

A simple mandibular bone cyst with remarkable tooth resorption

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ABSTRACT | This article reports an unusual case of a simple mandibular bone cyst with remarkable tooth resorption and pain. An 18-year-old woman was referred to our oral and maxillofacial surgery and trauma clinic, complaining of pain in the left third mandibular molar region. A radiographic exam revealed a well-demarcated radiolucent lesion in the left mandible with marked first molar distal root resorption. Based on the differential diagnoses, an incisional biopsy was performed through a bone window, under local anesthesia. Surgical exploration revealed a simple bone cavity filled with sanguineous fluid, with no cyst wall or tumoral tissue. Histopathological examination revealed thin, fibrous connective tissue with no epithelial coverage and permeated by extensive hemorrhage. Painful symptoms seemed to be caused by a caries lesion in the adjacent second molar and were resolved by tooth restoration with composite resin. A radiographic postoperative follow-up exam after two months suggests that there was complete bone healing and that tooth resorption had seemingly ceased. The final diagnosis was a simple bone cyst with unusual tooth resorption.

DESCRIPTORS | Bone Cysts; Tooth Resorption; Surgery, Oral; Pathology, Oral; Diagnosis.

RESUMO | **Reabsorção radicular extensa associada a cisto ósseo simples mandibular** • Este artigo relata um caso incomum de um cisto ósseo simples em mandíbula que apresentou reabsorção radicular e dor. Uma paciente do sexo feminino, com 18 anos de idade, foi encaminhada ao nosso serviço de cirurgia e traumatologia oral e maxilo facial com queixa de dor na região do terceiro molar inferior esquerdo. O exame radiográfico revelou uma lesão radiolúcida bem delimitada localizada na mandíbula esquerda com reabsorção acentuada da raiz distal do primeiro molar. Com base nos diagnósticos diferenciais, foi realizada biópsia incisional sob anestesia local por meio da exposição de uma janela óssea. A exploração cirúrgica da lesão revelou uma cavidade óssea simples preenchida com fluido sanguíneo, sem a presença de parede cística ou de tecido tumoral. O exame histopatológico revelou delicado tecido conjuntivo fibroso sem cobertura epitelial entremeado por extensa hemorragia. Os sintomas dolorosos pareciam ser provenientes de lesão de cárie no segundo molar. A queixa de dor cessou com a restauração do dente por meio de resina composta. O exame radiográfico após três meses de acompanhamento pós-operatório sugere completa reparação óssea e aparente interrupção da reabsorção da raiz dentária. O diagnóstico final foi cisto ósseo simples, acompanhado de incomum reabsorção do dente.

DESCRITORES | Cistos Ósseos; Reabsorção de Dente; Cirurgia Bucal; Patologia Bucal; Diagnóstico.

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INTRODUCTION

The simple bone cyst (SBC), also known as traumatic bone cyst, hemorrhagic bone cyst, solitary bone cyst, and idiopathic bone cavity, was first described by Lucas *et al.* in 1929,¹ and was classified under the name of simple bone cyst in the latest World Health Organization (WHO) classification (2005).² This lesion is described as an intraosseous, pseudocystic lesion lined with a thin fibrovascular membrane that is not surrounded by an epithelial lining, and that is usually either empty or filled with serous or sanguineous fluid. Radiographically, it is described as a well-defined, radiolucent lesion, surrounded by a thin radiopaque contour. The presence of scalloped margins extending between the dental roots is frequently observed. Clinically, these lesions appear as an empty cavity often filled with a serous or bloody fluid. Histopathological examination typically reveals only a tiny connective tissue sample permeated or not by hemorrhage. Fibroblasts and giant cell-like osteoclasts can sometimes be seen. The presence of congested capillary vessels is uncommon.³ The sites most affected by SBCs are the long bones (90%), with a high prevalence in the metaphyseal region of the humerus and femur (65% and 25%, respectively). Jawbone lesions are less common (1%–10%). The mandible is more commonly affected than the maxilla (4:1), with a predominance of the molar and premolar regions.⁴

CASE REPORT

An 18-year-old woman was referred to the oral and maxillofacial surgery and trauma service of the Luzia de Pinho Melo Clinical Hospital, in Mogi das Cruzes, São Paulo, Brazil. The patient's complaint was painless swelling in the left mandible body. Further inquiries revealed absence of previous trauma. Extra- and intraoral physical examinations revealed no abnormal findings around the lesion. Manual palpation of the swollen area revealed

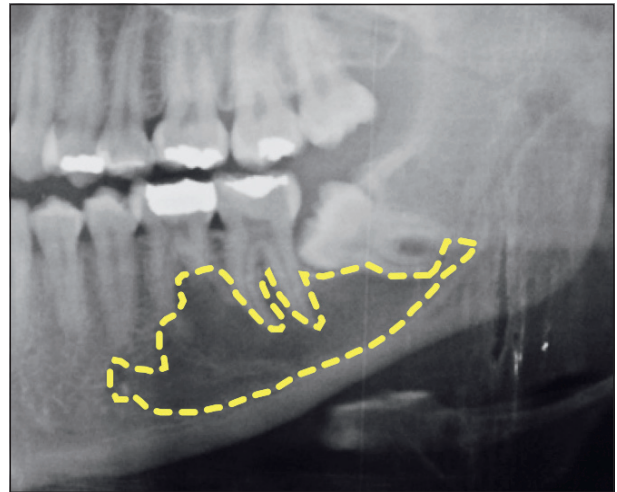


Figure 1 | Panoramic radiograph image showing a well-demarcated, radiolucent, unilocular lesion with scalloped margins in left mandible (yellow dashes). The lesion infiltrates the second molar region and causes a remarkable resorption in the distal root of the first molar.

pain. Painful lymph nodes were nonexistent. The general health status of the patient was good, and her family history was noncontributory. Routine laboratory values were within normal ranges. Radiographic panoramic images exhibited a relatively well-demarcated, radiolucent, unilocular lesion with scalloped margins, penetrating among dental roots associated with the inferior left third molar (Figure 1). A remarkable resorption of the distal root of the first molar could be observed. Differential diagnoses included odontogenic keratocyst, ameloblastoma, central giant cell lesion, unicystic ameloblastoma and other cystic lesions.

Lesion exploration through a bone window was performed under local anesthesia. After vestibular flap reflection and exposure of the external oblique ridge, a translucent lesion could be seen through an extremely thin bony wall. The bony wall membrane-like tissue was detached, and the cystic cavity was opened roughly. The lesion consisted of a bone cavity with no evidence of a cyst wall or tumoral tissue. Moreover, the crown of the left third molar was found to be impacted. Histopathologi-

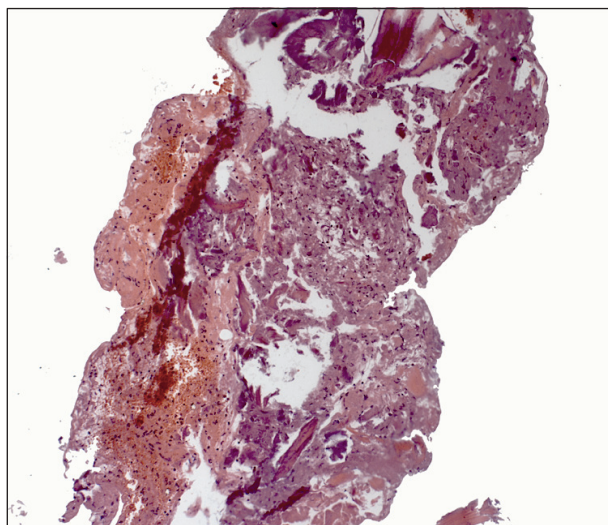


Figure 2 | Histopathological micrograph of the excised lesion exhibiting poorly collagenized tissue, bleeding and small fragments of bone.

cal examination showed the presence of a thin and poorly collagenized tissue, permeated with extensive hemorrhage. No epithelial lining could be observed surrounding the lesion. Focal bacterial colonies could also be observed (Figure 2). This description is consistent with an SBC.

The postoperative course was uneventful. Postoperative follow-up after 2 months showed satisfactory bone healing with partial regression of the lesion (Figure 3). An accurate pan-radiographic examination revealed the presence of a caries lesion in the inferior right second molar. This lesion was treated successfully with zinc oxide eugenol (ZOE), and definitive aesthetic restorative treatment was later performed. To date, no evidence of recurrence has been recognized.

DISCUSSION

SBC is a non-neoplastic bone lesion classified as an intraosseous pseudocyst by the WHO in 2005.² This lesion may be characterized by the presence of an osseous asymptomatic cavity with no epithelial lining that is usually an empty space or a space filled with serous fluid. It is classified by the WHO

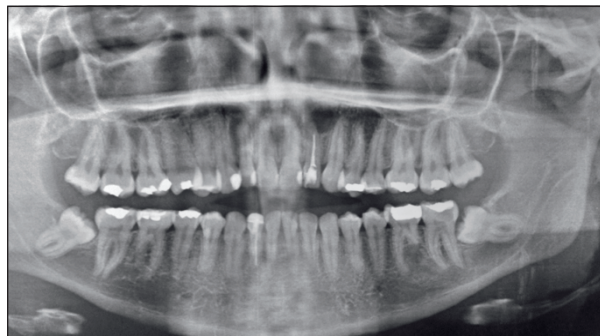


Figure 3 | Two-month postoperative panoramic radiograph reveals bone healing. Note the root defect in the left mandibular first molar.

as part of a group of bone lesions that include the ossifying fibroma, fibrous dysplasia, central giant cell lesions, aneurysmatic bone cysts, and cherubism. Ever since the SBC was first described by Lucas in 1929,¹ medical literature has extensively described the main characteristics and the recommended management for treating this lesion. Most of the cases described have been found in patients between the age of 20 and 30 years, and are usually radiolucent unilocular lesions with no or only slight bone expansion, followed by cortical thinning. Radiographic images show lesions with corticated scalloped margins extending between teeth roots. Rarely can tooth root divergence, bone cortical loss or root resorption be seen. Generally, SBCs do not cause cortical bone expansion.^{4,6,7} Although this case presented with an unusual swelling of the left mandibular body, Martins-Filho *et al.* (2012) revised 26 cases of SBC and found bone expansion only in 8% of cases.

Undoubtedly, the most distinguishing characteristic of the lesion presented was tooth resorption. Nakaoka *et al.* (2013) reported another unusual case of a simple mandibular bone cyst with remarkable tooth resorption presented in a 52-year-old woman.⁵ However, there are no similar reports of SBC with noticeable tooth root resorption. Tooth root resorption is a phenomenon frequently observed in both physiologic and patho-

logic processes. It occurs in primary teeth and has been reported in association with orthodontic tooth movements, occlusal traumatism, periodontal disease, periapical granulomas, and reimplanted teeth. Sometimes, a cyst or neoplasm may induce root resorption of the adjacent teeth.^{2,7,8}

In rare cases, SBC have been reported to heal naturally during the follow-up period.⁹ However, in addition to the SBC's own set of radiographic characteristics, some SBCs are difficult to distinguish from true cysts, resulting in SBC misdiagnosis and the patient's exposure to unnecessary surgical procedures. The employment of a magnetic resonance image (MRI), especially dynamic contrast-enhanced MRI (DCE-MRI), can provide useful information for distinguishing SBCs from other cysts.¹⁰

The confounding painful symptom in the present case seemed to result from a caries lesion in the adjacent teeth. This implies that an accurate physical examination associated with a reliable image exam evaluation should be performed. The

impacted inferior third molar in the left mandible was not removed in the surgical exploration, since it would augment the risk of pathological fracture from bone loss.

The recurrence rate of SBCs has been described as greater than 20%, especially in regard to abnormal SBCs.⁶ However, after a two-month postoperative assessment of the present case, no recurrence could be observed. Nonetheless, we believe that a careful, long-term follow-up is recommended for atypical SBCs, including this case.

CONCLUSION

A simple bone cyst is a frequent lesion, mostly found in women, between the age of 20 and 40 years. Tooth resorption or other complications related to these cysts are very rarely observed, and have been reported to self-resolve in some cases. This case reported an unusual manifestation of this pathological entity treated successfully with surgery.

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