Case Report

Oral verrucous carcinoma (Ackerman's tumor) with mandibular bone destruction and palpable lymph nodes

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سرطان الفم الثؤلولي (ورم أكرمان) مع تاكل عظام الفك السفلي وبروز الغدد الليمفاوية العنقية

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سرطان الفم الثؤلولي و سرطان الفم ذو الخلايا الحرشفية هما أورام مرتبطه باستخدام التبغ وقد يكون من المستحيل التمييز بينهما سريريا في بعض الاحيان. هذا تقرير حالة لسرطان فم ثؤلولي في مريض ذكر يبلغ من العمر 68 عاما ويتعاطى التبغ (التمباك) والكحول و يعاني من تاكل عظم الفك السفلي واسع النطاق مع وجود غدد ليمفاوية متعدده بالعنق. كان التشخيص السريري نحو سرطان الفم ذو الخلايا الحرشفيه و كشفت النتائج النسيجية المرضية انها سرطان فم ثؤلولي . تم إجراءعملية استئصال الفك السفلي مع تشريح رقبه تم فيه ازالة الغدد اللمفاوية انتقائيا.

Abstract

Oral verrucous carcinoma (OVC) and oral squamous cell carcinoma (OSCC) are tobacco associated neoplasms and sometimes it is impossible to distinguish between both clinically. This is a case report of OVC in snuff dipper and alcoholic 68 year-old male with massive mandibular destruction and multiple palpable lymph nodes where the clinical presentation was toward OSCC and the histopathological results revealed an OVC. A marginal mandible resection in OVC patient was performed with diagnostic selective neck dissection.

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Introduction

Oral verrucous carcinoma (OVC) is a low-grade variant of oral squamous cell carcinoma (OSCC), Reported first by Ackermann 1948 as a spit tobacco associated malignancy. Various names are used in the literature to describe this entity, including Ackerman‟s tumor, Buschke-Loewenstein tumor, florid oral papillomatosis, epitheliomacuniculatum, and carcinoma cuniculatum[(1,2)].

Many verrucous carcinomas arise from the oral mucosa in people who chronically use chewing tobacco or snuff, typically in the area where the tobacco is habitually placed[(3)]. In Sudan a local form of snuff (smokeless tobacco) called Toombak commonly use and the most common site for snuff dipping is the lower labial sulcus[(4)].

Oral verrucous carcinoma grows slowly and locally invasive in nature and unlikely to metastasize. It also has a heavily keratinized, or parakeratinized, irregular clefted surface with parakeratin extending deeply into the clefts. The prickle cell layers show bulbous hyperplasia, but, for a considerable time at least, the tumor has a well-defined lower border and basal lamina. Atypia is minimal, and there is usually a sub-epithelial inflammatory infiltrate[5,6].

Regional lymph nodes were often tender and enlarged because of inflammatory involvement, simulating metastatic tumor[5]. Although, the clinical presentation of OVC mimicking OSCC,its treatment modalities may differ.

A case of OVC will be presented in which the patient is snuff dipper and alcoholic male with massive mandibular destruction and multiple palpable lymph nodes.

Case report

A 68-year-old male was referred from a rural hospital to oral and maxillofacial surgery department, Khartoum Teaching Dental Hospital complaining of swelling at the anterior region of the mandible for the last five months. The patient stated that he noticed a sudden severe mobility of the anterior teeth and painful swelling projections at the lower labial vestibule that interfere with eating

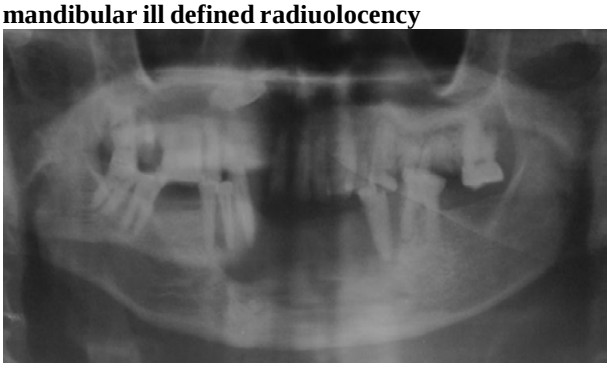
A review of the patient medical history doesn‟t reveal any significant finding. He is alcoholic and snuff dipper (tobacco) for the last 40 years. He used to snuff all the time even when he is sleeping and drinking alcohol on a daily basis (more than one bottle a day).

On examination, the patient looked well. Clinical examination revealed facial asymmetry on the lower third of the face due to the presence of the lesion (Fig 1). There were bilateral palpable submandibular lymph nodes which were mobile but not tender. Intraorally, there was fungating tender swelling extending from the third molar area on the left side to the second molar area on the right side with mobile canine on the right side (other mandibular anterior teeth was missing).

Fig 1: Preoperative clinical picture showing the exophetic growth with multiple mobile teeth

The whole gingivolabial vestibule was involved, and the lesion was extending to the floor of the mouth without any interference with the tongue movements. Computerized tomography (CT) scan and Orthopantomography (OPG) revealed destruction of the anterior mandibular alveolar bone with floating canine and first premolar on the right side and second premolar on the left side (Fig 2).

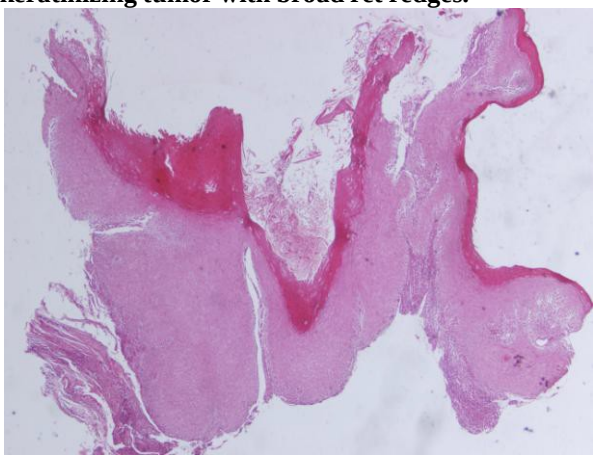
Fig 2: Orthopantomograph showing anterior mandibular ill defined radiuolocency



An incisional biopsy was done and revealed a verrucous carcinoma. Since the clinical presentation was more toward squamous cell carcinoma (SCC); re-biopsy was requested to confirm the diagnosis and came up with verrucous carcinoma again (Figs 3 & 4).

Fig 3: An intraoperatvie view of the resectedspecimen

Fig 4: This section shows a deeply infiltrative keratinizing tumor with broad ret redges.



Then the patient was planned for surgical excision of the lesion under GA.

Intraoperatively, a marginal resection of the lesion was done from wisdom tooth area on the left side to the right side leaving the wisdom tooth. The inferior border of the mandible was intact (Fig 5). Bilateral submandibular and submental lymph nodes were harvested and taken as diagnostic lymph nodes for possible metastasis. The excisional biopsy was sent to histopathological investigation, and the result confirmed the incisional biopsy finding which verrucous carcinoma (Fig 6).

Fig 5: This section shows parakeratosis with extensive inflammatory infiltrate in the underlying stroma

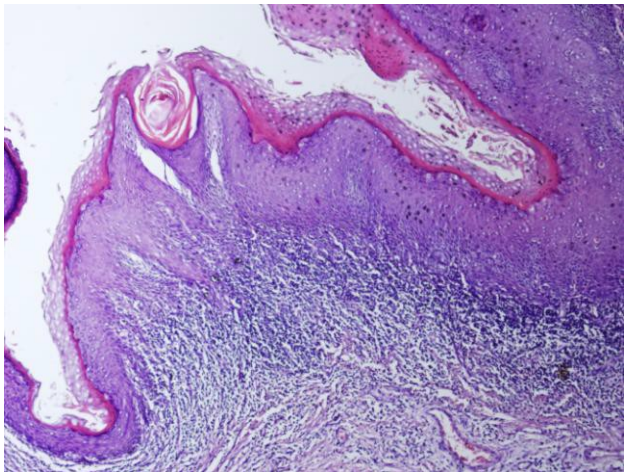
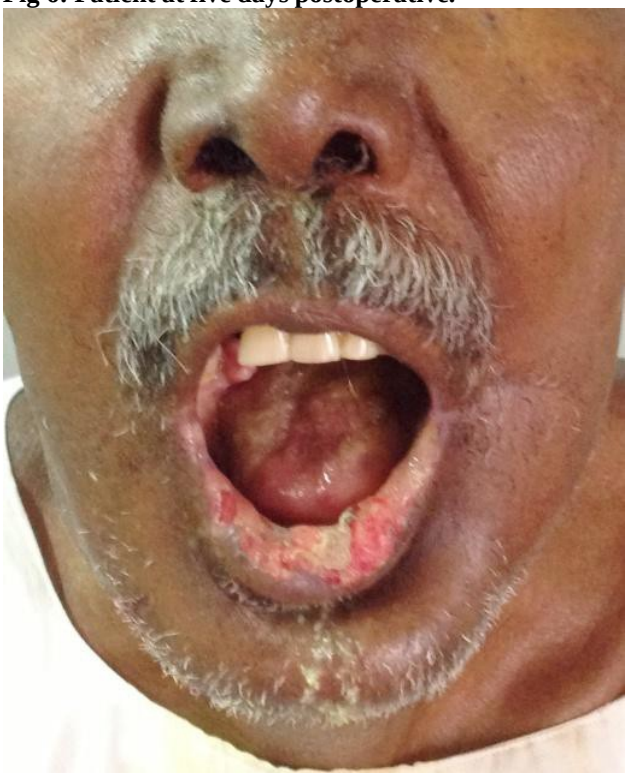


Fig 6: Patient at five days postoperative.



In 18months follow-up, the patient was well and disease-free (Figs 7 & 8).

Fig 7: Postoperative OPG showing the mandibular marginal resection.

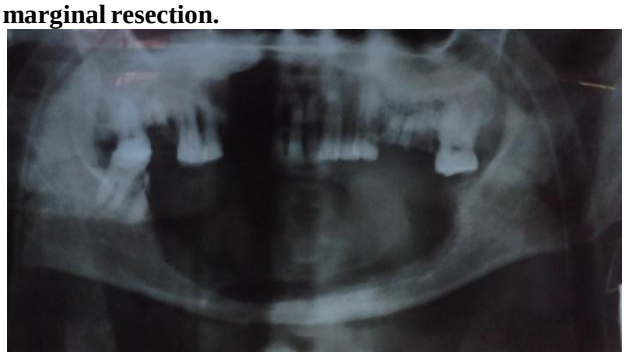


Fig 8: The patient at 18 months follow up.

Discussion

Oral verccous carcinoma is a rare malignant tumor which has been considered as a variant of well differentiated SCC. Alkan et al reported only 12 cases in 10 years duration[7]. Furthermore, Idris et al in their epidemiological study, there was no report for such tumor which reflect the rarity of this lesion[4].

The etiology of OVC is unclear. However, studies have shown strong associations with tobacco use, including inhaled as well as smokeless tobacco, alcohol, and opportunist viral activity associated with human papilloma virus (HPV)[8]. In this case report, the patient has a long history of two predisposing factors which are smokeless tobacco and alcohol abuse.

OVC is predominantly seen in males over the sixth decade[9]. Clinically, it has exophetic growth pattern with hyperkeratotic whitish warty surface all of which are evident in this report.

OVC is slow growing tumor but can become locally aggressive. However, even with local tumors progression, it is interesting that regional or distant metastasis is rare.

This patient was treated with surgery alone (marginal mandibular resection). Selective regional lymph node dissections were performed as part of the initial surgical treatment due to clinically palpable nodes. All of the neck specimens were negative for metastasis after detailed pathological examination.

Surgery is considered as the treatment of choice[7,10]. Some investigators have reported that radiation therapy was not only ineffective in many cases, but also caused anaplastic transformation of the neoplasm leading to rapid metastatic dissemination. In Demian et al study, 30% of patient with OVC whom received a curative dose of radiotherapy as definitive treatment, they developed a more anaplastic tumors[11]. This anaplastic transformation following radiotherapy favored the surgical treatment modality. Also, there is recommendation; that verrucous carcinoma should be treated as SCC with the treatment modality determined by effectiveness of control without regarding the potential risk of its developing into a far more aggressive lesion after irradiation[10]. Recent reports found that patients with OVCs had similar responses to radiotherapy as those with well differentiated SCC[6]. According to these controversial opinions; the patient was not sent for other treatment modalities.

In general, the prognosis of a patient with verruocus carcinoma is very good when compared to SCC[7]. Eighteen months of follow up, the patient is well with no evidence of recurrence

In conclusion, oral SCC and OVC are known tobacco associated neoplasm and sometimes it is difficult to distinguish between both clinically. Bone and lymph nodes involvement is usually considered to be associated with

SCC since the aggressive nature of it. In this patient; the clinical presentation was toward SCC and the histopathological results revealed an OVC. Surgery as single treatment modalities is proven to be effective in this case.

Competing interests

No competing interest.

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