Case Report

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Non Dentigerous Type Unicystic Ameloblastoma in Canine-Premolar: Rare among Common: Case Report

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Abstract

Ameloblasoma comprises 1-2 % of all cysts and tumors occurring in jaw. Unicystic ameloblastoma is a less aggressive variant of ameloblastoma mainly seen in younger age group with impacted tooth in mandibular ramus area. In present case young male patient developed asympotomatic swelling with no impacted tooth in the region of canine – premolar with histological features of mural and intraluminal proliferation pattern of unicystic ameloblastoma.

Keyword: Ameloblastoma; Odontogenic; Mural.

Introduction

The unicystic ameloblastoma, a variant of ameloblastoma developing within the lining, lumen or wall of a cyst as well as an invasive ameloblastoma that has a single cystic space rather than multi cystic spaces.[1]

It shows ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation.[2] They are seen in a younger age-group than solid tumors.[3] It is considered as less aggressive variant.[4]

The present case intraluminal and mural proliferation pattern is seen in canine-premolar region with no impacted tooth, which has been seen in very few reported cases.

Case Report

22 years old male patient had complaint of swelling in the lower left back jaw since 1 month with gradual progression & mild pain. On extra oral examination a diffuse swelling of approximately 2 cm in lower anterior region of left side of mandible (Fig 1), firm in consistency, non tender with no discharge. Intra-orally, swelling was in relation to 33, 34 and 35 region. Surface appeared smooth and well defined margins with no color change. A clinical diagnosis of odontogenic cyst and

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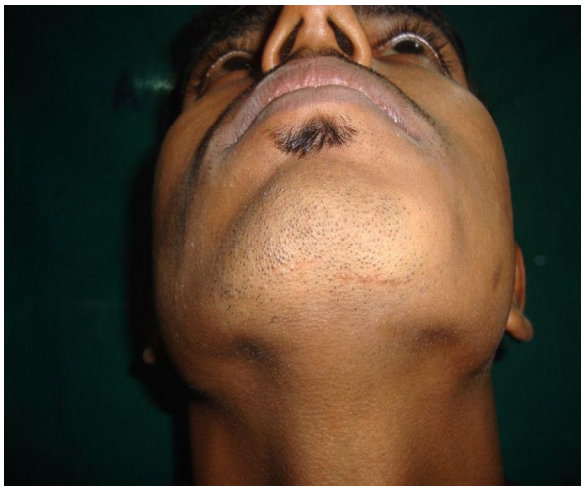
Fig 1: Diffuse swelling of 2 cm (approx) in lower anterior region of left side of mandible



benign odontogenic tumor was considered.

Orthopantomograph(OPG) and occlusal view (Fig 3 & 4) revealed well defined radiolucent lesion in relation to 33, 34 and 35 with lingual extension of the lesion. Aspiration suggested a cystic lesion. Excision of the lesion under general anesthesia was done. Gross examination of the excised specimen revealed cystic lining with nodular growth within lumen.

Histopathological examination show fibrous cyst wall with a lining that consists of ameloblastic epithelium. Epithelium shows basal layer of columnar cells with hyperchromatic nuclei that show reverse of polarity & basilar cytoplasmic vacuolization. Overlying epithelium in some areas resemble

Fig 2: Intraoral findings show small elevation with no color change

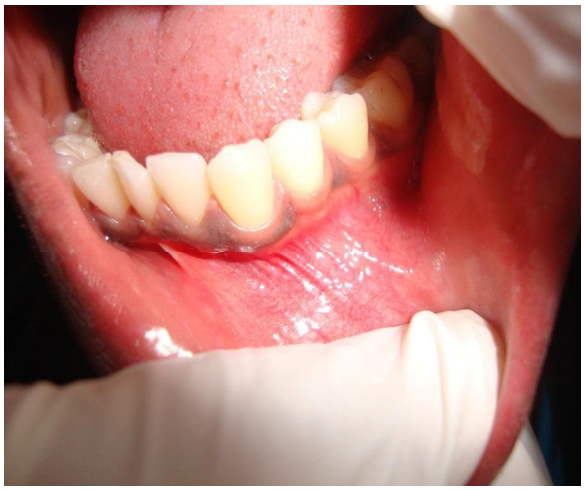


Fig 3: OPG shows unilocular cystic space below 33, 34 & 34

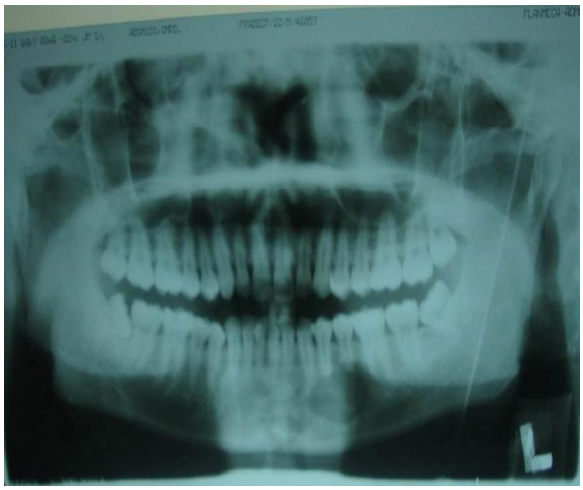


Fig 4: Occlusal view shows radiolucency extended to lingual side

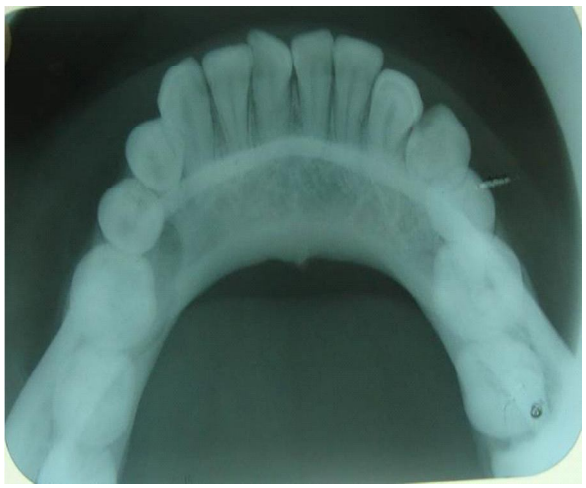


Fig 5: Gross specimen

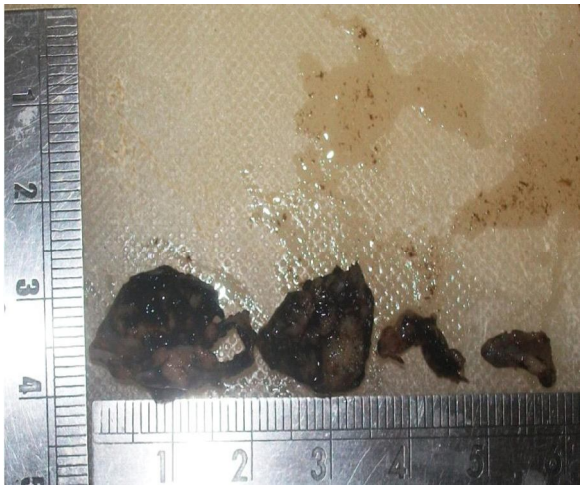


Fig 6: Shows mural proliferation

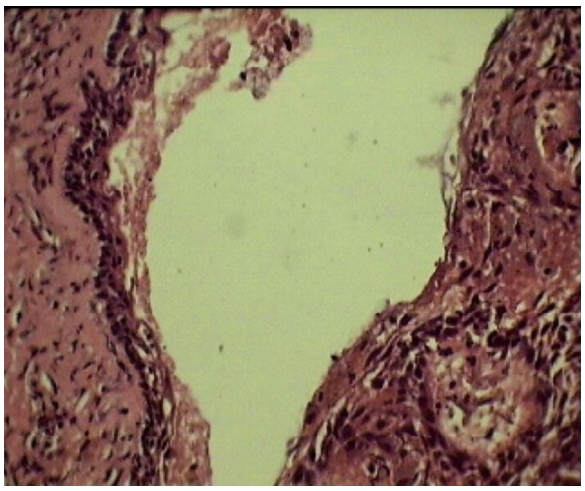


Fig 7: Shows intraluminal proliferation

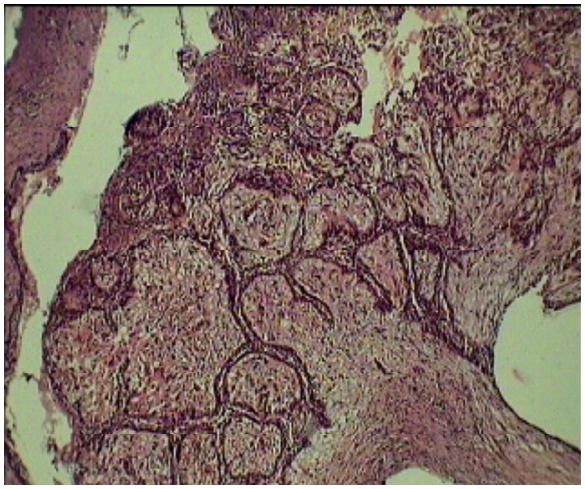
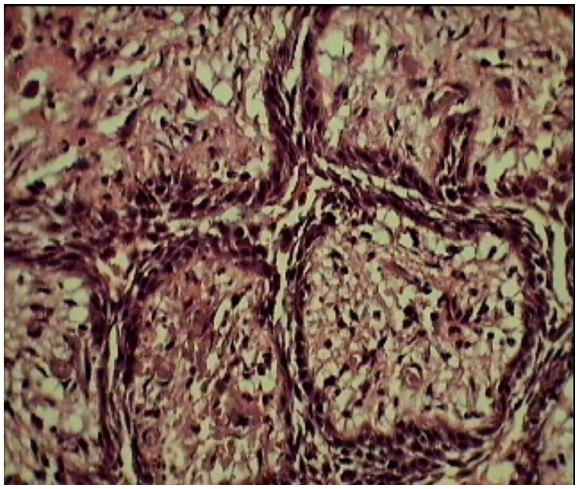


Fig 8: Shows Plexiform pattern with central stellate reticulum



stellate reticulum and shows mural and intraluminal proliferation in the plexiform pattern. Few areas showed subepithelial hyalinization. A diagnosis of unicystic ameloblastoma with mural and intraluminal proliferation was made.

Discussion

Unicystc ameloblastoma (UCA) is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas and diagnosed histo-pathologically as unicystic ameloblastoma, if a single cystic sac lined by odontogenic (ameloblastomatous) epithelium, often seen only in focal areas.[5]

The neoplasm originates within the mandible or maxilla from epithelium that is involved in the formation of teeth. Potential epithelial sources include enamel organ, odontogenic rests (cell rest of Malassez, cell rest of Serre) reduced enamel epithelium and epithelial lining of odontogenic cyst especially dentigerous cyst.[6]

Mandibular 3rd molar is mostly associated with impacted tooth in unicystic ameloblastoma (dentigerous type),[3] but a few cases are not associated with impacted teeth which are called non dentigerous variant. According to Konouchi H *et al*, mean age of non-impacted tooth related cystic ameloblastoma was 35 years in comparison to 16.5 years for the impacted tooth related variant.[7]

Leider *et al* proposed three pathologic mechanisms for evolution of unicystic ameloblastoma.[8]

a. The reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development.
b. Ameloblastomas arise in dentigerous or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining.
c. A solid ameloblastoma undergoes cystic

degeneration of ameloblastic islands with subsequent fusion of multiple micro cysts and develops into a unicystic lesion.

Ackermann classified this entity into the following three histologic groups:

Group I: Luminal UCA (tumor confined to the luminalsurface of the cyst)

Group II: Intraluminal/plexiform UCA (nodular proliferationinto the lumen without Infiltration of tumor cells into the connective tissue wall)

Group III: Mural UCA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

Another histologic subgrouping by Philipsen and Reichart[9] has also been described:

Subgroup 1: Luminal UCA

Subgroup 1.2: Luminal and intraluminal

Subgroup 1.2.3: Luminal, intraluminal and intramural

Subgroup 1.3: Luminal and intramural

UCA should be differentiated from other cysts because the former has a higher rate of recurrence than the latter.[5] Dentigerous cysts, odontogenic keratocyst, residual cysts, adenomatoid odontogenic tumour, giant cell lesions and sometimes solid ameloblastoma can be considered as differential diagnoses for a unilocular lesion with or without a 'dentigerous' relationship occurring within the jaws. However, keratocyst seldom shows cortical expansion, residual cysts are associated with missing teeth that have been extracted, adenomatoid odontogenic tumour has a predilection for anterior maxilla, central giant cell granuloma often arises anterior to first mandibular molar and solid ameloblastoma is seen less commonly in patients less than 30 years of age.[10]

Dense inflammatory infiltrates may induce hyperplastic epithelium and also cause intercellular oedema within cystic epithelium lining which appeared to be stellate reticulum and mimic ameloblastic proliferation.[11] In view of the reported ameloblastic potential of dentigerous cyst,[12] it is thus important to recognise true ameloblastomatous epithelium from ameloblastomatomatous-like epithelium. [13] Hence careful examination is must in radicular and dentigerous cyst.

The mural variety is seen to be more often associated with the 'non-dentigerous'type of these lesions, while the intraluminal proliferations are more than twice as frequent in UCAs of the 'dentigerous' type.[7] However in our case we observed both type of proliferation (mural and intraluminal) which make it a finding least documented.

Treatment is usually based on histology of unicystic ameloblastoma, if intraluminal then enucleation is sufficed but, if mural proliferation of ameloblastic epithelium then bony resection is mandatory. It is generally removed as dentigerous cyst without preoperative biopsy and Isacsson and associates considered biopsies of cystic lesion not to be recommended; all of the tissue must be included.

Conclusion

Our case showed proliferation of both mural and intraluminal which make it a finding least documented. We suggest that checking content of the lesion before doing removal of lesion whether cystic or solid; if possible CT scan and MRI are useful to know the extent of the lesion to prevent injury to neurovascular lesion.

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