

# Unicystic Ameloblastoma Mimicking Radicular Cyst

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## Abstract

Ameloblastoma is a slow-growing, persistent and locally aggressive neoplasm of epithelial origin with frequent occurrence in the mandible. The unicystic ameloblastoma (UA) is a less encountered variant of ameloblastoma, the clinical and radiographic presentation of which mimics a jaw cyst. Histologic examination shows a typical ameloblastomatous epithelium lining part of the cyst cavity with or without luminal and/or mural tumor proliferation. We present a case report of luminal variant of UA mimicking radicular cyst with a purpose to emphasize the significance of histopathological examination when clinical and radiologic suggestions may be misleading.

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## Introduction

This report presents a case of luminal variant of unicystic ameloblastoma (UA), which mimicked a radicular cyst. The UA a less encountered variant of ameloblastoma accounting for 5-15% of all ameloblastomas<sup>[1]</sup>. Clinically and radiographically, it shows the characteristics of a jaw cyst but histologic examination shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation<sup>[2]</sup>. This lesion is clinically less aggressive variant of ameloblastoma with a variable recurrence rate<sup>[1,3]</sup>. It presents three histopathologic subtypes – luminal, intraluminal, and mural<sup>[3]</sup>. The aim of this case report of UA is to emphasize the significance of histopathological findings to a histo-pathologist in absence of accompanying clinical and radiological records, and when clinical and radiological findings are innocuous and specious.

## Case Presentation

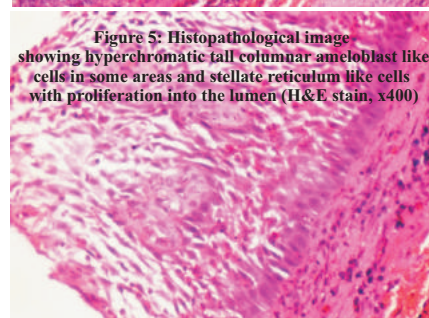
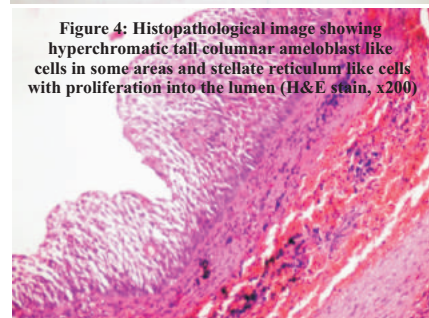
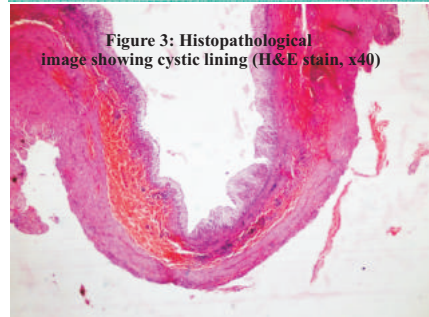
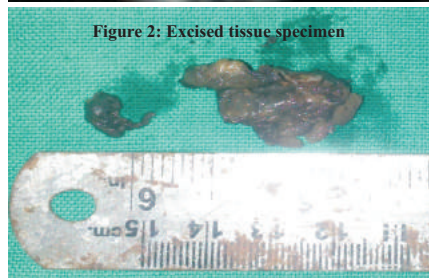
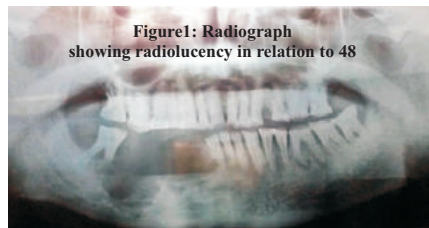
A female patient aged 48 years, presented with the complaint of pain and swelling in the right lower back tooth region since 2 months. Patient was apparently asymptomatic 2 months back. She noticed swelling in the mandibular right posterior tooth region, which increased in size associated with pain that subsided on taking medication. Intraoral examination revealed a swelling in relation to 48 with occlusal caries, which was tender on percussion. The swelling was round and hard in consistency. Radiographic examination showed a unilocular, well-circumscribed radiolucency in relation to 48. [Fig 1]. A provisional diagnosis of radicular cyst was made.

The gross examination of the biopsy specimen revealed a single soft tissue measuring 11 x 20 x 5 mm in size, irregular in shape, brownish black in colour and firm in consistency [Fig 2] and tooth 48 with occlusal caries.

Histopathological examination showed, under scanner view, cystic lining enclosing connective tissue stroma [Fig 3]. On higher magnification, the cystic lining showed hyperchromatic tall columnar ameloblast like cells in some areas and stellate reticulum like cells with proliferation into the lumen [Fig 4, 5].

The juxtaepithelial connective tissue stroma showed condensed collagen fibres, fibroblasts, mild inflammatory infiltrate, endothelial lined blood vessels and extravasated RBCs. A final diagnosis of luminal variant of unicystic ameloblastoma has been made.

(Figure 1-5)



## Discussion

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm with variable clinical expression accounting for 1% of all cysts/tumors of jaws and 18% of all odontogenic neoplasms. It is typically slow growing, locally aggressive and rarely metastasizes<sup>[4]</sup>. It has tendency to spread by infiltration through the medullary spaces in deep extension and a high rate of recurrence (55-90%) if not removed adequately<sup>[4,5]</sup>. Although this behavior is more common in the multicystic pattern, some unicystic lesions can present a similar biological profile<sup>[1]</sup>.

The UA, a subtype of intraosseous ameloblastoma, has long been delineated as a prognostically distinct entity<sup>[6]</sup>. It usually occurs in younger patients, predominantly in the second to third decade of life<sup>[6]</sup>. A slight male predilection (male to female ratio 1.6:1) has been reported. But in case of no associated unerupted tooth, male to female ratio is 1:1.8<sup>[7]</sup>.

Posterior mandibular region is most commonly involved (mandible to maxilla ratio being 13.1:3), with a large percentage of the lesions (>90%) in the third molar region<sup>[6,8]</sup> followed by the parasymphysis region, the anterior maxilla, and the posterior maxilla<sup>[8]</sup>. 50-80% of cases are associated with an impacted tooth and clinical and radiographic findings suggest the lesion to be an odontogenic cyst, particularly dentigerous cyst. Those not associated with impacted teeth are called nondentigerous variant<sup>[6,4]</sup>.

The UA is usually asymptomatic, although a large lesion may cause painless swelling of the jaws with facial asymmetry<sup>[9,10]</sup>. Mucosal ulceration is rare but may be caused by continued growth of the tumor. Small lesions are more often detected on radiographic examination for other cause or as a result of other symptoms such as tooth mobility, occlusal alterations and failure of eruption of teeth produced by the tumor<sup>[11]</sup>. In the present case, carious tooth, pain, swelling and radiologic features suggested a radicular cyst

Only histological examination can provide a definitive diagnosis of UA<sup>[4]</sup>. For a lesion to be diagnosed histologically as UA, demonstration of odontogenic epithelium, often in focal areas, lining a single cystic sac is necessary. UA should be differentiated from odontogenic cysts because the recurrence of UA is higher<sup>[12]</sup>. Ackermann classified this entity into the following three histologic groups<sup>[13]</sup>

**Group I:** Luminal UA (tumor confined to the luminal surface of the cyst)



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**Group II:** Intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall), and

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

Another histologic subgrouping by Philipsen and Reichart has also been described<sup>[14]</sup>:

Subgroup I: Luminal UA

Subgroup 1.2: Luminal and intraluminal

Subgroup 1.2.3: Luminal, intraluminal and intra-mural

Subgroup 1.3: Luminal and intramural

Treatment planning depends on the histological type of UA. Subgroups 1 and 1.2 UAs may be treated conservatively (careful enucleation), whereas Subgroups 1.2.3 and 1.3 should be treated aggressively<sup>[7]</sup>. The histological typing of the current case was 1.2 and the lesion was treated conservatively with careful enucleation. The recurrence rate for UAs, after conservative surgical treatment (curettage or enucleation), is generally reported to be 10–20% and on average, less than 25%<sup>[7,15]</sup>. The patient has been on follow-up and there is no report of recurrence till date.

**Conclusion**

The confirmatory diagnosis of UA can only be made by means of histological examination and it cannot be predicted preoperatively on clinical or radiographic grounds. As it may be locally aggressive and it has the potential for recurrence, it must be treated carefully with long term follow-up of the patient. Clinical and radiological findings provide important clue and guidance but the final diagnosis always rests upon histopathological examination.

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