An unusual Mandibular Solitary Bone Cyst Mimicking Residual Cyst

Belmehdi Akram[1], El Harti Karima[2], El Wady Wafaa[3]

ABSTRACT

Introduction: Solitary bone cyst (SBC) is a non neoplastic pattern defined as an intraosseous cyst having a tenuous lining of connective tissue with no epithelium. It is an asymptomatic lesion, which is often diagnosed accidentally during routine radiological examination commonly present in the posterior mandible as a unilocular radiolucency.

Case report: We report a case of a 56-years-old female which clinical and radiographic characteristics as well as the surgical and histopathological findings were in favor of an unusual SBC.

Conclusion: SBC is rare jawbone pathology with an unclear etiology. It mostly discovered during routine radiographic examination and it is generally associated to a good prognosis and a low rate of recurrence.

Keywords: Unusual Case Report, Solitary Bone Cyst, Simple Bone Cyst, Traumatic Bone Cyst, Mandible

INTRODUCTION

First described by Lucas and Blum in 1926, solitary bone cyst (SBC) is an uncommon pseudocyst of jaws, yet their diagnostic criteria were only established by Rushton in 1946. SBC represent approximately 1% of all jaw cysts.[1]

While the term 'traumatic bone cyst' is more widely used in the literature, the international histological classification adopted by the WHO for odontogenic tumours uses the term 'solitary bone cyst'.[2]

According to WHO this unusual cyst is known as asymptomatic, intra osseous, slow growing, benign, non expansile, empty or fluid filled cavity having a tenuous lining of connective tissue without epithelium.[3]

Traumatic bone cyst had several terms used by different authors. These include solitary bone cyst, simple bone cyst, hemorrhagic bone cyst, progressive bone cyst, idiopathic bone cyst, and unicameral bone cyst. Overall, more than 95% of these cases involve the long bones such as the proximal humerus and femur.[4]

The origin of lesion remains unclear. Several hypotheses for the pathogenesis of this lesion have been postulated[4] which proposed that the cyst develops because of a lack of collateral lymphatic drainage of venous sinusoids which results in the entrapment of interstitial fluid causing resorption of the bony trabeculae and cyst development.

The present case report describes the clinical and radiographic characteristics as well as the surgical and histopathological findings of SBC in an older female patient as an unsual presentation.

CASE REPORT

A 56-years-old female patient reported to the department of oral surgery for evaluation of an asymptomatic unilocular radiolucency of posterior right mandible which was discovered as part of routine radiographic screening for restoration of her oral cavity. The patient had noticed that she has controlled diabetes which was confirmed by *HbA1 value.*

Physical examination showed no evidence of lymphadenopathy, paresthesia or facial swelling. On local examination, 46 and 47 were found to be missing in the concerning without buccal or lingual expansion and the overlying mucosa was intact (Figure 1).

Panoramic radiography revealed a well circumscribed trapezoidal unilocular radiolucency extending into the toothless area of the right mandibular posterior region, and away from the mandibular canal. A radiological differential diagnosis of residual cyst was made (Figure 2).

The patient was scheduled for surgical exploration under local anesthesia. A buccal mucoperiosteal flap was made along the posterior border of the mandible.

A bony window was made at that site, but no cystic capsule or contents were found except for a thin tissue lining which lead to a diagnosis of SBC. The bony wall was curetted to stimulate bleeding and to remove thin connective tissue lining, irrigated well and primary closure was done (Figure 3). Clinical diagnosis of solitary bone cyst was made and the small amount of tissue obtained was sent for histopathological examination. Histological examination of wall fragments revealed bone without specific characteristics.

Histopathological examination revealed densely inflamed connective tissue stroma and loose vascular fibrous tissue adjacent to the bone. No epithelial lining was found.

Postoperative 6-month radiographical evaluation showed evidence that process of bone repair is in progress with a partial new bone tissue formation (Figure 4). The patient is still under regular follow-up.

DISCUSSION

A solitary bone cyst (SBC) or simple, traumatic, and hemorrhagic bone cyst is defined as an empty or fluid-filled intra-osseous cavity that is devoid of an epithelial lining,

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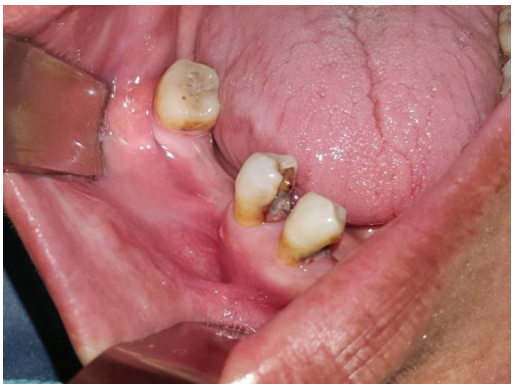

Figure-1: Intraoral picture of the lower posterior right region

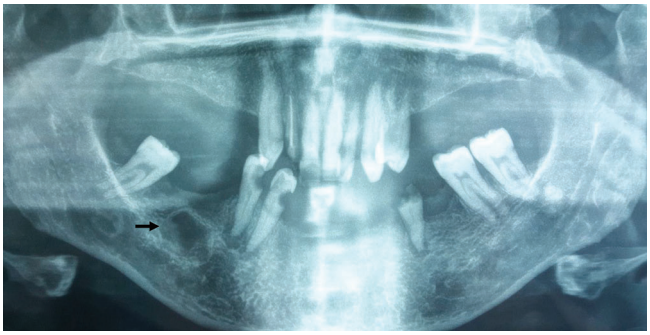

Figure-2: Preoperative panoramic radiography of the patient showing the radiolucent lesion under the toothless area

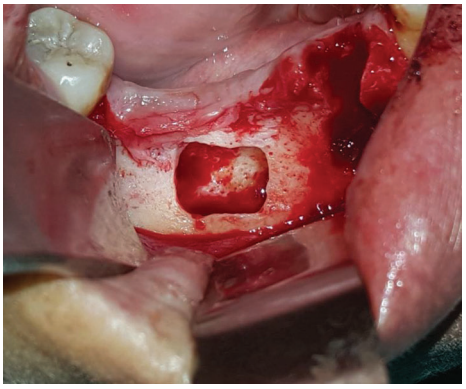

Figure-3: intraoperative picture demonstrating an empty cavity in the bone

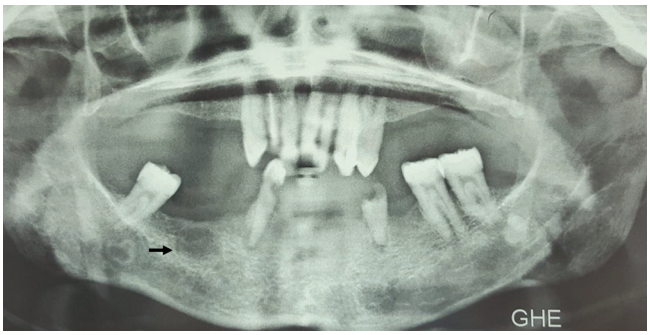

Figure-4: Follow-up orthopantomogram taken 6 months after treatmenta

that's why is not considered as a true cyst. It rarely occurs in the jaws with a percentage about 1% of all the jaw cysts.[5] The lesion is mostly diagnosed in patients below 30 years of age with an approximate mean age of 20 years. Though some studies have found no gender predilection, some state a masculine predominance.[6] However, the finding of the current case do not support research asserting that atypical cases are usually associated with an older age group or with fibro-osseous lesions.[1]

SBC usually affect the posterior premolar-molar area of the mandible, including the present case, although cases involving the symphysis, the mandibular condyle and rarely the maxilla have been reported.[3,7]

SBCs are commonly asymptomatic and painless without signs which are detected in the routine radiographic examination incidentally. There might be swelling and cortical expansions, but that occurs less commonly. The majority of these cysts are reported as single, distinct and isolated lesions of the mandible, although multiple lesions have been detected too.[8]

Pathogenesis of SBCs remains unclear, but several theories included a degeneration of bony tumors, deficient calcium metabolism, low-grade infection, disturbance in bone growth and excessive osteolysis. The most accepted theory advocates intramedullary hemorrhage following a bony trauma, clot liquefaction, enzymatic degradation of the surrounding bone and finally cystic cavitation. The presence of multiple enzymes and metalloprotienases also are favorable to the above mentioned theory of vascular osteolysis.[5]

Dental extractions might cause SBCs as a traumatic factor, and yet literature reveals that clinical history of trauma is elicited only in about 2% of the patients, which is very similar to our case where there was no relevant history of trauma to the teeth or jaws.[5,9]

Literature review has shown that traumatic bone cyst occurred together with osseous dysplasia. Cystic degeneration has been reported in patients with fibrous dysplasia which resulted in a non epithelial lined cavity. However, in the jaw bones and extracranial bones, fibrous dysplasia accompanied anevrysmal bone cyst rather than traumatic bone cyst.[6]

The radiographic features are seen as multilocular or unilocular radiolucent lesions. Borders of the cavity are variable from well defined to ill-defined. Radiolucency is the characteristic feature of this lesion, and scallop between the roots when several teeth are included. SBC radiographies could be mistaken to other lesions like the keratocystic odontogenic tumor because of little expansive growth and scalloped borders.[6,8]

The radiographic appearance here didn't correspond to the above mentioned features, our orthopantomogram showed a trapezoidal unilocular radiolucent lesion under a toothless area, which classically suggests a residual cyst as a first diagnosis.

Histological presentation is a vacant cavity of cancellous bone usually unlined or very occasionally lined with a thin connective tissue layer with a scant liquid content and numerous collagen fibers. The hall mark of traumatic bone cyst is the absence of epithelial lining.[6]

One of the reviews[10] have shown that in only 9.52% of the cases could a histological evaluation be made of the material obtained, revealing the presence of vascular connective

tissue without evidence of an epithelial component. This suggests that the absence of epithelial tissue is one of the most characteristic features of these lesions.

The diagnosis of SBC is more radiographic, complemented with the clinical findings during surgical management.

Various treatment modalities are suggested for SBC. Follow up of an asymptomatic lesion and waiting for spontaneous regression, or 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. In our patient, curettage of the bone wall was the chosen treatment which allowed a progression of bone repair.

Other options as the application of gel foam, injection of methylprednisolone acetate solution for treatment of long bone cases, allogenic bone grafting with platelet-rich plasma (PRP), injection of blood with hydroxyapatite and bone chips have also been proposed to have good results.[5,7]

Recurrences are rare, and usually occur within three months of surgery. Cases of multiple cysts or those associated with florid osseous dysplasia have high recurrence rates respectively about 71% and 75%26.[17]

In the present case, careful curettage of the lesion itself favored a progressive starting of bone formation and healing.

CONCLUSION

SBC is a rare jawbone pathology with an unclear etiology. It mostly discovered during routine radiographic examination and it is generally associated to a good prognosis and a low rate of recurrence. The lesion seldom causes any complications, but the possibility of pathologic fracture in larger lesions cannot be completely ruled out.

An adequate postoperative follow-up is recommended for this pathology for at least 3 years at yearly intervals, along with the corresponding radiographic control.

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