

Unusual imaging appearance of unicystic ameloblastoma

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Abstract

Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastous epithelium lining part of the cystic cavity, with or without luminal and/or mural tumor growth. It accounts for 5-15% of all intraosseous ameloblastomas. We report a case of UA in a 35-year-old female with an unusual large multilocular (tennis racket) appearance on the right body of mandible and illustrate the importance and complexity of differential diagnosis with a brief review of recent literature.

Keywords: Differential diagnosis, multilocular appearance, tennis racket appearance, unicystic ameloblastoma

Introduction

Ameloblastomas have been categorized into three biologic variants: Cystic (unicystic), solid (multicystic), and peripheral. The literature indicates that the cystic variant is biologically less aggressive and has a better response to enucleation or curettage than the solid ameloblastoma.^[1] Multicystic ameloblastoma is the most common variety and represents 86% of cases. Peripheral tumors are odontogenic tumors, with the histological characteristics of intraosseous ameloblastoma that occur solely in the soft tissues covering the tooth-bearing parts of the jaws. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts (DCs).^[2] It is a less aggressive variant and it has a low rate of recurrence, although lesions showing mural invasion are an exception and should be treated more aggressively.^[3]

Here, we report a case of unicystic ameloblastoma (UA) in a 35-year-old female with an unusual large multilocular

radiographic appearance on the right body of mandible. Multilocular appearance, though not uncommon in UA, the appearance of thin straight septa creates confusion in diagnosing the lesion. This case report illustrates the importance and complexity of differential diagnosis, as the management differs widely for all multilocular lesions.

Case Report

A 35-year-old female patient presented with a chief complaint of swelling in the right lower jaw back region since 3-4 months. Patient gave history of extraction of right mandibular second and third molars 2 years before the swelling appeared. On examination, extraorally, a non-tender diffuse swelling with variable consistency was present on right mandibular angle region. Intraorally, a well-defined swelling was present extending from mesial surface of right first molar to retromolar region, involving the edentulous region in relation to the right mandibular second and third molar teeth [Figure 1]. On palpation, it was non-tender, variable in consistency. Bicortical expansion was present. Based on these findings, provisional diagnosis of odontogenic keratocyst (OKC) was made. The lesion was then aspirated; a thin straw color fluid was obtained from the lesion, which was sent for protein analysis. The protein content of cystic fluid was estimated to be 3.9 mg%.

Orthopantomograph revealed [Figure 2], a huge well-defined multilocular radiolucency on the right body of the mandible, extending from the mesial surface of right mandibular first molar to ramus region. The periphery of the lesion is well defined, not surrounded by any sclerotic border. The internal aspect of the lesion is multilocular with large size locules. The septa within the lesion are straight and thin- etched and placed perpendicularly at the periphery, giving an appearance of a tennis racket like pattern. The lesion is causing resorption of both mesial and distal roots of right mandibular first molar. The inferior border of the mandible is intact. The cortices of mandibular canal also are intact.

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Multilocular lesions such as ameloblastoma, odontogenic myxoma, and central giant cell granuloma were considered under differential diagnosis based on radiological picture.



Figure 1: An intra-oral swelling involving right edentulous ridge in relation to mandibular second and third molar region with bicortical expansion



Figure 2: Orthopantomograph showing a well-defined multilocular lesion on right angle – ramus region. The septa within the lesion are thin and straight and placed perpendicularly at the periphery giving an appearance of tennis – racket. Resorption of first molar roots seen

However, positive aspiration of the lesion is contrast to these lesions. Based on aspiration, OKC, and DC were also considered under differential diagnosis. Residual cyst is considered as one of the differential diagnosis due to positive history of extraction prior to swelling. UA is considered as one of the differential diagnosis based on site, clinical picture and positive aspiration.

Incisional biopsy was performed and sent for histopathological analysis and a diagnosis of UA with mural proliferations was made based on histological picture [Figure 3]. Later the lesion was surgically excised and bone grafting was done to the patient. The excised specimen was also subjected to histopathological analysis, which confirmed the diagnosis of UA with mural proliferations. Post-operative follow-up of the patient had been satisfactory [Figures 4 and 5].

Discussion

Many benign lesions cause mandibular swellings,

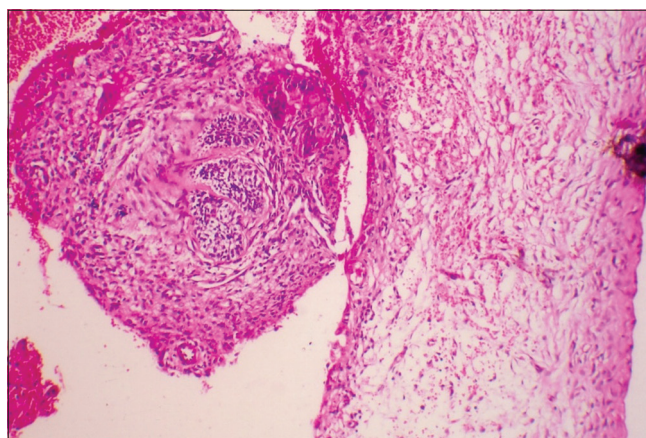


Figure 3: Histopathology picture showing an ameloblastomatous epithelium, with a cystic lumen into which the epithelial proliferations are seen



Figure 4: Immediate post-operative extraoral image of the patient



Figure 5: Immediate post-operative intraoral view of the patient with adequate mouth opening

whose origin can be, attributed to odontogenic or non-odontogenic causes. The most commonly encountered are ameloblastomas, radicular cysts, DCs, OKCs, central giant cell granulomas, fibro-osseous lesions and osteomas.^[4] The most common tumor of odontogenic origin is ameloblastoma, which develops from epithelial cellular elements and dental tissues in their various phases of development.^[2] The unicystic type of ameloblastoma is one of the least encountered variant of the ameloblastoma accounting for about 10-15% of all ameloblastomas.^[4-6]

Unicystic type appears more frequently in a younger population (3rd decade) than its solid counterpart (4th decade).^[7] In our case, the patient was 35-year-old. UAs were most commonly encountered in posterior mandible, followed by the parasymphysis region, anterior maxilla and the posterior maxilla.^[7] The ratio of the maxilla: Mandible is 1:7 for the dentigerous variant, versus 1:4.7 for the non-dentigerous type.^[8] In our case also the lesion was non-dentigerous type and was present on right body of the mandible extending up to the ramus region. Gender distribution shows a slight male predilection with a male: female ratio of 1.6:1. However, when the tumor is not associated with an unerupted tooth, the gender ratio is reversed to a male to female ratio of 1:1.8.^[4] Most of the UAs are associated with an impacted tooth, the mandibular third molar being involved most often.^[4] In our case, patient is a female, and the lesion is not associated with any unerupted/impacted teeth.

They are often asymptomatic until they are seen on a routine radiograph. As the lesion slowly enlarges, a slight, non-tender swelling becomes apparent on clinical examination. This swelling is the result of an expansion of the cortical plates of the jaw and can be identified by palpation as hard and bony.^[9] Our patient also showed a non-tender swelling with bicortical expansion.

Radiographically, UAs have been divided into two main patterns: Unilocular and multilocular. UAs have clear preponderance for the unilocular pattern.^[4] The radiological features of UA are typically unilocular and there is a round area of radiolucency. Therefore this lesion is often misdiagnosed as an OKC/a DC.^[5,10] It has been suggested that six radiographic patterns for UA can be identified ranging from well-defined unilocular to multilocular.^[11] Eversole *et al.*, identified predominant radiographical patterns for UA: Unilocular, scalloped, macromultilocular, pericoronal, interradicular, or periapical expansile radiolucencies.^[8] where as our case showed atypical multilocular appearance with some straight septa within the lesion.

Our case should be differentiated from other multilocular

lesions like solid ameloblastoma, central giant cell granuloma, odontogenic myxoma, and cystic lesions like OKC, DC, and residual cyst. Based on fluctuant consistency and positive aspiration all the multilocular lesions have been ruled out. OKC produces thick cheesy material instead of thin straw color fluid which is seen in our case. DC is ruled out as there is no impacted tooth present. Residual cyst also is ruled out because it will not produce so much of expansion as seen in our case.

Hemimandibulectomy was performed along with bone grafting. The specimen was sent for histopathological evaluation, which confirmed the diagnosis of UA.

In conclusion, UA, a type of ameloblastoma, presents with a variety of clinical, radiological and histopathological features. Hence, it presents as a challenge both for its diagnosis and treatment. There is always an on-going debate regarding the origin of UA. Immunohistochemical studies help us to know the nature of the lesion and also to differentiate the same from other cysts of odontogenic origin. Hence, it is essential that studies should be conducted on a large scale in order to know the origin and nature of the lesion.

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