

UNICYSTIC AMELOBLASTOMA: AN UNCOMMON VARIANT OF COMMON BENIGN TUMOUR, REPORT OF TWO CASES

Case Report

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ABSTRACT

Ameloblastoma is the common tumor of odontogenic origin. It is a neoplasm of enamel-organ tissue that has not undergone differentiation to the point of hard tissue formation. It is usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent, frequently seen in mandible. A wide range of multicystic ameloblastoma is the most frequent subtype while unicystic ameloblastoma can be considered as a variant of the solid or multicystic. This subtype is considered as a less aggressive tumor with a variable recurrence rate. However its frequency is often underestimated. Here in my case series, I am presenting 2 cases of ameloblastoma, where both of them are having histopathology showing unicystic in nature.

INTRODUCTION:

WHO (1992) defined ameloblastoma as “a benign locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium which usually has a follicular or a plexiform pattern lying in a fibrous stroma.”⁽¹⁾ Ameloblastoma represents 13–54% of all jaw tumors.⁽²⁾ Ameloblastomas are of three biologic variants: Cystic (unicystic), solid (multicystic), and peripheral.⁽¹⁾ Unicystic ameloblastoma (UA) was first described as a distinct variant of ameloblastoma by Robinson and Martinez in 1977.⁽³⁾ The term Unicystic ameloblastoma (UA) refers to cystic lesions that show clinical, radiographic, and gross features of jaw cyst, but on histological examination shows a typical ameloblastomatous epithelium lining part of the cystic cavity with or without luminal and/or mural tumor growth.⁽⁴⁾

The growth pattern of the unicystic ameloblastoma is seen in approximately 6% of all ameloblastomas.⁽⁵⁾ Occur in the second and third decade of life, predominantly in the posterior region of mandible.⁽⁴⁾ It has been suggested that epithelial adhesion or intrinsic production of proteinases; enzymes that normally degrade

central zone of the enamel organ after tooth development may be the cause of cystic degeneration of neoplasm.⁽¹⁾

Here, we present two rare cases of Unicystic Ameloblastoma with different Radiographic presentation in young female patients.

CASE REPORT:

Case 1

A 22 years old female reported with the Chief complaint of swelling on left side of face since 3 months. Patient has a medical history of swelling on same side before 4 years, extraction of 38, curettage done, drainage was placed swelling decreased. Which reappears before 3 months.

Extraorally, On inspection A Single well defined swelling was seen, 2 cm from ala to tragus antero- posteriorly. From cantho-meatalline to 2cm above Lower border of mandible. It was approximately 6*4 cm in size.

On palpation, Swelling was non tender, firm to hard in consistency. It was fixed with underlying tissue. The overlying skin was pinchable. No discharge while palpation was present. Lymph nodes were not palpated.

Intraorally, On inspection Single well-defined

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Figure 1: Extra-oral examination



Figure 2: Intraoral Examination

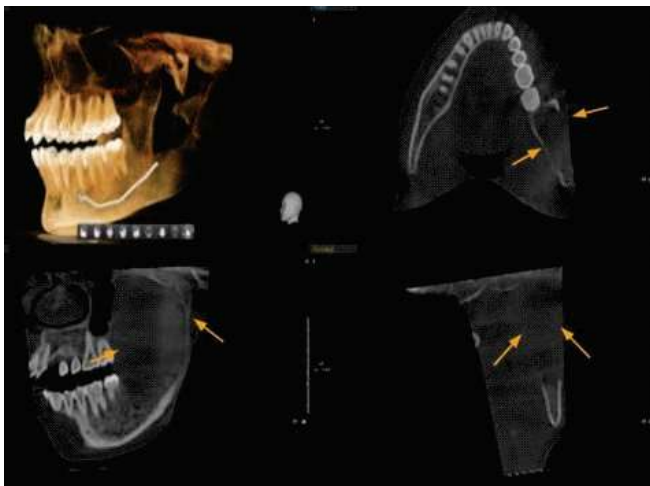


Figure 3 & 4: Radiographic examination

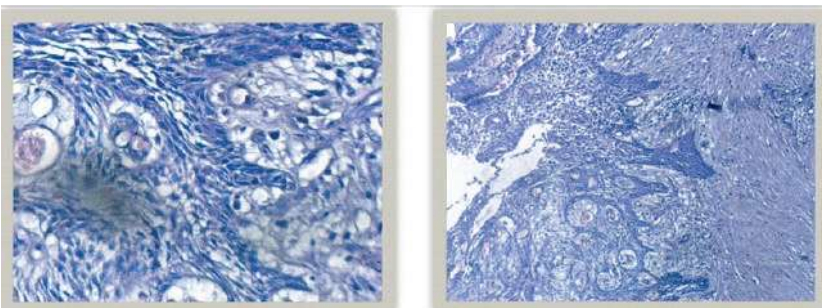
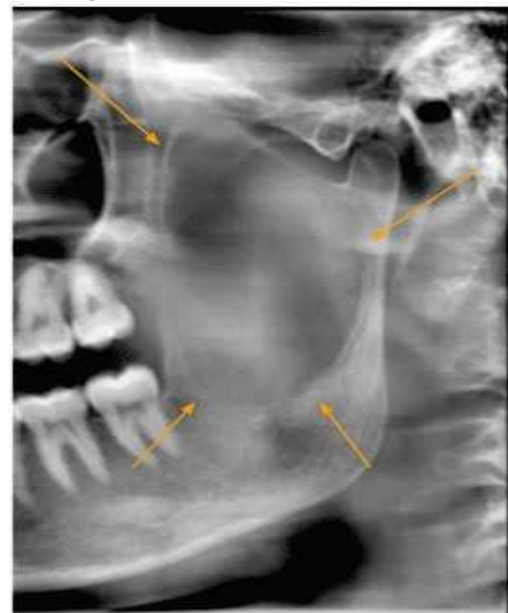


Figure 5: Given H&E stained section shows cystic lining and connective tissue capsule. Lining shows proliferation consisting of peripheral Ameloblast like cells and central stellate reticulum like cells. Connective tissue capsule shows RBS filled dilated blood vessels and chronic inflammatory cells.

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swelling present on left buccal mucosa near tuberosity region. Bulge extending from tuberosity to retromolar pad area. Medio-laterally lesion is involving buccal & lingual cortical plates. posterior part of the lesion is not traceable.

On palpation, Consistency—hard. Non tender, Fixed with underlying tissue, No discharge on palpation. On **Radiographic** examination, in orthopantomogram, Single Well defined unilocular radiolucency is seen. Internal structure is radiolucent, it is unilocular in appearance, no septa or locule seen. Surrounding structure shows expansion in Coronoid & sigmoid notch region. No root resorption or tooth displacement seen. Mandibular canal is traceable till 1 cm away from apex of 2nd molar.

In cone beam computed tomography, coronally it shows destruction of anterior border of the ramus and thinning but intact posterior border. Sagittally, shows mandibular canal passing just beneath the lesion near 2nd molar Axially, shows bucco-lingual expansion of the lesion, where more of buccal expansion can be seen.

Provisional diagnosis is Ameloblastoma and differential diagnosis is Odontogenic Keratocyst, Dentigerous cyst, Intra Osseous Hemangioma. Final Diagnosis is Intraluminal Unicystic Ameloblastoma. For treating it, Enucleation is performed.

CASE 2

A 16 years old female reported with the complaint Chief complaint of swelling on lower right face region since 5 days. Patient has a medical history of swelling on same side before a month, pain and mobility in 46, curettage done, visited a clinic, aspiration done, antibiotics given, swelling subsided. Which reappears before 5 days.

Extraorally, On inspection A Single well defined swelling was seen, Anterio-posteriorly – from commissure of mouth to 1.5 cm anterior to angle. Superio-inferiorly - 1.5 cm below ala tragus line to beyond 1 cm of lower border of mandible, 8*4 cm in size.

On palpation, Swelling was non tender, firm to hard in consistency. It was fixed with underlying tissue. The overlying skin was pinchable. No discharge while palpation present. Lymph nodes were palpated. Intraorally, On inspection Single well-defined swelling present on left buccal mucosa Anterio-posterior–mesial of 44 to distal of 47. Medio-laterally involving right buccal vestibule. It is 4*1.5 cm in size, Displacement of 45 lingually and 38 is absent.



Figure 6: Extraoral Examination



Figure 7: Intraoral Examination

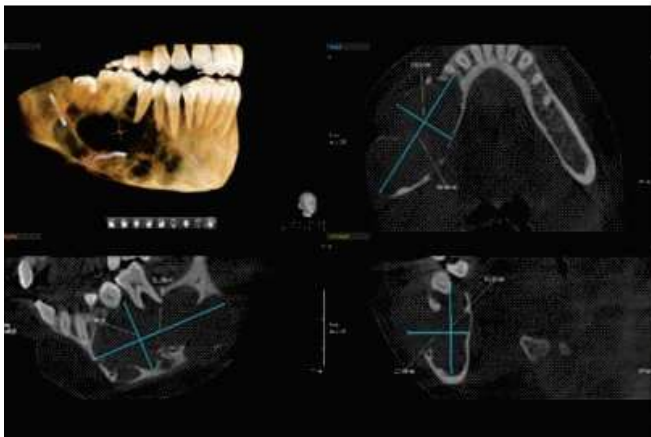
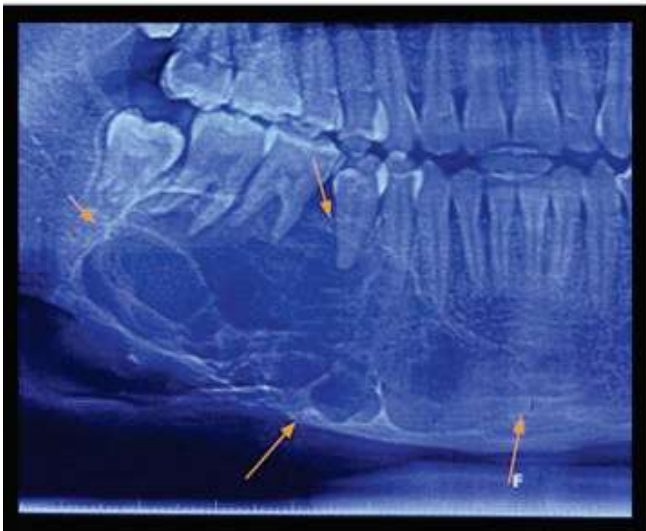


Figure 8: Radiographic examination

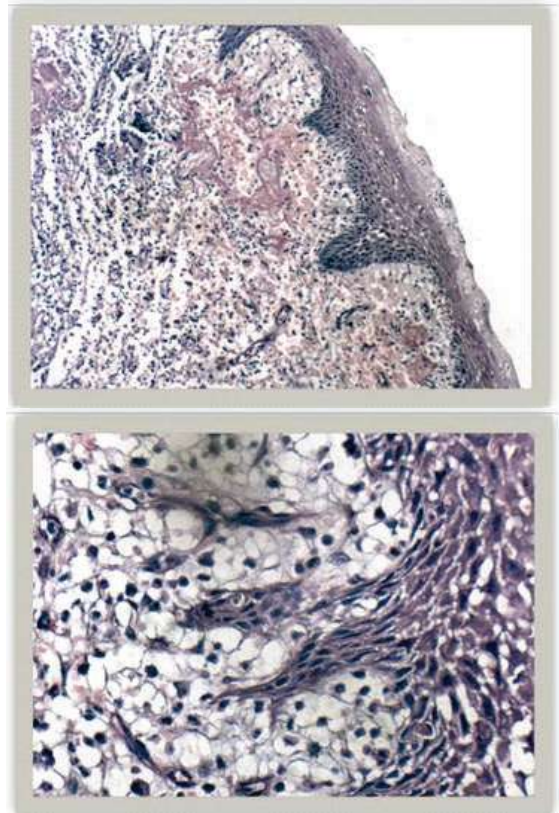


Figure 9: Microscopic Examination: Given H&E-stained section shows ameloblastic epithelium with fibrous cyst wall. Ameloblastic epithelium shows tall columnar ameloblastlike cells with hyperchromatic nucleus. The cells overlying epithelium are stellate reticulum like star shaped.

On palpation, Non tender, soft below 45,46 region, firm in surrounding area. Egg shell crackeling was present on palpation. Lesion was fluctuant in the center, fixed with underlying tissue. No discharge was present. Lymphnode was not palpated.

On Radiographic examination, in orthopantogram, Well defined multi locular radiolucency from 31 to Distal of 47 anterior posteriorly. Sup-inferiorly alveolar crest to lower border of mandible. Internal structure is radiolucent , having locules& septa creating compartments. Ant small size locales while posteriorly large size showing soap bubble appearance. Mandibular canal is appreciated till distal of 48. In Periphery, Having corticated borders, Curved at alveolar crest side and scalloped

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at lower border causing thinning of cortex. The effect on surrounding structure, Knife edge root resorption present I.r.t 45,46,47. Displacement i.r.t 45,46

In cone beam computed tomography, coronaly it shows destruction of buccal cortex and thinning of lingual cortex. Sagittaly, Expansion & thinning of lower border as well as mandibular canal is traceable in some area. Axially, shows bucco-lingual expansion of the lesion, where more of buccal expansion can be seen.

Provisional diagnosis is Ameloblastoma and differential diagnosis is Odontogenic Keratocyst, Lateral periodontal cyst and traumatic bone cyst. Final Diagnosis is Unicystic Ameloblastoma. For treating it, Enucleation is performed.

DISCUSSION

Unicystic Ameloblastoma(UA) is a rare benign odontogenic tumour commonly occurring in the 2nd - 3rd decades of life and has recurrence rate of less than 25%.⁽¹⁾ in our cases 1st patient was 22 years old and 2nd patient was 16 years old. The patients were female where literature suggest no sex prediction.

The complaint of both the patients were swelling & the lesions were asymptomatic. In the 2nd case associated symptoms were mobility, root resorption & displacement of tooth. Commonly associated symptoms literature shows painless swelling with expansion of cortical plates, unilocular lesion with sclerotic borders, impacted teeth, displacement, tipping, external root resorption, and rootdivergence. Involvement of the mandibular posterior region is the commonest site, in the 1st case lesion was involving ramus and in the 2nd case lesion was involving body of the

mandible.⁽⁶⁾

The radiographic appearance of Unicystic Ameloblastoma is peculiar, with a circumscribed radiolucency . It appears as a single cyst-like, unilocular radiolucency in the mandibular posterior region, as present in 1st case. However, it may also appear as a multilocular radiolucency, as present in 2nd case.⁽⁵⁾ The commonly investigations advised are OPG, CBCT , FNAC & BIOPSY is the gold standard

The differential diagnosis pertaining to the present cases include , Odontogenic keratocyst, Dentigerous cyst, Traumatic bone cyst and lateral periodontal cyst . Odontogenic keratocysts does not cause cortical expansion, as seen both the cases. Dentigerous cyst is associated with an unerupted tooth in 1st case extraction of impacted 38 done & in 2nd case lesion was extending till mesial aspect of 48and not assotiated with impacted teet. Traumatic bone cysts have scalloping border and intact lamina dura seen. Lateral periodontal cyst is small in diameter seen in between two roots of the teeth, does not cause root resorption as present in 2nd case.

1st case is having intraluminal type of unicystic ameloblastoma. Based on the clinicopathologic study performed in 57 cases of UA, Ackermann classified this entity into 3 histologic groups, namely, luminal, intraluminal/plexiform, and mural.[3,8] In another histologic subgrouping (modified after Ackermann et al.) by Philipson and Reichart, UA was classified into 4 subtypes, namely, subtype 1 luminal UA, subtype 1.2 luminal and intraluminal UA, subtype 1.2.3 luminal, intraluminal, and intramural UA, and Subtype 1.3 luminal and intramural UA.⁽⁴⁾

Here in both these cases enucleation has been

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performed, there are different treatment modalities for UA which include enucleation, curettage, marsupialization and marginal resection. The present scientific literature recommends that UA should be treated in a conservative manner (enucleation, curettage, and marsupialization) when compared to solid or multicystic ameloblastomas on account of their less aggressive biological behavior. The response of the UA to conventional treatment is favorable. Subtypes of UA 1 and 1.2 can be treated conservatively (careful surgical enucleation), whereas subtypes 1.2.3 and 1.3 should be treated aggressively (radical resection).⁽³⁾

Because UA is cystic, well-localized and surrounded by fibrous capsule, the prognosis is good. However, when it breaches the periphery of the fibrous capsule it behaves more aggressively. Because the recurrence of UA is long delayed, long-term postoperative follow-up is required.⁽³⁾ Unicystic Ameloblastoma (UA) although less aggressive than multicystic ameloblastoma, should be included in the differential diagnosis of swellings of maxillofacial region.

CONCLUSION

In most cases, UA has been reported to cause jaw expansion of cortical plates and dentists should familiarize themselves with such cases to prevent their under-diagnosis in clinical practice. Complete clinical, radiological, and histological examination contributes to the final and prompt diagnosis of the lesion. Although conservative treatment is preferred for patients, long-term follow-up of patients is mandatory to prevent recurrence of lesion and for better prognosis.

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