

Calcifying cystic odontogenic tumor radiographically mimicking a lateral periodontal cyst: clinical case report

Tumor odontogênico cístico calcificante mimetizando radiograficamente o cisto periodontal lateral: relato de caso clínico

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Abstract

The calcifying cystic odontogenic tumor (CCOT) is a rare odontogenic lesion derived from the remaining odontogenic epithelium of the maxilla or mandible. Its occurrence in the maxilla and mandible is similar, however, the canine region is the most affected when compared with other regions. The aim of this paper is to relate the case of a patient who presented a CCOT in the left region of the maxilla, between the canine and the first premolar, characterized by an increase in volume that was painful on palpation. The lesion presented a well circumscribed radiolucent radiographic image, however, presented no root resorption and radiopacities suggestive of dystrophic calcification, common signs in this tumor. With the diagnostic hypothesis of a lateral periodontal cyst, enucleation of the lesion was performed. The patient has been followed-up for around 15 months without signs of recurrence.

Descriptors: Odontogenic cyst, calcifying; Periodontal cyst; Maxilla/radiography

Resumo

O tumor odontogênico cístico calcificante (TOCC) é uma lesão odontogênica rara derivada do epitélio odontogênico remanescente da maxila ou mandíbula. Possui ocorrência semelhante em maxila e mandíbula, porém a região de canino é a mais afetada quando comparada com outras regiões. O objetivo deste estudo é relatar um caso de um paciente que apresenta um TOCC em região esquerda de maxila, entre o canino e o primeiro pré-molar, caracterizado com aumento de volume doloroso à palpação. Radiograficamente a lesão apresentava imagem radiográfica radiolúcida bem circunscrita, no entanto não apresentava reabsorção radicular e radiopacidades sugestivas de calcificação distrófica, sinais comuns neste tumor. Com hipótese diagnóstica de cisto periodontal lateral, foi feita enucleação da lesão. O paciente está em acompanhamento há cerca de 15 meses sem sinais de recorrência.

Descritores: Cisto odontogênico calcificante; Cisto periodontal; Maxila/radiografia

Introduction

The calcifying cystic odontogenic tumor or Gorlin's Cyst is an uncommon lesion with a variable clinical behavior and considerable histopathologic diversity. The calcifying odontogenic cyst (COC) was first described as a distinct entity by Gorlin *et al.*¹ in 1962, as a "calcifying epithelial odontogenic cyst". Only in 1971 was it recognized by the World Health Organization as a distinct pathologic entity. At present the WHO defines Gorlin's cyst as an odontogenic tumor, including all its variants²⁻⁴.

Clinically it presents as asymptomatic, slow developing, with predilection for the anterior segment (incisor / canine area), however, the mandible and maxilla appear to be affected with the same frequency⁵. It generally affects young adults in the third and fourth decades of life, without predilection for gender⁵⁻⁶. Radiographically, in the majority of cases, it presents an osteolytic, unilocular, radiolucent, well defined aspect, although it occasionally presents as a multilocular lesion. Radiodense structures are present in 50% of cases in variable quantities and forms⁷. Usually they may be associated with included and impacted teeth. The cortical bone plates are frequently thin and expanded, and may become perforated by the lesion, which may occasionally lead to dental displacement and resorption of the adjacent tooth⁷.

Histologically, the CCOT normally presents as a cystic cavity composed of a fibrous capsule, lined with an odontogenic epithelium formed by basal cells of the columnar or cuboid type with a cellular aggregate reminiscent of ameloblasts⁸. Below it there is the presence of a tissue reminiscent of the star-shaped reticulum of the enamel organ. A remarkable characteristic of this lesion is the presence of wide, circular cells, without nuclei and lightly eosinophilic, so-called ghost cells. In addition to this dysplastic dentin

can be observed, and occasionally, the cyst is associated with an area of hard dental tissue formation, which resembles an odontoma⁵. Lesions may also be found in association with other odontogenic tumors, such as ameloblastomas, ameloblastic fibroodontomas, calcifying odontogenic epithelial tumors and adenomatoid odontogenic tumors⁹⁻¹⁰.

The lateral periodontal cyst is another uncommon cystic lesion of odontogenic origin. The aim of this report was to describe a case of CCOT with a radiographic aspect similar to that of the lateral periodontal cyst.

Case report

The patient, a 26-year-old woman, presented at the Oral Diagnosis Service of the University Paulista, relating the chief complaint of "growth and pain in the gingiva" that had been developing for approximately 12 months.

On extraoral physical exam, discrete asymmetry was found in the region of the nasolabial sulcus with slight increase in volume on the left side. The intraoral exam showed an expansion in the region of the alveolar ridge on the left side of the maxilla, between the canine and second premolar, with a firm consistency, presenting curvature of the vestibular bone cortical (Figure 1). The patient related pain on palpation of the lesion; the teeth involved presented pulp vitality.

Radiographic exams were requested, in which a radiolucent, unilocular lesion, with well defined limits was found between the roots of teeth 23 and 24 (Figure 2). It was possible to observe root displacement, however, no root resorption and radiopacities suggestive of dystrophic calcification were found. With the diagnostic hypothesis of a lateral periodontal cyst, enucleation of the lesion was



Figure 1. Clinic exam showed an expansion in the region of the alveolar ridge on the left side of the maxilla, between the canine and second premolar



Figure 2. Radiographic exams showed an unilocular lesion, without root resorption and radiopacities suggestive of dystrophic calcification were found

performed. By histopathological analysis it was possible to observe the presence of cystic liquid with cholesterol crystals, an epithelial lining of which the basal layer cells presented a cubic aspect, in addition to a large quantity of ghost cells, confirming the final diagnosis as a calcifying cystic odontogenic tumor (Figure 3). The patient has been followed-up for around 15 months without signs of recurrence (Figure 4).

Discussion

The calcifying cystic odontogenic tumor is a benign cystic neoplasia and constitutes 2% of all benign odontogenic lesions. The lateral periodontal cyst is another uncommon cystic lesion of odontogenic origin. It represents less than 1% of odontogenic cysts. It generally presents as localized, restricted to the height of the middle third and lateral to the root of a vital tooth, and occasionally vestibular gingival tumefaction may occur. Radiographically a well cir-

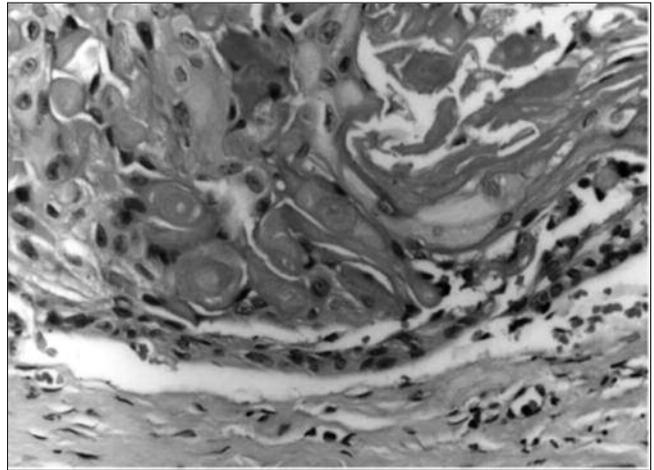


Figure 3. Microscopic analysis – it was possible to observe the presence of an epithelial lining of which the basal layer cells presented a cubic aspect, in addition to a large quantity of ghost cells



Figure 4. Clinic aspect of the region about 40 days after the surgery

cumscribed radiolucent area is observed. Normally the lateral periodontal cysts is localized between the premolar and canine (corresponding to 65% of cases), the male gender being most affected, in which the most predominant age ranges between the 5th and 6th decades of life¹¹⁻¹³.

The classical radiographic presentation of CCOT consists of a unilocular, radiolucent lesion, with radiopaque points within it, and it may be associated with an impacted tooth. In the reported case, the radiographic characteristics of the tumor did not present as classical, and mimicked those of a lateral periodontal cyst, since this normally presents a small radiolucent lesion, localized at the height of the middle third and lateral to the root of a vital canine or premolar.

There is consensus among authors with regard to the treatment recommended for CCOT, which would be surgical enucleation, presenting a very low rate of recurrence, being rate. However, if the CCOT is associated with pathology, the treatment must take into consideration the presence of the associated lesion¹⁴.

Conclusion

It is very important for the dentist to know how to recognize cystic lesions of the gnathic bones, as detailed exams as well as

the use of biopsy are necessary for an early diagnosis. Although the CCOT and lateral periodontal cyst are rare lesions, they present distinct behaviors and prognoses, and require distinct treatments.

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