Unicystic Ameloblastoma presenting as Dentigerous Cyst
Niladri Sekhar Bakshi, Srijon Mukherji, Yogendra Chauhan

ABSTRACT
Unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histological examination show a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation. Unicystic ameloblastoma is a less encountered variant of the ameloblastoma and believed to be less aggressive. As this tumor shows considerable similarities with dentigerous cysts, both clinically and radiographically the biologic behavior of this tumor group was reviewed. Moreover, recurrence of unicystic ameloblastoma may be long delayed and a long-term postoperative follow-up is essential for proper management of these patients. Here, we present a case of unicystic ameloblastoma in a 12-year-old female patient.

Keywords: Unicystic ameloblastoma, Dentigerous cysts, Luminal and/or mural.

INTRODUCTION
Unicystic ameloblastoma is a tumor of young age group (second decade), typically unilocular radiographic appearance, macroscopically cystic nature and most important, as its relatively better response to conservative treatment make it a distinguishable entity. It accounts for 10 to 15% of all intraosseous ameloblastoma.1 Although most commonly found in association with the crowns of impacted teeth, it may be found in inter-radicular, periapical, or edentulous region.2 Commonly associated manifestations include painless swelling, facial asymmetry, unilocular lesion with defined sclerotic borders, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference and extrusion of tooth.3,4 This distinct prognostic entity is predominantly observed in the mandibular molar-ramus region. The posterior region of maxilla is considered to be rare and atypical. The ratio of mandibular to maxillary unicystic ameloblastoma has been reported to be 13:1.5,8 The present case report describes unicystic ameloblastoma in mandibular molar-ramus which mimic dentigerous cyst.

CASE REPORT
In this article, we report the case of a patient diagnosed with unicystic ameloblastoma histologically. A 12-year female presented to our institute with complaints of right facial swelling from 3 to 4 months. The patient was apparently healthy and her past medical history was noncontributory. Extraoral examination revealed a single diffuse solitary, oval swelling on right side of mandible. The swelling was hard in consistency with smooth surface, diffuse margins and is nontender. The color of overlying skin was normal (Fig. 1).

Intraoral examination showed right side mandibular ramal swelling. A orthopantomogram revealed a 5 × 5 cm well defined radiolucent image involving right mandibular ramus, with deep pushed wisdom tooth. Inferior border of the mandible was seen to be intact (Fig. 2). Differential diagnosis included dentigerous cyst, unicystic ameloblastoma, odontogenic keratocyst. The presence of pushed unerupted tooth as directing the diagnosis more toward dentigerous cyst in the given age of the patient.

Fig. 1: Right facial swelling
Fig. 2: Orthopantomogram showing radiolucent lesion with impacted tooth
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In this case, we performed careful enucleation of the cyst along with the teeth within it. Inferior alveolar nerve was carefully preserved (Fig. 3). A very thin buccal cortical plate was preserved so that it can scaffold bone healing and remodeling.

The surgical specimen was macroscopically described as solitary, thick cystic lining attached to cervical region of an impacted molar (Fig. 4). Diameter of cyst lining is 2.77 cm. Specimen shows crown of a molar tooth and mural proliferation on inner wall of cystic lining. Microscopically described as very thin cystic epithelium resembling reduced enamel epithelium. Cystic lumen shows interconnecting strands of odontogenic epithelium with peripheral ameloblast like cells and central stellate reticulum like cells.

Histopathologically, diagnosis of plexiform type unicystic ameloblastoma was confirmed.

A 6 weeks postoperative orthopantomogram shows a well healed surgical site (Fig. 5).

DISCUSSION

Unicystic ameloblastoma shares common clinical and radiographical manifestations with other odontogenic lesions making diagnosis difficult. 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.

Solid ameloblastoma is multilocular and is seen uncommonly in patients less than 30 years of age.

Differentiating unicystic ameloblastoma from dentigerous cyst possess great challenge. In dentigerous cyst, expansion of the buccal cortex occurs as cyst grows toward most dependent part, i.e. buccally. Ameloblastoma usually grows buccally and lingually. In present case, only buccal cortical expansion was present with an impacted tooth favoring the diagnosis of dentigerous cyst clinically, radiologically and surgically.

Various treatment modalities for unicystic ameloblastoma have been recommended, such as enucleation and curettage, marsupialization to reduce the size of the lesion, segmental or marginal resection. These treatments can be followed by adjunctive therapy, including cryotherapy, thermal or chemical cauterization and even radiotherapy or chemotherapy. The reported recurrence rate for unicystic ameloblastoma ranges from 10 to 25% with any of the recommended treatment. There is no adequate evidence to prove which treatment modality is more effective.

Resection of unicystic ameloblastoma results in the lowest recurrence rate (3.6%), if adequate bone margins are removed. Despite high success rate for resection of unicystic ameloblastoma more conservative treatment in order to optimize quality of life is generally favored. Marsupialization together with other adjunctive treatment resulted in an 18% recurrence. The treatment modality is not so popular method, but benefit is expected in very ill patients or those with a huge lesion.

Enucleation alone yielded the highest recurrence rate among all treatment (30.5%). Two possible explanations: firstly, cystic lining of the tumor is inadequately removed;
secondly, ameloblastic tumor cells can invade the cancellous bone to a certain extent.9-11 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 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.12-14

It has been suggested that, for all unilocular lesions, an excisional biopsy by enucleation should be carried out.

Ackerman et al, in their study of unicystic ameloblastomas, defined three subgroups. Group I (42%) consisted of a unilocular cyst with a nondescript but variable epithelial lining. Inactive odontogenic cell rests might be present in the fibrous wall, but there was no infiltration by neoplastic epithelium. Group II lesions (9%) featured intraluminal plexiform proliferation but no infiltration of the cyst wall. In group III lesions (49%), plexiform or follicular-type ameloblastoma, sometimes in continuity with the cyst lining, infiltrated the wall. Group III lesions need to be treated more aggressively similar to solid/multicystic ameloblastoma. The present case can be considered under Ackerman group II.

Ackerman type 1 or type 2 unicystic ameloblastoma, enucleation with curettage and regular follow-up policy are advocated till recurrence is noted. However, for a pathological diagnosis of Ackerman type 3 unicystic ameloblastoma resection in the forms of partial maxillectomy, marginal or segmental resection of mandible is recommended. The rationale for treatment without an incisional biopsy is that a small tissue may not reflect all types of Ackerman unicystic ameloblastoma; thus, the chance of under diagnosis is high.1

CONCLUSION

As unicystic ameloblastoma tends to affect young adolescent patients, the concern to minimize surgical trauma and permit jaw function and tooth development to proceed reasonably unimpaired. As patient age is 12 years, which is influencing factor related to the choice of treatment. We support the concept of enucleation and extraction of closely related teeth and regular follow-up in our case.

REFERENCES


ABOUT THE AUTHORS

Niladri Sekhar Bakshi (Corresponding Author)

Associate Fellow, Department of Oral Surgery, Calcutta Institute of Maxillofacial Surgery and Research, Kolkata, West Bengal, India

Phone: 03324989759, e-mail: dr.niladri@yahoo.in

Srijon Mukherji

Director and Chief Consultant, Department of Maxillofacial Surgery, Calcutta Institute of Maxillofacial Surgery and Research Kolkata, West Bengal, India

Yogendra Chauhan

Surgical Fellow, Department of ENT and Head and Neck Surgery Calcutta Institute of Maxillofacial Surgery and Research, Kolkata West Bengal, India