

## Case Report

### Unicystic Mural Ameloblastoma – An Unusual Case Report and Review of the Literature

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#### Abstract

Unicystic ameloblastoma show clinical, radiographic features of a jaw cyst and histologic features of ameloblastoma lining the cyst cavity, with or without luminal and/or mural tumor proliferation. UA is usually less aggressive with distinctly lower recurrence rate than conventional ameloblastomas.

Key words: Unicystic Ameloblastoma, Odontogenic tumor, Neoplasm.

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#### Introduction

Ameloblastoma is an odontogenic tumor with four distinct forms, solid/multicystic, peripheral, unicystic and desmoplastic.<sup>1</sup> Robinson and Martinez were first to describe Unicystic ameloblastoma (UA) as a separate entity in 1977.<sup>2</sup> Ackermann et al suggested to use the term UA to ameloblastomas with a single epithelial lined cystic cavity. According to them, UA histologically is categorized into three types, luminal, intraluminal and mural.<sup>3</sup> It makes about 10 to 15 percent of all intraosseous ameloblastomas.<sup>4</sup> It is chiefly

seen in younger individuals, almost exclusively in posterior mandible. Generally UA is asymptomatic, while larger lesions may cause a painless swelling of the jaws.<sup>5</sup> Radiologically UA represents a well-circumscribed radiolucency surrounding the crown of a unerupted third molar, resembling a dentigerous cyst. When compared to solid ameloblastomas, UA is less aggressive with lower recurrence rate.<sup>6</sup>

Here we describe a case report of UA in a 24 year male and its review of literature.

### Case Report

A 24 year old male patient reported to the department of oral medicine and radiology with a chief complaint of swelling on right side of face since 2 ½ years. The swelling was initially small in size and gradually increased in size to attain the present size. The patient consulted a local dentist for the above problem and the swelling did not subside on medication. On examination a solitary diffuse swelling was seen on right side of middle 3<sup>rd</sup> of the face, measuring approximately 8x8 cms, extending from infra orbital region to lower border of mandible; anterioposteriorly from nasolabial fold to tragus of ear. (Figure 1)



**Figure 1:** Extra Oral photograph showing diffuse swelling was seen on right side of middle 3<sup>rd</sup> of the face

There were no signs of ulceration or pus discharge. The swelling was bony hard in consistency. Intra oral examination revealed a solitary swelling in lower right retro molar area, approximately 5x5 cms extending from mesial surface of 47 to retro molar area. Both buccal and lingual cortical plate expansion was present. (Figure 2)

Radiologically a circumscribed unilocular radiolucency was seen surrounding unerupted right mandibular 3<sup>rd</sup> molar. (Figure 3) Surgical enucleation of the



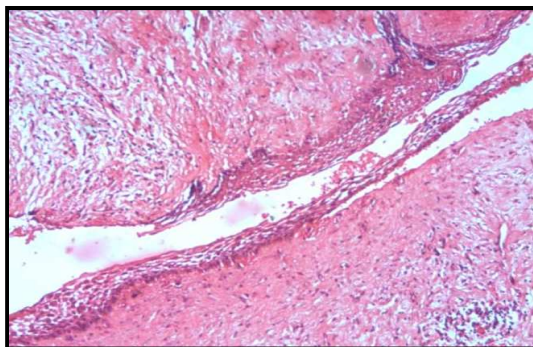
**Figure 2:** Intra oral examination revealing a solitary swelling in lower right retro molar area extending from mesial surface of 47 to retro molar area.



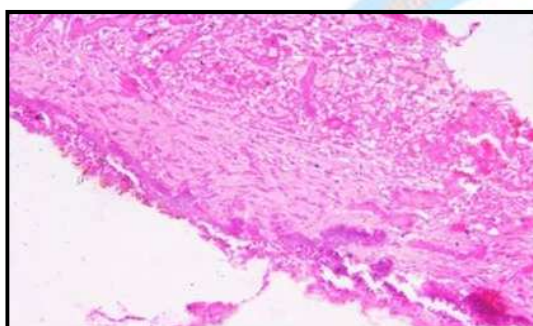
**Figure 2:** Orthopantomograph showing unilocular radiolucency surrounding unerupted right mandibular 3<sup>rd</sup> molar

tumor was done and the specimen was sent for histopathologic examination. Gross examination of the specimen revealed two irregular soft tissue specimen appearing as cystic lining was received, 1x2cm in size, firm and cream in color. Haematoxylin and eosin sections revealed a well defined cystic lumen bordered by odontogenic epithelial lining overlying a delicate to dense connective tissue stroma. Cystic epithelium revealed basal columnar to cuboidal cells with hyperchromatic nuclei and superficial loosely arranged stellate reticulum-like cells. Underlying delicate connective tissue stroma showed thin collagen fiber bundles arranged haphazardly with mild inflammatory component and few blood vessels. Focal isolated islands of odontogenic epithelium

were also evident in the connective tissue stroma containing peripheral columnar to cuboidal cells and central stellate reticulum-like cells. (Figure 4) Budding was evident from the epithelial lining into the connective tissue. (Figure 5)



**Figure 4:** Photomicrograph showing cystic epithelium and superficial loosely arranged stellate reticulum-like cells. (H& E; 10 X)



**Figure 5:** Photomicrograph showing budding from the epithelial lining into the connective tissue.

A diagnosis of UA (mural type) of right side of mandible was made. Post operative follow up for a period of one year was uneventful.

### Discussion

After extensive studies now UA is designated as a separate entity and has clinical and radiological features of cysts and histological features of ameloblastoma in their epithelial lining of cystic lumen.<sup>7</sup> Most of the cases are seen in males with aratio of 1.3:1, our patient was a male.

Generally UA is seen in younger individuals with about 50% in the second decade of life.<sup>8</sup> Most of the cases are seen in posterior mandible and associated with impacted molars as in our case.<sup>9</sup> In most of the cases, it appears as a well-circumscribed radiolucency that surrounds the crown of an unerupted third molar, resembling a dentigerous cyst.<sup>10</sup>

Ackermann et al, classified this entity into 3 histologic groups:<sup>11, 12</sup>

**Group 1:** Luminal UA, where epithelium in some areas shows ameloblastic transformation without infiltration into the connective tissue wall.

**Group 2:** Intraluminal/plexiform UA, where the lining epithelium shows a nodular proliferation of plexiform ameloblastoma into the lumen without infiltration of tumor cells into the connective tissue wall.

**Group 3:** Mural UA, where invasive islands of ameloblastomatous epithelium in the connective tissue wall are seen. Our case falls in this category.

The differential diagnosis includes dentigerous cysts, odontogenic keratocyst, residual cysts, adenomatoid odontogenic tumor.<sup>13</sup> Treatment of UA depends on the histological subtype. In luminal and intraluminal types, enucleation is done, whereas in mural type, bony resection is generally carried out.<sup>14, 15</sup>

### Conclusion

UA has specific clinical, radiologic and histological features when compared to solid ameloblastoma. To understand UA more number of cases should be followed for a longer period. UA should be included in the differential diagnosis in any lesion ranging from simple abscess to cysts to fibro-osseous lesions/neoplastic growths in posterior mandibular regions.

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