Keratoacanthoma of the oral cavity with osteomyelitis of the mandible

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ABSTRACT

Introduction- Keratoacanthoma is a benign proliferative disease of the epithelium. It is commonly seen in the exposed parts on the lips, cheek nose and dorsum of the hand. It is rarely seen in the oral cavity. Keratoacanthoma of the oral cavity with mandibular osteomyelitis has not been reported until now.
Case report- We report a case of keratoacanthoma of the oral cavity with mandibular osteomyelitis. 82 years old patient with oral lesion of 5 months duration with history of tooth extraction, biopsy was nonconfirmative for malignancy and the CT scan showed destructed mandible. He underwent excision and reconstruction with mandibular reconstruction plate and advancement flap. Histo pathology was reported askeratoacanthoma of the oral mucosa and osteomyelitis of the mandible.
Conclusion- Keratoacanthoma may be a differential diagnosis for well differentiated squamous cell carcinoma. Tooth extraction could be the aetiology for keratoacanthoma and the osteomyelitis of the mandible.

Key words: Keratoacanthoma of the oral cavity, osteomyelitis of the mandible.

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INTRODUCTION

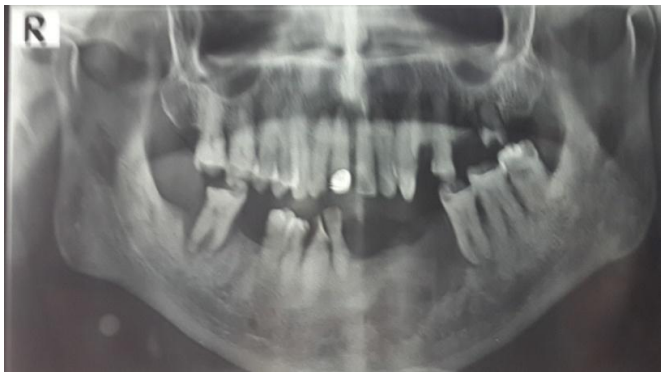
Keratoacanthoma is a benign self-limiting disease that proliferates from epithelium of the hair follicle.[1] Keratoacanthoma is commonly seen in the elderly male over the sun exposed areas like the cheek, nose, lips and dorsum of the hand.[2] The lesion appears as a crater which has keratin in the center. These lesions have clinical and histological features similar to well differentiated squamous cell carcinoma. Surgical excision is the treatment of choice, as it is required for the diagnosis.[3] Keratoacanthoma of the oral cavity is described to be a rare entity.[1] But keratoacanthoma associated with osteomyelitis of the mandible has not been reported.

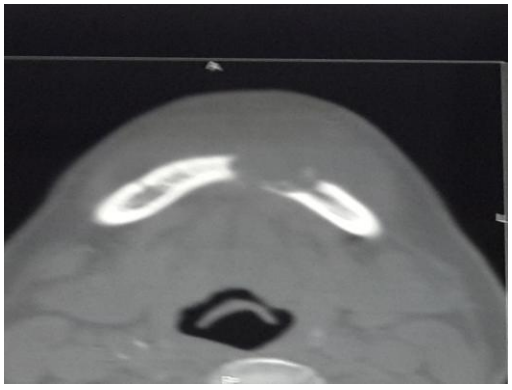
CASE REPORT

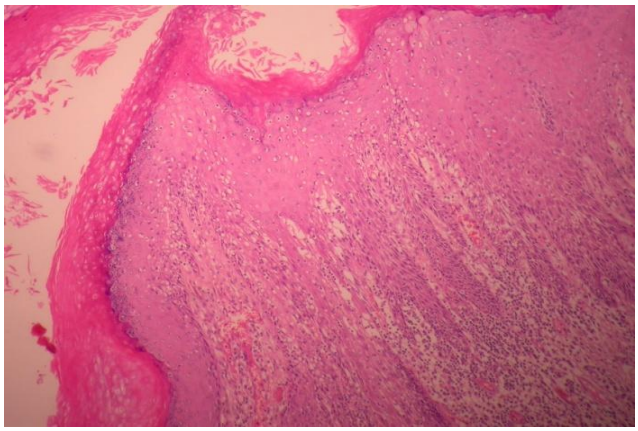
82 years old gentleman presented with non-healing, intraoral lesion of 3 months duration. He had history of tooth extraction 2 months back. The lesion was on the right side of the alveolus over the molar region, which was 5cm X 3cm in size, crateriform lesion with induration extending to the level of incisors. Level 1 and 2 groups of cervical lymph nodes were 2cm x2cm size, firm and tender. CT scan showed destructed mandibular destruction extending upto the opposite parasymphseal region. [figure 1,2,3]Fine needle aspiration cytology of his neck nodes showed signs of inflammation.

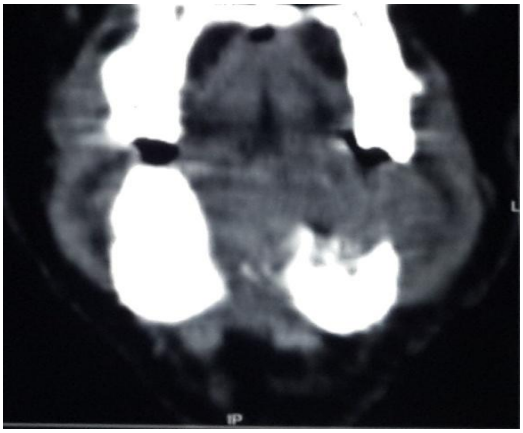
Repeat biopsies of the lesion were inconclusive for malignancy. In view of above clinical features and the CT scan showing destructed mandible, he underwent wide local excision of the lesion with supraomohyoid neck dissection. Mandible was reconstructed with a mandibular reconstruction plate and it was covered by advancement flap from the floor of the mouth. His post-operative period was uneventful. He had complaints of anesthesia over cheek and chin over the region supplied by mental nerve.

Microscopic examination showed features of keratoacanthoma (fig 4) with osteomyelitis of the mandible. HPE showed hyperkeratotic squamous epithelium with severe and prominent keratosis, acanthosis and papillomatosis was seen. At places dysplasia was liited to 2/3rd of the lining epithelium. Atypical squamoid cell clusters are seen in stroma. Subepithelialfibrocollagenoustissue show dense chronic inflammatory cell infiltrate. The cervical lymph nodes were reported as free from tumour.









DISCUSSION

Keratoacanthoma of the oral cavity is very rare. Yuk Kwan Chen has recorded possibly the twelth case of intraoral keraatoacanthoma, and the first keratoacanthoma of the tongue.[4] Keratoacanthoma of the oral cavity with osteomyelitis of the mandible has not been reported until now.

Keratoacanthoma are considered low grade squamous cell carcinoma which will regress in many cases. The causative factors include trauma, sunlight, immunosuppression, genetic causes, lichen planus[5,6,7].

Keratoacanthomas are commonly seen on the sun exposed skin, and especially over the vermillion border of the lips [1]. However, lesions have been described in literature, over the oral mucosa, lip and conjunctiva. The incidence of such extra skin lesions are rare [1,8]. Keratoacanthomas arise from the epithelium of the hair follicle [1], and the oral keratoacanthomas has been postulated to be from either ectopic sebaeceous glands or the overlying mucosa [9]. Keratoacanthoma of the mucosa and osteomyelitis of the mandible has not been reported in the English literature.

With age the bone gets less denser and also there is decrease in the height of the mandible. Osteomyelitis is caused by caries teeth, teeth extraction, open fractures, irradiation and other causes.[10] Tooth extraction might have led to osteomyelitis. The same trauma can be attributed to the development of the keratoacanthoma of the oral cavity. Most commonly, CT scan finding of the osteomyelitis of the mandiblie is a lytic type of lesion and if the bone is eroded, then it suggests involvement of the bone.[11]

Carcinoma cunnilata can also be considered as differential diagnosis .It is a rare well differentiated squamous cell carcinoma which is misdiagnosed frequently as osteomyelitis of mandible, and also shows destruction of the bone[12]. Our case had only atypia at some places and features were more suggestive of keratoacanthoma.

Evaluation could not differentiate between malignancy and a benign problem, which has been a problem with keratoacanthoma. Hence It is better to treat the patient as malignancy. The treatment of keratoacanthoma of the oral cavity is wide local excision and reconstruction.

Surgical excision is mostly for diagnosis and to rule out malignancy. Standard treatment for the present condition would be wide local excision and reconstruction with free fibula followed by osteointegrated implants. Considering the morbidity of the procedure in the old man, reconstruction with a plate and local flaps was done.

CONCLUSION

Keratoacanthoma of the oral cavity is a very rare disease. It can occur in conjunction with osteomyelitis. Tooth extraction may have been the cause for both the pathology. Surgical resection and histological examination can differentiate it from malignancy.

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