J. Oral Diag. 2019; 04:e20190023.

CASE REPORT

Unusual presentation of a simple bone cyst in the coronoid process of the mandible with xanthomatous cells

Adriana Caroline Leite [1]
Eduardo Dallazen [1]
Fabio Augusto Ito [1]
Hedelson Odenir Iecher Borges [1]
Cristian Statkievicz [1]
Heliton Gustavo Lima [1]
Ademar Takahama-Jr [1]*

Abstract:

Simple Bone Cyst (SBC) is an uncommon jaw lesion considered a "pseudocyst" because the lack of epithelial lining. In turn, xanthomatous cells are abnormal macrophages, also called histiocytes, characterized by the abundant presence of lipid content. They are mainly related to a soft tissue injury called xanthoma, but can also be found in pre-existing intraosseous lesions or in intraosseous xanthoma variations. Thus, the objective of this study is to report a case of a 16-year-old male patient, referred with an asymptomatic radiolucent lesion in the right coronoid process. The cone-beam computed tomography confirmed the presence of an osteolytic lesion and an exploratory surgery was indicated. During the surgical procedure, an almost empty cavity was found, with small fragments of soft yellowish tissue, which was removed. The microscopic analysis revealed the presence of xanthomatous cells. After 3 months a total bone healing was noted. Clinical and radiographic follow-up of the case was performed for 1 year, without signs of recurrence. It is very important the association of clinical and histopathological characteristics for the diagnosis, mainly in cases with atypical radiographical, clinical and histopathological presentation.

Keywords: Bone Cysts; Biopsy; Panoramic Radiography; Tomography

[1] Universidade Estadual de Londrina, Departamento de Medicina Oral e Odontologia Infantil - Londrina - SP - Brasil.

Correspondence to:
Ademar Takahama-Jr.
E-mail: ademartjr@uel.br

Article received on October 31, 2019.
Article accepted on December 9, 2019.

DOI: 10.5935/2525-5711.20190023

INTRODUCTION

The Simple Bone Cyst (SBC) is an uncommon jaw lesion described as a "pseudocyst" due to the lack of an epithelial lining. The etiopathogenesis is uncertain therefore it is known by variety of names, such as traumatic bone cyst, hemorrhagic cyst, solitary bone cyst and idiopathic bone cavity. Nevertheless, some causal factors are suggested: bone tumor degeneration, altered calcium metabolism, low-grade infection, local alterations in bone growth, venous obstruction, increased osteolysis, intramedullary bleeding, local ischemia, or a combination of such factors[1].

The SBC is more common in the posterior region of the mandible, usually asymptomatic and discovered through routine radiographic exams. Radiographically, it presents as a well defined radiolucent lesion which can project between the teeth roots without resorption[2,3]. During the surgical exploration, it is usually found an empty cavity or filled with blood, serous fluid or both[4,5]. The surgical exploration with curettage of cavity walls generally presents satisfactory results with bone formation, but the clinical and radiographic follow-up is mandatory[2].

The foam cells are abnormal macrophages characterized by the abundant presence of lipid content. They are mainly related to a soft tissue injury known xanthoma, however they can also be found in preexistent intraosseous lesions or in intraosseous variant of xanthoma[6].

Thus, we present a rare case of SBC in the coronoid process of the mandible with the presence of foam cells in the histopathologic analysis.

CASE REPORT

A 16-year-old male patient was referred to by his dentist for evaluation of a radiolucent lesion next to the coronoid process, discovered in the panoramic radiograph requested for evaluation of the third molars. On the physical examination, there was no asymmetry or swelling. On radiographic examination a circumscribed radiolucent lesion was observed next to the right coronoid process, measuring about 10mm x 10mm (Fig. 1).

The patient denied any symptoms in the region. Computed tomography was requested and revealed a well delimited hypodense lesion with discreet expansion of the outer cortical bone, without association with the inferior alveolar nerve (Fig. 2). With the main diagnostic hypothesis of odontogenic keratocyst or simple bone cyst an ambulatory curettage of the bone cavity under local anesthesia was performed. During the surgery, a bone cavity almost empty was noted, with small fragments of yellowish soft tissue in the lower part of the cavity that was removed (Fig. 3). Microscopically, fragments of connective tissue were found with presence of xanthomatous macrophages (Fig. 4).

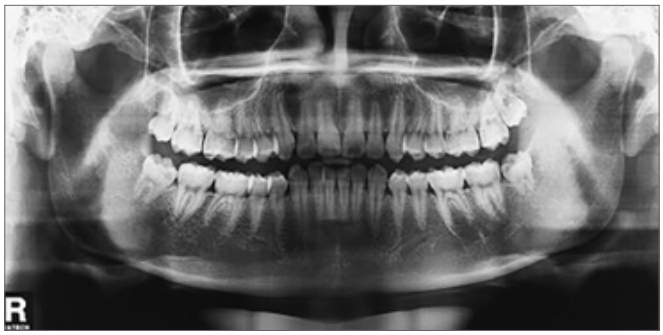

Figure 1. Panoramic radiograph showing a radiolucent circumscribed lesion in the right coronoid process of the mandible.

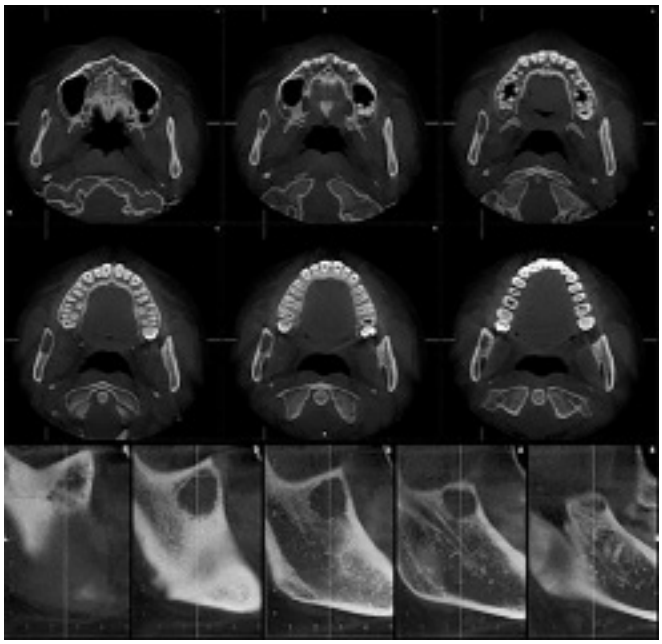

Figure 2. Cone beam computed tomography revealing a hypodense lesion.

Radiographic examination 3 months after the surgical procedure revealed bone healing of the region. In the follow-up of 1 year, no signs of recurrence were noted (Fig. 5). According to the clinical, radiographical, transurgical, histopathological and on the follow-up, the diagnosis of SBC with xanthomatous cells was stablished.

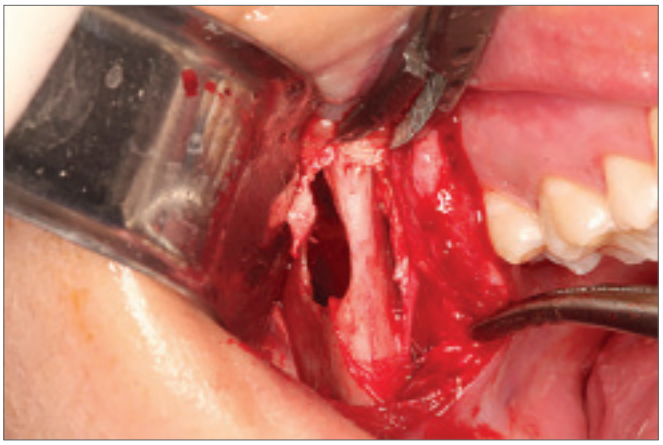

Figure 3. Bone cavity observed during the surgery.

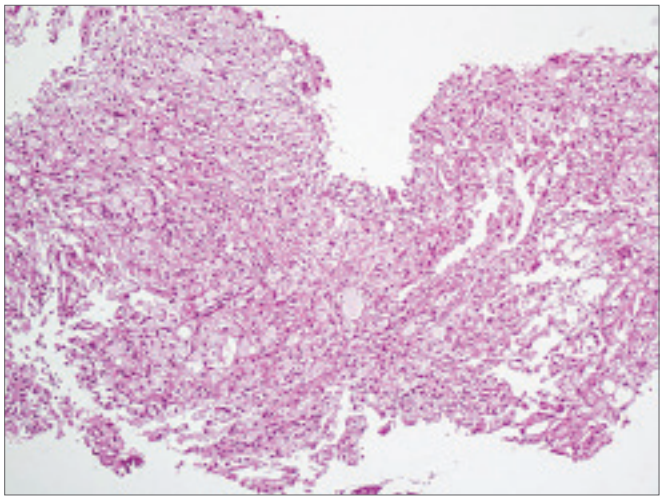

Figure 4. Histopathological aspect showing a connective tissue with predominance of xanthomatous macrophages. (HE 200x).

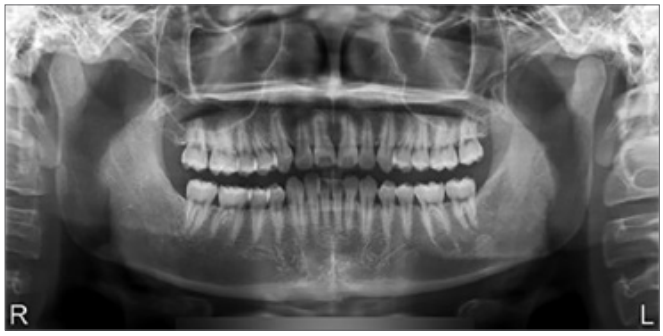

Figure 5. One year of radiographic follow-up showing total bone healing.

DISCUSSION

In a systematic review recently published by Chrcanovic & Gomez[7], analyzing 1253 previously reported cases of SBC, there were an equally prevalent in the two sexes, with a mean age 20.4 years, most of them located in the posterior region of the mandible, asymptomatic, radiolucent unilocular lesion without bone expansion, cortical bone thinning (but without cortical perforation), and pulp vitality of the involved teeth. They also found that only one in every four cases was preceded by a traumatic event.[7]

According to the literature, the most common localization of SBC is the posterior body of mandible. Strabbing et al.[8], reviewing 121 cases of SBC, found 8 cases affecting the mandibular ramus, but they didn't specify the precise location of the ramus. Another study of Zhang et al.[9] showed that mandibular ramus was affected in 13 reported cases in the literature. Two of these cases were located in condylar process and none in the coronoid process. As far as we know, there is no reported case of a simple bone cyst in the coronoid region. Sabino-Bezerra et al.[10] reported 6 cases of atypical presentations of simple bone cysts of the mandible, and none of them in the same location of our case, or either the atypical histopathological feature.

SBC generally presents an empty cavity or with blood, serous fluid or both inside[1,4,5]. The presence of foam cells inside of SBC has not been reported until now. When the foam cells were found in the histopathological examination of our case, a possible diagnosis of xanthoma was considered, which is also rare in these location However, xanthomas are usually present as a solid lesion[11-13]. The intraosseous xanthoma is uncommon and typically presents as osteolytic, expansive and painless process. It is extremally rare in the jaws being sparse reported in the literature. This lesion differs from the others more common in the jaws and don't have hyperlipidemia association like the xanthomas found in other bones, being considered by some authors as a distinct entity[11,14].

The xanthoma was encapsulated in 4 cases related by Daley et al.[11]. In cases reported by other authors, there is lack of information about the presence or absence of capsule or capsule was not found[11,15-17]. The xanthoma etiopathogenesis is uncertain and it is not determined if should be considerated an aggressive, reactive neoplasia or a low intensity neoplasic process. However, the commonly performed treatment was curettage of the surrounding bone and, in some cases, only partial excision[12,17]. Both treatments were satisfactory and did not present recurrence in the reported cases.

Some authors classify the xanthomas as: xanthomatous variant - when xanthomatous changes are found in preexisting bone lesions; secondary xanthoma

- lesions associated with hyperlipidemia; and primary xanthoma - those not associated with hyperlipidemia[18,19]. Following the suggestion of these authors, the reported case could be considered a xanthomatous variant of simple bone cyst.

CONCLUSION

Finally, it is very important the association of clinical and histopathological characteristics for the diagnosis. Cases with atypical radiographical, clinical and histopathological presentation, like ours, may need a close follow-up, and sometimes the final diagnosis depends on this to be performed.

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