

Computed tomography imaging features of multicystic granular cell ameloblastoma: a case report

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ABSTRACT

Multicystic ameloblastoma is a clinical variant of ameloblastoma. It is often locally aggressive with significant impact on patient morbidity and mortality. Ameloblastoma is often asymptomatic and is usually detected as an incidental finding during routine dental examinations. It presents slow and persistent growth; in some cases a gradual expansion of the jaw producing facial asymmetry may be noticed. Although the occurrence is rare, ameloblastomas can exhibit malignant behavior and metastasize. Clinical evaluation includes careful review of the patient's medical history and physical examination of the head and neck region. Radiographic imaging techniques are useful for locating the lesion, estimating its dimensions and assessing its effects on adjacent structures. Management of ameloblastoma is a challenge for all involved in the field of head and neck surgery. The aim of this paper was therefore to describe the tomographic features of a multicystic granular cell ameloblastoma in a 40-year-old white male patient.

Keywords: Ameloblastoma, Granular Cell Tumor, X-Ray Computed Tomography, Dental Radiography.

Aspectos tomográficos de ameloblastoma multicístico de células granulares: relato de caso

RESUMO

O ameloblastoma multicístico é uma variante clínica do ameloblastoma, muitas vezes localmente agressivo, com significante aumento de morbidade e mortalidade. O ameloblastoma é

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frequentemente assintomático, sendo identificado incidentalmente durante exames odontológicos de rotina. Trata-se de uma lesão de crescimento lento e persistente, podendo ser notada, em alguns casos, uma expansão gradual da mandíbula, produzindo assimetria facial. Ameloblastomas raramente apresentam um comportamento maligno. A avaliação clínica inclui uma revisão cuidadosa da história clínica do paciente, bem como exame físico da cabeça e pescoço. Modalidades de imagens radiográficas são úteis para localizar a lesão, avaliar suas dimensões e os efeitos sobre as estruturas adjacentes. O manejo da lesão representa um desafio para todos os profissionais envolvidos na área de cirurgia de cabeça e pescoço. Assim, o objetivo deste trabalho foi descrever as características tomográficas de um ameloblastoma multicístico de células granulares em um paciente do sexo masculino, leucoderma, de 40 anos de idade.

Palavras-chave: Ameloblastoma, Tumor de Células Granulares, Tomografia Computadorizada por Raios X, Radiografia Dentária.

INTRODUCTION

Ameloblastoma is a rare odontogenic tumor of the jaws that originates from the epithelium involved in teeth formation: enamel organ, odontogenic rests of Malassez, reduced enamel epithelium and odontogenic cyst lining (1,2). This a locally aggressive tumor that accounts for about 10% of odontogenic tumors (3) and 1% of all oral cancers (4). It is often asymptomatic and so discovered in routine radiographs, but swelling and expansion of the jaws are also common clinical manifestations (5). Radiographically, ameloblastomas are osteolytic, typically found in the tooth-bearing areas of the jaws, and may be either unilocular or multilocular. Radiographic margins are usually well defined and sclerotic, because ameloblastoma grows slowly and persistently (6).

Three types of ameloblastomas can be distinguished, mainly on the basis of clinical behavior and prognosis, as follows: multicystic, unicystic or peripheral (7). The multicystic type (also known as intraosseous or solid ameloblastoma) appears as multilocular or “soap bubble” radiolucencies. Unicystic ameloblastomas appear on radiography as unilocular radiolucent lesions and are often associated with an impacted tooth. Peripheral ameloblastomas occur in the gingiva and there is no bone involvement or radiographic features.

Clinical evaluation includes careful review of the patient’s medical history and physical examination of the head and neck. Imaging work includes both panoramic radiography and computed tomography (CT) (8). Panoramic images provide an overview of the dental arches and a close view of a large number of anatomical structures with low doses of radiation. In turn, CT is helpful when planning the surgery because it shows the entire tumor volume and the tumor’s relationships to surrounding structures, including soft tissue (9). Management of ameloblastoma poses a challenge for all involved in the field of head and neck surgery. Successful treatment requires not only adequate resection of the tumor but also reconstruction of the residual defect that is both functional and aesthetically acceptable (1,10). The aim of this paper is to describe the tomographic aspects of a multicystic granular cell ameloblastoma.

CASE REPORT

A 40-year-old white male patient was seen at the radiology clinic for evaluation of an asymptomatic swelling on the right side of the lower face. Panoramic radiograph showed radiolucency from the lower right third molar to the left central incisor (Figure 1). Inferiorly, bone resorption extended to the inner cortex of the lower margin of the mandible. A multilocular area with undulating borders and bony septa was identified. The location of the mandibular canal could not be determined. The Lower right premolars, first molar and second molar were missing. The lower central incisors and the right lateral incisor and canine exhibited external root resorption. Occlusal radiography showed minimal expansion of the bone cortex (Figure 2). Incisional biopsy findings suggested a diagnosis of granular cell ameloblastoma.



FIGURE 1 – Panoramic radiograph showing an extensive multilocular radiolucent lesion in the right mandible with tooth loss in the area.

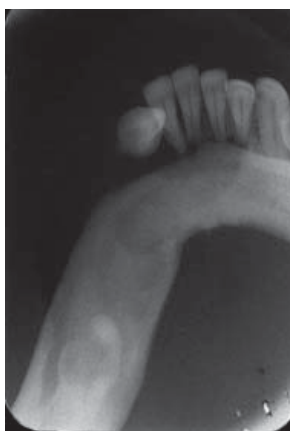


FIGURE 2 – Occlusal radiograph showing floating teeth in the anterior region, and discrete expansion of the bone cortex.

A helical CT scan (Philips Aura CT Scanner, Amsterdam, The Netherlands) was taken at 120 kV and 100 mA for surgical planning. Iohexol iodine based-contrast (Omnipaque 300; Sanofi Winthrop; Markham, Ontario, Canada) was injected intravenously for evaluation of soft tissue involvement. Multiplanar images were reconstructed with 1-mm-thick axial slices, a 0.5mm reconstruction interval and a matrix size of 512 x 512 (Figures 3 and 4). The three-dimensional reconstruction provided better definition of the limits of the lesion and improved understanding of the relationships between the structures involved.

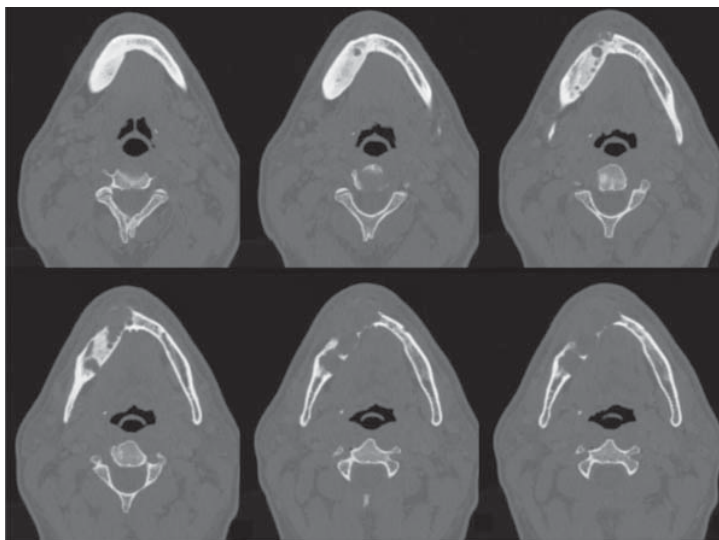


FIGURE 3 – Axial computed tomography sections.

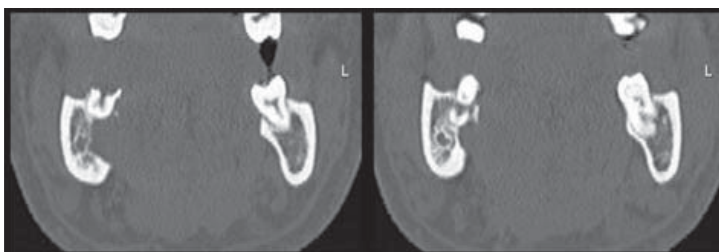


FIGURE 4 – Coronal computed tomography sections.

Features seen on CT imaging included a destructive lesion causing hyperostosis of the lower border of the mandible, with mutiloculated hypodense areas, and destruction of the buccal and lingual cortical plate. The buccal cortical plate was expanded and perforated. Soft-tissue involvement was not seen, since the lesion did not absorb contrast media. In coronal sections, we observed that the lesion extended to the distal root of the lower right third molar, adjacent to the lingual cortical plate.

DISCUSSION

Ameloblastoma is an epithelial odontogenic tumor with locally aggressive behavior and a high recurrence rate. It most often occurs in the lower molar region, and the mandible is affected five times more frequently than the maxilla (11). A study reporting on 60 cases of ameloblastoma showed that 69% arose in the angle of the mandible, 23% in the body of the mandible, and 8% in the symphyseal and parasymphyseal regions (12). In the present case, the lesion center was located on the right side of the mandibular body and extended from the midline to the lower right third molar. It has been reported that the mean age of patients with ameloblastoma is 36 with a range of 4-92 years. Our study is in line with these data, since the patient was a 40-year-old. In developing countries, ameloblastoma occurs in younger patients with equal sex distribution (11).

Although ameloblastoma has been described as an uncommon lesion in the scientific literature (1), one study in Sri Lanka analyzed 1677 cases of odontogenic tumors and found an occurrence of 48.7%. Furthermore, the multicystic type was the most common of the three types, with an occurrence of 63.7% (13). In a study investigating 340 Brazilian patients treated for odontogenic tumors between 1954 and 2004, there were 45.3% cases of ameloblastoma and 0.3% cases of ameloblastic carcinoma (14).

The radiographic and tomographic features of our case were fully compatible with multicystic ameloblastoma, which predominantly presents with one of two histological patterns: follicular or plexiform. Other histological variants include acanthomatous, desmoplastic, basal cell and granular cell (5). In this case, histological analysis revealed granular cell ameloblastoma, a variant first reported in 1918 by Krompecher (15). The current literature includes few reported cases of granular cell ameloblastoma, some of which eventually produced metastasis. Granular cell change is a rare histopathological entity (16), and the question of whether the presence of these cells is a degenerative process or is prognostic of an aggressive clinical course is still under discussion (17).

Rarely, ameloblastomas may exhibit malignant behavior including hematogeneous dissemination and can then be classified as metastasizing ameloblastomas or the more aggressive ameloblastic carcinomas. If an unlikely event such as pulmonary metastasis occurs, a chest radiograph should be considered (18). A number of different treatment approaches to ameloblastomas exist. Conservative surgical treatment includes enucleation, curettage, electrocauterization, excision and marsupialisation. Radical surgery depends on the tumor's histological and clinical presentation and includes marginal, segmental or total resection of the involved jaw with wide margins (2,12). Multicystic ameloblastoma has a high recurrence rate following simple enucleation (60-80%) (19). Surgical resection of ameloblastoma with safe margins is the best primary method for treating multicystic ameloblastomas to avoid recurrence (20).

FINAL CONSIDERATIONS

We have reported a case of multicystic granular cell ameloblastoma of the mandible in a 40-year-old male patient. Conventional radiographs were useful for detecting the lesion and giving an idea of its dimensions. CT provided reliable information about the location, shape, periphery and internal appearance of the Multicystic Ameloblastoma and its effects on surrounding structures. This imaging technique is a very good aid for establishing diagnosis and planning treatment and follow-up.

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