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Case Report: We report a case of 35year male presented with swelling in anterior mandible and recurrent gum bleeding an unusual site and unusual presentation.

Conclusion: Being a rare tumour SOT should be differentiated from other similar looking tumour i.e. acanthomatous ameloblastoma, SOT like islands arises from cystic wall and many others for definite therapy

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Squamous Odontogenic Tumour of Anterior Mandible – A Rare Case in Unusual Location

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I. Introduction

quamous odontogenic tumour (SOT) is rare intraosseous benign epithelial neoplasm with locally invasive nature. SOT was first described by Pullon et al. (1975). In ensuing 40 year less than 50 cases reported till now. SOT is defined as locally infiltrative neoplasm consisting of islands of well differentiated squamous epithelium in a fibrous stroma. The age range is between 8-74 years with mean of 38.7 years² with slight male predominance (M:F 1.4:1).

SOT occurs intraosseously and develops in the periodontal ligament between the roots of vital erupted permanent teeth. The tumour location is slightly more common in posterior mandible than anterior mandible¹. Usually patients present with asymptomatic gingival swelling or local pain, mobility of teeth, osseous expansion and mild gingival erythema.¹

Radiographically tumour present as well defined triangular radiolucency adjacent to roots of teeth.³ Occasional calcification and cystic degenerationcan occur.

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Histologically, the tumour composed of multiple islands of squamous epithelium surrounded by mature connective stroma without peripheral palisading. Occasionally calcification and central cystic degeneration also observed.

Conservative surgical treatment is sufficient and recurrences are rare.

II. CASE REPORT

A 35 year male presented with recurrent gumbleeding since last 10 days. He had history of gradually increasing gingival swelling, pain and mobility of lower incisors over anterior mandible from last one year.

Non contrast MDCT-Denta scan imaging study revealed a well-defined mixed radio opaque radiolucent space occupying lesion involving lower mandible measuring 27.8 x 25.3 x 19.8 mm. Periphery of the lesion was partly sclerotic and partly surrounded by radiolucent halo. The lesion was composed of granular septation with concomitant sign of multifocal calcification. Extensive expansion, distortion and destruction of the buccolingual cortex of bone and knife edge type of root resorption is also observed. [Figure1]

Previous incisional biopsy from the tumour tissue reported asacanthomatous ameloblastoma.

Anterior segmental mandibulectomy done. On gross examination a greyish white irregular solid-cystic structure measuring 5 x 3 x 2cm. Outer surface of which was smooth and covered with gingival mucosa. Inner surface was friable. Histopathological examination revealed presence of islands of squamous cells lined at its periphery by flattened cells without categorical pleomorphism or atypical mitotic figure surrounded by fibrous stroma. Occasional cells show clear cytoplasm. Palisading of cells and ameloblastic stroma is not noted anywhere with serial sections. Focal calcification and central cystic changes also observed. [Figure 2, Figure 3, Figure 4]

III. Discussion

In 1975 Pullon et al. first described 6 cases of previously unnamed oral lesions as squamous odontogenic tumour.⁴

Bansal et al descried a table of 44 cases showed that tumour location is slightly more common in mandible than maxilla. Posterior mandible is more prone

than anterior for the lesion and anterior maxilla is far more common location than the posterior maxilla. Only few cases were multicentric and only one case was bilateral in posterior maxilla.5 Though SOT in maxilla is more aggressive than mandible and required more radical surgery.^{6,7} Only one case is reported till now tumour localised between roots of central incisor of mandible⁵. In our case tumour was localised below all the incisors of mandible.

Clinically tumour presents as painless gradually increasing swelling of mandible or maxillary bone, mobility of teeth, pain and erythema of the lesion. Though SOT may be asymptomatic and detected in routine intraoral radiograph. Our case is unique it also presents with recurrent gum bleeding.

Radiographically tumour present as well defined triangular radiolucency adjacent to roots of teeth.3 Occasional calcification and cystic denegation can occur. The lesion is usually central but sometimes it may be peripheral³ which may produce some saucerization of the underlying bone- a result of pressure from tumour expansion rather than neoplastic infiltration.¹

Due to calcified material SOTs may be misdiagnosed as acathomatous ameloblastoma, desmoplastic ameloblastom, well differentiated squamous cell carcinoma or pseudoepitheliomatous hyperplasia. Other possible differential diagnosis may be "squamous odontogenic tumour like islands arising in the walls of odontogenic cyst". 1 SOT can differentiate from ameloblastomaby observing absence of peripheral palisading and cytoplasmic vacuolation. In addition, stellate reticulum like cells and ameloblastic stroma, which are always present in ameloblastoma, are never seen in SOT.8

CH Siar et al showed positive reactivity of varying intensity in the neoplastic epithelial for Notch1, Notch3, Notch4 and their ligands Jagged1 and Delta1. No immunoreactivity was detected for Notch2 and Jagged2.9

IV. CONCLUSION

It is a rare tumour mimicking other more common odontogenic tumour, intracystic squamous cell carcinoma and some benign proliferative lesions. It should be bear in mind that SOT is a locally aggressive tumour which is curative with careful surgery and should be differentiated from the other mentioned tumour and tumour like lesions for specific therapy.

Abbreviations

SOT – Squamous odontogenic tumour

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