

# Unicystic Ameloblastoma in Anterior Maxilla - A Rare Case

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**Abstract:** Unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity with or without luminal and/or mural tumor growth. They believed to be less aggressive and respond more favorably to conservative excision than the solid or multicystic ameloblastomas. We report a case of unicystic ameloblastoma of the anterior part of the maxilla that was treated by enucleation and use of Cornoy's solution under suspicion of lateral periodontal cyst. The nature of the lesion became evident only when the enucleated material was available for histologic examination.

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## Introduction

Ameloblastoma is the most common tumor of odontogenic origin. The World Health Organization (WHO) defines it as a locally invasive polymorphic neoplasm that often has a follicular or plexiform pattern in a fibrous stroma. It is a benign tumor whose importance lies in its potential to grow to enormous size resulting in bone deformity. The origin of the tumor is thought to be from sources that include residual epithelium of the tooth-forming apparatus, such as the epithelial cell rests of Malassez, enamel organ, reduced enamel epithelium and odontogenic cyst lining. Ameloblastomas are usually asymptomatic and found on routine dental radiographs; however, they may also present with jaw expansion. Radiographically, ameloblastomas can either be uni or multilocular with well-circumscribed margins. Its slow but relentless growth may cause movement of tooth roots or root resorption. Ameloblastomas are typically differentiated histologically into unicystic intraosseous, multicystic, solid intraosseous (80-90% of all ameloblastomas) or peripheral. The term 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 part of the cyst cavity, with or without luminal and/or mural tumor growth [1].

This report is a rare case of unicystic ameloblastoma of the maxilla that was treated by enucleation under suspicion of lateral periodontal cyst.

## Case Report

A 9-year-old male patient with non-contributory medical history sought dental evaluation due to trauma followed by painless swelling on the maxillary left anterior region. The patient described initial observation of the swelling approximately 2 months prior to presentation. Clinical examination revealed firm, blue color swelling extending from left central incisor to mesial of deciduous canine and associated mobility in 21 and 62 [figure 1, figure 2].



**Figure 1**



**Figure 2**

Panoramic radiograph disclosed an ill-defined unilocular radiolucent lesion (approximately 2 cm × 3 cm) in relation to primary maxillary left lateral incisor. The unilocular radiolucency extended from the distal surface of the permanent maxillary left central incisor to the mesial surface of deciduous canine [figure3,4].



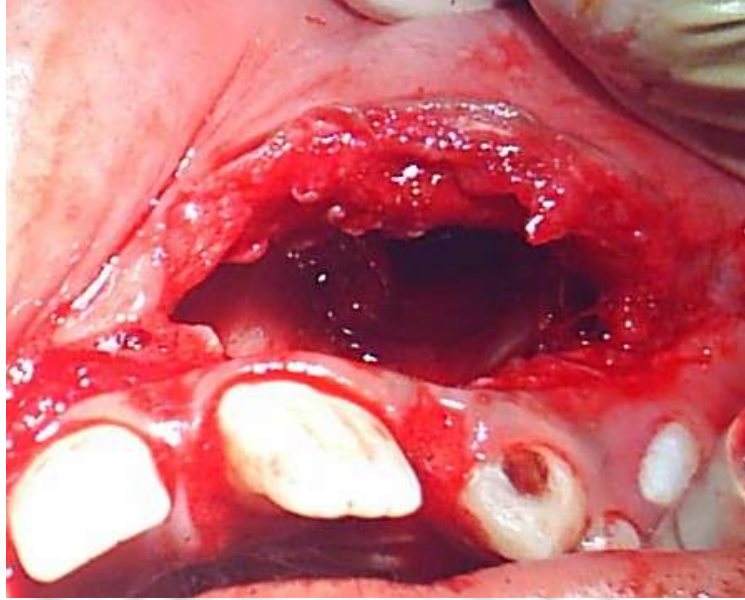
**Figure 3**



**Figure 4**

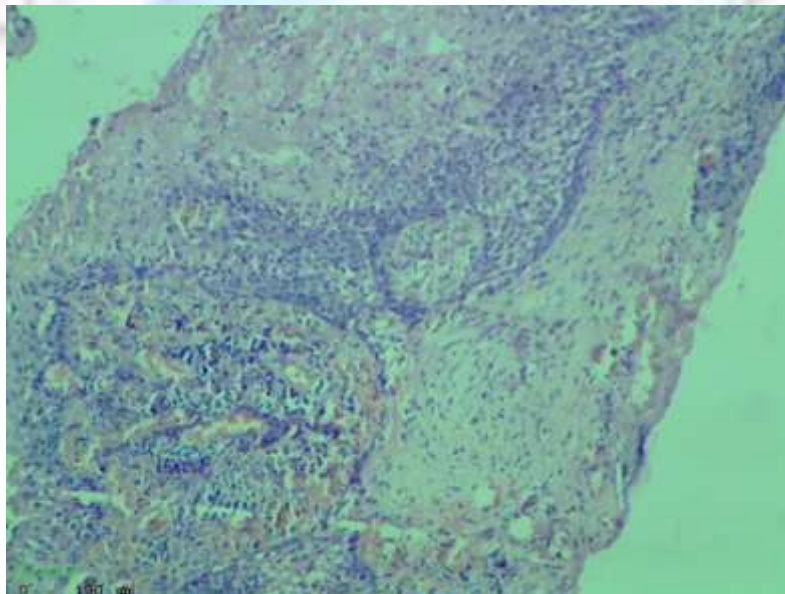
All anterior teeth were vital on examination. All anterior vital teeth and radiographic radiolucency in anterior maxillary region leads to provisional diagnosis of lateral periodontal cyst. On aspiration of cyst cavity, golden yellow colored fluid was noticed. Histological analysis of fluid showed numerous neutrophils, macrophage and few desquamated epithelial cells in an eosinophilic background which is the usual finding of inflammatory cysts hence, no conclusive evidence could be drawn from the same.

On the basis of age, location, clinical and radiographical features, differential diagnosis of lateral periodontal cyst and an inflammatory dentigerous cyst was made. Patient's parents were informed regarding the complexity of diagnosis, different treatment options, and recurrence. Taking into consideration the age and complexity of diagnosis, surgical excision was planned under local anesthesia. However during the surgery unexpected extensive bone loss was found and hence Carnoy's solution was applied to fix that area [figure 5].



**Figure 5**

Copious irrigation of bony cavity was done with normal saline, hydrogen peroxide and betadine solution. Deciduous lateral incisor of the affected side was removed during the surgery. Carnoy's solution was applied in the bone cavity for 3 min with cotton applicators. Excised lesion was sent for histological analysis. Histologic section reveals very thin cystic lining. Fibrous capsule shows few island of odontogenic epithelium with peripheral ameloblast like cell and central stellate reticulum that confirmed the diagnosis of ameloblastoma [figure 6]. Three months follow-up shows evidence of healing and improvement [figure 7, 8].



**Figure 6 - Fibrous capsule showing strands of odontogenic epithelium.**

Peripheral cells resembles ameloblast and central cell resembles  
ameloblast and central cell resembles stellate reticulum



Figure 7



Figure 8

## DISCUSSION

Unicysticameloblastoma is a tumor of young age group with slightly more than 50% of cases occurring in patients in the second decade of life, typically unilocular radiographic appearance, macroscopically cystic nature and most important, its relatively better response to conservative treatment make it a distinguishable entity[2]. It accounts for 10% to 15% of all intra-osseous ameloblastoma. The UA has an almost equal male to female distribution. More than 90% lesions are located in mandible[3]. Maxillary UA are very rare.

In a clinicopathologic study of 57 cases of unicysticameloblastoma, Ackermann classified this entity into the following three histologic groups[2]

- Group I— luminal UA (tumor confined to the luminal surface of the cyst);
- Group II— intraluminal/plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall);
- 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[4]:

- Subgroup 1— luminal UA;
- Subgroup 1.2— luminal and intraluminal;
- Subgroup 1.2.3— luminal, intraluminal and intramural;
- Subgroup 1.3— luminal and intramural.

Various treatment modalities for UA have been used such as segmental or marginal resection, more conservative treatment such as enucleation and curettage, marsupialization to reduce the size of the lesion, followed by second stage surgery[5]. These treatments can be followed by adjunctive therapy including cryotherapy, thermal or chemical cauterization, and even radiotherapy or chemotherapy. However Subgroup 1.2.3 and 1.3 has a high risk for recurrence, requiring more aggressive surgical procedure. This is because the cystic wall in these cases has an island of ameloblastoma tumor cells and there may be penetration into surrounding cancellous bone. This variant subgroup 1.3 is similar to our case. The reported recurrence rate after treatment for unicysticameloblastoma ranges from 10% to 25%. There is no adequate evidence to prove which treatment modality is more effective. Lau et al reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application and 18% by marsupialization followed by enucleation (where the lesion reduced in size)[6]. Enucleation followed by application of Carnoy's solution has resulted in a recurrence rate of 16.0% which is the best except for resection. The recurrence rate could even be lower than reported, if the closely related teeth with tumor are extracted. Because in an attempt to preserve the tooth without damage, tumor remnants may be left around the tooth apex or root and these may lead to recurrence[7]. In the present case reports, teeth in close relation of tumor were extracted before the application of Carnoy's solution.

Whatever surgical approach the surgeon decides to take, long-term follow-up is mandatory as recurrence of unicystic ameloblastoma may be long delayed. A conservative surgical approach usually results in high recurrence rate but radical surgery may lead to facial deformity. Despite high success rate for resection, particularly in mural type of unicystic ameloblastoma more conservative treatment in order to optimize quality of life is favored.

### **CONCLUSION**

In children, because the growth is not completed, choosing the most appropriate treatment for ameloblastoma is often more challenging and poses a special difficulty. Hence, we conclude that the surgical protocol must include the postoperative histopathologic examination for all lesions to rule out any ameloblastomatous changes, so that the patient can be followed up properly to take care of any recurrences happening. And conservative surgical approach can be a treatment of choice in children where growth of jaw has not been completed.

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