Aggressive verrucous carcinoma in hard palate: A rare case report

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Abstract
Verrucous Carcinoma is a rare, slow growing, well-differentiated squamous cell Carcinoma. In this case, we report the situation of aggressive, verrucous carcinoma in the hard palate beyond the midline and extends to the maxillary sinus.

Keywords: Verrucous carcinoma, proliferative projection, squamous cell carcinoma and maxillectomy

Introduction
Ackerman first described Oral Verrucous Carcinoma in 1948 [1]; OVC constitutes 2-12% of all carcinomas in the oral cavity, and buccal mucosa is the most region to occur V.C. following the hard palate, the floor of the mouth and gingiva [2]. Chewing tobacco, smoking, infection with papillomavirus PHV, and chronic irritation are V.C.s’ potential causes [3]. This tumor is more frequently in men, with a male: female ratio of 5/1 [4]. It clinically appears as a white-grey, warty growth, exophytic mass which looks like cauliflower [5]. Histologically appearance is as papillary surface epithelium, intense Parakeratinized elephant feet grow inward, pushing underlying connective tissue with intact basement membrane and slight cellular atypia [6]. Verrucous Carcinoma is described as a benign lesion with minimum aggression. It does not give regional and distant metastases, but in some cases, it becomes aggressive and penetrations skin, fascia, cartilage and even bone [1, 7].

Case report
A 55-year-old woman patient consulted our Department of Oral and Maxillofacial surgery with a chief complaint of a large mass in the hard palate. Seven months ago, patient suffered from a minor, painless mass; she reported to the dentist, who diagnosed it as an ulcer and gave her a course of antibiotics. The mass gradually grew until it reached this size. Intra-oral examination shows painless, whitish, no bleeding, exophytic mass which looks like cauliflower. Histologically appearance is as papillary surface epithelium, intense Parakeratinized elephant feet grow inward, pushing underlying connective tissue with intact basement membrane and slight cellular atypia [6]. Verrucous Carcinoma is described as a benign lesion with minimum aggression. It does not give regional and distant metastases, but in some cases, it becomes aggressive and penetrations skin, fascia, cartilage and even bone [1, 7].

Oral health was poor, and tooth examination shows mobility in premolar, canine and molars. The lesion protrudes inside the mouth and puts pressure on the tongue, as shown in Figure (1).

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Fig 1: Clinical appearance of the tumor shows papillary exophytic growth
The patient has no familial history of oral neoplasms; she was not a smoker or alcoholic. Her history is good; no disease exists. Extra oral examination shows asymmetry of the face and swelling on the right cheek was clear, clinical examination of Lymph nodes was natural, and no enlargement was seen. Computer tomography C.T scan examination appears a destructive lesion bone, extending to the maxillary sinus. The inferior orbital wall was intact, as shown in the figure (2).

Magnetic Resonance Imaging (MRI) was preformed after injection contrast material, it shows mass destructing the right upper jaw and extends to the maxillary sinus, nasal cavity, touch the lower horn, and there is no enlargement of lymph nodes in the neck as in Figure (3).

The clinical and radiological findings had suggested Oral Verrucous Carcinoma; the clinical differential diagnosis was Verrucous Carcinoma, squamous cell Carcinoma, and Verrucous hyperplasia. Under local anesthesia, a biopsy was taken. It shows under microscopy acanthosis and hyperkeratosis of the covering epidermis invasion into the deep dermis, forming irregular islands with slight cellular atypia Figures (4, 5, 6) show the biopsy's microscope photo.
All findings and histological examination confirmed Verrucous Carcinoma. The treatment plan was surgical excision of the tumor under general anesthesia, patient in a supine position, careful aseptic condition, and mouth intubation. Weber-Ferguson approaches and partial maxillectomy were made from the left lateral incisor, the tumor was resected with safety edges 1 C.M, and the palatine tonsil was removed, as shown in Figure (7). However, lymph node curettage was not indicated in this case.

Reconstruction of the defect was made with an obturator fabricated by a 3D printer using PEEK materials, as shown in (8).

The maxillary defect was filled with medicated gauze. The obturator was temporarily fixed with ligated wires to the zygomatic arch and alveolar bone. The obturator stays in place for ten days until healing soft tissue, and then the gauze packing is removed. The grossly was sent to a histological examination, which confirmed the diagnosis. The resected specimen and palatine tensile margins were negative for V.C. the lesion was staged as T4N0M0. The patient did not have radiotherapy or chemotherapy after surgery. The follow-up evaluation 2 year after surgery showed the absence of recurrence or metastasis.

**Discussion**

Oral Verrucous Carcinoma is a rare tumor subtype of Squamous Cell Carcinoma (SCC); its consulates 2-12% of all SCC in the oral cavity [2]. It is low grade, well differentiated from SCC, and clinically demonstrated as a painless, warty, exophytic mass like cauliflower. It is a characteristic slow growing, locally invasion with rare regional or distant metastatic potential [8], but sometimes it can be aggressive, grow very large and destroy adjacent tissue such as bone and cartilage [7].

The oral cavity and Larynx are the most common site of V.C. in the head and neck. In oral cavity, it commonly arises from buccal mucosa following, hard palate, lip floor of the mouth and gingiva [9]. The etiology of V.C. is unclear, however, there is a correlation between the incidence of V.C. and snuff, chewing tobacco, poor oral hygiene, human papilloma virus (HPV) infection and other irritation such as betel nut chewing [10], OVS can also arise from leukoplakia, verrucous leukoplakia and erythroplakia [11]

The V.C. occurs between the fifth and the sixth decade and tends to occur in older males more than females [4]. However, other studies report equal sex distribution [12].

V.C.’s histological diagnosis is considered a challenge; it requires an experienced pathologist and an adequate biopsy [13]. The microscopic examination shows the pathological feature of OVC as a hyperplastic epithelium with intense keratinized superficial projection and Parakeratinization

Elephant foot like down growth compressing underlying connective tissue with intact basement membrane [14]. The acanthosis with broadened rete ridges causes a decrease of blood splay to distant epithelial cells, which become swelling and gradually die, which cause clefts. The parakeratin lining of the clefts with parakeratin plugging, which is believed to be characteristic of this tumor, there is infiltration of lymphocyte and plasma cells in connective tissue, with no significant degree of cellular atypia [6, 15].

Oral verrucous lesions are considered as challenge to pathologist, V.C must be differentiated from verrucous hyperplasia (VH), both have similar clinical and pathological features [16], Verrucous hyperplasia and Verrucous Carcinoma are identical in characteristic parakeratosis superficially, however, in Verrucous hyperplasia there is no invasion of the hyperplastic epithelium into connective tissue compared with Verrucous Carcinoma. While Verrucous Carcinoma epithelial overgrowth with wide and projection rete ridges shows a pushing-bordered into connective tissue, Verrucous hyperplasia is superficial and does not extend through tissue [16, 17].

Also, V.C. can be differentiated from SCC histologically, which different with clinical behavior and method of treatment [14]. The vital feature distinguishing V.C. from SCC is the lack of cytologic criteria for malignancy in squamous epithelia in V.C. and the margins of tumor pushing connective tissue rather than invasion underlying tissue as in SCC [18].

A full thickness biopsy and an excellent clinical description should permit the pathologist to distinguish Verrucous Carcinoma from verrucous hyperplasia and other forms of squamous cell carcinoma [16].

Many authors consider surgery with safety margins of 1 cm as an effective treatment for V.C. [12, 19]. However, others considered histological margins above 5mm sufficient to decrease the risk of recurrence [20].

Radiotherapy alone or in combination with surgery in the treatment of V.C. is controversial. Suresh et al. report in their study that contributes radiotherapy and surgery in the treatment of V.C. in the oral cavity have worse O.S. (overall survival) and DDS (disease-specific survival) [21].

Ferlito et al. refer in their survey study to resistant V.C. to irradiation than conventional SCC and portability plastic transformation after irradiation [9]. However, other studies recommended radiotherapy for particular clinical situations, depending on the cancer site, such as V.C. in the Oropharynx, Hypopharynx and Superagilities retromolar region, also in an
advanced stage where surgery is complicated [19, 22]. Chemotherapy for OVC treatment is arguably, except in situations where surgery or radiotherapy treatment is a contraindication because of extensive lesions, poor clinical condition [23].

Cervical Lymph node metastasis is very rare in V.C [24]. McClure et al. reported that neck dissection is unnecessary even in a large tumor because lymphadenopathy in V.C. is inflammatory rather than metastatic [25].

Walvekar et al. suggested selective neck dissection, particularly in clinical lymphadenopathy, even though all his patients who underwent neck dissection had a negative node [11]. Franklin et al., in their retrospective review study of patients, suggested avoiding routine lymphadenectomy in oral Verrucous Carcinoma [9]. V.C has a good prognosis, but there is a risk of recurrence or developing second primary cancer Long-term follow-up Studies of V.C reported a highly variable recurrence rate ranging from 0-66% at five years, related to the tumor stage, patient age and treatment method [26, 22, 27].

Conclusion

We reported a large maxillary Verrucous Carcinoma case classified as a rare group of tumors in the oral cavity. Verrucous Carcinoma has a good prognosis for early diagnosis and treatment with surgery, and there is no risk of cervical or regional metastasis. A close follow-up is necessary for the high incidence of local recurrence and possibly developing second primary cancers.

References