

Case report

An Atypical Presentation of Solitary Bone Cyst in Jaw: A Case Report

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How to cite this article:

Nidhi K. An Atypical Presentation of Solitary Bone Cyst in Jaw: A Case Report. Int J Histopathol Interpret 2022;11(1):10-14 Received:11-05-2022; Accepted:13-06-2022; Web Published:21-06-2022

Abstract:

A solitary bone cyst (SBC) is an uncommon, benign cyst that primarily occurs in children and adolescents. They are pseudocysts primarily affecting long bones, with an incidence of 1% incidence among all the cysts of jaws. These cysts are generally detected during routine radiography, being asymptomatic. In this case report, we are presenting a case of SBC in mandible giving an unusual presentation.

KEY WORDS: Mandible, Solitary Bone cyst, Trauma, Scalloping, Multilocularity.

INTRODUCTION :

Solitary Bone Cyst (SBC) is a fluid filled/empty intraosseous lesion found most commonly in metaphyseal region of long bones, e.g, proximal humerus and femur. They are basically empty cavities, without an epithelial lining, hence termed pseudocysts, their etiology being unknown.

Most of the lesions occur in the posterior mandible, especially in the premolar–molar region/body of mandible and anterior maxilla¹.

SBC are normally asymptomatic and discovered during routine radiographic examination¹. The presence of pain, oedema, paraesthesia, displacement and root resorption has been reported on rare occasions².

OPG & CBCT are more precise imaging examinations in the diagnosis of SBC. Radiographically, they appear as well delimited radiolucent defects. When multiple teeth are involved in the cystic lesion, the radiolucency

frequently shows scalloping between the dental roots, which is the characteristic feature of SBC.¹

In this case report, we are describing a case of SBC in mandible showing unusual presentation.

Case Report:

A 17 years old male patient reported to our hospital with complaint of pain and swelling on the lower right back teeth region for 2 years with no associated fever or paraesthesia. The patient's medical history was not significant.

The swelling, extra orally, extended from the corner of mouth to the angle of mandible on the right side which was 7*4 cm in size approximately [Figure 1(a)]. The swelling was firm and tender on palpation. On intraoral examination, it extending from 44 to 48 region, obliterating the buccal and lingual vestibule, without any sinus opening or tooth mobility [Figure 1(b)].

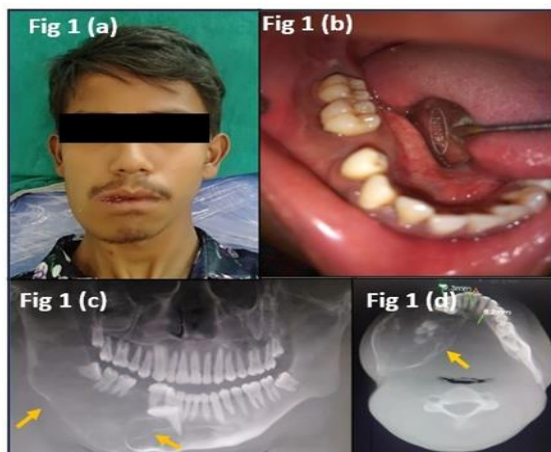


Fig 1 (a) Extraoral swelling on right side of mandible; Fig 1 (b) intraoral swelling obliterating buccal and lingual Vestibules; Fig 1 (c) OPG Showing radiolucency extending from 3 region involving ramus; Fig 1 (d) CBCT showing bucco-lingual expansion of bone

Radiographically, OPG revealed a well-defined multilocular radiolucency interspersed with thin radiopaque lines (septa) seen on the right side of mandible [Figure 1(c)]. It extended from 31 till ramus, accompanied with thinning of lower border of mandible. Upward displacement of 46, 47, and 48 and anterior displacement of 45(impacted) was well appreciated. Furthermore, CBCT revealed buccal and lingual expansion of bone [Figure 1(d)].

On the basis of radiological and clinical findings, differential diagnosis consisted of odontogenic keratocyst, dentigerous cyst, ameloblastoma and central giant-cell granuloma.

Aspiration biopsy yielded a blood tinged clear fluid and multiple soft tissue bits were received for the histopathological examination [Figure 2(a,b)].

Histopathological examination of the incised specimen revealed cystic lumen with thin strip of connective tissue which was fibro-cellular showing

globules of eosinophilic amorphous material. Peripheral areas of cystic wall exhibited focal bony trabeculae suggestive of reactive bone [Figure 2 (c,d)].

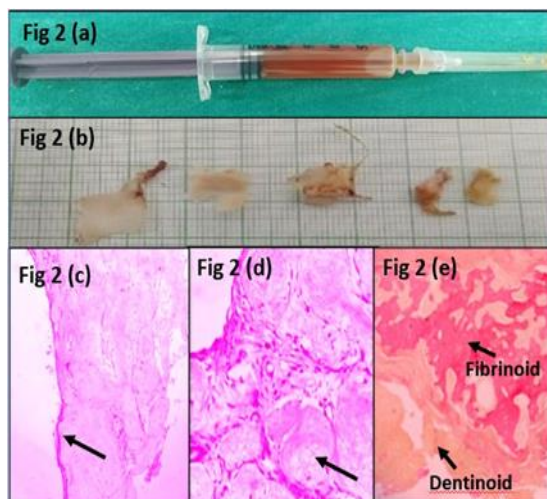


Fig 2 (a,b) Blood tinged clear fluid gross specimens; Fig 2 (c)Fibrocellular connective tissue wall; fig 2 (d) Globules of Eosinophilic amorphous material; Fig 2 (e) Von Gieson stain differentiating fibrinoid and dentinoid material

Discussion:

SBC is an uncommon, nonepithelial lined, intraosseous bone cavity of the jaws, first described by Lucas in 1929. They are referred with variety of names: Simple Bone Cyst, Haemorrhagic bone cyst, Extravasation cyst³.

The etiology of SBC is still uncertain, but some investigators have suggested an association with trauma, i.e, trauma-haemorrhage theory. It suggests that trauma to a bone unable to cause fracture result in an intraosseous hematoma that does not organize

and repair, it liquefies, leading to the formation of a cystic defect. Other etiological theories include the incapacity of interstitial fluid to exit bone, inadequate venous drainage, local disturbances bone growth, ischemia necrosis of bone marrow and altered metabolism resulting in osteolysis, benign neoplastic degenerative lesions, altered calcium metabolism, low-grade infections, and bone tumors undergoing cystic degeneration⁴.

The mandibular body is the most frequent site of SBC, as seen in our case; however, cases involving the symphysis region, mandibular ramus, condylar area, and anterior maxilla have been reported. Cases have also been reported where SBC was present in the zygomatic bone^{1,5}.

SBCs are mostly diagnosed fortuitously on routine radiographic examination as they are usually asymptomatic. They appear as a unilocular radiolucent area with an irregular but definite edge and slight cortication. Bony septa may be present and lesions are sometimes interpreted as multilocular as observed in our case [Figure 1c)]. The lesion may extend between the roots of the erupted teeth, producing a characteristic jagged/scalloped contour as evident in this case. But it may cause resorption of roots along with it which makes the radiolucency showing a linear contour along the roots. The radiographic features are not definitive for the diagnosis, with the need for a combined analysis of clinical and histopathological findings^{4,6}.

Otherwise, SBC might be confused with other odontogenic and non-odontogenic lesions of the jaws which exhibit radiolucency radiographically.

The following differential diagnoses can be considered: periapical cyst or radicular cyst, odontogenic keratocyst, lateral periodontal cyst, ameloblastoma, cherubism, central giant-cell granuloma, ghost cell lesions, cementifying fibroma and aneurysmal bone cyst¹.

Histopathological examination of SBC consists of loose vascular fibrous tissue membrane of variable thickness with no epithelial lining. Sometimes, areas of haemorrhage are associated with necrotic tissue or tissue showing myxoid appearance⁷. In our case, histological examination showed fragments of fibrovascular connective tissue, numerous fibroblasts and dilated blood vessels. At places, eosinophilic amorphous material showed calcifications and entrapment of cells resembling woven bone formation. Peripheral areas of cystic wall exhibited focal bony trabeculae gave the confirmation of the existing lesion [Figure 2(c,d)]. These histopathological results ruled out the presence of OKC, lateral periodontal cyst because of absence of an epithelial lining. The absence of giant cells ruled out the diagnosis of central giant cell granuloma. Further, Van Gieson's staining was performed to differentiate collagen and smooth muscles in tumors. In our case, it was done to differentiate fibrinoid from dentinoid material and

ghost cells [Figure 2(e)] which was helpful in ruling out cementifying fibroma or any ghost cell lesion.

SBCs are usually treated by opening the cavity, evacuating the cystic contents followed by curettage of the cavity to stimulate bleeding. This leads to formation and organization of a clot and healing by new bone formation⁸. Cases have been reported where SBC heal spontaneously without any intervention. Patients are kept under continuous observation and periodic follow ups and we have adopted the same measure for our case.

Conclusion:

Solitary bone cysts are a very rare uncommon pseudocysts with an uncertain etiology. A thorough clinical examination, history of patient, radiological & histopathological evaluation will be helpful for final diagnosis and treatment plan. Recurrence after surgery is extremely rare and hence it shows an excellent prognosis.

Funding: Self-funded

Conflict of Interest: Nil

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