first surgical operation was performed only for diagnostic purposes and it was not the definitive one. One month later, additional surgery was performed – hemiglossectomy, with oncologically safe resection margins according to NCCN 1,10,15,20 mm. The second excision was radical in the 3D space, therapeutic and oncologically definitive. The excision wound was closed primarily. Anti-inflammatory drugs and painkillers were prescribed.



Figure 1: Clinical presentation of the VC on the right side of the tongue: exophytic growth, broad-based, papillary (cauliflower-like) surface with no superficial ulcerations

3. Discussion

VC is a rare neoplasm, predominantly found in males (males: females – 3:1) over the fifth decade [14]. It is described by Ackerman [1] as a variant of squamous cell carcinoma with unique behavior, typical clinical course and histopathologic findings. VC is a large lesion, slow growing, rough, exophytic with pebbly mamillated surface and usually locally aggressive with low incidence of metastasis. It often represents a unique challenge in both the establishment of an initial diagnosis and the determination of an appropriate treatment plan. However, squamous cell carcinoma is considered much more aggressive when compared to VC [13].

In the present case, a lesion on the right side of the tongue is described, with typical clinical presentation of carcinoma. It is reported in the literature that squamous cell carcinoma of the tongue is usually located in the marginal area, followed by the ventral surface [12].

VC, as well as the squamous cell carcinoma is usually associated with poor oral hygiene, smoking, tobacco chewing, ill-fitting dentures, etc. The untreated longstanding leukoplakia and the verrucous hyperplasia are found by some investigators to be a predisposing factor in VC development [2]. The presence of leukoplakia may correlate with the high incidence of local tumor recurrence, because of the susceptibility for multi-centricity and so called field cancerization. Long term follow up is recommended in these patients. In our case no history of smoking, neither longstanding leukoplakia were found.

VC is described as painless growth. In the present study, oral discomfort and mild pain on swallowing was noted in the anamnesis. These symptoms have not been reported in the literature as regular standing signs when describing the clinical presentation of oral VC. No palpable neck nodes were found in our case. VC is often associated with significant stromal reaction and most enlarged lymph nodes are reported to be reactive to secondary infection or inflammation rather than metastasis [3],[17]. However, we find neck dissection reasonable in case of uncertainty

regarding clinically suspicious lymphadenopathy and the histopathologic findings.

The histologic features in the present clinical case demonstrate a lack of low-differentiated tumor cells and no signs of local infiltration or aggressive tumor behavior (fig. 2 A, B). Elephant foot-like rete ridges can be seen, which are consistent with VC but not with squamous cell carcinoma. There are no islands of dysplastic epithelium in the underlying connective tissue, usually seen in squamous cell carcinoma.

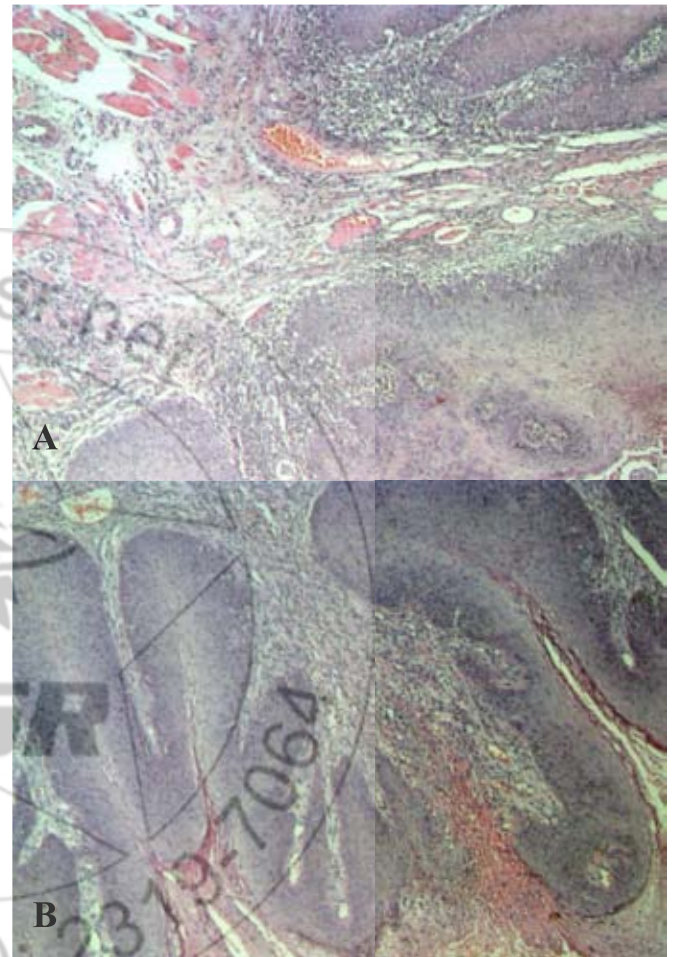


Figure 2 A, B: Histopathologic findings of VC: well-differentiated uniform cells with minimal atypia; papillary surface, with superficial epithelial keratinization; "pushing borders" at the tumor front with lymphoplasmacytic infiltration in the underlying connective tissue, associated with strong inflammatory reaction.

VC is usually classified as a "benign lesion". However, the tumor may be locally destructive and may be able to invade underlying structures. Distinguishing VC from other benign and malignant formations could be difficult in some cases. There are tumors described in the literature having all features of VC with small foci of squamous cell carcinoma, named by some authors as "hybrid" variants [7]. The coexistence of VC with squamous cell carcinoma is expected to be associated with increased risk of metastasis.

The problems in diagnosing VC are broadly discussed. The initial biopsies, especially small superficial biopsies, often are interpreted as benign hyperplasias or as hyperkeratotic,

acanthotic squamous proliferations [6]. Misinterpretation may be associated with the failure of the surgeon to submit an adequate tissue specimen for biopsy, followed by inappropriate histopathologic findings. Therefore, in our case a marcoexcision was performed initially to allow correct diagnosis establishment and determination of an adequate treatment plan. An excellent collaboration between the clinician and the pathologist is needed.

Treatment of VC requires special attention. When taking into account the biologic behavior, the high recurrence risk and considering the possibility of hybrid tumor formation, probably the most accurate choice of treatment is wide field surgical excision. More conservative surgery in some cases may compromise the outcome. If contiguous structures (i.e. bone, cartilage) are involved marginal/bone resection and decortication, or even more extended surgery may be considered. According to Medina [7] and Tharp [16], surgical treatment as a single therapeutic method is successful in 82-85% of cases for primary tumor removing, and in 94% for recurrence tumor removing. The effect of radiation therapy in the VC management remains controversial. Some investigators deny the role of irradiation in VC treatment, while others support the combination of radiotherapy and surgical treatment for better clinical outcomes. Although, there are reports in the literature revealing increased recurrence and anaplastic transformation of VC cells after radiotherapy [4],[10]. The effect of the chemotherapy is still uncertain and there is a lack of sufficient data in the literature. Tanaka [15] reported the better outcome in advanced VC of the tongue, when chemotherapy is applied preoperatively. In our case, the tumor was thoroughly removed with adequate clear surgical margins to prevent persistence of tumor cell clusters capable of malignant transformation. Chemo- or radiotherapy was completely excluded of our treatment plan.

The presence of squamous cell carcinoma components in VC is related to poor prognosis and high malignant potential. In all cases where VC pathological diagnosis is established a meticulous investigation of the whole tumor mass is mandatory to ascertain a squamous cell carcinoma – hybrid tumor coexistence [14]. When coexistence is found, squamous cell carcinoma foci are usually found in the deep areas of invasive front, which determines the need of an appropriate biopsy taking. We assume the hybrid tumors treatment should be performed according the principles of squamous cell carcinoma surgical treatment.

4. Conclusion

There is limited number of clinical cases in the current scientific literature presenting the biology, histopathologic features, treatment and prognosis of VC in the oral cavity. Although, it is a rare tumor with good prognosis, VC requires careful examination. The neoplasm must be distinguished from the squamous cell carcinoma and so called “hybrid” tumors. Currently the most common and recommended treatment is surgical excision. The effect of radiation therapy remains controversial. The postsurgical period requires long term follow up to prevent tumor regrowth.

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