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CASE REPORT

Huge Solitary Bone Cyst located in Posterior Mandible

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ABSTRACT

The term 'solitary bone cyst' defines several lesions such as simple bone cyst, hemorrhagic cyst, traumatic bone cyst, and idiopathic bone cavity. Solitary bone cysts are not true cysts, they lack an epithelial lining and their etiology is uncertain. Solitary bone cysts are common lesions affecting long bones and less frequently, the jaws, especially the mandible. Lesions usually presents a typical radiographic appearance. The goal of this paper is to report a case of a solitary bone cyst in a 19-year-old female mimicking multiloculer benign neoplasm and review the literature. The lesion was treated by surgical curettage. No additional treatment, such as a surgical reconstruction or bone graft was needed.

Keywords: Solitary bone cyst, Mandible, Platelet rich fibrin.

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INTRODUCTION

A solitary bone cyst is a benign cavity in bone. Despite its name, it is a pseudocyst and lacks epithelium. It is known by numerous names. Etiology of this entity is uncertain. There are many theories in literature regarding etiology of these lesions. These lesions have been encountered in almost all bones of the skeleton. These pseudocysts more frequently affect patients between 10 and 20 years of age. The prevalence of solitary bone cysts is higher among men compared to women. Solitary bone cysts are usually asymptomatic. They can be diagnosed during routine dental exams on radiographs. Radiographically, solitary bone cysts are variably sized radiolucent lesions with irregular or scalloped but well-defined borders. Sclerotic margins are sometimes seen. The aim of this paper is reporting a case of solitary bone cyst with unusual appearance along with review of current literature to assist better understanding of this under-explored lesion.

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CASE REPORT

A 19-year-old female patient applied to our hospital for routine dental examination. During radiographic examination on panoramic radiograph and cone beam computed tomography (CBCT), a large multilocular well-defined radiolucency was detected in right posterior mandible (Fig. 1). The associated teeth tested vital and the patient could not recall any history of pain, swelling, or trauma to the area. The superior border of the lesion appears to scallop, between the roots of teeth. However, no root resorption was detected (Fig. 2). She was referred to oral and maxillofacial surgery department for definitive diagnosis and treatment. After first evaluation, the provisional diagnosis was a benign odontogenic tumor, such as unicystic ameloblastoma or keratocystic odontogenic tumor.

Clinical examination was unremarkable. There was no evidence of lymphadenopathy, swelling, or asymmetry. Intraoral examination did not reveal any soft tissue abnormality or bony expansion. There was no periodontal problem with no evidence of gingivitis, periodontal pocketing, or tooth mobility. To receive biopsy, patient was taken to operating room. After reflection of mucoperiosteal flap from buccal sulcus, bone was removed by bur. The lession was exposed. Surprisingly, the lesion was nothing but an empty cavity of bone. The only unusual finding was bulky body of inferior alveolar nerve (Fig. 3). It was understood that the pseudomultilocular appearance was caused by remaining cortical bone tissues of partially resorbed mandibular canal. Curettage was performed on walls of cavity to provide bleeding and recive biopsy. Then empty cavity

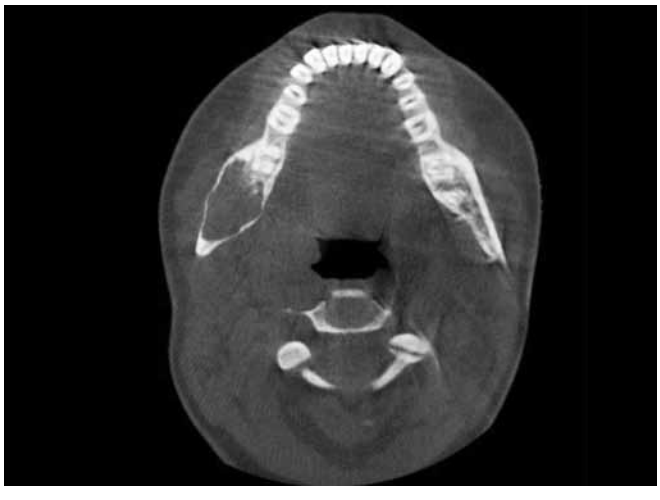

Fig. 1: Cone beam computed tomography showing multilocular and large lesion

was filled with platelet rich fibrin. Histological examination demonstrated fragments of fibrovascular connective tissue and numerous red blood cells. It was clear that there was no epithelial lining. The definitive diagnosis was 'solitary bone cyst'. Three months later, patient was called back for follow-up (Fig. 4). The bleeding caused by curettage was adequate to provide initial healing and bone formation at the site was satisfactory. The patient was evaluated in our clinic two more times, 6 months and 1 year after surgery. The lession manifested excellent recovery.

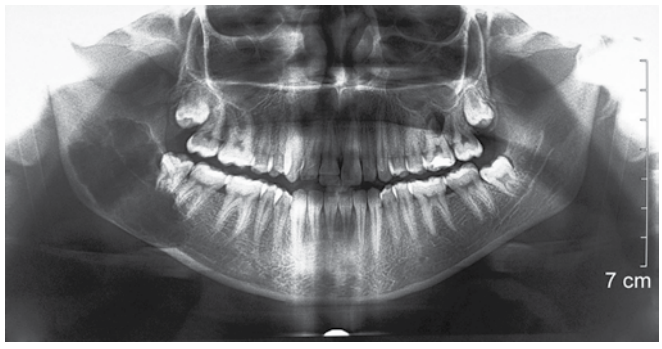

Fig. 2: Panoramic radiograph showing a large radiolucent, multilocular lesion of the right posterior mandible with scalloped margins

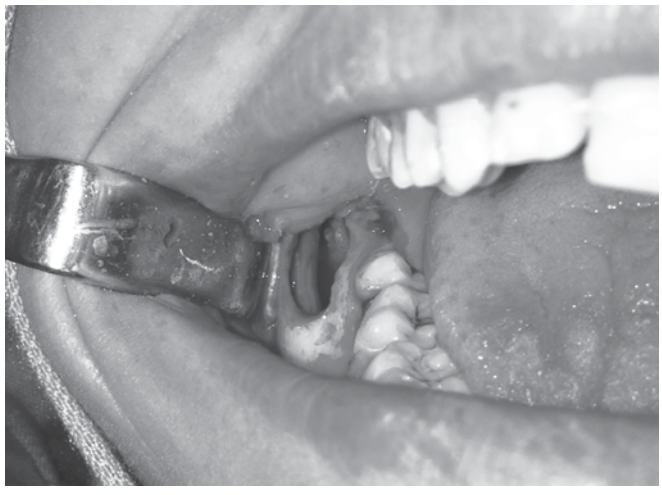

Fig. 3: Intraoperative photo showing empty bone cavity and inferior alveolar nerve

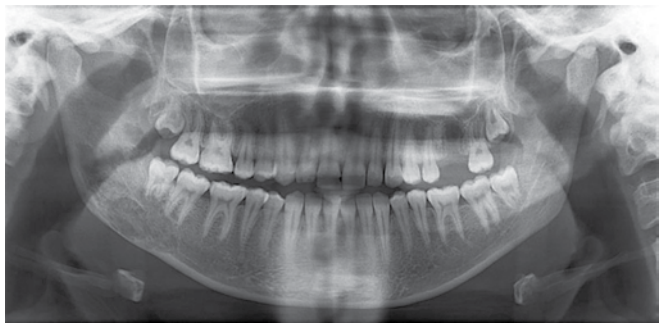

Fig. 4: Panoramic radiographic after 3-months of follow-up

DISCUSSION

Solitary bone cysts presented in jaws were first found in 1926 by Lucas and Blum under the name traumatic bone cyst.[1] Rushton established diagnostic criteria in 1946 as a single cyst that has no epithelial lining, has an intact bony wall, is fluid filled and has no evidence of acute or chronic inflammation.[2] Solitary bone cysts are pseudocysts of bones. It is known by numerous other names, however, the World Health Organization has designated the name 'solitary bone cyst' for these entities in 1992. These lesions can be found in literature under different names as hemorrhagic cyst, traumatic cyst, pseudocyst, simple bone cyst, extravasation cyst unicameral bone cyst, progressive bone cyst and idiopathic bone cavity.[3]

Etiology of this entity is uncertain. There are many theories in literature regarding etiology of these lesions such as; a sequela of intraosseous hematoma, alterations in calcium metabolism, mild infectious conditions, local bone growth alteration, orthodontic treatment, venous obstruction, and a localized alteration of bone metabolism resulting in the development of an area of osteolysis.[4] However, some of the following theories are predominant in literature: (1) an abnormal growth of the jawbone, (2) a process of tumor degeneration, and (3) a factor that triggers a bleeding trauma. The third theory is most common among researchers. This theory depends on intramedullary hemorrhage, followed by hematoma formation. The hematoma causes venous stasis leading to necrosis and osteoclastic resorption.[5] This theory could apply to the jawbone due to the multiple microtraumas undergone by the teeth and alveolar processes.[5] However, this mechanism have been questioned by many authors since trauma history is not present in most cases.[6] Most recent theory for development of this lesion is synovial cyst theory. This mechanism depends on intraosseous trapment of synovial tissue during fetal or early infant period. This new theory is consistent with cases seen in adolescents and cavities filled with fluid.[7] Despite the number of theories, none of these mechanisms are conclusive. Some authors belive in multifactorial etiology.[5] Solitary bone cysts associated with other pathologies, such as fibrous dysplasia or cement dysplasia are also reported.[4] Since only few cases are associated with trauma, some author suggest that the name 'traumatic bone cyst' should no longer be used.[8]

These lesions have been encountered in almost all bones of the skeleton. Over 90% of solitary bone cysts are located in long bones, most commonly the proximal humerus and femur. Involvement of the jaws is less frequent.[9] These pseudocysts more frequently affect young patients in second and third decades of life. The prevalence of solitary bone cysts is slightly higher among women compared to men.[10] In the maxillofacial region, lesions appear most frequently in the mandible (61%). Most involved areas in mandible are posterior body of mandible. Involvement of ramus is rare but possible.[10] When maxilla is affected, lesions appear in

anterior part of this bone. It is possible that the maxillary sinus makes radiographic evaluation more difficult.[11] Involvement of uncommon regions, such as zygoma, coronoid process and condyle, is also reported.[12]

Solitary bone cysts are usually asymptomatic. They can be diagnosed during routine dental exams on radiographs. These entities lack characteristic symptomatologies. However, mild pain can be encontered in areas of tooth displacement.[13] Associated teeth are usually vital and there is no external tooth resorption.[10] There are only few cases of pathological fractures of jaws associated with this lesion is present in literature.[13] However, pathologic fractures in long bones as a result of this lession is common. Radiographically, solitary bone cysts are variably sized radiolucent lesions with irregular or scalloped but well-defined borders. Sclerotic margins are sometimes seen.[10] Solitary bone cysts are usually soliter entities but synchronous development is found in literature.[6] Tooth resorption is rare which distinguishes this lesion from odontogenic tumors. While most cases manifest unilocular appearance, multilocular or lobular display is possible.[14] Computed tomography is a valuable tool to determine exact location of lesion. Magnetic resonance imaging enables analysis of interior of lesion showing presence and absence of fluid.[15] This entity should be considered during the differential diagnosis of radiolucent lesions with unerupted teeth.[14]

A solitary bone cyst is a benign cavity in bone and it can be either empty or contains some fluid. Cystic fluid may have hematic or serous content.[4] Although these lesions are lacking a true epithelial cystic lining, the walls of the cyst might be lined with a thin layer of vascular fibrous connective tissue. This lining may contain areas of vascularization, fibrin, erythrocytes, and sometimes giant cells adjacent to the bone surface.[14] Deposits of new bone formation, collagen deposits, and giant cells can also be seen in pathologic examination.

Curettage of cyst walls and provide blood accumulation inside cavity is usually enough to provide recovery. There is no evidence supporting grafting of the cavity. Spontaneous resolution is possible but since some lesions tends to show aggressive behavior it should be considered carefully.[13] Since, recurrence is possible, follow-up is indicated. In old literature, recurrence of this lesion was reported as 2%. However, recent studies reports recurrence incidence as high as 26%. It usually takes 3 to 6 months to observe initial stages of healing. Recurrence can be detected early because initial bone healing is slower in cases of recurrence compared to cases of complete healing.[3] Complete healing, however, takes up to 2 years following surgery. Multilocular or lobular lesions have a higher tendency to recure; therefore, those lesions requires further follow-up. Suei et al[3] states that follow-up should be continued for 3 years and until recovery is confirmed by radiographically. However, recurrence after 4 years following surgery is also reported. In cases associated with cemento-osseous dysplasia, follow-up should be done more carefully because these lesions have higher incidence of recurrence. Recurrence is also higher in multiple lesions. Then again, the lession usually has excellent prognosis compared to other lesions of maxillofacial region.[13] The orthopedic literature describes intralesional injection of steroids (methylprednisolone) as a treatment method.[16] Another minimal invasive method is intralesional injection of bone marrow in cavity. These conservative treatments can only be performed in maxillofacial region when diagnosis is confirmed with several imaging methods and aspiration shows air-filled empty cavity.[17]

CONCLUSION

The authors of this paper believe that, follow-up of such patients annually is recommended in the first years after surgery, until complete ossification of the defect is reached, since recurrence is possible and the area is weakened with high-risk of fracture.

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