Verrucous Carcinoma Involving the Lower Lip: A Case Report

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Abstract: Oral Verrucous Carcinoma is a premalignant lesion which has a potential to transform into oral squamous cell carcinoma. It is also called as Ackerman’s tumor, Buschke Loewenstein tumor, florid oral papillomatosis, epithelioma cuniculatum, and carcinoma cuniculatum. The commonest site is oral cavity followed by pharynx. It is difficult to differentiate the lesion from verrucous leukoplakia and verrucous hyperplasia. In this case report we are reporting such a case of verrucous carcinoma which is involving the lower lip.

Keywords: Ackermann’s Tumor, Oral Verrucous Carcinoma, Verrucous Hyperplasia

INTRODUCTION
Oral Verrucous Carcinoma (OVC) is a distinctive clinicopathologic entity. It is characterized by an exophytic, wart-like macroscopic appearance. The most commonly affected sites are buccal mucosa, gingiva and alveolar ridge. Its histological appearance is relatively benign and does not seem to change by time and surgical attempts [1]. Lymph node and distant metastases are rare during any stage of this tumor. Hence, the lesion's biologic activity places it between conventional (non-verrucous) carcinomas and non-autochthonous hyperplasias of squamous epithelium [2] Whenever possible, surgical removal is recommended over radiotherapy [3].

CASE REPORT
A 40 years old male patient reported to the department with a chief complaint of a growth on his lower lip since 2 months. He stated that the growth was insidious in onset, small in size to begin with and has gradually progressed to the present size over 2 months duration. He did not give any positive history of pain or bleeding from the lesion. He reported that he has habit of chewing betel nut since 20 years and keeps the quid on the right labial mucosa and vestibule for approx. 30 min and then spits out. He also has habit of chewing pan since 20 years and drinking alcohol since 10 years. On general physical examination, he was moderately built and well oriented to time, place and person. On extra-oral examination, bilateral submandibular lymph nodes were palpable. They were solitary, oval in shape, measuring approx. 1cm in diameter, firm and mobile. There was hypoesthesia over right side of lower lip. Encrustation was seen on the right side of lower lip at the vermilion border (figure 1). On intra-oral examination, solitary, well-demarcated, roughly oval shaped growth measuring approx. 5cm in diameter was located on right side of lower lip vermilion border and labial mucosa (figure 2,3). It was extending from vermilion border till 5cm over the labial mucosa and mesio-distally from level of 31 till 33. The margins were raised and well-defined. The surface was white and pink with papillary projections. On palpation, all the inspectory findings were confirmed. It was firm in consistency and the margins were indurated. It was nontender. There was hypoesthesia over the lesion and over the surrounding mucosa. There was also generalized blanching of the oral mucosa. Palpable vertical bands were present over the buccal mucosa bilaterally, soft palate and faucial pillars. The uvula was shrunken.
On the basis of history and clinical examination, a provisional diagnosis of ulceroproliferative lesion on the lower lip vermilion and lower labial mucosa and oral submucous fibrosis was given. The differentials were listed as proliferative verrucous leukoplakia and verrucous carcinoma. He was then referred to the department of oral surgery for the surgical excision of the lesion (figure 4).
The histopath picture revealed mucosa adjacent to lesion composed of well differentiated squamous epithelium with hyperkeratotic exophytic surface papillae and broad, blunt, downward pushing rete ridges. There is minimal cytologic atypia and minimal lamina propria with few adherent skeletal muscle bundles and adipose tissue. Features were consistent with verrucous carcinoma (figure 5).

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and later versions. Informed consent was obtained from the patient for being included in this report.

DISCUSSION

Oral Verrucous Carcinoma (OVC) is a rare variant of oral squamous cell carcinoma (OSCC), first described by Ackermann, and henceforth also known as Ackermann's tumor [4]. The classic clinical, topographic, and histologic features of verrucous carcinoma have been well documented and corroborated by numerous workers since Ackerman's original description of the entity in 1948. Shear and Pindborg provided additional clarification of this lesion and further concluded that benign verrucous hyperplasia may represent an early stage in the continuum of eventual verrucous carcinoma development [5, 6].

It is responsible for a proportion of oral SCCs varying from 4.5-to-9.5%. Use of tobacco is considered the most common etiological factor for OVC. Shear and Pindborg reported that out of 28 patients with verrucous lesions, 24 (86%) used tobacco, and one was an arecanut quid chewer. Along with it, viral etiology; mostly human papilloma virus (HPV), is also sometimes suspected [6]. OHPV is responsible for roughly 28% of cases, usually types 16 and 18, but also
2 and 11. Viral carcinogenesis is most likely caused by the suppression or mutation of gene p53, which is responsible for cellular tumor suppression activity. Even constant irritation to the mucosa is also an etiological factor.

This lesion is generally seen in elderly patients with age range of 60-70 years. Males are predominantly affected than females. In majority of cases the carcinoma affects the buccal mucosa, gingiva or alveolar ridge followed by palate, lip and floor of mouth occasionally. Even in our case the lower lip and labial mucosa was involved. The neoplasm is chiefly exophytic and has papillary projections sometimes covered by white leukoplakic film. It has a well-demarcated margin and a warty surface. They form rugae like folds with deep clefts in them. In mandible the lesion can get fixed to the periosteum and destroy the underlying bone. Vidyasagar and colleagues reported that mandibular bone involvement was associated with 19.6% of VCs [7]. Pain and difficulty in mastication is commonly associated with lesion but bleeding is rare.

The biologic activity of VC distinctly differs from that of conventional carcinomas. Regional lymph node involvement is uncommon in VC, while adjacent structures are often involved with time and growth of the primary tumor [3]. Verrucous carcinoma and verrucous hyperplasia are difficult to be differentiated clinically. Verrucous hyperplasia (VH) is considered to be precursor of verrucous carcinoma.

The classical histopathological features of OVC are an intact basement membrane, with preservation of stratification and broad rete pegs of carcinoma cells, which appear to punch into the underlying tissue, often called as “elephant feet” [8]. The parakeratin plugging also occurs extend ing into the epithelium. The parakeratin lining the clefts with parakeratin plugging is the hallmark of verrucous carcinoma. When lesions get infected, focal intraepithelial abscesses are often seen [9]. But unfortunately diagnosis of OVC is difficult even with generous biopsy specimen. Often the diagnosis of verrucous carcinoma is established only after multiple biopsies [10]. Only differentiating feature between verrucous carcinoma and hyperplasia is that verrucous hyperplasia is superficial while verrucous carcinoma is deeply embedded.

The differential diagnosis (DD) of OVC is one of the most challenging issues among oral diagnosticians and pathologists. Verrucous hyperplasia, proliferative verrucous leukoplakia (PVL) etc can be given as clinical DD. PVL is defined as proliferative, irregular white patch or plaques that progress slowly and multifocally. Their chance of malignant transformation is more compared to OVC also they show dysplastic changes histologically. Clinico-histopathological similarities between the wide spectrum of verruciform lesions like VH to noninvasive OVC and invasive well-differentiated OSCC make the diagnosis difficult. Verrucous hyperplasia and VC are similar both clinically and histopathologically [11, 12].

It is most commonly managed by surgical mode of treatment. Radiotherapy alone have not shown significant effect in treatment of OVC and also caused anaesthetic transformation of the neoplasm [13]. According to Koch et al., all patients suffering from OVC should be surgically treated first [14, 15]. Even in our case we have selected the surgical mode of treatment and it has been successful also. Other treatment modalities like cytostatic drugs may be preferred when surgery is not an option. Various dosages of cytostatic drugs have been proven to show beneficial effects in reducing tumor size; α-interferon (IFN) seems to support the therapy by delaying the growth of the tumor but does not take the place of surgery alone [16]. The prognosis of verrucous carcinoma is better than that of other kinds of life-threatening malignant tumors. According to study conducted by Candau-Alvarez A et al, the recurrence in 2-year follow-up has been 7.69% and in 5-year follow-up has been a skewed 0–66.7%. The survival rate in verrucous carcinoma is excellent at 93.65% [17].

CONCLUSION
Oral verrucous carcinoma (OVC) is a neoplasm that is less aggressive variant of SCC. It affects elderly individuals more commonly. The clinical differentiation of VC is difficult as it resembles verrucous hyperplasia (VH), verrucous leukoplakia and therefore histological examination is required for this. Multiple biopsies are to be carried out to confirm the diagnosis.

REFERENCES

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