CASE REPORT

Unicystic Ameloblastoma Associated with Primary Mandibular Second Molar: A Case Report

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Abstract

Background: Unicystic ameloblastoma is a rare, benign, locally invasive odontogenic neoplasm of young age that shows clinical, radiographic, or gross features of an odontogenic cyst but histologically shows typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth.

Aim: To report a case of an asymptomatic unicystic ameloblastoma in a 12-year-old child, along with its management and follow-up.

Case description: A 12-year-old boy presented with swelling with respect to the left body of the mandible. The orthopantomogram (OPG) and computed tomography scan revealed a large unilocular radiolucency in the left mandible associated with the primary second mandibular molar. Complete enucleation of the cyst and extraction of the associated primary teeth and underlying permanent teeth were done under general anesthesia. Carnoy's solution was applied in the bone cavity for 3 minutes with cotton applicators. Postoperative healing was uneventful. Prosthetic rehabilitation was done during the follow-up period.

Conclusion: Unicystic ameloblastoma is rarely seen in younger children, so a pediatric dentist must be cautious while diagnosing an intraoral swelling. Timely intervention and conservative surgical treatment, along with a proper follow-up, improved the treatment outcome and prevented potential complications in the future.

Clinical significance: This report highlights the salient features of unicystic ameloblastoma to be able to accurately diagnose and manage the lesion.

Keywords: Carnoy's solution, Case report, Surgical enucleation, Unicystic ameloblastoma.

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Introduction

Ameloblastoma is the most common benign odontogenic tumor of the jaws that constitutes about 1% of all cysts and tumors of the jaws.[1,2] It is generally a painless, slow-growing, locally aggressive tumor causing expansion of the cortical bone, perforation of the lingual or the buccal cortical plate, and infiltration of the soft tissues. It has a peak incidence in the third and fourth decade of life but can be found in any age group with equal gender predilection (1:1).[1–5] The relative frequency of mandible to maxilla is reported to be varying from 80–20% to 99–1%. In the mandible, the majority of ameloblastomas are found in the molar ramus region.[1,3]

In a conventional radiograph, ameloblastoma can be presented as either unilocular or multilocular corticated radiolucency; the bony septa results in a honeycomb or soap bubble appearance or tennis racket pattern. In some places, cortical plates are spared and expanded, as in other regions, they are destroyed; root resorption is a common finding.[6] Buccal and lingual cortical plate expansion is more common in ameloblastoma than in other tumors.[7]

The challenge in managing ameloblastoma is achieving complete excision and reconstruction of the defect when the tumor is large.[1] Ameloblastoma is treated by enucleation, curettage, or surgical excision, depending on the size and type of the lesion. The rate of recurrence ranges from 17.7% for en bloc resection to 34.7% for conservative therapy. Wide resections with a safety margin of healthy bone to prevent local recurrence were preferred.[2]

This paper reports the case of a unicystic ameloblastoma associated with a primary mandibular second molar, which was successfully managed with cyst enucleation along with surgical removal of the permanent successor.

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Case Description

A 12-year-old male patient was referred from a private clinic to the Department of Pediatric and Preventive Dentistry for the management of mild diffuse swelling in the lower left back tooth region for 3 weeks (Fig. 1). The swelling was insidious in onset and gradually increased to the present size. There is a history of pulp therapy done on the same tooth by a general practitioner when the child was 10 years old. There was no history of toothache or decrease in the size of the swelling, or any discharge from the swelling.

Extraoral examination revealed a diffuse swelling in the lower left side of the mandible, without loss of facial symmetry, and the mucosa overlying the swelling was normal. Local examination revealed restored teeth 75, a swelling of approximately 2 × 3 cm in size extending from mesial of 34 to distal of 36, obliterating the vestibule. On palpation, the swelling was non-tender and hard in consistency. The teeth were nontender on percussion. Thus, a provisional diagnosis was made as a radicular cyst/dentigerous cyst. Intraoral periapical radiograph (IOPA)/orthopantomogram (OPG) was taken to confirm the diagnosis (Figs 2 and 3).

Fine needle aspiration cytology was performed under conscious sedation in the next appointment, where the aspirate contained blood and inflammatory cells. It was reported that it contained inflammatory cells (Figs 3A and B). So, after obtaining written consent from the parents, the procedure was performed under general anesthesia (Figs 4A and B). The buccal flap was raised, the cyst enucleation was done, and 75 and 35 teeth were extracted along with it (Fig. 4C). Copious irrigation of the bony cavity was done with betadine solution and normal saline. Carnoy's solution was applied in the bone cavity for 3 minutes with cotton applicators. The specimen was sent for histopathological examination. The bony cavity was rinsed with saline, and sutures were placed.

Postsurgical healing was satisfactory and uneventful. The patient was discharged the next day with medications, and oral hygiene instructions were given. The patient was advised to report to the department for suture removal and follow-up. IOPA was taken on 75 regions during the 3-month follow-ups, where it was noticed that the radiolucency was reduced (Fig. 5). For space management, a removable partial denture was delivered with respect to the 35th region (Fig. 6).

Histopathological examination (Fig. 7) revealed a nonkeratinized cyst lining the epithelium in association with fibrovascular connective tissue. In some areas, the lining epithelium shows ameloblastomatous changes like basal columnar cells with hyperchromatic nuclei and reversal of polarity. Suprabasilar cells are stellate reticulum-like cells. The underlying connective tissue shows dense connective tissue fibers, moderately dense chronic inflammatory cells, capillary vessels, and extravasated RBCs. In some foci, the epithelium shows hyperplastic changes adjacent to the inflamed connective tissue. The diagnosis was confirmed as "infected luminal unicystic ameloblastoma."

Discussion

Unicystic ameloblastoma is a tumor affecting the young age group (the 20s), typically unilocular radiographic appearance, macroscopically cystic nature, and, most importantly, its relatively better response to conservative treatment makes it a different entity. It accounts for 10–15% of all intraosseous ameloblastoma.[8] Although most commonly found in association with the crowns of impacted teeth, it may be found in interradicular, periapical,

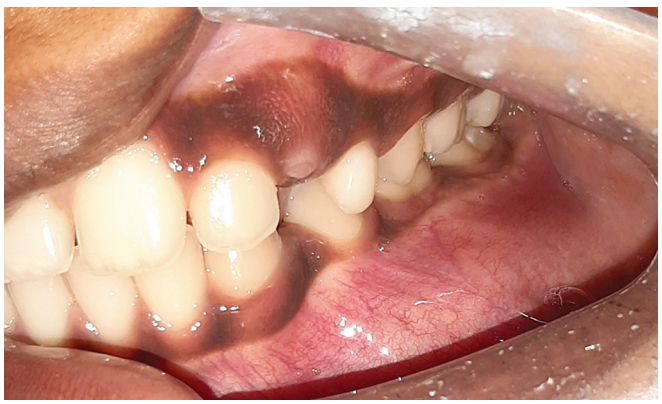


Fig. 1: Intraoral view showing an expansible lesion in the left mandibular posterior region

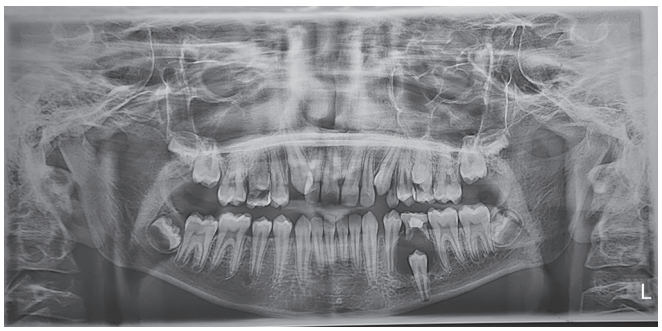
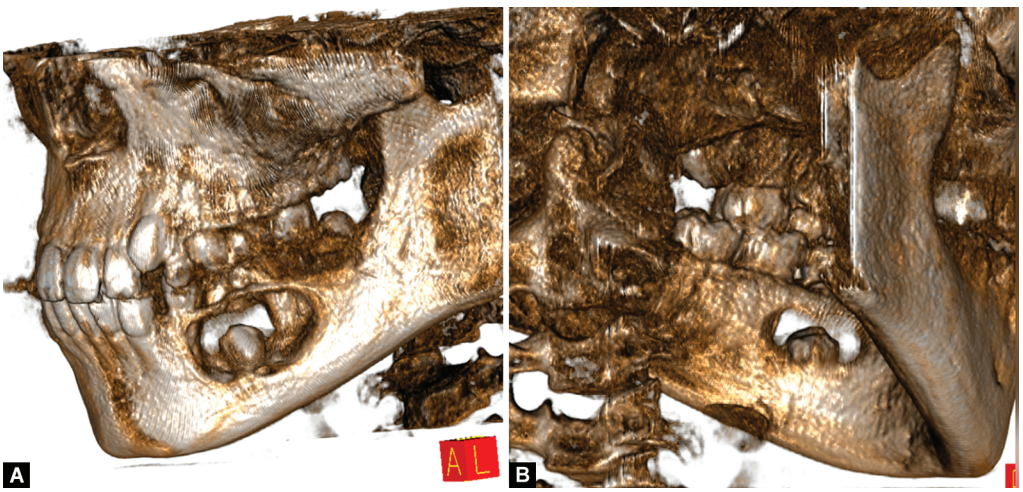
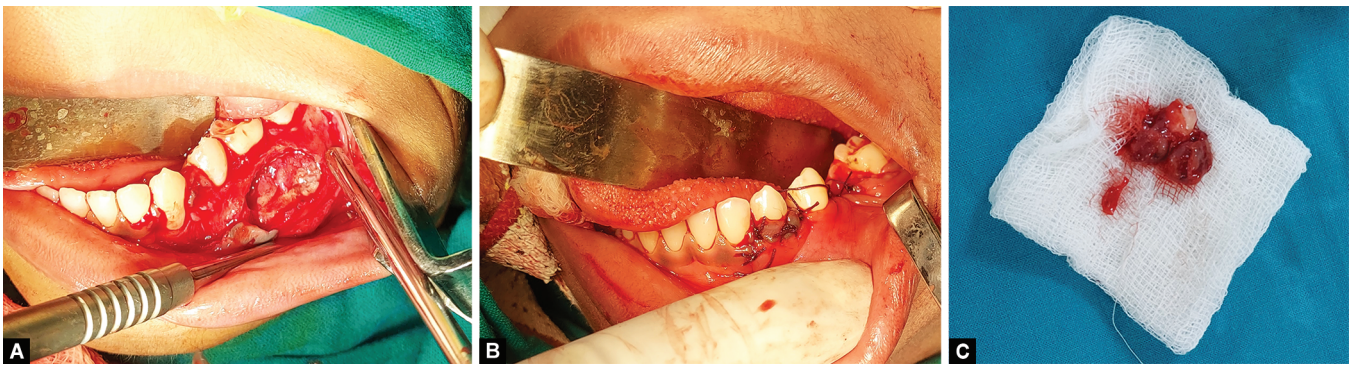


Fig. 2: Panoramic view showing radiolucency associated with 75 (preoperative)



Figs 3A and B: (A) Cone-beam computed tomography (CBCT) buccal view showing involvement of tooth 35 and buccal cortical plate erosion; (B) CBCT showing lingual cortical plate erosion



Figs 4A to C: (A) Procedure done under general anesthesia (GA); (B) Surgical area after suture placement; (C) Excised lesion along with permanent tooth bud of 35

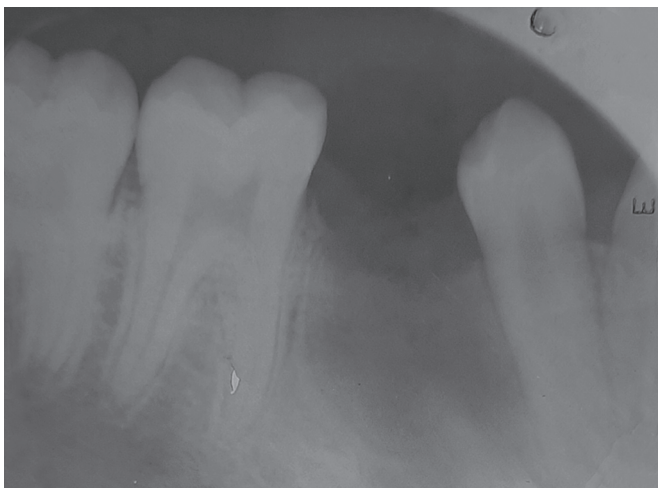


Fig. 5: Intraoral periapical radiograph (IOPA) showing a reduction in radiolucent lesion after 3-month follow-up

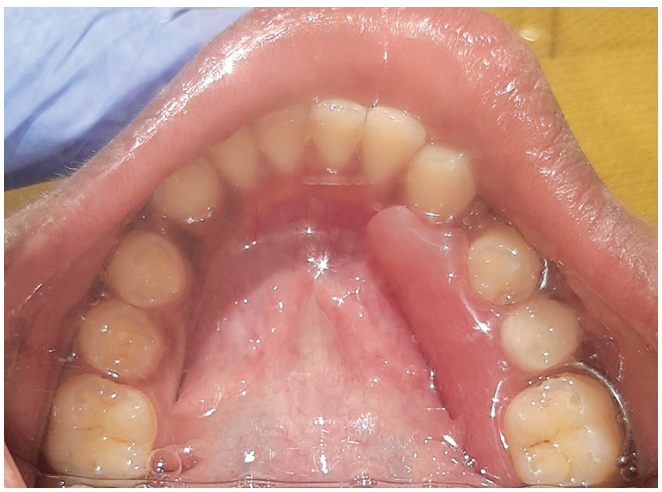


Fig. 6: Removable partial denture (RPD) with respect to the 35th region

or edentulous regions.[9] Common manifestations include painless swelling, unilocular lesions with defined sclerotic borders, facial asymmetry, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference, and extrusion of the tooth.[10] It is predominantly observed in the mandibular molar-ramus region. The ratio of mandibular to maxillary unicystic

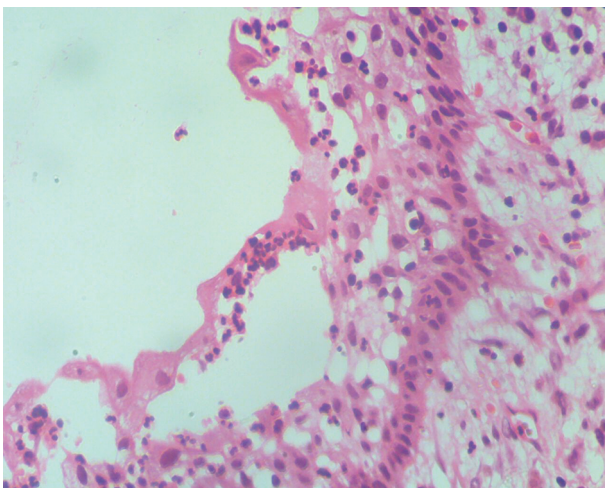


Fig. 7: Showing the nonkeratinized cyst lining epithelium in association with a fibrovascular connective tissue

ameloblastoma has been reported to be 13:1.[11] The present case report describes the unicystic ameloblastoma of mandibular molar-ramus regions mimicking an inflammatory dentigerous cyst.

Unicystic ameloblastoma shares common clinical and radiographical manifestations with other odontogenic lesions, which makes it difficult to diagnose. Dentigerous cyst, odontogenic keratocyst, residual cyst, adenomatoid odontogenic tumor, giant cell lesion, and sometimes solid ameloblastoma are some of the possible differential diagnoses for unicystic ameloblastoma (UA). Keratocyst usually spreads anteroposteriorly and sometimes shows cortical expansion, and on aspiration, it shows a large amount of keratin.[12] Residual cysts are associated with missing teeth that have been extracted. Adenomatoid odontogenic tumors are commonly seen in the anterior maxilla, whereas a central giant lesion often arises anterior to the first mandibular molar.[13] Solid ameloblastoma is multilocular and is seen uncommonly in patients <30 years of age.[14] Great difficulty exists in differentiating dentigerous cysts from UA. Some of the features that help in differentiating UA are defects in the wall of a cyst, unilocular cystic lesion extending into the ramus, and 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),[10] presence of erythematous and granulomatous tissue at the marginal gingival (mucosal ulceration) with the absence of the bony cortex, and associated healthy primary dentition.[12] In the present study, the histopathological findings favored the diagnosis of UA.

There are various treatment modalities for UA, such as segmental or marginal resection, more conservative treatment such as enucleation and curettage, and marsupialization to reduce the size of the lesion, followed by second-stage surgery.[15,16] These treatments can be followed by adjunctive therapy, including cryotherapy, thermal or chemical cauterization, and even radiotherapy or chemotherapy.[17,18] The reported recurrence rate after treatment for unicystic ameloblastoma ranges from 10 to 25%.[15] There is no reasonable evidence to prove which treatment modality is more effective.

Enucleation alone yielded the highest recurrence rate among all treatments (30.5%). Two probable explanations: firstly, the cystic lining of the tumor is inadequately removed; secondly, ameloblastic tumor cells can invade the cancellous bone to a certain extent.[19] 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 be even lower than reported if the closely related teeth with tumors are extracted. 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.[18,19] In the present case report, teeth in close relation to the tumor were extracted. Carnoy's solution, a powerful fixative, penetrates the cancellous spaces and thus fixes the remaining tumor cells. Usually, Carnoy's solutions are applied for 3–5 minutes. However, Frerich et al.,[20] suggested that the application of Carnoy's solutions should not exceed 3 minutes and should not be directly applied over the nerve as it could lead to nerve impairment.

It has been suggested that for all unilocular lesions, an excisional biopsy by enucleation should be carried out. If the histopathological diagnosis shows Ackerman et al.[8] type 1 or 2 unicystic ameloblastoma, then follow-up and a wait-and-see policy is advocated till recurrence is noted. However, for a pathological diagnosis of Ackerman et al.[8] type 3 unicystic ameloblastoma, resection in the forms of partial maxillectomy, marginal, or segmental resection of the mandible is recommended. Therefore, we support the concept of applying Carnoy's solutions for 3 minutes following enucleation and extraction of closely related teeth.

Conclusion

Dentists should be aware of the unilocular radiolucencies of the jaws as it can be unicystic ameloblastoma. Timely intervention and conservative surgical treatment, followed by the application of Carnoy's solution and the extraction of closely related teeth, may improve treatment outcomes and potential complications associated with larger resection.

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