Conservative Management of Unicystic Ameloblastoma: A Case Report

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Abstract

Ameloblastoma represents 1% of all tumors in the oral cavity. It is an odontogenic tumor, benign in nature, usually presents in jaw bone. The unicystic ameloblastoma (UA) presents itself as a cystic cavity without solid neoplastic features, which differs from the solid ameloblastoma with regard to the age of the patients and the rate of recurrence. It shows clinical and radiologic characteristics of an odontogenic cyst. A case of UA is presented here, where lesion was pain free with continuous expansion of the mandible. It was removed by curettage and chemical cauterization done with Carnoy's solution under GA.

Keywords: Unicystic ameloblastoma, Odontogenic cysts, Curettage, Intramural.

Introduction

Ameloblastoma is a benign odontogenic tumor, usually presents in the jaw bone. The origin can be the embryonic remains of odontogenic cysts, the enamel organ, or the stratified squamous epithelium of the oral cavity.[1] It accounts for approximately 1% of oral tumors.[2] In 1977, Robinson and Martinez first used the term "Unicystic ameloblastoma (UA)," but it was also named in the second edition of the international histologic classification of odontogenic tumors by the WHO as "cystogenic ameloblastoma."

UAs are a rare variant of ameloblastomas, which usually occur in younger populations. They are characterized by slow growth and being relatively locally aggressive, with the main site of origin at the posterior portion of the mandible.[3]

There can be three pathologic mechanisms for the evolution of UA: [4]

1. The reduced enamel epithelium associated with the developing tooth undergoes ameloblastic change with subsequent cystic transformation.
2. Ameloblastomas arise in dentigerous or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic
3. stratified squamous lining.
4. Solid ameloblastoma undergoing cystic degeneration of ameloblastic islands with subsequent fusion of multiple microcysts and then into a unicystic lesion.

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Case Report

A systemically healthy 16-year-old male patient reported at the Department of Pediatric Dentistry at Government College of Dentistry, Indore, complaining of asymptomatic swelling on lower left side of face for 2 months which was initially small and was increasing gradually to attain the present size. There was no significant medical history or known allergies reported.

On extraoral examination, a diffuse swelling was seen which is measuring about 9 × 5 cm in size. Overlying skin was normal. No visible pulsations and discharge were seen. On palpation, swelling was firm in consistency and tenderness present (Figure 1). It extends anteroposteriorly from the body region of mandible to angle of mandible and superoinferiorly from the maxilla to lower border of mandible region. On intraoral examination, 37 was missing and pale color mucosa seen with respect to 37 region. There was no history of extraction. A clinical provisional diagnosis of odontogenic cyst/tumor was made.

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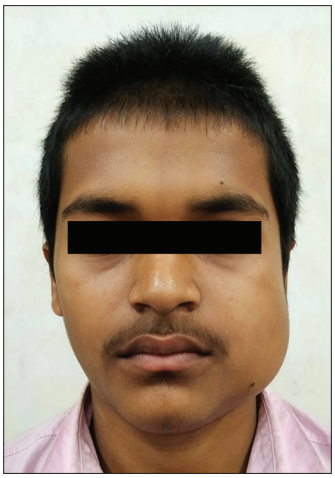

Figure 1: Pre-operative extraoral view

On panoramic radiograph examination, a unilocular radiolucent lesion of size 5 × 7 cm extending from 36 to the mid ramus region, with 37 and 38 embedded in the lesion (Figure 2). Aspiration yielded mixed blood. An incision biopsy was done. Tooth 38 with soft tissue was taken and sent for biopsy. Histopathologically, the lesion was characterized as UA (with mural proliferation) (Figure 3). Taking in consideration about patient's age and the size of the lesion, a conservative treatment was planned.

Under general anesthesia, a crestal incision was given from 35 region extending up to anterior border of ramus. After layerwise dissection, cystic lining was exposed, curetted completely and extraction of tooth 36 and 37 tooth was done. Chemical cauterization was done with Carnoy's solution. After thorough irrigation, hemostasis was achieved and dressing of cavity done with betadine and soframycin roller gauze. Closure of the remaining cavity was done with 2-0 Vicryl suture. The patient was recalled after 1 month for removal of intermaxillary fixation. The patient was recalled for follow up after 7 months also (Figure 4).

Discussion

Ameloblastoma is a neoplasm of odontogenic epithelium, mainly of enamel organ type tissue that has not undergone differentiation to the point of hard tissue formation. It accounts for about 1% of all oral tumors and about 9–11% of odontogenic tumors.[1] In general, it is a slow growing but locally invasive tumor.[3] With no gender predilection, its peak incidence is in the 3–4 decades of life.[1] Major occurrences in the mandibular molar ramus area with slight male predilection were elicited.[5] The World Health Organization (1991) defined ameloblastoma as a benign but locally aggressive tumor with a high tendency to recur, consisting of proliferating odontogenic epithelium lying in a fibrous stroma. Ameloblastoma is classified, according to the WHO and the International Agency for Research on Cancer, 2003, as a benign tumor with odontogenic epithelium, mature fibrous stroma, and without odontogenic ectomesenchyme.

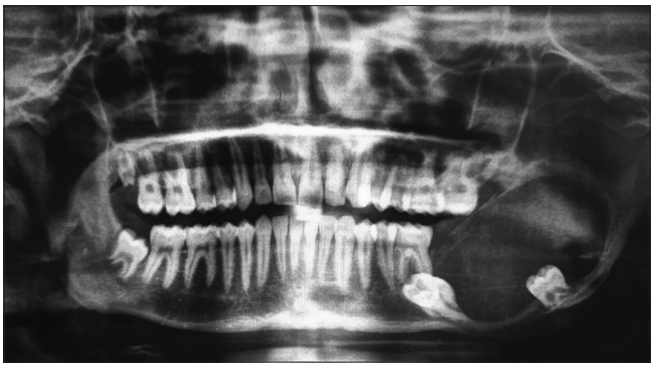

Figure 2: Pre-operative OPG radiograph

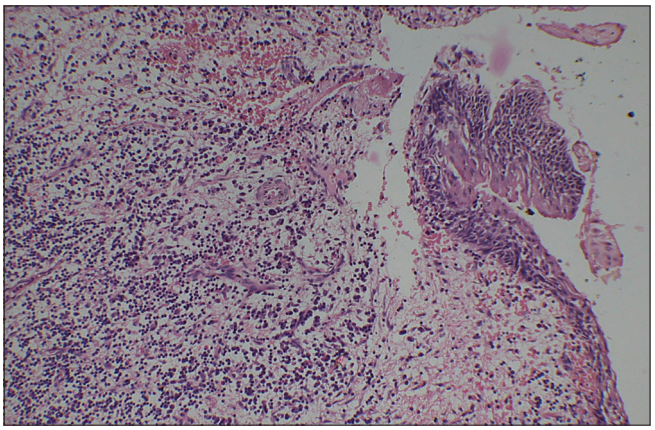

Figure 3: Mural proliferation (histopathology image)

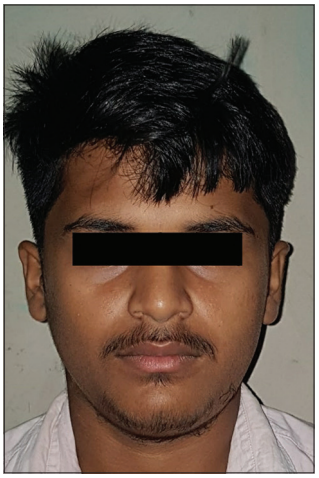

Figure 4: Post-operative extraoral view

Ameloblastoma is further classified into:

- Solid/multicystic
- Extraosseous/peripheral
- Desmoplastic ameloblastoma
- Unicystic.

Unicystic type of ameloblastoma shows clinical and radiological features of an odontogenic cyst. In histologic examination, it shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor

proliferation. About 5–15% of all ameloblastomas are of the unicystic type. UA with an unerupted tooth occurs with a mean age of 16 years as opposed to 35 years in the absence of an unerupted tooth. The mean age is considerably lower than that for solid/multicystic ameloblastoma with no gender predilection. In 90% of cases, the UA is located in the mandible with 77% located in the molar ramus region. Mandible-to-maxilla ratio is 13:1.

Patients commonly complain of swelling (typically asymptomatic) and facial asymmetry. Pain is an occasionally presenting sign. Asymptomatic swelling often indicates a long duration lesion and of significant size. Continued growth of the tumor may cause ulceration of the mucosa overlying the lesion. Minor lesions tend to be discovered more often on routine radiographic examination or as a result of local effect produced by tumor such as tooth mobility, occlusal alterations, and failure of eruption of teeth. Maxillary UAs are very rare. In 1987, Gardener *et al.* reported first case of maxillary UA in a 12-year-old boy in the molar area.

Radiographic appearance presents with unilocular to multilocular patterns with predominance for unilocular configuration. Six radiographic patterns for unicystic ameloblastoma were identified by Eversole *et al.*, ranging from well-defined unilocular to multilocular appearances. Thick enhancement of the tumor wall and small intraluminal nodules as characteristic features of this type lesion was detected only by contrast-enhanced magnetic resonance imaging.[4]

Histologically, the minimum criterion for diagnosing this lesion is the expression of a single cystic sac lined by odontogenic epithelium. UA should be differentiated from odontogenic cysts because of high recurrence rate of former than the later.

UA is a prognostically distinct entity with a recurrence rate of 6.7–35.7%.[1] Minimum recommended follow-up period by most of the authors is up to 5 years after the treatment, but recurrences can occur up to 21 years.[6]

Ackermann classified it into three following histologic groups:[7]

- Luminal UA
- Luminal and intraluminal UAs
- Luminal, intraluminal, and intramural UAs
- Luminal and intramural UAs.

Histologic subgroups given by Philipsen and Reichart: [7]

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

Treatment of UA includes both radical and conservative surgical excision, curettage, chemical and electrocautery, radiation therapy, or combination of surgery and radiation.[1] The treatment of ameloblastoma poses a special problem in children, as the growth of the jaw is not completed. Conservative treatment of ameloblastoma seems to be acceptable in childhood. Curettage followed by marsupialization is most common procedure.[8] *Isacsson et al.* suggested conservative treatment of UA in lower jaw, but contraindicated in the maxilla due to the risk of infiltration.[9]

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