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Case Report

The live and let live pseudo-cyst: A Classic Rare Case of the Simple Bone Cyst.

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ABSTRACT

Background and Setting: In 1992, WHO proposed the term "solitary bone cyst" (SBC) for lesions first stated by Lucas in 1929 and by Blum in 1932. Hemorrhagic cyst, traumatic cyst, pseudocyst, simple bone cyst, extravasation cyst and idiopathic bone cavity are other synonyms for this lesion. SBC represents approximately 1% of all jaw cysts. They are especially common in the long bones (90%), mainly the humerus and femur, in proximity to the epiphyseal plate, and very few (10%) cases occur in the jaws. The mandible is more commonly affected than maxilla; the area most prone being the body of the mandible between canine and third molar.

The lesion is common in young males, especially during the second decade of life. The simple bone cyst is a pseudocyst devoid of an epithelial lining and is usually empty or contains blood or straw-colored fluid.

The present case reviews a solitary/ simple bone cyst in the mandible, of an 18-year-old otherwise healthy male; who had a trauma. The lesion was asymptomatic and was discovered on routine radiographic examination. Surgical curettage of the lesion was done, following which the patient has recovered well, and has been on regular follow-ups.

INTRODUCTION

Simple bone cyst (SBC) is a pseudocyst affecting the bony skeleton. A pseudo cyst by definition is a pathological cavity without an epithelial lining; with clinical and radiographic similarities to true cysts, and relies on the histo-pathological findings for diagnosis. [1] The pseudo cavity will almost always reveal a space, but may occasionally be filled with straw-colored or clear fluid. Simple bone cysts are also called by various names, and the unicameral bone cyst is one of them. [2, 3] It was first described by Lucas in 1929. [4] Rushton in 1949 recognised the diagnostic features of this condition. [1]

The aetiology of the simple bone cyst is unknown. SBC is commonly seen in males below 30 years of age. [1]The lesion is discovered on routine radiographic examination. The involved teeth exhibit no pathologies such as pulpitis, root resorption, or displacement. The present case report discusses a simple bone cyst in an otherwise healthy, 18-year-old male.

CASE REPORT

An 18-year- old healthy male patient, reported to the private dental practitioner, with a history of trauma, during a football game; about 1 month ago. He was subjected to an orthopantomogram which revealed a radiolucent lesion in the left lower jaw, of which, the patient was unaware. The patient was further referred to the present institution for evaluation and management of the lesion. The patient's medical, dental, surgical, and personal history was non-contributory. No abnormalities were detected on general and extra-oral examination. Intra-orally, on hard tissue examination, a partially erupted 38 was noted, (Figure 1) and no abnormality was detected in relation to the other teeth, of the left mandibular quadrant. A mild cortical expansion was noted in the 37, 38 regions involving both buccal and lingual plates. The mucosa over the expansion appeared normal; it was non-tender and bony hard on palpation. On pulp vitality testing, all teeth in the mandibular left quadrant were found to be vital.

The Orthopantomogram showed a well-defined unilocular radiolucency in the left posterior body of

FIGURE 1: *Intraoral picture was showing cortical expansion*

mandible measuring about 4 centimetres x 2 centimetres in size. (Figure 2)

Antero-posteriorly, the lesion extended from the mesial aspect of 36 to about 1 cm distal to 38, also involving the ramus. Supero- inferiorly, it extended

FIGURE 2: *Panoramic radiograph showing the extent of the SBC*

from the periapical region of 36, 37 up to the inferior cortex of mandible. The patient was subjected to other radiographic investigations such as lateral occlusal radiographs, postero anterior (PA) mandible view and computed tomography (CT).

A lateral occlusal radiograph revealed the thinning of the buccal cortical plate in relation to 36, 37 and 38 (Figure 3).

The Postero- Anterior (PA) mandibular view revealed the thinning of the medial surface of the ramus (Figure 4).

The computed tomography image revealed a unilocular, single, expansile

FIGURE 3: *Lateral occlusal radiograph*

lesion around the apices of 36 and 37 in the sagittal section (Figure 5).

The axial section revealed smooth borders of the lesion, which was well-scalloped and corticated, growing along the length of the mandible.

FIGURE 4: *PA Mandible View*

The following radiographic differential diagnoses were considered:

Keratocystic odontogenic tumor (KOT): KCOT occurred in the younger age group; showed a tendency towards male predilection, and normally exhibited a presentation of an extensive mandibular involvement in the posterior body of mandible and ramus. Radiographically, it showed a well-defined radiolucency with a corticated borders associated with mild cortical expansion as the tumour grew through the medullary spaces. The histopathological features ruled out this tumor.

Ameloblastoma: A unilocular radiolucent lesion with well-defined corticated borders. It is often associated with a partially or unerupted tooth and found mostly in the posterior mandible. However, significant expansion, perforation with soft tissue infiltration, multilocular appearance, and root resorption of the associated teeth, are characteristic features which did not favour this tumor

FIGURE 5: *Axial section of the lesion on a computed tomograph*

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Odontogenic myxoma: An asymptomatic tumor that is found in the 10-30-year-old patients that present as a lytic osseous pathology, of various sizes, with cortical expansion. Scalloping of the lesion in between the roots of the teeth is seen. Although the above-mentioned features were in favor of the present lesion, there were some factors such as, the multilocular appearance with honeycomb pattern; root resorption and root displacement which ruled out the tumor

Aneurysmal bone cyst: A benign, expansile tumour whose radiographic appearance is a well-defined sharply osteolytic lesion, with a thin sclerotic margin; mostly seen in the posterior mandible.

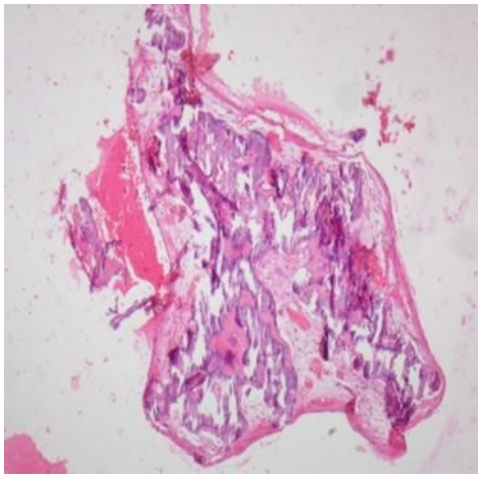

Figure 6: *Hematoxylin and Eosin sections showing scattered neutrophils and absence of epithelium*.

However factors such as its occurrence in children and resorption of associated teeth and the typical soap bubble appearance; ruled out this lesion.

Simple bone cyst: A typical unilocular radiolucent lesion with scalloped margins extending between the roots of involved teeth; well-corticated borders; absence of resorption or displacement of the associated teeth. It mainlypresents as an asymptomatic, painless lesion with a tendency to occur in the posterior mandibular region in young males; and is usually associated with a previous history of trauma. [5, 6] Preparation of a bony window in the 35, 36 regions, followed by a fine needle aspiration cytology revealed a straw colored fluid. Curettage of the entire lesion was done. Histopathology revealed areas of haemorrhage and vital host bone. Few scattered neutrophils and lymphocytes were seen. The epithelial lining was not present in the given tissue, therefore suggesting a pseudo cyst for, e.g., the simple bone cyst. (Figure 6)

DISCUSSION

The World Health Organization (WHO) has included the TBCs, in the group of bone-related lesions, along with the aneurysmal bone cyst, ossifying fibroma, osseous dysplasia, fibrous dysplasia, cherubism and central giant cell granuloma. [7]

Solitary bone cysts exhibit non-epithelial lined cavities occurring in many parts of the skeleton. They are frequently encountered in long bones with 90% incidence and have an incidence of 5% in the jaw bones. [8] The simple bone cyst is an uncommon pseudocyst of the maxillo-mandibular region. Several causative factors have been proposed:

- tumor-produced bone degeneration,
- calcium metabolism modifications,
- local changes in the bone growth,
- venous blockage
- increased osteolysis
- Bleeding intramedullary
- local ischemia,
- the mild infectious process,
- infection in the bone marrow,
- ischaemic necrosis in bone marrow
- exsanguination due to hemangioma or lymphoma,
- lymphaticdrainage obstruction-sinusoidal veins resulting in trabecular bone resorption,
- synovial fluid incorporation into bone in developmental anomalies
- interstitial fluid's normal drainage-blocked
- osteoblastic differentiation
- or amalgamation of the said factors.

Any trauma may result in this cyst, including dental extractions or undue force applied during orthodontic treatments. [1]

The most accepted among these is the trauma-hemorrhage theory: which also states that the trauma that is insufficient to cause a bone fracture results in an intra-osseous hematoma. [1]

In the jaws, it has been suggested that the mandible is more commonly affected than the maxillae because it is subjected to more micro-traumatic events, particularly in the premolar areas. [9] SBC's have also been reported in the symphyseal region as well. [8] Presumably, this correlates with the increased incidence of SBCs here. Simple bone cysts have also been reported at unusual sites, such as the condylar and coronoid processes and zygomatic arch. [1] Involvement of the maxilla is rare (1%). [1] This results in a cavitation following necrosis that may contain blood and/or serous fluid and that may have a lining of fibrous connective tissue. In the jaws, SBCs may contain nothing at all. [6] SBCs affect patients of young age. However, in a case series by Ballester et al., [1] patients above 40 years have been reported. [1] The lesion is more prevalent in men compared to women with a male to female ratio of 3:2. [9] Most studies

describe no gender differences, but Peñarrocha et al. and Ballester et al. [1] have reported a definite female predilection. These differences may be associated with epidemiological variations in the different parts of the world. [3] They are asymptomatic and are most commonly detected during the routine radiological assessment.

SBCs are asymptomatic;though rarely symptoms such as pain, paraesthesia, failure of eruption of permanent teeth and pathological fractures have been reported. [1] In a case series by Luana Flores et al., [5] patients reported with mild discomfort in the jaws, together with pain.

SBCs present as unilocular radiolucencies ranging from 1 to 10 centimetres in diameter. [2] SBC's usually present as a unilocular radiolucency, with an occasional multilocular appearing radio-transparency, associated with a cortical expansion and tumefaction of slow growth. [11] The scalloping effect is characteristic of SBCs, where it extends between the roots of the teeth. Cases with smooth margins of the lesion have also been reported. The teeth associated with these lesions usually have an intact lamina dura, but cases with loss of lamina dura have also been observed. [1] Associated teeth are usually vital with no signs of resorption. Cystic expansion may increase the root pressure due to the traumatic force. [1]

Usually, an empty cavity is found on surgical intervention that may be filled with blood or sero- hematic or serous fluids. [9]

Histology of these lesions exhibits a cancellous bony cavity that may be empty, with the absence of an epithelial lining; but they may present with abundant giant cells and osteoclasts, or may present as a thin connective tissue layer with a scant liquid content. In any case, the absence of an epithelial lining is a constant characteristic feature of this lesion. [1]The present case presented with the classical: clinical, radiographical, and histological features. The lesion was asymptomatic, unilocular, well-defined lesion with corticated borders; with a buccal and lingual cortical expansion; with no resorption or loss of vitality of associated teeth; with a scalloped appearance of the lesion in between the roots, and a histopathology, showing the absence of an epithelial lining.

Aspiration of the contents and curettage of the lining is the preferred mode of treatment; followed by packing with gel foam saturated with thrombin and penicillin to promote healing. Periodic follow-up and radiographic examination are mandatory. If asymptomatic, the lesion must be kept under observation. [8] Recurrence is rare (22.6%), However, in a case series which evaluated the prognosis of simple bone cyst in 31 cases, resorption of the lamina dura, scalloped margin, nodular bone expansion, root resorption, sclerotic mass or multiple cavities were associated with higher rate of recurrence. [8]

The present case had a fair prognosis.

CONCLUSION

Surgeons and clinicians must be familiar with the presentations in diagnostic imaging and all available clinical information about the SBCs. Being aware of only the morphologic analysis far-removed from the clinical and radiographic setting may lead to misdiagnosis.

Traumatic/simple bone cyst should be considered in the differential diagnosis for unilocular radiolucent lesions of the posterior mandible.

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