

## CASE REPORT

# Solitary bone cysts—A rare occurrence with bilaterally symmetrical presentation

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**ABSTRACT**

Solitary bone cysts (SBCs) are bone cavities that lack a true epithelial lining. They are more commonly seen during the first 2 decades of age. Very few cases have been reported over 40 years of age. SBCs are usually discovered as an accidental coexisting finding during a routine radiologic examination or during another unrelated dental complaint. They present as a unilocular or multilocular radiolucent lesion associated with vital teeth with mild or no cortical expansion. Bilateral presentation is however very rare. We present a case of 52-year-old female patient with bilateral presentation of SBCs.

**Key words:** Solitary bone cyst, simple bone cyst, traumatic bone cyst

## INTRODUCTION

A solitary bone cyst (SBC) or simple, idiopathic, traumatic, hemorrhagic or osteodystrophic bone cyst is an empty or fluid-filled intrabony cavity that is devoid of an epithelial lining.<sup>[1]</sup> It manifests primarily in the long bones and rarely presents in the jaws.<sup>[2]</sup> It was first recognized by Virchow and later described by Jaffe.<sup>[3]</sup> Its designation as a pseudocyst may be attributed to its cyst like radiographic appearance or an intraoperative presentation as an intrabony cavity.<sup>[1]</sup> Occurrence of SBC is generally seen during the first 2 decades of life with higher male predilection.<sup>[2]</sup> SBC commonly are unilateral in occurrence, which has been classified as a classical distinguishing feature by few. However, bilateral presentations have been rarely documented.<sup>[4]</sup> The region between the canine and the third molar in the mandible is the most common site of presentation with the mandibular symphysis being the second most common site. Very rarely it occurs in the maxilla.<sup>[5]</sup> Radiographic features include a well-defined, unilocular radiolucency with scalloped margins. The radiolucency may be periapical or may involve the interdental and interradicular spaces without compromising the pulpal vitality of the associated teeth.<sup>[6]</sup> Very rarely expansion of the cortical plate is noticed. Diagnosis is generally confirmed only after surgical management. Intraoperatively, the cavity is

empty and no epithelial lining is present. In most of the cases, histopathological diagnosis revealed normal bone and fibrous tissue. Occasionally, erythrocytes and giant cells are seen.<sup>[5]</sup> The case discussed in this report elicits a marked change in the age of presentation and bilateral occurrence in the posterior mandible.

## CASE REPORT

### Clinical presentation

A 52-year-old female patient came to our private practice with a complaint of pain in the left lower jaw and teeth. On examination she revealed the presence of a large disto-occlusal carious lesion on the left lower second molar which was tender on percussion. This was indicative of periapical periodontitis of the above mentioned tooth. A routine orthopantomogram was suggested to assess the extent of the carious lesion and assess the general dental status of the patient.

### Radiographic features

Orthopantomogram revealed the presence of bilateral radiolucencies with clearly defined borders in the periradicular region of the mandibular third molars, which were roughly 3 × 2 cm in dimensions [Figure 1]. The radiolucencies involved the interdental and interradicular spaces of the mandibular second and third molars with no resorption of involved roots. There was no evidence of cortical expansion clinically and hence was not investigated by imaging. The radiograph also confirmed the presence of a disto-occlusal carious lesion of the left mandibular second molar with involvement of the pulp and periapical region.

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## Surgical management

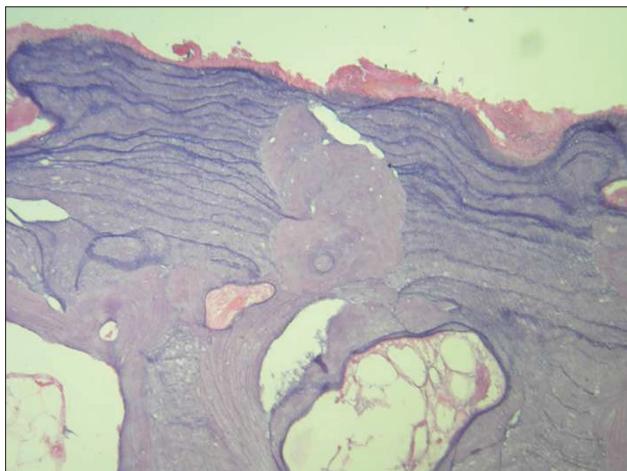
A fine-needle aspiration was performed for the patient, which provided a negative yield on both the sides and the patient was planned for enucleation under procedural sedation. The second and third molars were extracted bilaterally which gave adequate exposure to the cystic cavity. Intraoperative examination of the cystic cavity revealed empty spaces with no lining [Figure 2]. A provisional clinical diagnosis of traumatic bone cyst was made and the lesion was thoroughly curetted to induce fresh bleeding. The curetted material with a small bone specimen and extracted teeth were submitted for histopathological examination. The wound was closed primarily with absorbable sutures.

## Histopathology

Very less amount of tissue were available for the microscopic study. Histopathological examination of sections stained with hematoxylin and eosin showed cystic cavity with hemorrhagic lining and bone suggestive of solitary or traumatic bone cyst (SBC) [Figure 3].



**Figure 1:** Orthopantomogram (OPG) demonstrating bilaterally symmetrical periradicular cystic radiolucencies in relation to the mandibular third molars



**Figure 3:** Photomicrograph showing a cystic cavity with a hemorrhagic lining and bone demonstrating reversal lines representing normal surrounding bone. (H&E stain, x100)

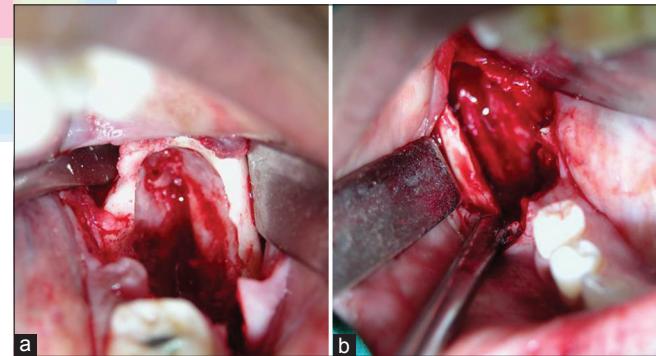
The specimen demonstrated the presence of bony trabeculae exhibiting reversal lines, thin fibrous connective tissue and extravasated red blood cells (RBCs) suggestive of normal bony wall. No cystic lining epithelium was evidenced in the section which is characteristic of solitary bony cyst.

## Follow-up

The patient was followed-up for a period of 3 years with a radiograph taken once a year. The orthopantomogram taken after 3 years of surgery, revealed complete resolution of the lesions bilaterally with good bone regeneration and obliteration of the cystic spaces [Figure 4].

## DISCUSSION

SBC, is an uncommon cyst representing about 1% of all the jaw cysts. They are osteolytic lesions forming a cavity with either a geodic or polymorphous shape and may be empty or filled with blood, serum or serohematic liquid.<sup>[2]</sup> The definition of SBC by Rushton still holds good in the description of the same as a single cystic lesion with no epithelial lining, no signs of acute or chronic infection and may be empty or fluid-filled. Clinically the cysts generally appear as isolated, nonsymptomatic, painless cystic lesions in the bone. Rarely, they present as multiple lesions where more than one cyst is seen. The most common age



**Figure 2:** (a and b) intraoperative picture demonstrating bilaterally empty cavities in the bone



**Figure 4:** Post-operative OPG taken after 3 years showing complete healing of the bony cavities with no signs of relapse

of presentation is in the first 2 decades of life, with the mandibular body being the most common anatomical site of presentation.<sup>[7,8]</sup> The posterior mandible follows next with few occurrences reported in the maxilla.<sup>[9,10]</sup> Some patients demonstrate a swelling as a presenting complaint and may have cysts associated with cortical expansions.<sup>[7]</sup> There are numerous theories regarding its pathogenesis including degeneration of bony tumors, faulty calcium metabolism, low-grade infection, disturbance in bone growth and excessive osteolysis. The most widely accepted theory by Pommer advocates intramedullary hemorrhage secondary to trauma to bone, clot liquefaction, enzymatic degradation of the surrounding bone and finally cystic cavitation. The presence of multiple enzymes and metalloproteinases also are favorable to the above mentioned theory of vascular osteolysis. It is speculated that the amount of fluid in the cavity diminishes with age of the lesion until the cavity eventually gets filled with gas, which in turn causes cyst enlargement.<sup>[2,11]</sup> SBC could also be associated with the presence of florid cementoosseous dysplasia, aneurysmal bone cysts and cementomas.<sup>[3]</sup> Traumatic events including dental extractions<sup>[11]</sup> have been believed to be the main cause of SBCs and yet literature reveals that clinical history of trauma is elicited only in about 2% of the patients,<sup>[12]</sup> which is very similar to our case where there was no relevant history of trauma to the teeth or jaws. A radiographic study which evaluated the characteristics of SBCs gave a clearer picture of traumatic events leading to their presence, where almost 30% of the lesions showed X-ray evidence of trauma including incomplete fractures, hairline fractures, unhealed alveolus, extractions and disimpactions of teeth.<sup>[8]</sup>

Most often SBC is diagnosed on routine radiographic examination. It classically presents as a homogenous unilocular radiolucency with cone-shaped condensation of the bone at the periphery of the lesion and may extend interdentally between the roots of the teeth showing scalloping.<sup>[3,13]</sup> Radiographic characterization of 44 lesions revealed that 64% of the lesions were conical, while 16% each were either irregular or oval and 4% were round.<sup>[8]</sup> The role of imaging modalities in diagnosis of SBCs may be augmented with the use of MRIs which show homogeneous intermediate signal intensities which are indicative of fluid in the cavity. Furthermore dynamic MRIs are suggestive of fluid movement from the surrounding medulla into the cyst.<sup>[14]</sup> The diagnosis of SBC is more radiographic, complemented with the clinical findings during surgical management. However, histopathology, if any tissue is retrieved and clinical use of pulp vitality testing of associated teeth provide valuable information in understanding the lesion.<sup>[10]</sup> SBCs are commonly managed by curettage of the bone wall and induction of fresh bleeding into the cavities. Spontaneous resolution of the lesion due to organization of the formed blood clot is proposed as the normal sequel leading to complete resolution. Application of gel foam, allogenic bone grafting with platelet-rich plasma (PRP), injection of blood

with hydroxyapatite and bone chips have also been proposed to have good results.<sup>[3,13]</sup>

Presentations of multiple SBCs, cortical expansions and evidence of clinical symptoms of neurological deficits like hypoesthesia have been reported as atypical presentations of SBCs. It has been reported in a large series of multiple presentations that two out of 34 presentations were asynchronous and 20% showed presence of three cysts. It also revealed that there was a 1:1.8 predilection for females, 44% showed cortical expansions and 65% of the cysts were empty cavities with 35% showing intracavitory fluid.<sup>[12]</sup> Recurrence is more common with cysts showing cortical expansions with rates ranging between 1.6 and 26% and average time for recurrence being 24–30 months mandating a yearly follow-up for a minimum of 3 years. Bone healing is generally complete by 12–24 months and is usually uneventful with the exception of a single case report discussing evolution and expansion of the cyst over a period of 4 years, which was managed with a similar modality of surgical exploration with a radiological follow-up for 3–4 years post-surgery.<sup>[15]</sup>

Variation in presentation is always a diagnostic enigma and mandates a thorough differential diagnosis. SBCs have been diagnosed rarely with pain and swelling as presenting complaints with some of the lesions showing cortical expansions. Variations in the form of unusual anatomical locations, where two instances of SBCs in the zygoma have been reported. A rare case of recurrence of the lesion has also been reported.<sup>[14]</sup>

## CONCLUSION

Though simple in their manifestation and indicating extremely good prognosis, SBCs still form a group of lesions which may exhibit challenges in diagnosis. The occurrences of multiple, synchronous lesions are rare and when present are seen more often in the first 2 decades of life and generally in the body of the mandible. Our report discusses atypical presentation of this cyst in a female patient in her 6<sup>th</sup> decade of life which presented as bilaterally symmetrical lesions in the posterior mandible. When identified early and managed with surgical exploration and radiological follow-up, these lesions show complete resolution.

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