

Unicystic Ameloblastoma: A Quandary Diagnosis

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Abstract: - Ameloblastoma is a tumour of odontogenic origin that can occur on the mandible especially in the posterior region classified as a benign type of tumour. It can surface from the tooth germ ectodermal epithelium, Odontogenic cyst epithelium, stratified squamous epithelium as well as the enamel organ epithelium. Typically, it is the 2nd most common variant of all odontogenic tumours. They represent about 1% of all jaw tumours. Among the variants of ameloblastoma, Unicystic ameloblastoma is considered the least aggressive. Unicystic ameloblastoma also comprises subtypes namely - mural, luminal and intraluminal subtypes out of which the mural subtype is taken under review with special consideration.

Keywords:- Benign Tumor, Odontogenic Cyst, Unicystic Ameloblastoma, Mural Subtype

I. INTRODUCTION

Ameloblastoma is a true neoplasm of enamel organ type tissue that does not undergo differentiation in the sense that enamel development does not occur. Robinson depicted it as unicentric and nonfunctional yet intermittent in growth but persistent when observed clinically.¹

Unicystic ameloblastoma is an odontogenic tumour variant that mimics a cyst clinically and radiographically to the point where grossly it also echoes cystic elements. However, histologically, it showcases a cystic cavity lining with ameloblastic epithelium. The lining epithelium can be accompanied with or without luminal and/or mural tumour growth.² Unicystic ameloblastoma lesions originate from reduced enamel epithelium or a dentigerous cysts or from solid ameloblastoma lesions that endure cystic degeneration.¹ Clinical signs are primarily asymptomatic but can introduce a swelling or enlargement on the jaw that is conventionally painless accompanied with a resultant facial asymmetry. Radiologically, a common predominant observation noted is a unilocular radiolucency; however, multilocular appearances may also be observed sporadically.³

Unicystic ameloblastoma was initially considered as a unique and separate entity by Robinson and Martinez, representing 10-15% of all ameloblastomas having an almost equal male to female distribution upon occurrence commonly occurring between the demographic group of 20 - 30 years of life.⁴

II. CASE REPORT

A 40-year-old male patient arrived with a chief complaint of pain inside the mouth in the lower left back tooth region for about 3 months. On acquiring his history of events it was revealed that the pain was sudden in onset, dull aching, intermittent, moderate in intensity, which radiates to the left side of face, aggravates on having food and relieves on its own. The pain was associated with a swelling for 2 months which was initially small in size and progressed steadily to observed size. There was also a prior history of intermittent low-grade fever over the same period. Patient noticed fluid discharge from the affected region which was of cream colour with blood.

History of tobacco chewing for 12 years twice daily for about 10 -15 minutes & keeps it on the right and left back tooth region.



Fig 1: Extraoral Examination of Patient Displaying Slight Lower 3rd Left Asymmetry

On extraoral examination, facial asymmetry [Figure.1] noted on the left lower third of face with mild and diffuse of size approximately (4×2) cm extending superiorly-inferiorly from an imaginary line joining corner of mouth and tragus of ear to the inferior border of the mandible with indistinct borders.

On palpation consistency appears to be soft. A sinus tract opening was noted 0.5 cm below the inferior border of the mandible in the left Parasymphysis region [Figure.2]. Pus admixed with blood discharge noted.



Fig 2: Sinus Tract Opening Observed towards the Region Below Inferior Border of Mandible

On intraoral examination, a solitary diffuse swelling was noted on the left mandibular vestibular region extending from to 31 to 36 regions of size approximately (5× 2 cm) [Figure.3], with indistinct borders, overlying mucosa and surrounding mucosa appearing normal. Buccolingual expansion noted from 34 to 36 regions [Figure.4].

On palpation it appears tender and the consistency is soft. Grade I mobility noted with respect to 34,35,36. No decortication noted, no discharge was noted [Figure.5].



Fig 3: Display of Solitary Diffuse Swelling from Region of 31 - 36



Fig 4: Buccolingual Expansion Noted from Region of 34 – 36



Fig 5: Confirmation of Findings on Palpation Including Soft Consistency

Radiographic investigations involved an orthopantomogram [Figure.6] which revealed a multilocular well-defined radiolucent lesion of size (6×3) cm noted on the mandible extending from mesial aspect of 45 to mesial aspect of 37. Impacted 15, 13, 23, 38, 32, 48, retained deciduous 53 and missing 41.[Figure.7] A well-defined radiopaque lesion was noted at the right angle of mandible of size (5×2) cm approximately with well-defined borders, *suggestive of Sialolith* [Figure.8]



Fig 6: Orthopantomogram Reveal of Multilocular Lesion on Mandible from Region of 45 to 37



Fig 7: Radiographic view Indicating Missing 32 and 41 and Impacted 38



Fig 8: Radiographic View Indicative of Sialolith on the Right Angle of the Mandible

CBCT [Figure.9] revealed a multilocular radiolucent lesion noted on the mandible extending from mesial aspect of 45 to mesial aspect of 37 region [Figure.11]. Buccolingual and labial cortical plate expansion is noted along with destruction of lingual and labial cortical plates [Figure.10], additionally root resorption is noted wrt 34 [Figure.12].

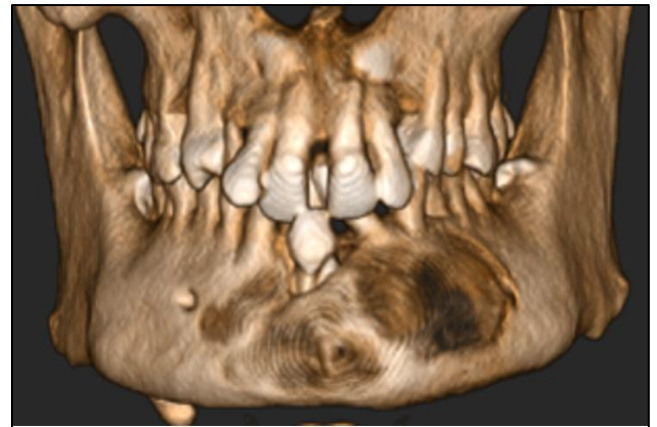


Fig 9: CBCT View of Multilocular Lesion on Mandible Extending from Region of 45 to 37

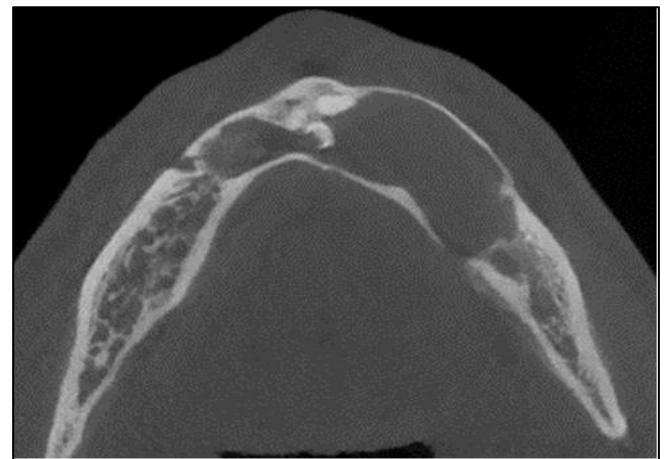


Fig 10: Buccolingual and Labial Cortical Plate Expansion Noted along with Destruction of Lingual and Labial Cortical Plates

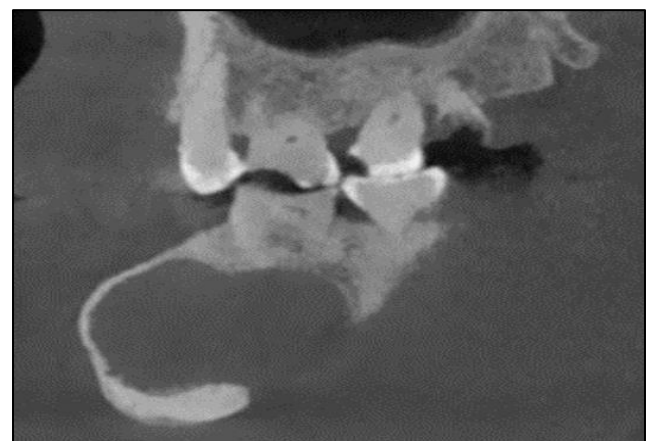


Fig 11: Multilocular Radiolucencies Noted Across Quadrants

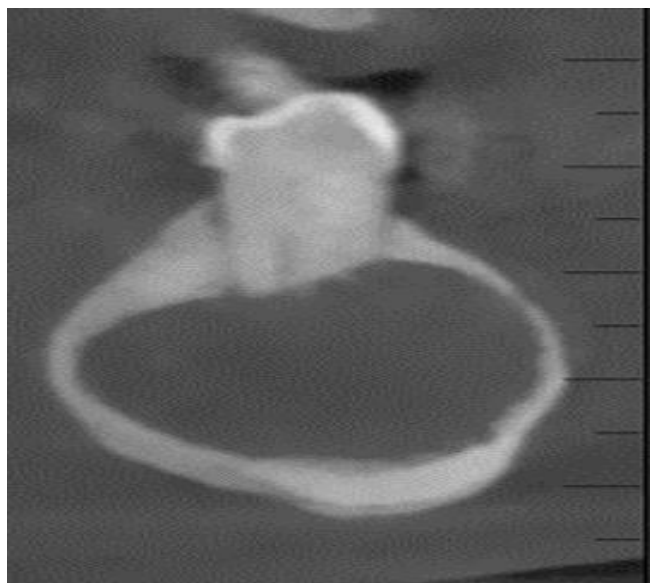


Fig 12: Radiographic View of Root Resorption with Regards to 34

An incisional bone biopsy on histopathological examination exhibited a lumen lined by non-specific odontogenic epithelium. Hyalinized fibrosis and cellular connective tissue was also appreciated, suggestive of Aggressive odontogenic cyst. [Figure.13].

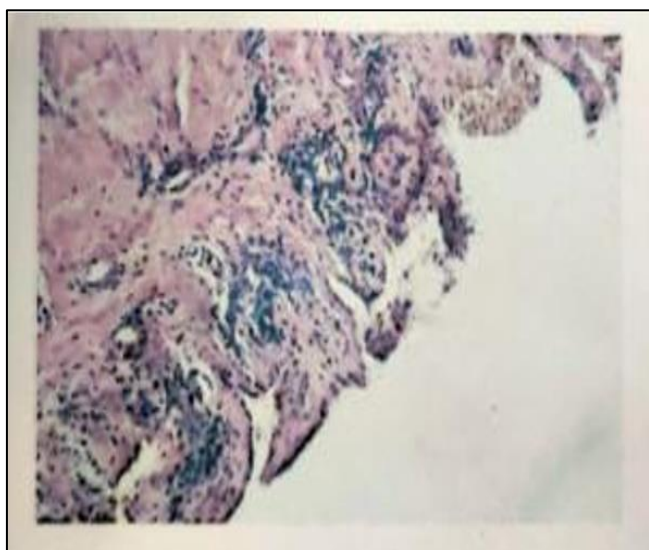


Fig 13: Histological Presentation of Hyalinized Fibrosis and Cellular Connective Tissue

The patient was planned for a mandibulectomy with free fibula reconstruction and removal of right and left submandibular gland.

Grossly, a cut section across the resected specimen of bone revealed a unicystic lesion. Histopathology on excisional biopsy revealed, cyst wall lined with a palisading basal layer with nuclei and stellate reticulum. A few smaller cysts with denuded epithelial lining and areas of squamous metaplasia with focal keratinization. Correlating the clinical, radiographic, gross, and histopathology features, the lesion was concluded as Unicystic ameloblastoma of mural type.

III. DISCUSSION

Unicystic ameloblastoma subgroups that are associated with mural components are often assessed as the most aggressive type. They represent 10%–15% of ameloblastomas and commonly occur between the 2nd to 3rd decade in terms of years of age with no clear bias for an exclusive gender.

If the lesions are associated with an impacted tooth they will frequently be addressed as the dentigerous type with the ones that aren't associated with impactions referred to as non dentigerous types. The most typical spot of manifestation is usually the posterior portion of the mandible.⁵ This was the situation in this case where facial asymmetry was observed due to a swelling formed on the lower left third, extraorally. The mural variant being the most aggressive has to be treated with utmost concern with other variants of ameloblastoma being open towards a more conservative approach.

Histopathology of the present case on incisional biopsy presented a lumen lined by non-specific odontogenic epithelium along with hyalinized fibrosis and cellular connective tissue. The cyst wall would be lined with a palisading basal layer with nuclei and stellate reticulum on excision biopsy reveal.

In order to diagnose a lesion as unicystic ameloblastoma through its histological characteristics its histologic criteria of identification is invested by the observation of a cystic structure lined by ameloblastic epithelium with a columnar basal layer, subnuclear vacuoles, reverse polarity of hyperchromatic nucleus, and a thin layer of oedematous, degenerating stellate reticulum-like cells on the surface.⁶

Radiologically, they can be diversified into two main patterns: unilocular and multilocular with a habitual affinity towards the unilocular pattern. Eversole *et al.*, identified predominant radiographic patterns for Unicystic ameloblastoma: Unilocular, scalloped, macro multilocular, pericoronal, interradicular, or periapical expansile radiolucencies.⁷ Our current case displayed a rather multilocular pattern with bone plate expansion also being noted. The aggressiveness of the lesion can also be determined through any resorptions that might be noted; in the present case as observed with regards to the tooth 34.

Unicystic ameloblastoma can be managed using varied methods like enucleation, marsupialization, resection just to name a few. Certain treatment protocols observe a varied recurrence rate.

A recurrence rate of 3.6% is observed with the resection method which is significantly lower in comparison to a moderately high of 30.5% seen with procedures deploying enucleation.⁵ The mural variant, however, has recurrence rates similar to the conventional types if treatment is limited to enucleation and curettage. When the tumor manages to invade the cystic wall, it is in principle believed to have a solid component which overshadows the cystic component with respect to treatment planning.⁸

IV. CONCLUSION

The diagnosis of the lesion was based on the correlation of the observations noted based on the clinical, radiographic, gross, histopathology features, after which the lesion was determined to be a Unicystic ameloblastoma of mural type. Unicystic Ameloblastoma is a lesion with a strong inclination towards recurrence. In the event of adjacent tissue penetration from the cyst wall, there is a strong possibility of recurrence. In order to prioritize effective long term treatment it becomes necessary for careful postoperative analysis to rule out any chances of recurrence.

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