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Surgical management of a huge oral verrucous carcinoma: A case report and review of the literature

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Abstract

Verrucous carcinoma is a well-recognized low-grade variant of squamous cell carcinoma. Cutaneous, oral, and anogenital forms exist. Exposure to persistent chronic irritation, inflammation, and repeated injury, as well as carcinogenic agents such as human papillomavirus infection, smoking, and alcohol use, are established risk factors. These neoplasms occur mostly in the oral cavity. The usual extraoral sites include the larynx, esophagus, genitals, and perineum. It is an extremely uncommon site of occurrence for the extraoral chin region. This unusual location makes the index case unique. Other uncommon sites reported include finger and foot. Case studies of verrucous carcinoma with huge tumor sizes are rare. Although it can be destructive locally, verrucous carcinoma typically does not spread to distant sites. Wide surgical excision with free margins is the most common treatment approach with a favorable prognosis. These tumors are likely to recur if they are incompletely excised, and recurred lesions tend to be more aggressive clinically as compared to their original counterparts. Herein, the authors describe a case of a huge oral verrucous carcinoma localized on the chin of a 43-year-old female patient. The clinical course, diagnostics, and proposed treatment have been discussed with the existing available literature.

Keywords

Verrucous carcinoma, huge size, surgery, case report

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Introduction

Verrucous carcinoma (VC) is a low-grade form of squamous cell carcinoma with specific clinical, morphological, and cytokinetic features.[1] It is among the most infrequent forms of oral carcinoma.[2] VC affects more males than females, and older individuals (average 50–70 years old) are mostly affected compared to the younger age group; 75% of VC patients are above 60 years old.[3] The cause of VC is essentially unknown. However, tobacco, alcohol, and betel nut consumption are the most associated risk factors. In addition, chronic inflammation or irritation and repeated trauma, particularly in the oral cavity, have also been implicated as risk factors. Similarly, VC can arise in chronic inflammatory conditions such as lichen sclerosus, burn scars, chronic ulcers, leishmaniasis, and low-risk human papillomavirus (HPV).[4] However, the role of HPV is controversial as diagnostic thresholds vary, but if histologic criteria are rigorously applied, low-risk HPV is only rarely positive. VC can arise in association with other lesions, including syringocystadenoma papilliferum, congenital venous malformation, and cutaneous horn.[5]

VC is a well-recognized low-grade malignant tumor with three distinct variants, namely cutaneous, oral, and anogenital subtypes.[3] VC is known to be a slow-growing tumor that presents predominantly as an exophytic growth with pebbly, micronodular surfaces. Clinically, VC presents as a tan-white outgrowth with a broad base attachment, producing a cauliflower-like warty lesion that is locally aggressive but well circumscribed. Adjacent cervical lymphadenopathy may be associated, but regional and distant metastases rarely occur in this tumor, even in advanced cases.[4] Morphologically, VC displays unique features. These include epithelial exophytic

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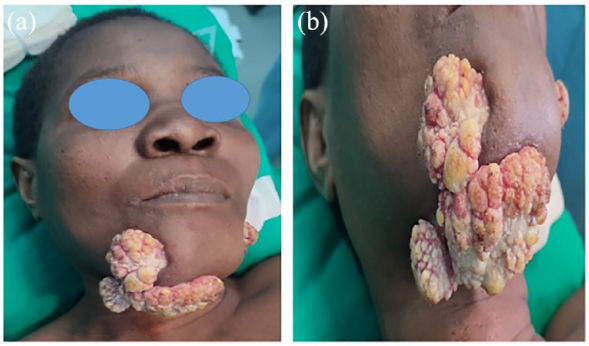

Figure 1. Photographs of the patient before surgery showing an extraoral ulcerating mass in the chin region extending to the submandibular area (a) and a cauliflower or verrucoid nature of the lesion (b).

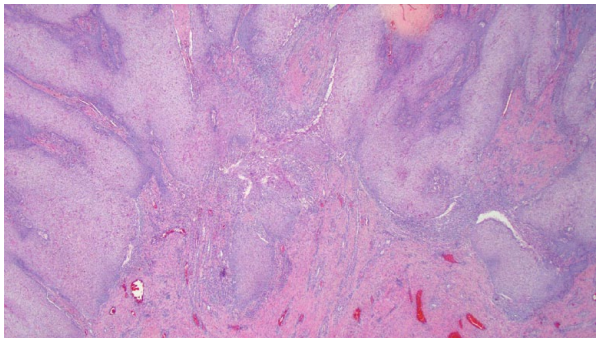

Figure 2. Photomicroscopic image of verrucous carcinoma highlighting a well-differentiated squamous proliferation with papillomatosis and minimal cellular nuclear atypia, deep bulbous process with a pushing margin, hematoxylin and eosin (H&E) staining 20× original magnification.

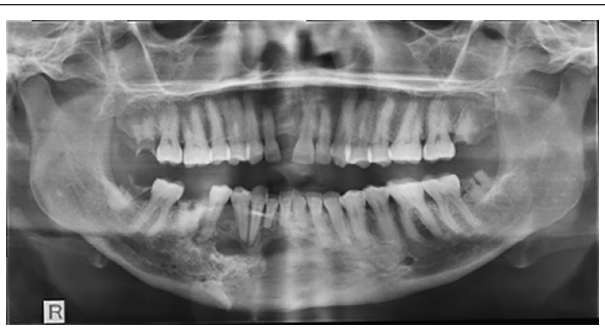

Figure 3. An orthopantomogram showing normal findings.

projections forming high ridges with keratin-filled invaginations and down-growth as blunt papillas that seem to push instead of infiltrating the underlying connective tissue. The tumor cells have minimal or no cytological atypia.[1,2] A complete surgical excision is the main treatment approach. The survival rate of patients with VC is very good, with low rates of recurrence. Herein, we report a case of VC on the chin in a 43-year-old female patient with delayed presentation who was successfully treated. A brief review of the literature is also presented.

Case presentation

A 43-year-old female reported to the oral and maxillofacial surgery clinic of our institution in January 2023 because of a painless, slow-growing ulcerated mass on her chin for 3 years. It started as a small nodule on the chin, progressively increasing in size over time. Later, the nodule ruptured to form an open ulcer. No relieving or aggravating factors were reported, and the lesion was associated with mild discomfort during eating. The patient denied a history of trauma. She was an alcohol consumer but a nonsmoker with a healthy general condition. On examination, there was no facial asymmetry, and lymph nodes were not enlarged. Speaking and swallowing remained undisturbed. Intra-orally, the presence of an ulcer on the lower border of the mandible in the region of the chin and submandibular area with an uneven surface was noted. Extraorally, the lesion was fungating, whitish-pink with a cauliflower shape, measuring 8 × 6 cm on its greatest dimension. The lesion was locally destructive, predominantly localized extraorally in the submandibular region (Figures 1(a) and (b)). A clinical diagnosis of oral VC was entertained, with differential diagnoses of giant condyloma, warts, pseudoepitheliomatous epidermal hyperplasia, keratoacanthoma, and squamous cell carcinoma.[6,7]

Histopathology of an incisional biopsy demonstrated a verrucous lesion consisting of well-differentiated squamous cell proliferation with exophytic and endophytic components. The tumor displayed blunt projections with bulbous processes and pushing margins. Individual cells had minimal nuclear atypia (Figure 2). The findings were in keeping with oral VC, with differential diagnoses of kerathoacanthoma and well-differentiated squamous cell carcinoma. An orthopantomogram (OPG) (Figure 3) and chest x-ray were normal. The patient was counseled for wide-ranging local excisional surgery. The procedure was scheduled after obtaining her authorization.

Surgical procedure

The surgical procedure was performed under general anesthesia. Under aseptic conditions, a wide local tumor excision was performed (Figure 4(a)). The tumor removal was approached extraorally with at least 1 cm of free surgical margins. The surgical wound was primarily closed with the advancement of nearby muscles and skin in layers in a watertight fashion (Figure 4(b)). The patient's postoperative course was uneventful. The excised lesion was sent for histopathological examination. The final diagnosis report was

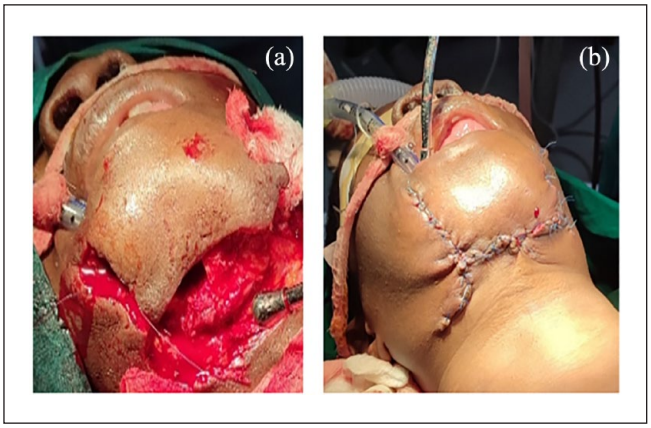

Figure 4. Presentation of patient immediately after the tumor excision (a) and after surgical reconstruction of soft tissue (b).

similar to the previous incisional biopsy, with free surgical margins. To date, the patient remains symptom-free. No recurrence has been observed after a follow-up of almost 12 months (Figure 5(a)–(d)).

Discussion

VC is a variant of squamous cell carcinoma with specific clinico-morphological and cytokinetic features.[1] The etiology of VC is uncertain. However, the tumor usually affects elderly males with adverse habits of tobacco and alcohol. In addition, the etiology of this neoplasm has been linked to oncogenic HPV infection of the mucosa and genitals. But this may represent incidental colonization.[2,3] Younger individuals, as was mirrored in our patient, can also be affected. Our patient is a middle-aged female with a history of alcohol use. VC is a low-grade tumor with a good prognosis when affecting the skin.

These neoplasms primarily affect the oral cavity. Common extraoral locations include the oropharynx, anogenital, esophagus, and perineum. It is a very uncommon occurrence in the extraoral chin region. The index case stands out due to its odd location. Other extremely unusual locations reported include the foot, finger, and kidney.[6] It may also arise in association with inflammatory and neoplastic conditions. The high chances of its occurrence are associated with proliferative lesions that develop from a benign precursor. The clinical behavior of VC can be destructive, apart from its deceptively benign microscopic occurrence. The most commonly affected sites are the buccal mucosa, gingiva, and alveolar ridge,[4] but in our patient, the lesion was located on the skin around the lower jaw. As evidenced in our patient, these lesions may grow very large and can extensively destroy adjacent tissues, including cartilage and bone.[2] In our case, regardless of the huge size of the lesion, there was no bone involvement. The tumor grows slowly and locally, invasive with fewer chances to metastasize. It appears as a painless, thick white plaque resembling a cauliflower.[5,6] This is similar to our case;

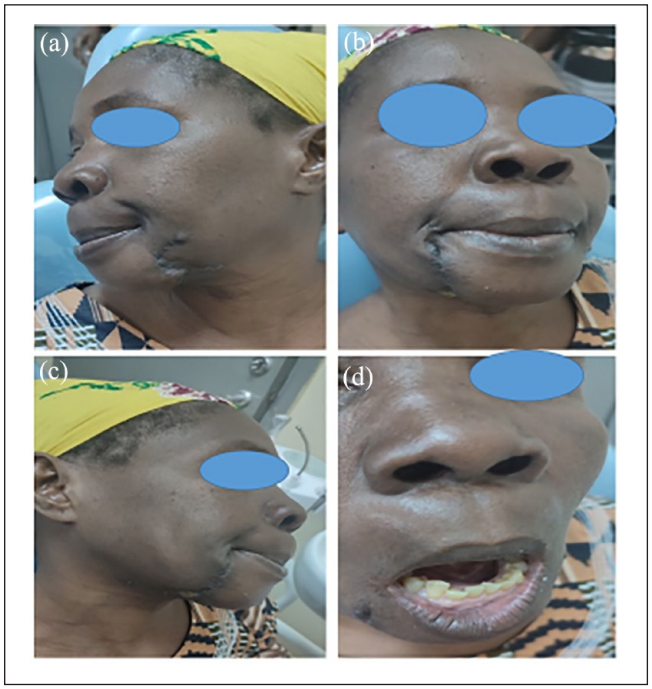

Figure 5. Photographs of the patient 12 months after surgery, demonstrating a satisfactory functional (a) and cosmetic appearance (b); the patient remains symptom-free (c), with no local recurrence (d).

regardless of the long duration and size of the lesion, there was no distant metastasis.

Diagnosis of VC involves both examination of the lesion in the patient and vigilant histologic study. As observed in the index case, a suitable biopsy must show not only a well-differentiated hyperkeratotic warty superficial surface but also an equally well-differentiated bulbous rete-ridge pattern at the bottom of the lesion.[7,8] The differential diagnoses include giant condyloma (Buschke-Löwenstein tumor), verrucae (warts), pseudoepitheliomatous epidermal hyperplasia, keroacanthoma, and squamous cell carcinoma.[8] The diagnosis can be delayed due to bland histopathological features and mimicked reactive processes. Caution should be exercised when making a diagnosis in superficial or fragmented biopsies. As it was mirrored in our patient, VC tumors display hyperkeratotic warty growths that are longstanding, progressive, and may or may not be locally destructive (Figure 2). Notably, the macroscopic and microscopic appearances and the biological behavior of this tumor are distinctive; it is a cancer of low-grade malignancy up to date.[9,10]

Management of VC, especially of the head and neck region, supports surgical excision as the primary treatment. Nevertheless, carbon dioxide laser, cryosurgery, chemotherapy, intralesional or iontophoretic methods, photodynamic therapy, systemic retinoid therapy, and radiotherapy modalities have been recommended.[8,10] However, contradicting ideas exist regarding the value of radiation therapy. Some

investigators have stated that radiation therapy has no role in the treatment of VC, while others support the use of radiation therapy alone or as an adjunct to surgery.[2] Progression to aggressive squamous cell carcinoma after radiation or chemotherapy has been reported. The significance of positive margins emphasizes the need for surgical resection with adequate margins.[10] VC has an excellent prognosis with surgical management.[11,12] In our case, wide tumor excision was done with negative free margins, and there was no recurrence after 12 months of follow-up. The overall 5-year survival rates of 61.1% and 77.9% have been observed in previous studies.[2,13,14] The low 10-year observed survival rate (40.0%) in some studies is likely due to additional high mortality from other causes that are associated with advanced age.

A potential caveat for our case study is the lack of essential information such as preoperative investigations, including blood tests to rule out HPV and computed tomography (CT) scans partly because of the patient's financial constraints. Similarly, we did not take intra-oral preoperative photographs.

Conclusion

VC is a rare variant of well-differentiated squamous cell carcinoma. The current case study describes a report of a giant VC of the chin in a 43-year-old female. The tumor grows gradually, has a tendency toward local invasion, and seldom metastasizes. VC is treated by surgical removal with free margins to ensure safety of the neighboring anatomical structure, and regular patient follow-up is critical.

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Author contributions

K.A.N. Conceptualization; methodology, data curation; writing—original draft; D.S.R. Data curation, writing—review and editing; D.C. Data curation; writing—review and editing; C.J.B. Data curation; writing—review and editing; R.T.P. Conceptualization; investigation; writing—review and editing; A.M. Conceptualization; data curation; methodology, investigation, writing—original draft, writing—review and editing.

Data availability statement

No data was generated from this study.

Declaration of conflicting interests

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Ethics approval

Ethical approval for this study was waived by Kilimanjaro Christian Medical Centre Research Ethics Committee.

Informed consent

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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