

Case report**Verrucous Carcinoma: A Case Report and Review of Literature**

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Abstract: Ackerman first recognized verrucous carcinoma (VC) as a distinct entity in 1948. Although it occurs in other anatomic sites, most intraoral cases involve buccal mucosa, alveolar mucosa and gingival. Verrucous carcinoma usually presents as a diffuse, largely exophytic, superficial spreading, highly keratinized, warty form of well differentiated squamous cell carcinoma that is unlikely to metastasize. Here we present a case of an old man with a chief complaint of an intraoral exophytic growth, which on clinical, radiologic and histopathological examination revealed to be a oral verrucous carcinoma. This article also enlightens on the review of literature in detail on verrucous carcinoma.

Key words: Verrucous; Carcinoma; Papillary; Exophytic; Ackerman tumor; Papillomatosis.

Introduction:

Also known as Snuff dipper's cancer. It is a form of epidermoid carcinoma of the oral cavity which was defined as an entity by Ackerman in 1948.¹ It is a relatively rare tumor constituting 4.5 to 9% of oral SCC as SCC is 25 times more likely to develop in spit tobacco users than this low grade variant. The only epidemiologic assessment of this tumor in a western culture reported an annual incidence rate of one oral lesion per 1 million populations each year.

It has a white, warty appearance forming a well circumscribed mass raised above the level of the surrounding mucosa. Based on site of occurrence they are classified into oral, anourologic and palmoplantar type. This article presents a case report on verrucous carcinoma and also highlights in brief the review of literature.

Case report:

A 42 year old male patient reported to the department of oral medicine & radiology at Rama Dental College Hospital and Research Centre Kanpur with chief complaint of growth in left side of his mouth since 6 months & swelling on left lower face since 7 days (Fig. 1). Patient gave history of exophytic growth on left buccal mucosa since 6 months. Initially the growth was small in size and gradually increased to attain the present size. The growth was associated with pain, pain was sharp,

intermittent, non-radiating aggravates on chewing, eating along with the burning sensation. Patient also gave the history of extraoral swelling on left lower face since 7 days.

The swelling was gradually increasing to attain the present size. The patient was asthmatic since 10 years and under the treatment of Deriphyllin. Patient gave history of chewing tobacco in form of Khaini 5 to 6 pouches per day along with Bidi smoking (4 to 5 bundles per day). On examination patient was moderately built, nourished and his vital signs were within normal limits.

Extraoral examination reveals Facial asymmetry was present due to swelling over the parasymphysis & body of the mandible on left side measuring about 4x 3cm which was present since 7 days (Fig 1). The swelling was oval in shape & extending from the parasymphysis region posteriorly just anterior to masseter region & superiorly from occlusal line to the lower border of mandible (Fig. 1).



Figure 1: Extraoral examination: Reveals swelling on left lower half of face

The swelling was sessile & endophytic & overlying skin was red in color with increased temperature. The swelling was firm in consistency & tender on palpation. Bilateral submandibular lymphnodes were palpable, firm in consistency, mobile & non-tender.

Intraoral examination reveals Patients periodontal condition was poor with generalized periodontal pockets & gingival recession. Papillary & verruciform surface projections are present over the left buccal mucosa (4 in number) & attached gingiva in relation to 47-48 (2 in number). Initially 2 papillary projections were present but since 2 months there is rapid increase in size & number. There is restricted mouth opening due to the lesions since 5 months (Fig. 2).



Figure 2: Intraoral examination : reveals Papillary & verruciform surface projections present over the left buccal mucosa & attached gingiva in relation to 47-48.

There are 6 separate papillary projections are present, 4 over the left buccal mucosa along the occlusal line & 2 over the attached gingiva irt 47-48. The borders of the projections are regular & definable. The projections were sessile & exophytic which are tender on palpation & firm in consistency. There is also pus discharge from gingiva in relation to 46 & 47.

Based on the history & clinical examination patient was provisionally diagnosed as a case of verrucous carcinoma irt left buccal mucosa & attached gingiva and Chronic periodontal abscess irt 36.

All the blood investigations were under normal physiological limits except ESR which was raised to 22 in Wintrobe's method. The radiographs revealed a diffuse radiolucency irt 36-37 with bone resorption (Fig. 3, 4 & 5).



Figure 3: Intraoral periapical radiograph: depicts diffuse radiolucency irt 36-37 with bone resorption

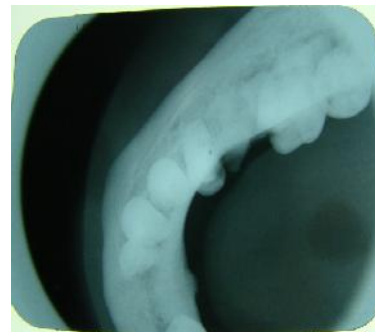


Figure 4: Occlusal radiograph: reveals bone loss in region of 36-37



Figure 5: Orthopantomograph reveals diffuse radiolucency irt 36-37 with bone resorption

Incisional biopsy was performed and report revealed the presence of stratified squamous epithelium & supporting connective tissue stroma. The elongated bulbous rete pegs are invading deep into the connective tissue stroma. Parakeratin plugging is seen at places with basilar hyperplasia showing numerous mitotic figures. Connective tissue stroma is fibro-cellular with dense chronic inflammatory cells.

Based on the histopathology report patient was finally diagnosed as a case of verrucous carcinoma of left buccal mucosa & gingiva.

Discussion:

Verrucous carcinoma can be defined as a diffuse, largely exophytic, superficial spreading, highly keratinized, warty form of well differentiated squamous cell carcinoma that is unlikely to metastasize². It has a characteristic white, warty appearance forming a well circumscribed mass raised above the level of the surrounding mucosa. VC occurs in most of the cases arise from the oral mucosa in people who chronically uses chewing tobacco or snuff. Extra oral sites include laryngeal, vaginal & rectal mucosa & skin from the breast, axilla, ear canal & soles of the feet.³

Males show more prevalence as compared to females. Approximately 5 % of all intra oral SCC. Buccal mucosa accounts for more than half of the cases & the gingiva is the location for nearly one third of the cases. Patients usually affected are > 50 yrs. Lesions are typically white but may appear erythematous or pink. The colour depends on the amount of keratin production & the degree of host inflammatory response, leukoplakia or tobacco pouch keratosis may be seen in adjacent mucosal surfaces.⁴

HPV infection is thought to facilitate or cause VC. HPV 16 has been identified frequently in genital and peri-ungual VC. Identification of intra tumor HPV DNA adds support for a role of this virus in tumor development.⁵ Chronic inflammation may lead to the development of VC. Associations with VC have been found in patients who chewed tobacco and betel nuts and dipped snuffs.

Based on site of occurrence are as follows:

Oral Verrucous Carcinoma

(Ackerman tumor, oral florid papillomatosis) Early lesions appear as white, translucent patches on an erythematous base. Ulceration, fistulation, and invasion locally into soft tissues and bone (eg, mandible) may occur. Common locations include the buccal mucosa, alveolar ridge, upper and lower gingiva, floor of mouth, tongue, tonsil, and vermilion border of the lip. Painful nonmalignant lymphadenopathy can be seen with concurrent infection.⁶

Anourologic type (Buschke-Loewenstein tumor) This type most commonly occurs on the glans penis, mainly in uncircumcised men.

Palmoplantar verrucous carcinoma

(epitheliomacuniculatum) These tumors most commonly involve the skin overlying the first metatarsal head, but they also occur on the toes, heel, medioplantar region, dorsum, and amputated stumps. Exophytic tumors with ulceration and sinuses draining foul-smelling discharge cause pain, bleeding, and difficulty walking.

VCS of other sites (eg, trunk, extremities, scalp, face) have been reported.⁶

Grinspan has divided verrucous carcinoma based on histopathologically into four types
Type I A: Acanthosis, papillomatosis, leukoedema, moderate ortho or parakeratosis, hypertrophic interpapillary crests and stratification of the basal layer.

Type I B: cryptic depression of the epithelial surface, invagination of the epithelium and fistulous tendency.

Type II: Areas with the characteristics of type IA or IB and areas with hyperchromatic nucleus and atypical mitosis.

Type III: Areas of type I or type II and features of squamous cell carcinoma. Anaplastic cells and metastasis are frequently observed in this type.⁷

Staging:

Most VCs are non-metastatic and are staged based on size, as follows.⁶

1. T0 lesions - In situ
2. T1 lesions - Less than 2 cm in diameter
3. T2 lesions - Between 2 and 4 cm in diameter
4. T3 lesions - Greater than 4 cm in diameter
5. T4 lesions - Invasive of muscle or bone

The treatment includes complete tumor extirpation at first presentation. Because VC is a lesion that can recur, metastasize, and ultimately cause death. Recurrent VC carries a relatively poor prognosis.⁸

Cryosurgery using liquid nitrogen is a safe and low-cost procedure for the ablation of selected verrucous carcinoma and is well tolerated by patients. Cryotherapy has provided a high cure rate for select well-circumscribed superficial VC. Because of no histologic control, close follow-up is necessary. This procedure is the least likely to result in cure and is the least preferred intervention.

Curettage and Electrodesiccation:

Cure rates of 96-99% have been quoted in several large studies for destruction of T0 and T1 VC (ie, in situ lesions and invasive lesions <2 cm in diameter). This high cure rate was affected by careful patient selection. The main disadvantage of curettage and electrodesiccation is a lack of margin control; nonetheless, the procedure is

minimally invasive, well tolerated, and effective for in situ lesions without deep involvement.⁸

Excision with conventional margins:

Simple excision is most valuable in the treatment of small VCs of the trunk and extremities and in areas in which tissue sparing is not essential. Cure rates following simple excision of well-defined T1 lesions may be as high as 95-99%. A 4mm margin of normal tissue is recommended for straightforward lesions. Standard excision with permanent conventional sections is a highly effective treatment for many VCs.

The depth of the excision should include the subcutaneous fat because even small VCs may extend into the subcutaneous fat. The disadvantages of excision with an arbitrary margin are that, in some cases, the pathology reveals a subclinical positive margin, requiring further surgery. Additionally, more normal tissue may be excised than is necessary.⁹

Mohs-surgery:

A dermatologic surgeon usually offers Mohs micrographic surgery (MMS). The main advantage of MMS over simple excision in the extirpation of cutaneous VC is the ability to examine all excision margins (deep and lateral) and to carefully map residual foci of invasive carcinoma. MMS provides a cure rate for VC of 94-100% and has been of particular value in curing VC with perineural invasion. MMS offers the added benefit of preserving normal tissue, thus facilitating reconstruction. MMS is performed routinely in an outpatient setting under local anesthesia.

A multidisciplinary approach employing MMS performed in conjunction with plastic surgery, otolaryngology, and radiation oncology may allow for the complete removal of deeply invasive VC, preserve vital structures, and facilitate the reconstruction of a large operative defect. Because of its many advantages, MMS is the procedure of choice for VC where tissue preservation is needed. Furthermore, surgery for VC

using MMS may be an integral component in the management of certain VCs that otherwise would be beyond the experience of the cutaneous surgeon.¹⁰

Radiation therapy: Radiation therapy offers the potential advantage of avoiding the trauma and deformity of a surgical procedure. Radiation therapy for patients with VC commonly is administered 5-12 times over a 5- to 6-week period with fractionated doses of 400-800 cGy.

Cure rates for T1 lesions range from 85-95%. Although the initial cosmetic result following radiation often is good, the long-term result frequently is poor, with atrophy, hypopigmentation, and telangiectasia. Some patients treated with radiation also develop radiation necrosis. This risk increases over time. Radiation therapy is not advocated for use over bony structures because of the risk of osteoradionecrosis.

Radiation therapy is not advocated for patients who are young or middle-aged because of the small, but real, risk of a radiation-induced cutaneous carcinoma or sarcoma later in life.¹⁰

Most patients with VC have a good prognosis. Local recurrence following definitive treatment is not uncommon, but distant metastasis is rare. Patients with oral VC may be at an increased risk of a second primary oral squamous cell carcinoma, which carries a poor prognosis.

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