

Radiographic imaging pattern of ossifying fibroma mimicking ameloblastoma: a case report

• **Jefferson Rocha Tenório** Department of Oral Pathology, University of São Paulo, São Paulo, Brazil • **Solange Kobayashi-Velasco** Department of Oral Pathology, University of São Paulo, São Paulo, Brazil • **Fábio Daumas Nunes** Department of Oral Pathology, Special Care Dentistry Center, University of São Paulo, São Paulo, Brazil • **Marcelo Gusmão Paraiso Cavalcanti** Department of Oral Pathology, University of São Paulo, São Paulo, Brazil

ABSTRACT | Radiographic examinations complement the anamnesis and physical exam with the purpose of reaching diagnosis, prognosis and treatment planning. In this case report, a 48 year-old male Caucasian patient was referred to an oral and maxillofacial surgeon by a general practitioner after a panoramic radiography for treatment planning; the implant surgery follow-up portrayed a multilocular radiolucent image at the left posterior mandible. Based solely on the panoramic radiography, the diagnostic hypothesis was ameloblastoma. The surgeon decided to perform an incisional biopsy. However, during the procedure, the professional noted that the lesion was easily detached from the adjacent bone and opted for the total removal of the lesion, thus altering its diagnostic hypothesis to central ossifying fibroma (COF). The histopathological result confirmed the diagnostic hypothesis provided by the surgeon, i.e. COF. Although multilocular presentation is not common, COF should be considered in the scope of multilocular radiolucent lesions of the jaws. In addition, computerized tomography imaging exam complemented by surgical and histopathological aspects should be considered for establishing the final diagnosis and conducting the therapeutic approach.

DESCRIPTORS | Ossifying Fibroma; Radiology; Diagnosis.

RESUMO | **Padrão de imagens radiográficas para fibroma ossificante mimetizando ameloblastoma: um relato de caso** • Os exames radiográficos complementam a anamnese e o exame físico a fim de estabelecer o diagnóstico, o prognóstico e o planejamento do tratamento. Neste relato de caso, um paciente do sexo masculino, de 48 anos, foi encaminhado a um cirurgião bucomaxilofacial por um clínico geral após uma radiografia panorâmica para o planejamento do tratamento; o seguimento da cirurgia de implante retratou uma imagem radiotransparente multilocular na mandíbula posterior esquerda. Baseado apenas na radiografia panorâmica, a hipótese diagnóstica foi ameloblastoma. O cirurgião decidiu realizar uma biópsia incisional. No entanto, durante o procedimento, o profissional notou que a lesão era facilmente separada do osso adjacente e optou pela retirada total da lesão, alterando sua hipótese diagnóstica para fibroma ossificante central (FCO). O resultado histopatológico confirmou a hipótese diagnóstica do cirurgião, ou seja, o COF. Embora a apresentação multilocular não seja comum, o FCO deve ser considerado em casos de lesões radiotransparentes multiloculares das mandíbulas. Além disso, o exame de tomografia computadorizada complementado por aspectos cirúrgicos e histopatológicos deve ser considerado para estabelecer o diagnóstico final e conduzir a abordagem terapêutica.

DESCRITORES | Fibroma Ossificante; Radiologia; Diagnóstico.

CORRESPONDING AUTHOR | • **Jefferson da Rocha Tenório** Department of Oral Pathology, University of São Paulo, São Paulo, Brazil • **Av. Prof. Lineu Prestes, 2227** São Paulo, SP, Brazil • **05508-900** E-mail: jeffersonrtenorio@usp.br

• Received Dec 12, 2018 • Accepted Jan 15, 2018
• DOI <http://dx.doi.org/10.11606/issn.2357-8041.cldr.2019.152635>

INTRODUCTION

The decision-making process in oral and maxillofacial lesions depends on a diagnosis based on clinical and radiographic information and in many cases, a biopsy. Clinical data is obtained thorough anamnesis and physical exam. When a biopsy is performed, the surgical procedure complemented by the macroscopic aspect of the lesion that was removed may contribute for the suggestion of diagnostic hypotheses.¹

Radiographic exams such as periapical or panoramic radiographs are the most frequent exams employed by dental clinicians mainly due to their low cost. These exams may occasionally influence the professional towards a certain diagnostic hypothesis, and consequently, a specific clinical conduct.² However, radiographs may occasionally mislead the clinician. In some cases, a cone beam computed tomography (CBCT) exam may add important information in terms of surgical planning.³ Many lesions that are frequently described as expressing certain radiographic aspects may have different histopathological diagnoses than the initial hypothesis based on clinical and radiographic evaluations.⁴

Ameloblastoma is an odontogenic benign tumor that is originated from odontogenic epithelium. According to the World Health Organization (WHO) classification (2017), ameloblastomas are either conventional or unicystic. In a panoramic radiography, the conventional type is frequently described as an intraosseous multilocular radiolucency.⁵ Central ossifying fibroma (COF) is a benign fibro-osseous lesion that presents neoplastic behavior. It may appear as unilocular or multilocular, radiopaque, radiolucent or mixed radiopaque-radiolucent in a panoramic radiography.⁶ Even though they depict distinct histological characteristics, depending on the radiographic aspect, ameloblastoma may be included as differential diagnosis for COF.⁷

The purpose of this report is to describe a case of COF that mimicked an ameloblastoma on panoramic radiographic examination and address the radiographic aspects of these two lesions.

CASE REPORT

A 48 year-old male Caucasian patient was referred to an oral and maxillofacial surgeon by a general practitioner after a panoramic radiography for treatment planning; implant surgery follow-up portrayed a multilocular radiolucent image at the left posterior mandible. The lesion was located in the left mandible, extending from the alveolar ridge to the base of the mandible. This lesion had caused external root resorption in the left inferior second premolar and in the left inferior second molar (Figure 1).



Figure 1 | Panoramic radiograph. Multilocular image located at the left mandible, from tooth 34 to tooth 38, and from the jaw base to the alveolar ridge. The lesion induced external root resorption in teeth 34, 35 and 37.

In anamnesis, the patient reported no health problems and mentioned that he did not take any medicines. He described no discomfort nor pain. In the physical exam, the mucous membrane had normal characteristics and a slight increase in volume at the buccal aspect of the mandible.

Based on the clinical examination and panoramic radiography, the diagnostic hypothesis of ameloblastoma was suggested, and an incisional biopsy with intraoral surgical access was planned. A CBCT exam was not performed due to the nature

of the procedure (incisional biopsy). During the procedure, however, the surgeon overturned his initial hypothesis due to macroscopic characteristics in the involved tissue. The lesion presented a great cleavage plane and separated easily from the adjacent healthy bone tissue; the intraoperative characteristic led the surgeon to perform the complete removal of the lesion. The pathological tissue had a brownish color, fibrous consistency, and its fragments were easily detached from the osseous tissue, thus resembling a fibro-osseous lesion.

The histopathological examination of the specimen showed an intense proliferation of mesenchymal stem cells that were fusiform, had ovoid shapes, and produced significant amount of calcified material. This calcified material was mostly basophilic, spherical and acellular, compatible with cement, but areas of osteoid-like material could also be visualized. The lesion also had an important hemorrhagic component. With these characteristics, the diagnosis of COF was set (Figure 2).

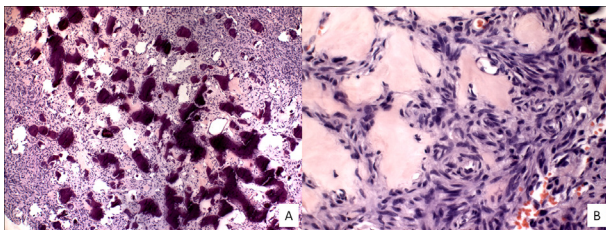


Figure 2 | Photomicrographs. A. (H&E, ×100) Area with extensive vascularization and presence of multiple basophilic calcifications of circumferential aspect that occasionally coalesce. B. (H&E, ×400) Mesenchymal stem cells with monotonous aspect, indistinct cell borders and oval nucleus. Absence of mitosis or anaplasia.

A clinical and radiographic follow-up with six-month intervals has been conducted for the past two-years with no evidence of relapse.

DISCUSSION

Several jaw bone lesions present typical imaging characteristics. However, many of them demonstrate similar imaging findings, which often makes diagnosis difficult and may hide the true biological

behavior of the underlying lesion. For this reason, some information are very important for narrowing the possible differential diagnoses, among them: age and sex of the patient, location of the lesion, relation with the involved teeth, margins of the lesion, and cystic or solid nature of the lesion and symptoms reported.⁸ Thus, because of the broad spectrum of pathological processes affecting jaw bones, there is a considerable overlap of imaging aspects at a panoramic radiography and CBCT exams³ and biopsy are often required to make the final diagnosis.⁹

Slotweg and El Mofty defined COFs as a “well-demarcated lesion composed of fibrocellular tissue and mineralized material of various appearances” present in the jaw bones of adult individuals, with histological variants more common in younger individuals such as juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma.¹⁰ The COF radiographically shows a well-circumscribed unilocular image that is initially radiolucent and, as it progresses, shows a mixed lesion composed of radiolucent and radiopaque areas. Mature lesions usually appear to be predominantly radiopaque involved by a radiolucent halo.¹¹

This study shows an unusual radiographic presentation of COF, which was characterized as a multilocular radiolucent lesion. A group of Brazilian researchers performed a study with the objective of analyzing the clinical-radiographic and microscopic features of a series of COF cases and, regarding the radiographic characteristics, the authors verified that although there were differences as to the density of the lesions, all the cases studied were unilocular lesions.¹²

COF lesions may present various radiographic aspects. A case described by Ramos-Perez et al. reported a patient with pain in the lower canine region. The periapical radiography indicated endodontic treatment with a radiolucent lesion at the periapical region. The clinician opted for the excision of the periapical lesion. During the surgical procedure, the clinician observed macroscopic characteristics of

fibro-osseous lesion, removed it and carried out a three-year follow up with no signs of recurrence.¹³

Misdiagnoses may also occur with ameloblastoma. In another case, a periodontal defect in the periapical radiography was treated as a persistent endo-periodontal lesion, as described by the authors. When the clinicians decided to perform an enucleation and send the material for a biopsy exam, the histopathological exam result was solid ameloblastoma. After one year, a recurrence focus resulted in a more invasive surgical procedure performed on the patient.¹⁴

Radiology examinations are an auxiliary method to orientate the clinician towards the right path in terms of diagnosis and treatment plan. Still, radiographs occasionally mislead the professional because of the superimposition of structures or diversity of aspect of the same lesion throughout its development.^{13,14} A series of three-dimensional imaging exams that complement conventional radiographs can be employed, since they overcome the aforementioned limitations and provide more specific information in terms of diagnosis and therapeutic options. The most frequently used imaging 3D exams are multislice computed tomography (CT) and CBCT. Also, magnetic resonance imaging (MRI), positron emission tomography combined with CT (PET/CT) and more recently positron emission tomography combined with MRI (PET/MRI) may be considered.²

High-resolution CT and CBCT play a major role for the assessment of lesion margins and their relationship with important anatomic structures, such as the inferior alveolar nerve.² A limitation of the present case is the absence of a more specific imaging exam that would bring valuable information to the surgical planning, such as CT, since the change of conduct and diagnosis was altered during the intraoperative period.

Ameloblastoma usually requires a long follow-up and recurrences may occur especially after

conservative treatment, while COF recurrence rate is lower due to the nature of the lesion and follow-up is still recommended.^{15,16} In this report, the patient is in follow-up without signs of recurrence.

CONCLUSIONS

In our case report, the panoramic radiograph depicted an ameloblastoma-like lesion that was recognized as a COF during the surgical procedure. Although multilocular presentation is not common, COF should be considered in the scope of multilocular radiolucent lesions in the jaws. In addition, CT imaging exam complemented by surgical and histopathological aspects should be considered for the establishment of the final diagnosis and conduction of therapeutic approach.

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