

## UNICYSTIC AMELOBLASTOMA OF THE MANDIBLE- A CASE REPORT WITH HEMIMANDIBULECTOMY

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**ABSTRACT:** Ameloblastoma is a true neoplasm of odontogenic epithelial origin. Its incidence, combined with its clinical behavior, makes ameloblastoma the most significant odontogenic neoplasm. Unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation. As this tumor shows considerable similarities with dentigerous cysts, both clinically and radiographically the biologic behaviour of this tumor group was reviewed. Unicystic type has a considerably better overall prognosis and a much reduced incidence of recurrence compared with conventional ameloblastoma.

**KEYWORDS:** Ameloblastoma, Unicystic, odontogenic, dentigerous, Hemimandibulectomy.

### INTRODUCTION

Odontogenic tumors represent a spectrum of lesions ranging from malignant (rare) and benign neoplasms to dental hamartomas, all arising from odontogenic epithelia and/or ectomesenchyme with variable amount of dental hard tissues. The most common tumor of odontogenic origin is ameloblastoma, which develops from epithelial cellular elements and dental tissues in their various phases of development. It is a slow-growing, persistent, and locally aggressive neoplasm of epithelial origin. Its peak incidence is in the 3rd to 4th decades of life and has an equal sex distribution. It is often associated with an unerupted third molar. The vast majority of ameloblastomas arise in the mandible, and the majority of these are found in the angle and ramus region. There are three forms of ameloblastomas, namely multicystic, peripheral (extra osseous) and unicystic tumors. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cyst. The goal of treatment of ameloblastoma is to achieve complete excision and appropriate reconstruction

#### Case report

A 16 year old male patient reported to the Department of Oral and Maxillofacial Surgery with a swelling on right side of lower jaw since 3 years and there was no associated pain, difficulty in opening the mouth and chewing.

On extra oral examination a well circumscribed, hard, non tender swelling of size 7cm x 4cm is present on the right side of face, arising from the right side of the mandible, involving the condyle, coronoid process, ramus, angle and body upto the right lower lateral incisor tooth. On intra oral examination, on inspection, no swelling was noticed and entire mucosa was normal. On palpation, there is expansion of buccal and lingual cortical plates extending from the body and ascending ramus on right side of mandible. (**Fig.1**)

The patient was sent for routine blood investigations which were found to be normal. Radiographic examination revealed a well defined unilocular radiolucency with an impacted 3<sup>rd</sup> molar tooth extending from condyle, coronoid process upto lateral incisor region, involving lower border of mandible. Root resorption was noticed in the teeth related to the tumor. (**Fig.2**)

On aspiration, light yellow color fluid was noticed. Incisional biopsy was performed and sent for histopathological examination (**Fig.3**). Histopathological examination revealed fibrous connective tissue capsule with odontogenic epithelium. Odontogenic epithelium exhibits basal cuboidal cells with hyperchromatic nucleus. Over the basal cells, stellate reticulum like cells are vident. Epithelial mesenchymal interface is flat. Connective tissue



Fig.1. Preoperative photographs

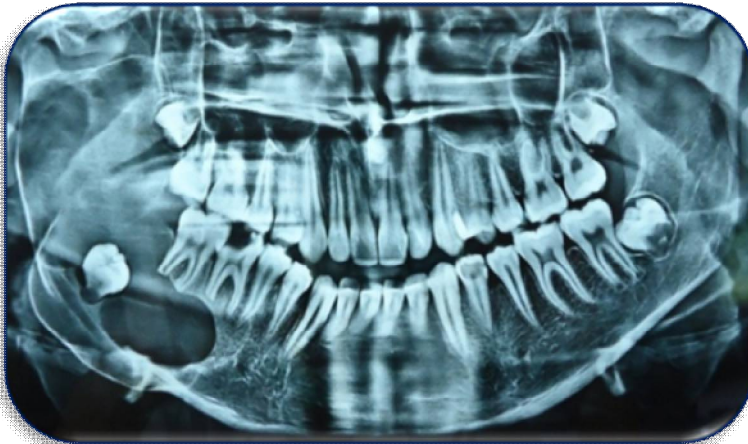


Fig.2. Orthopantomograph showing cystic lesion



Fig.3. Incisional Biopsy

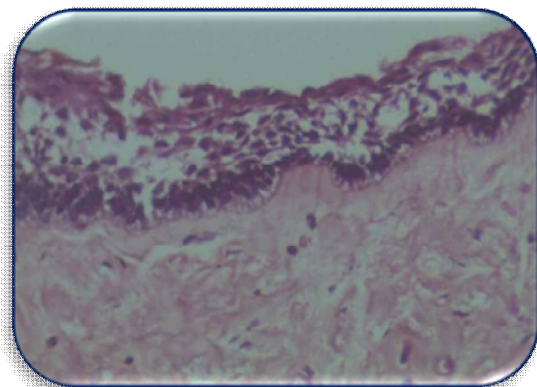
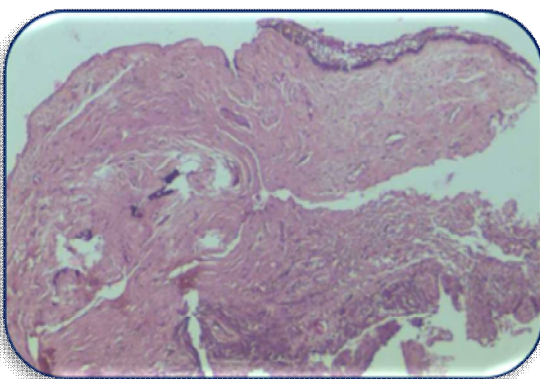


Fig.4. Photomicrographic picture.

stroma exhibits mild chronic inflammatory cell infiltration predominantly lymphocytes and plasma cells suggestive of **unicystic ameloblastoma. (Fig.4).**

The patient was taken up for surgery, planned for right hemimandibulectomy under general anesthesia. Lower lip split incision was given in the midline. Tumor for resection was approached from the labial and buccal side and further the intra oral incision was extended from the retromolar to vertical ramus and careful lingual dissection was performed to separate the tumor from adjacent tissues. **(Fig.5).** Osteotomy cuts are given between lateral and central incisor region on right side of mandible. The entire tumor was removed in pieces.**(Fig.6).** Reconstruction plate was used to maintain the contour of mandible.**(Fig.7).** Wound was closed in layers. Utmost care was taken to suture the midline split of the lower lip to avoid scar formation. Primary closure was achieved. **(Fig.8).** Healing was normal and recovery was uneventful**(Fig.9).**

### Discussion

Unicystic ameloblastoma, a variant of ameloblastoma first described by Robinson and Martinez in 1977<sup>1,3,8</sup>, refers to those cystic lesions that show clinical and radiologic characteristics of an odontogenic cyst but in histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation<sup>9</sup>. Unicystic ameloblastoma accounts for 10-15% of all extraosseous ameloblastomas in various studies<sup>1, 2,10</sup>.

It is essential to distinguish between the three clinical types of ameloblastomas<sup>7</sup>, the intraosseous solid or multicystic lesion; the well-circumscribed unicystic type; and the rare peripheral extraosseous ameloblastoma, because they require different forms of treatment.

Unicystic ameloblastomas are most often seen in young patients<sup>2</sup>, with about 50% of such tumors diagnosed during the second decade of life<sup>4</sup>. More than 90% of unicystic ameloblastomas are found in the mandible, usually in the posterior region<sup>3</sup>. The ratio of mandibular to maxillary unicystic ameloblastoma has been reported to be 13:1<sup>(2)</sup>. The lesion is often asymptomatic, although a large lesion may cause painless swelling of the jaws, facial asymmetry, unilocular lesion with defined sclerotic borders, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference, and extrusion of tooth<sup>2</sup>.

In the present case, radiologic presentation suggested a dentigerous cyst appearance with an impacted third molar, which is pushed to one side, and root resorption is noticed in the teeth related to the tumor, indicating the aggressive nature of the lesion.

Histologically, the minimum criteria for diagnosing a lesion as unicystic ameloblastoma is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas.

In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann<sup>1</sup> classified this entity into three histologic groups:

- Luminal unicystic ameloblastoma (tumor confined to the luminal surface of the cyst)
- Intraluminal/Plexiform unicystic ameloblastoma (nodular proliferation into lumen without infiltration of tumor cells into connective tissue wall)
- Mural unicystic ameloblastoma (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium).

Another subgrouping by Philipsen and Reichart<sup>1, (4)</sup> has also been described as follows:

**Subgroup 1:** luminal unicystic ameloblastoma

**Subgroup 1.2:** luminal and intraluminal

**Subgroup 1.2.3:** luminal, intraluminal and intramural

**Subgroup 1.3:** luminal and intramural

The diagnosed unicystic ameloblastoma as subgroups 1 and 1.2 can be treated conservatively, whereas subgroups 1.2.3 and 1.3 showing intramural growths require radical resection<sup>6</sup>.

Unicystic ameloblastoma shares common clinical and radiographical manifestations with other odontogenic lesions making diagnosis difficult<sup>2</sup>. Dentigerous cyst, odontogenic keratocyst, residual cyst, adenomatoid odontogenic tumor, giant cell lesion, and sometimes solid ameloblastoma can be the possible differential diagnosis for unicystic ameloblastoma.

Great difficulty exists in differentiating dentigerous cyst from unicystic ameloblastoma. However, following manifestations favors unicystic ameloblastoma: unilocular cystic lesion extending into the ramus, expansion of both the buccal and lingual cortex (tumor usually grows buccally and lingually, whereas the cyst grows toward most dependent part, i.e. buccally)<sup>2</sup>.

Choice of treatment has to be considered in conjunction with other clinical and pathologic factors such as the size, location and growth pattern of the tumor<sup>5</sup>. Whatever surgical approach the surgeon decides to take, long-term follow-up is mandatory, as recurrence of unicystic ameloblastoma may be long delayed.

The treatment modalities of ameloblastomas according to Bataineh<sup>7</sup> can be broadly classified as conservative and radical. Feinberg and Steinberg mentioned enucleation and curettage as conservative approaches.

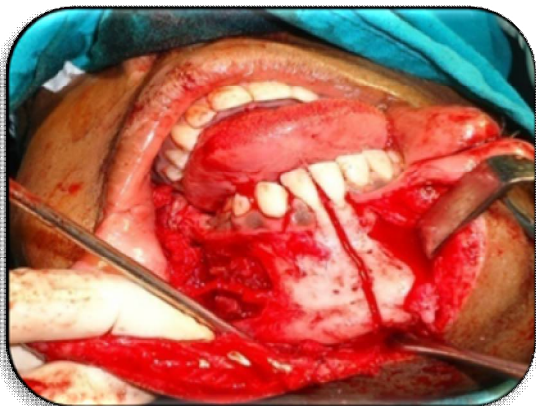


Fig. 5. lip split approach



Fig.6. Resected specimen

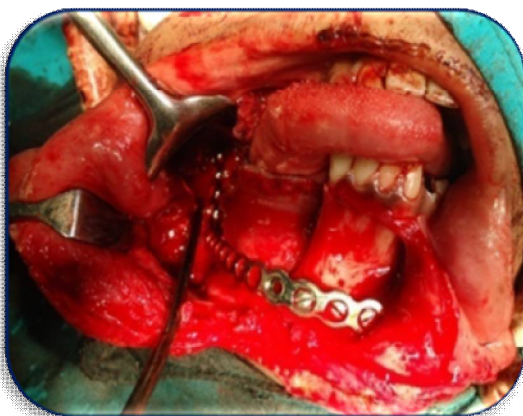


Fig. 7. reconstruction



Fig.8.primary closure

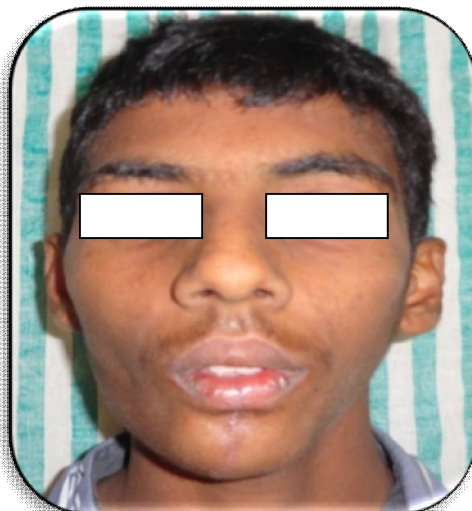


Fig.9.post operative photograph



Fig.10.Post operative orthopantomograph

Gardner and Pecak<sup>7</sup> stressed the fact that unicystic ameloblastoma, in which the tumor has proliferated into the lumen, with fibrous connective tissue wall completely surrounding the tumor can be expected to be cured by enucleation, but when the connective tissue wall is involved, should be treated by marginal resection.

Feinberg and Steinberg<sup>7</sup> suggested that the tumor should be treated aggressively when it involves the periphery of connective tissue wall of the cyst. Nakamura et al<sup>7</sup> stated that wide resection of the jaw is usually the recommended treatment for ameloblastoma, should priority be given to recurrence rate. In this patient, in view of aggressive nature of tumor and the fact that the tumor involved the adjacent tissues hemimandibulectomy seem to be justified.

The probable reason for a bad prognosis is that the unicystic ameloblastoma is generally cystic, well localized and surrounded by a fibrous capsule. However, once the tumor has breached the periphery of the capsule, it can infiltrate the surrounding cancellous bone and therefore may behave more aggressively. Preoperative diagnosis of unicystic ameloblastoma can be difficult or sometimes impossible<sup>10</sup>.

The treatment of ameloblastoma itself is considered an important prognostic factor, as suggested in several studies<sup>5</sup>. Recurrence of this tumor reflects the inadequacy or failure of the primary surgical procedure. A recent systematic review showed that enucleation of unicystic ameloblastoma resulted in a highest recurrence rate 30.5% and the lowest recurrence rate was associated with resection of tumor 3.6%<sup>2,4</sup>. Therefore, proper preoperative diagnosis of these kinds of lesions and long-term follow-up is a must<sup>3,9</sup>.

## CONCLUSION

Preoperative diagnosis of unicystic ameloblastoma can be difficult or sometimes impossible because this variant of ameloblastoma shares significant clinical and radiographic similarities with odontogenic cysts and tumors and because incisional biopsy may not be able to reflect the true nature of the lesion. Unicystic ameloblastoma is a tumor with a strong propensity for recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. Long-term follow-up is mandatory because recurrence may appear years after tumor removal.

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**\*\*\*\* pl. note that the original title "UNICYSTIC AMELOBLASTOMA OF THE MANDIBLE- A CASE REPORT" is changed to " UNICYSTIC AMELOBLASTOMA OF THE MANDIBLE- A CASE REPORT WITH HEMIMANDIBULECTOMY' to avoid conflict with the title.**