Case Report

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Dentistry Section

Ackerman's Tumour - A Case Report

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ABSTRACT

Ackerman's tumour or Verrucous Carcinoma (VC) is a distinct form of Squamous Cell Carcinoma (SCC). It is predominantly seen in elderly men and strongly associated with the use of tobacco. In oral cavity it occurs predominantly in buccal mucosa followed by gingiva, palate and floor of the mouth. Clinically it manifests as proliferative finger like projections resembling a cauliflower which is characteristic of its diagnosis. Histologically, papillary or verruciform surface and parakeratin plugs between the surface projections are seen with an intense infiltrate of chronic inflammatory cells in the connective tissue thus it requires enormous expertize for diagnosis. Although VC is illustrated as a benign lesion with minimum aggressive potential over a period of time it can evolve into SCC. Hence most appropriate management of Ackerman's tumour is early diagnosis and surgical excision of the lesion. With this above background, we hereby report an enticing case of verrucous carcinoma in a left retromolar trigone extending into lower alveolar ridge in a 52-year-old male patient.

Keywords: Proliferative papillary like projections, Para keratin plugs, Squamous cell carcinoma, Verrucous carcinoma.

CASE REPORT

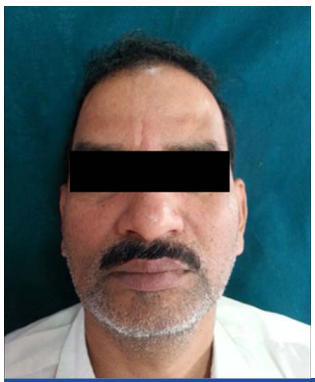
A 52-year-old male patient, reported to the Department of Oral Medicine and Radiology with a chief complaint of pain and swelling in the lower left retromolar trigone since 10 days [Table/Fig-1]. Patient noticed a small, painless growth over the left retromolar trigone six months back, which gradually grew to the present size. Patient gave history of pain associated since one month which was initially mild and intermittent but has aggravated since 10 days. Patient consulted a dentist 20 days back with the same complaint for which he was prescribed a course of antibiotics (Amoxicillin 500 mg thrice daily for five days) and analgesics (Diclofenac Sodium 50 mg thrice daily for three days) deleterious habit history revelaed holding smokeless tobacco quid for 15 minutes in the lower left retromolar trigone since 20 years, 8-9 times/day following which he used to spit out the contents.

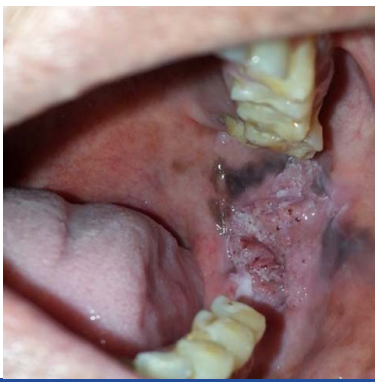
No relevant medical or family history were contributory. On examination all vital signs were within normal limits and no gross asymmetry of the face was noted. Ipsilateral left submandibular lymph node was mobile, non-tender and firm on palpation.

Intraoral soft tissue examination revealed solitary exophytic sessile growth with keratotic proliferative finger like projections over the left retromolar trigone measuring approximately 4x3 cm extending anteriorly to lower alveolar ridge of 36,37 region [Table/Fig-2]. All inspectory findings were confirmed on palpation. Hard tissue examination revealed missing teeth in relation to 36,37 and dental caries in relation to 48. With all the above findings a working diagnosis of proliferative verrucous leukoplakia in relation to left retromolar trigone was made and a differential diagnosis of verrucous carcinoma and verrucous hyperplasia were considered. Subsequently patient was subjected to routine radiographic and blood investigations.

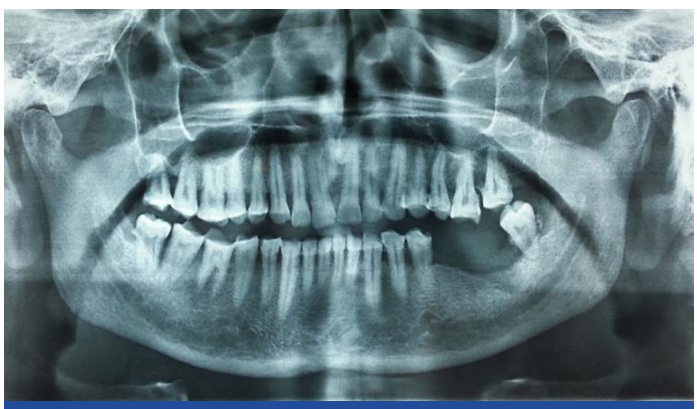
Panoramic radiograph [Table/Fig-3] revealed missing teeth (36,37) and sparse trabecular pattern were noted in relation 36,37 region. Based on these features patient was subjected to Computed Tomography (CT) to evaluate further extension and nature of lesion of mandibular left retromolar trigone. Additional finding revealed irregular polypoidal submucosal thickening of 6mm noted in the left retromolar trigone [Table/Fig-4a]. No evidence of lytic/ destructive lesion noted in underlying bone [Table/Fig-4b]. There were multiple level 1B lymph nodes on left side, largest measuring 1 cm in sort axis [Table/Fig-4c]. The CT findings were suggestive of an irregular polypoidal submucosal thickening of left retromolar trigone with lymphadenopathy.

All haematological investigations were found to be within normal limits. Under local anaesthesia, incisional biopsy was performed which was suggestive of verrucous carcinoma. Under strict aseptic conditions and general anaesthesia, nasal intubation was performed. Supraomohyoid neck dissection was done. Facial artery and facial vein were ligated. Pedicled submental flap was raised and prepared. Primary lesion present on the left retromolar trigone extending till alveolar process was segmentally resected [Table/Fig-5]. Thorough irrigation was done. Lymphnode level IA, IB, IIA, IIB and III were

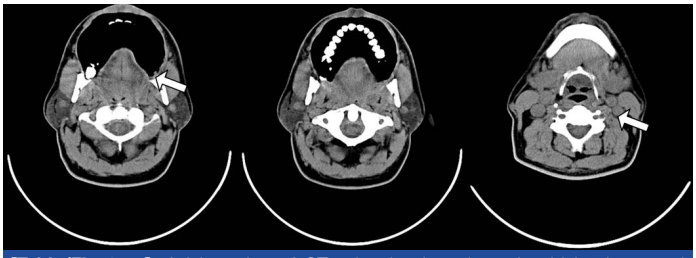




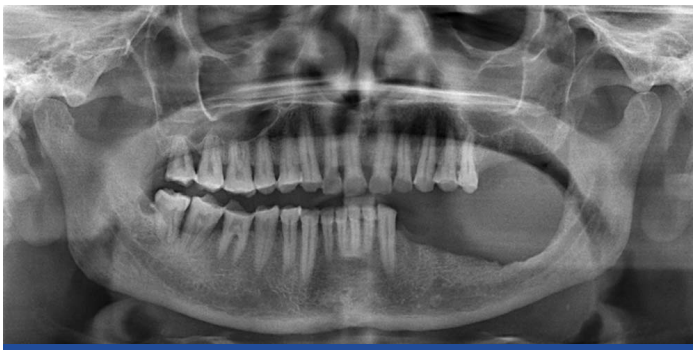
[Table/Fig-1]: Frontal view of the patient. **[Table/Fig-2]:** Exophytic sessile growth with keratotic proliferative finger like projections over the left retromolar trigone



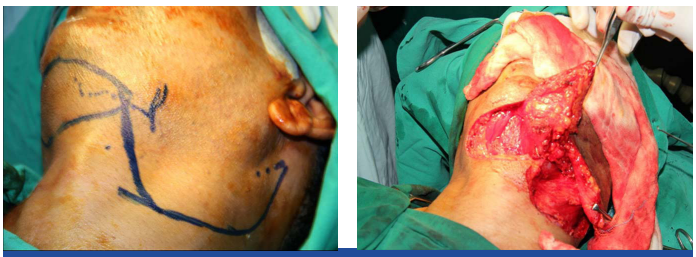
[Table/Fig-3]: Orthopantomogram (preoperative)



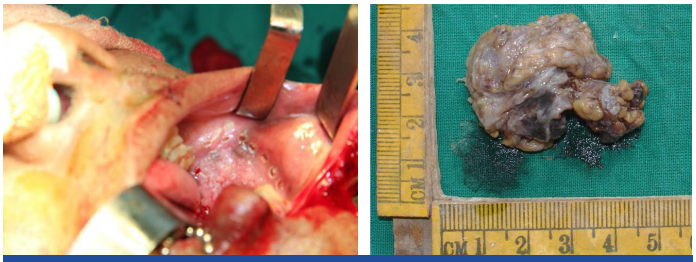
[Table/Fig-4a-c]: Axial section of CT showing irregular polypoidal submucosal thickening of left retromolar trigone, with No evidence of lyitc/destructive lesion in underlying bone and multiple level 1B lymph nodes on left side



[Table/Fig-5]: OPG reveals segmental resection of posterior mandible irt (36,37)



[Table/Fig-6a]: Intraoperative markings over the neck and submental area to plan incision **[Table/Fig-6b]:** Pedicled submental flap raised and prepared

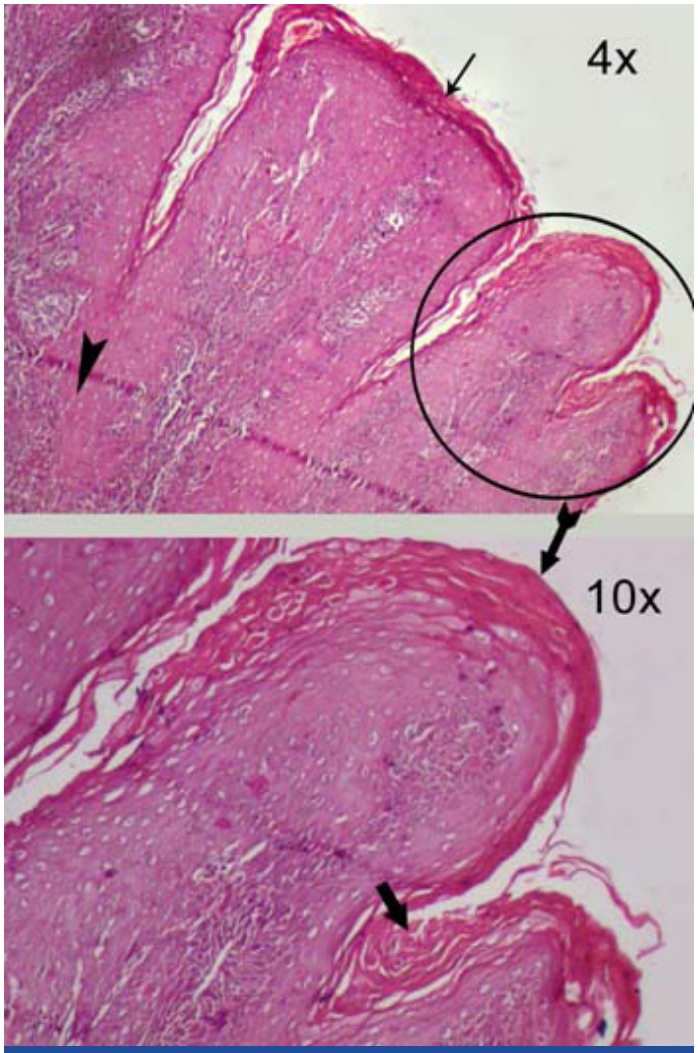


[Table/Fig-6c]: IReconstruction done using submental flap
[Table/Fig-7]: Gross specimen

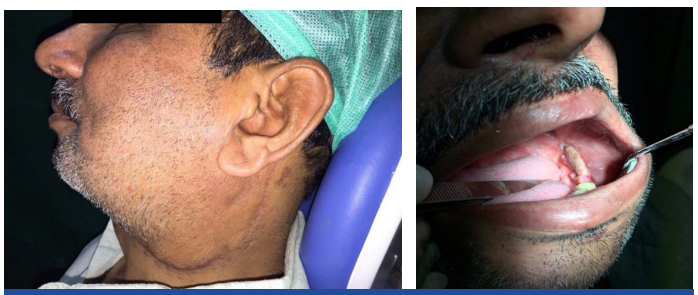
cleared. Wide excision was performed and reconstruction was accomplished by using submental flap [Table/Fig-6a-c]. The specimen [Table/Fig-7] was sent for histolopathological evaluation. H&E section revealed hyperplastic parakeratinised stratified squamous epithelium with papillary projections and keratin plugging with cleft formation [Table/Fig-8a] and wide, elongated rete ridges with pushing margins into underlying connective tissue [Table/Fig-8b]. These features reconfirmed earlier histopathological diagnosis. Hence considering clinical, radiographic and histopathological findings, a final diagnosis of verrucous carcinoma was given. Following treatment, patient is under followup since last six months [Table/Fig-9a&b] and prognosis is good.

DISCUSSION

Verrucous Carcinoma (VC) originally described by Lauren V Ackerman which is a unique variant of Squamous Cell Carcinoma (SCC) [1] and constitutes 1-10% of cases of SCC [2]. It has a distinct mode of behaviour and clinicopathological features. It can be distinguished from SCC by slow growth, rare dysplasia and lack of metastasis [3]. VC prone to occur in the age of 6th-7th decade as



[Table/Fig-8a]: Photomicrograph showing hyperplastic parakeratinised stratified squamous epithelium with papillary projections (thin arrow); keratin plugging with cleft formation and pushing margins into connective tissue (arrow head) (4X)
[Table/Fig-8b]: Photomicrograph revealing higher magnification (10X) of marked area wherein wide and elongated rete ridges with keratin plugging (thick arrow) are evident



[Table/Fig-9a]: Postoperative view (Lateral view)
[Table/Fig-9b]: Postoperative intraoral view (after 6months)

per Koch et al., in the current case it occurred in 5th decade [2]. VC is predominantly seen in elderly men and strongly associated with the use of tobacco as seen in our case [4,5]. Various aetiological factors includes chewing betel nut, poor oral hygiene and human papilloma virus infection [3,4]. Above mentioned factors can prompt individuals to develop potentially malignant lesions such as leukoplakia, oral submucous fibrosis and erythroplakia [6]. Ackerman noticed the most common site of occurrence in the oral cavity is the buccal mucosa (61.4%) followed by lower alveolus (11.9%) whereas in the current case it occured in the retromolar area extending into the lower residual alveolar ridge [7].

VC are slow-growing, exophytic, well-demarcated hyperkeratotic lesions as per Schrader et al., and Jordan [8,9] Clinically, the surface shows an exophytic growth with keratotic proliferative papillary projections resembling a cauliflower and sometimes interspersed

with red areas [3]. Most cases are symptomatic where in patients complain of pain and discomfort [3]. In early stages, the lesion is soft in consistency and as the lesion progresses induration may be seen. Lymph nodes are often tender and enlarged due to inflammation. Metastasis is very rare [3]. Current case clinical findings were consistent with the above mentioned features. VC should be differentiated from large papillomas, papillary SCC and verrucous hyperplasia and venereal warts [10].

Histopathologically VC has a deceptively benign microscopic appearance. It is characterized by wide and elongated rete ridges that appear to "push" into the underlying connective tissue. Abundant keratin production, papillary or verruciform surface and parakeratin plugs between the surface projections are seen with an intense infiltrate of chronic inflammatory cells in the connective tissue which were in accordance to histological features of our case. The histopathologic diagnosis of VC requires an adequate wide margin for incisional biopsy. Individual cells are not very dysplastic, thus the pathologist must carefully evaluate overall histomorphologic configuration of the lesion to arrive at an appropriate diagnosis [3]. Differential diagnosis of VC includes: (i) SCC showing verrucoid features, (ii) Proliferative verrucous leukoplakia, (iii) epithelial hyperplasia, (iv) pseudoepitheliomatous hyperplasia, (v) verruca vulgaris; (vi) keratoacanthoma [11].

Diverse treatment modalities for VC are as follows: surgery [12], radiotherapy [13], photodynamic therapy, interferon [12] and chemotherapy [14], out of which surgical excision is the most preferred [15] which was planned and implemented in our case. VC incidence of bone invasion was not found in case series of Oliveira et al., which is in concurrent with our case [15]. In treatment planning for VC, neck dissection plays a vital role to prevent locoregional control of the disease and its recurrence. VC is an extremely challenging pathological diagnosis and often even an adequate biopsy may miss areas of squamous differentiation [7]. Given the low incidence of pathological bone involvement, more conservative surgical options such as marginal mandibular resection may be considered while planning surgical therapy as done in our case. It has excellent prognosis due to its slow growth and lack of metastasis [12]. Alper et al., observed local reccurence rate (57%) in VC, thereby long term follow up is required [10].

CONCLUSION

We would like to conclude, by stating that although VC is known as a benign lesion with minimum aggressive potential it may further progress to SCC in untreated cases, hence early diagnosis, surgical excision and long term follow up is required.

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