

Solitary Bone Cyst of Posterior Maxilla: A Rare Presentation

Abstract

Solitary bone cyst (SBC) is an uncommon, nonneoplastic osseous lesion that mainly affects metaphysis of long bones and rarely presents in jaws. Due to the lack of true epithelial lining, it is considered as a pseudocyst. It is generally asymptomatic and often discovered incidentally during routine radiographic examination as well-defined unilocular or multilocular radiolucent lesion in the posterior mandible mainly in the first two decades of life. Here, we report a very rare case of a 15-year-old female patient having a lesion in the posterior maxilla with clinical, radiological, and histopathological presentations of SBC.

Keywords: Maxilla, solitary bone cyst, unilocular radiolucency

Introduction

Solitary bone cyst (SBC) is an osteolytic lesion forming a cavity with either a geodic or polymorphous shape and may be empty or filled with blood, serum, or serohematic liquid.^[1] SBC has different synonyms such as traumatic bone cyst, simple bone cyst, idiopathic bone cyst, hemorrhagic bone cyst, progressive bone cavity, or unicameral bone cyst suggesting its different etiopathogenesis.^[1,2] While the term “traumatic bone cyst” is more widely used in the literature, the International Histological Classification adopted by the World Health Organization for odontogenic tumors uses the term “solitary bone cyst.”^[3] SBCs are generally asymptomatic and found incidentally on radiographic examination. However, some cases have reported with pain, swelling, paresthesia, and tooth sensitivity.^[3,4] SBC is most commonly seen in the mandible as compared to maxilla. Few cases which have been reported in maxilla almost all are found in the anterior maxilla region.^[4] None of the cases of SBC in the posterior maxilla has been reported in the literature. The author thus presents here a case of SBC found in a rare location of the right posterior maxillary region.

Case Report

A 15-year-old female patient reported to the Department of Oral Medicine and Radiology

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with the chief complaint of fractured and discolored tooth in the upper front teeth region of the jaw for 8 years [Figure 1]. The patient revealed no contributory medical and dental history but reported a minor trauma to the upper jaw 8 years back. The patient never had any symptoms related to the region. Previously, she had visited a private dental practitioner with the same complaint. She was advised extraction of the tooth, followed by implant placement for which the patient had undergone cone-beam computed tomography (CBCT) scanning of the maxillary anterior region (5 mA, 90 kV, and 360°) for implant planning. In that scan, the dentist encountered a large cystic lesion in the upper right posterior region. For further evaluation of the lesion, the dentist referred the patient to our department.

On clinical examination, maxillary right central incisors, right lateral incisor, left central incisor, and left lateral incisor were vital and they responded normally to both thermal and electrical pulp testing. No swelling or draining sinuses were noticed, and the overlying mucosa was normal on the buccal as well as palatal aspect in the maxillary right posterior region [Figure 2a and b]. Apart from this, there was Ellis Class III fracture with maxillary left central incisor.

On radiographic examination, panoramic view showed a 2 cm × 1.5 cm well-defined unilocular radiolucent lesion with sclerotic

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border extending from the apical region of the upper right second premolar to the second molar. Radiolucency between the teeth showed scalloping with intact lamina dura of all involved teeth. Mild root resorption was evident with the upper right second premolar, upper right first molar, and upper right second molar, and altered root morphology was seen with the upper right second premolar. In addition, there was a fracture of 21 along with external and internal resorption [Figure 3]. CBCT of maxilla (5 mA, 90 kV, and 360°) also revealed single, large, well-defined, expansile radiolucent lesion of size 2.8 cm × 2.1 cm on the right side of the maxilla. The lesion showed scalloped borders along with the displacement of the floor of maxillary sinus [Figure 4a–d]. There was no evidence of expansion of the buccal or lingual cortical plates in the CBCT of maxilla. Based on the location of the lesion, differential diagnoses included were keratocystic odontogenic tumor, ameloblastoma, and solitary (traumatic) bone cyst.

Fine-needle aspiration was carried out which showed clear, thin, red-colored aspirate with few epithelial remnants.[Figure 5] Incisional biopsy was also done under local anesthesia and the incised tissue was sent for histopathological examination. Microscopic section revealed hemorrhagic areas, scanty fibrous tissue, and bony fragments. There was no evidence of epithelial lining in the present sections [Figure 6a and b]. Thus, the

diagnosis of solitary or traumatic bone cyst was obtained after correlating clinical, radiological, and histopathological features. The patient was advised to undergo for conservative surgical treatment, but she did not turn up for further follow-up.

Discussion

SBC is not a common lesion. It is seen more commonly in young individuals with a range of 2–35 years and about 78% of the cases are found in the second decade. The lesion has a male predilection with a male-to-female ratio of 1.6:1.^[2,4] However, some researchers suggested equal sex distribution, while others have suggested male predominance.^[2,3] Howe’s analysis found that the lesion was more common in Asian and black females.^[4] It occurs more commonly in the mandible and seldom in the



Figure 1: Profile view



Figure 3: Panoramic radiograph showing a unilocular radiolucency apical to maxillary right second premolar, first molar, and second molar

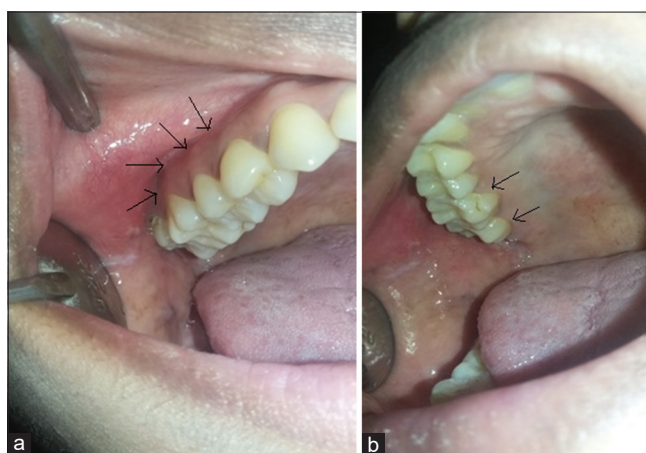


Figure 2: (a) Intraoral photograph showing no evidence of swelling or draining sinuses with normal overlying mucosa on the buccal aspect in the maxillary right posterior region (arrows). (b) Intraoral photograph showing no evidence of swelling or draining sinuses with normal overlying mucosa on the palatal aspect in the maxillary right posterior region (arrows)

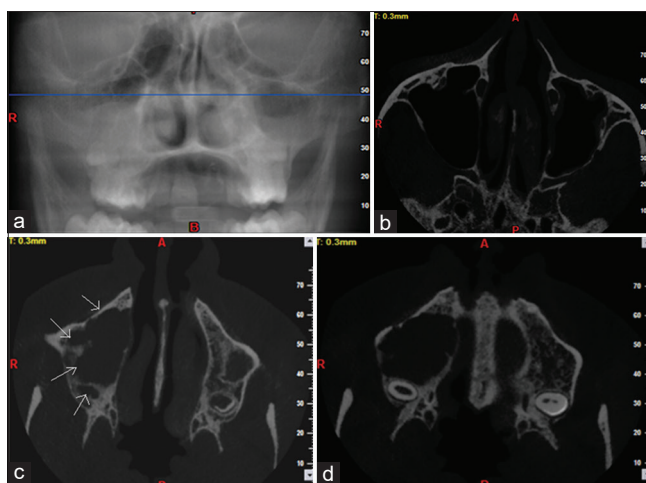


Figure 4: (a) Cone-beam computed tomography revealing a large expansile radiolucent lesion on the right side of the maxilla (b-d). Cone-beam computed tomography revealing single, large, well-defined, expansile radiolucent lesion of size 2.8 cm × 2.1 cm on the right side of the maxilla. The lesion showed scalloped borders along with the displacement of floor of maxillary sinus



Figure 5: Fine-needle aspirate showing clear, thin, red-colored aspirate with few epithelial remnants

maxilla. In mandible, 95% of the cases occurred in the body and symphyseal area and some in ramus and condylar area. In the body of mandible, the second premolar is the most common location.^[5] In some studies, one-fifth of the reported cases showed bilateral lesions.^[4,6] In maxilla, all cases reported have been in the anterior region. Posterior maxilla is a rare location for SBCs. Thus, the case reported by the authors here is an important contribution to the data as it is reported in the posterior maxilla region. The exact etiology is not known. However, the most accepted theory is that trauma leads to intramedullary hemorrhage. In normal circumstances, the hemorrhage will result in clot and thus no further bleeding occurs. However, in SBC, there is failure in early organization of hematoma in marrow spaces which subsequently leads to liquefaction of clot and further causing necrosis of bone marrow. This, in turn, results in trabecular resorption by osteoclastic activity. This theory is supported by the fact that most of the lesions develop after an injury or previous history of trauma and the fact that it occurs in areas where spongy bone-containing hemopoietic marrow enclosed in a heavy compact cortical layer explains why most of the lesions are found in mandible and in young individuals. Furthermore, the breakdown products of hemolysis also produce an increase in osmotic pressure leading to transudation and cyst formation.^[4,7] Now, trauma which can induce SBC is believed to be of insufficient force to cause a fracture of healthy bone. Iatrogenic trauma caused by tooth extraction or orthodontic treatment has also been suggested to incite SBC formation.^[8] Some cases are associated with fibro-osseous dysplasia or cemento-osseous dysplasia which could be due to cystic degeneration of existing bony tumor.^[9]

In 60% of the cases, there are generally no symptoms and are only found by chance on a radiographic examination.^[2,4,5] Symptoms if present are swelling and pain and in some cases, labial paresthesia has been reported. About 50% of the patients report a history of trauma with the affected region. The time lag between injury and diagnosis ranges from 1 month to 20 years. Swelling is found more commonly in the buccal or labial aspect, and the related teeth are vital.^[2,4]

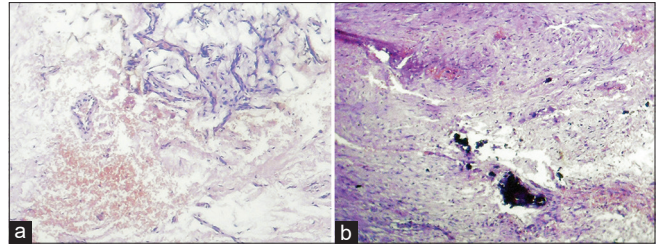


Figure 6: (a and b) Microscopic slide showing histological features of solitary bone cyst

Panoramic radiographs mostly show a unilocular radiolucent area with an irregular but definite edge and slight cortication. An occlusal radiograph will show radiolucency extending along the cancellous bone with no effect on the buccal or lingual plate. There may be some degree of condensation on the margins but not as sharp or opaque as seen in a radicular cyst. Posterior mandible is the most common location, and the cyst is found enveloping the roots of unerupted teeth. Radiographically, SBC mimics other common jaw lesions posing difficulty in diagnosis. Scalloping is an important radiographic feature of SBC and lamina dura may or may not be lost. Occasionally, root resorption and bony septa may be found.^[2,4,9,10]

Aspiration is done to support the diagnosis of SBC. Most lesions yield a bloody aspirate or yellow liquid which represents long-standing clots. For larger cyst, two needles are used to avoid negative pressure. If blood aspirate is yielded, the supernatant plasma and fluid should be tested for high levels of bilirubin.^[4] The acid and alkaline phosphatase activities in fluid aspirated from SBC have shown increase in the concentration of acid phosphatase.^[10] Surgical exploration in the most cases finds an empty cavity, but the rest shows a blood or serosanguinous or serous fluid within the cavity. There is no visible lining seen in SBC. Few cases may show thin membrane or granulation tissues or blood clot. Histopathological features include loose vascular fibrous tissue membrane of variable thickness with no epithelial-lining fragments of fibrin. Hemorrhage or hemosiderin pigments are commonly found. Adjacent bone shows evidence of osteoclastic resorption on the inner surface.^[2,4,10]

A diagnosis of SBC is given when the cyst is single, has no epithelial lining, shows no evidence of acute/chronic infection, contains principally fluid and not soft tissue and walls should be bone which is hard and thin in part.^[4] The treatment of SBC is mainly focused on establishing bleeding in the lesional cavity. This is done with raising suitable flap and outlining the outer bony wall which is removed and sent for histopathological examination to confirm diagnosis. Rest of the inside should be scraped to avoid damage to the neurovascular bundles and lightly packed to achieve hemostasis and closure of wound. Once the bleeding stops, there is absorption of the fluid in the cavity which leads to regression of the cavity.^[4,10,11] Intralesional injection of corticosteroid (methyl prednisolone) has been described as

a treatment modality for SBC in the long bones. It has been proposed that the healing is not solely a response to the corticosteroid treatment but rather results from mechanical disruption of the cavity.^[10,11] No evidence has been found the use of this modality in the jaw lesions. The recurrence rate of SBC is low, but SBCs which are associated with cemento-osseous dysplasia or other lesions have a high probability of recurrence.^[10]

The most interesting part of the case was the location of the SBC. A PubMed search for the location did not show a single SBC reported in the posterior maxilla region. A possible explanation could be that lesions in the posterior maxilla may not be easily visible on a conventional radiograph due to maxillary sinus. This makes the case report an important addition to the data available for SBC.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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