Case Report

Mural Unicystic Ameloblastoma mimicking Odontogenic Cyst

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Abstract

Unicystic ameloblastoma is considered a less aggressive variant of ameloblastoma with clinical and radiographic features mimicking a cyst. Herein, we present a case of unicystic ameloblastoma in a 20-year-old female who was clinically and radiographically diagnosed as an odontogenic cyst. Unicystic ameloblastoma and its subtypes are reviewed with special emphasis on the mural variant. The main aim of this case report is to highlight (1) the need of including an odontogenic tumor in the differential diagnosis of the unilocular, well-circumscribed radiolucent lesion by the clinicians (2) importance of serial sectioning of the specimen to identify the mural component if any present, and (3) following resection as the mode of treatment.

Keywords: Mural ameloblastoma, odontogenic cyst, unicystic ameloblastoma

Introduction

Unicystic ameloblastoma (UA) is an odontogenic tumor but resembles a cyst clinically and radiographically. Its gross specimen features also resemble a cyst. Histologically, it exhibits ameloblastic epithelium lining a cystic cavity. The lining epithelium is accompanied with or without luminal and/or mural tumor growth.[1] UA was categorized as a separate group by Martinez and Robinson.[2] The lesion UA arises from reduced enamel epithelium or a dentigerous cyst or from solid ameloblastomas that undergo cystic degeneration.[3] UA was grouped by Philipsen and Reichart as luminal (subgroup 1); luminal and intraluminal (subgroup 1.2.); luminal, intraluminal, and intramural (subgroup 1.2.3); luminal and intramural (subgroup 1.3).[4]

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Case Presentation

A 20-year-old female patient came to the outpatient clinic with a chief complaint of painful swelling inside the mouth in the right lower back tooth region for about 3 months. History revealed that the swelling was initially small in size and had gradually increased to the present size. The pain was intermittent and dull and the patient was under medication.

On extraoral examination, no swelling was noticed and the mouth opening was normal. There were no signs of paresthesia in the lower lip. On intraoral examination, a swelling measuring 4 × 3 cm was noticed in the buccal aspect of 45 to 47 region obliterating the gingivobuccal sulcus with lingual cortical expansion. There was no associated discharge. Tooth 48 was clinically not present. On palpation, the swelling was tender

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with eggshell crackling. On fine-needle aspiration, blood-tinged fluid was obtained.

Orthopantomogram [Figure 1] revealed a unilocular well-circumscribed radiolucency with sclerotic border measuring 3 × 3.5 cm extending from the mesial root of 46 till the mesial root of 47. Inferiorly, the lesion extended 0.5 cm above the lower border of the mandible while 48 was absent. Absence of impacted tooth (48) and features like buccolingual cortical expansion, unilocular radiolucency together with the absence of cheesy, yellow aspirate ruled out the possibility of the dentigerous cyst, and odontogenic keratocyst.

An incisional biopsy on histopathological examination exhibited cystic lining epithelium associated with the connective tissue wall. The basal cells of the cystic lining epithelium were hyperchromatic with stellate reticulum like cells superficially. The lesion was histologically diagnosed as cystic ameloblastoma.

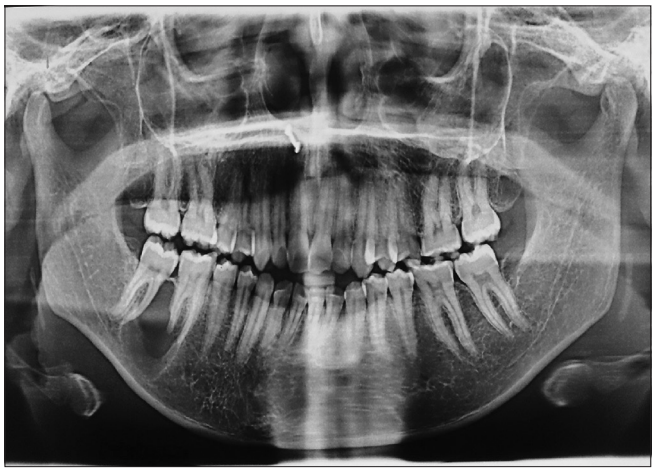

Figure 1: Panoramic radiograph representing well-defined unilocular radiolucent lesion in the right mandible concerning 46 and 47

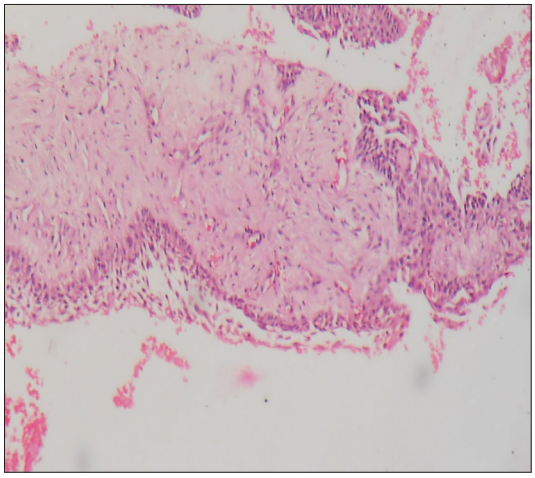

Figure 3: Photomicrograph showing cystic lining epithelium with connective tissue wall. (H and E staining, 4×)

The patient was planned for a segmental resection with continuity defect and reconstruction with non-vascularized iliac crest bone graft under general anesthesia.

Grossly, a cut section across the resected specimen of bone revealed a unicystic lesion [Figure 2]. Histopathology revealed cystic lining epithelium in association with connectives tissue wall [Figures 3-5]. The lining epithelium exhibited hyperchromatic basal cells and loose stellate reticulum-like cells on the surface. In the connective tissue wall, and ameloblastic follicle with tall columnar peripheral cells exhibiting subnuclear vacuolization and central stellate reticulum with areas of cystic degeneration was seen. Correlating the clinical, radiographic, gross, and histopathology features, the lesion was concluded as UA of mural type. (Subtype 1.3).

Discussion and Literature Review

UA subgroups with mural component are considered as the most aggressive type.[5]

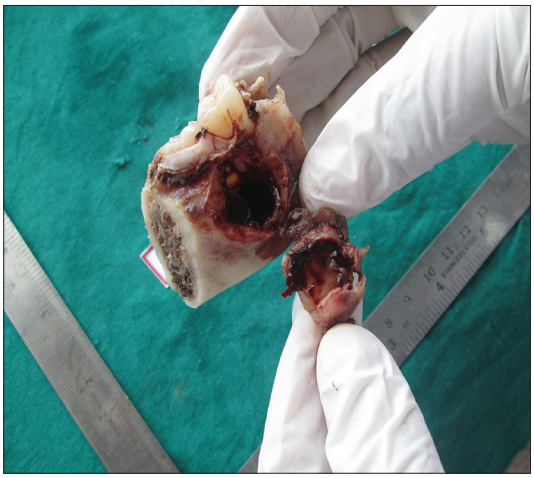

Figure 2: Gross specimen of the lesion

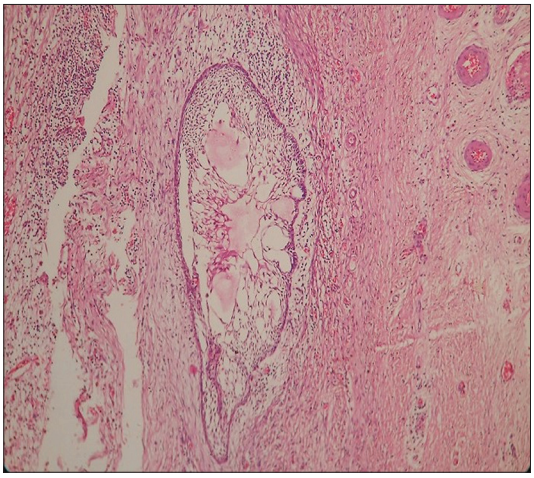

Figure 4: Photomicrograph shows ameloblastic island as follicle in the connective tissue. (H and E staining, 10×)

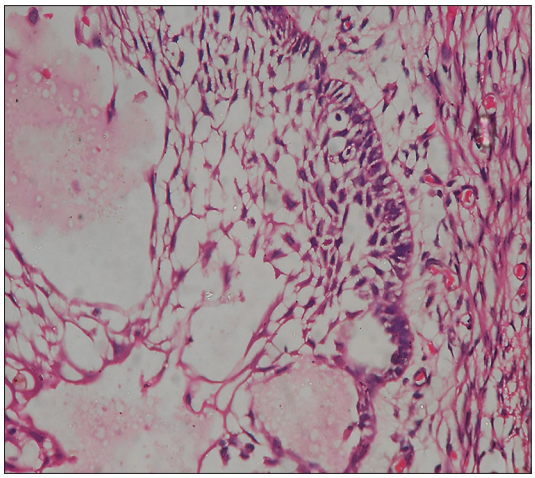

Figure 5: Photomicrograph showing peripheral cells exhibiting reversal of polarity, hyperchromatic nuclei with stellate reticulum in the islands. Areas of cystic degeneration also present (H and E stain, 40×)

It represents 10%–15% of ameloblastomas[2] and manifests commonly from 11 to 30 years of age with equal gender predilection.[2] Majority of the lesions are associated with an impacted tooth (dentigerous type) and others are non-dentigerous type. Smaller lesions are asymptomatic while larger lesions result in facial asymmetry and in some cases ulceration of the overlying mucosa.[6] The most frequent site of occurrence is the posterior mandible.[1,2] The ratio of occurrence in maxilla and mandible is 1: 7 (dentigerous type) and 1: 4.7 (non-dentigerous type).[7]

Rarely UA mural type has been associated with varied histological features. UA (subgroup 1.2.3) with mural proliferation exhibiting follicular and acanthomatous patterns along with desmoplastic type have been recorded.[5] Though on incisional biopsy the case was diagnosed as cystic ameloblastoma the lesion was typed as UA only after excisional biopsy.[2] The segmentally resected mandible on gross examination encompassed a unicystic lesion signifying the importance of gross examination before typing the lesion as unicystic as some solid-multicystic ameloblastomas can also have areas of cystic degeneration.

It is important to note that in this case only upon histological examination of the excisional specimen the ameloblastic follicle was seen in the connective tissue wall and the lesion was identified as subgroup 1.3. Since the subgroups with the mural component are aggressive and their recurrence is 35.7%,[4,5] it is suggested that unless UA lesions are extensive and pose esthetic problems in young patients, resection can be employed as the line of treatment than enucleation and marsupialization to prevent recurrence and second surgery.

The subtypes with the mural component are the most aggressive type of UA.[5] Increased expression of CD 34, MMP 2, and MMP 9 in the mural variant than in luminal and the intraluminal type explains the proliferative potential and the aggressiveness of the variant along with the necessity of radical treatment for the lesions.[8]

UA is treated by different methods like enucleation, marsupialization, and resection. Lower recurrence rate (3.6%) is observed with the resection method and highest (30.5%) with procedures employing enucleation.[9]

In a study of 116 cases of UA, recurrence associated with marsupialization was 12%. The study indicated that presence of mural histology in ameloblastoma, cortical bone resorption, or root resorption can imply aggressiveness of lesion which would result in recurrence following marsupialization in the early stage.[10]

Conclusion

The main aim of the article is to emphasize that UA resembles a cyst clinically and radiographically, thus warranting its inclusion in the differential diagnosis of odontogenic cysts by the clinicians. Similarly, incisional and excisional biopsy specimens of UA should be subjected to periodic sampling and serial sectioning to determine the presence or absence of mural component. This will enable precise diagnosis of the lesion and will also facilitate the surgeons to decide on the treatment plan. Aggressive treatment like resection should be considered to prevent repeated surgeries. Post surgically, cases of mural UA should be reviewed periodically for recurrences.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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