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Unusual Aggressive Radiographic Appearance of Simple Bone Cyst (A Case Report and Review of Literature)

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Authors' contributions

This work was carried out in collaboration between both authors. Both authors read and approved the final manuscript.

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Case Report

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ABSTRACT

Aim: This study reports a rare case of simple bone cyst (SBC) with a radiographic behavior similar to malignancies and a mixed internal structure in a 43 year-old woman who presented to a dental office with clinical symptoms.

Presentation of Case: A 43 year-old woman presented to a dental office with chief complaint of pain at left mandibular molars for almost 2 months. She was wearing a cervical collar for some spine problems. The panoramic x-ray showed a radiolucent lesion with well defined non-corticated borders in periapical region of the mandibular left first molar (tooth 36). The tooth was vital. Cone Beam Computed Tomography (CBCT) images showed a lytic lesion with well defined non-corticated borders that destructs the lingual cortical wall without any sign of expansion to soft

tissues. The upper border of the mandibular canal was unclear and seemed destructed by the lesion. The internal structure of the lesion seemed mixed. Three months later, the patient reported numbness of her lower lip. CBCT showed an enlargement of the lesion and the presence of buccal cortical perforation was seen. On biopsy the lesion was diagnosed as a Simple Bone Cyst (SBC). **Discussion:** SBC is often a benign lesion which appears as a radiolucent lesion with scalloped corticated borders. Adversely, current case showS a mixed lesion with buccal and lingual cortical perforation and rapid growth .the concomitant pain, the lower lip numbness and the radiographic appearance suggest the probability of an aggressive local condition or a malignancy **Conclusion:** SBC may have various radiographic presentations, unusual SBCs (or Traumatic bone cysts TBCs) may show aggressive radiographic characters. In these cases, biopsy and pathological features are necessary for diagnosis.

Keywords: Simple bone cyst; traumatic bone cyst; aggressive; cortical perforation.

1. INTRODUCTION

The etiology of simple bone cyst (SBC), also known as traumatic bone cyst, is unknown [1,2]. It is assumed that SBC is related to traumainduced hematoma [3, 4]. The early diagnosis of SBC can be reached through receiving the history, physical examination, and radiography. The lesion should be differentiated from other lesions, such as central giant cell granuloma through radiography [5] Other lesions, including aneurysmal bone cyst [6,7] must be taken into account due to their clinical and radiological similarities. SBC of jaws is primarily treated through exploration surgery [8]. Some of such lesions relapse after treatment [9]. male/female ratio for SBC is 3 to 2. Patients with SBC are mostly 10-20 year-old [10,11], and SBC mostly appears in posterior mandible [12]. The uncommon and atypical areas for SBC are the condyle, coronoid, and zygoma [13]. Most of such lesions do not have clinical symptoms [14,15]. Symptoms such as pain and paresthesia have been observed in few cases [16], and must lead to investigations to establish a diagnosis and eliminate malignancies. Most of SBC are completely radiolucent [17] often with no cortical perforation. This study reports a rare case of SBC with an unusual radiographic appearance and an aggressive behavior in a 43 year-old woman who presented to a dental office with clinical symptoms.

2. CASE PRESENTATION

A 43-year-old woman presented to a dental office with the chief complaint of occasional vague pain at left mandibular molars for almost 2 months. She did not mention any specific family problem. Past medical history wasn't significant except for a trauma the patient had almost a year ago, when a car door hit the left side of her

face. Intraoral and extraoral examination did not show any anomaly, and lymph nodes of the area were also normal. The gum and mandible were normal in both observation and palpation. Panoramic radiography was ordered for further analysis of teeth and jaws and regarding the presence of vast teeth restoration. It showed a radiolucent lesion with well defined noncorticated borders in the periapical region of the mandibular left first molar (tooth 36). Periapical radiography was performed for more accurate analysis of the tooth 36, restoration, and research of signs of necrotic pulp. The vitality of the tooth 36 was confirmed by an endodontics specialist. The patient was referred to an oral and maxillofacial surgeon for further analysis of the lesion. In the Cone Beam Computed Tomography CBCT images he requested, a lytic lesion with uncorticated borders was observed in the periapical region of tooth 36. The lesion destroyed the lingual cortical wall without any sign of expansion and spread to the buccal cortical wall. The thinning of the buccal cortex could be observed. The upper border of the mandibular canal was unclear and seemed to be destructed by the lesion. In the axial view, the extension of the lesion from lingual cortex to buccal cortex was obvious. The lesion was 7.3 × 8.8 mm (Figs. 1,2). The internal structure of the lesion seemed mixed with cementum-like calcifications. Regarding all these elements, medical staff proposed to follow-up the patient to observe the progression of the lesion Due to the aggravation of the symptoms, the feeling of tingling and numbness of lower lip in its left part the patient returned to the dental office after three months. The follow-up CBCT showed the destraction of alveolar nerve canal boundaries and an increase in size of the lesion, further thinning of the buccal cortical wall with an appearing perforation and an enlargement of the lingual cortical perforation, and presence of

areas of buccal cortical perforation. The size of the lesion increased to 7.72×13.52 mm (Figs. 3,4). Moreover, the internal structure of the lesion seemed more mature and opaque. Considering the rapid destructive growth of the lesion in these 3 months, the differential diagnosis of aggressive benign tumors or a malignancy seemed more probable. A biopsy was performed to rule out the malignant lesions such as plasmacytoma, eosinophilic granuloma, metastasis and other intraosseous malignancies. The surgery report mentioned an empty space with no tissue at the site of the lytic lesion. The surgeon reported it as SBC because there was not any evidence of cystic lining or any solid mass in the cavity. He realized bone curettage Samples were addressed for histopathological examination that found no epithelium lining or tumoral cells and confirmed the presumptive diagnosis of SBC.

6 month postoperative CBCT showed healing of the surgery site with granular bone (Figs. 5,6). No evidence of recurrence was seen after 6 month. Imaging was recommended for further follow up.

3. DISCUSSION

SBC of jaws is a pseudocyst without any epithelium. The etiology of SBC is unknown, however most of the suggested etiologies are trauma-induced hemorrhage. Sometimes, the lack of trauma and areas less prone to trauma, such as the posterior mandible where SBC is frequent, weaken the above etiology [18]. Another theory states that SBC is a developmental cyst [19]. Most of SBCs are asymptomatic. The radiographic presentation of SBC is often radiolucent with scalloped borders and may spread between roots [3,13]. Radiographic features are not sufficient for diagnosis of SBC, surgical findings and histopathological results are required for the final diagnosis [14]. Although the bone cavity is often smooth and shiny, curettage should be performed in order to draw samples for ruling out the risk of other diseases. The healing often begins 6-12 months after the curettage [18].

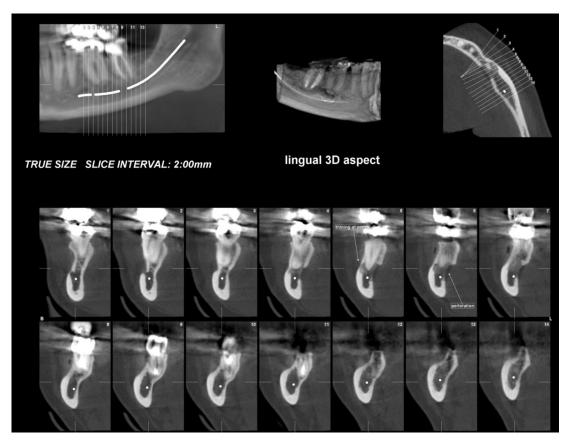


Fig. 1. First evaluation CBCT

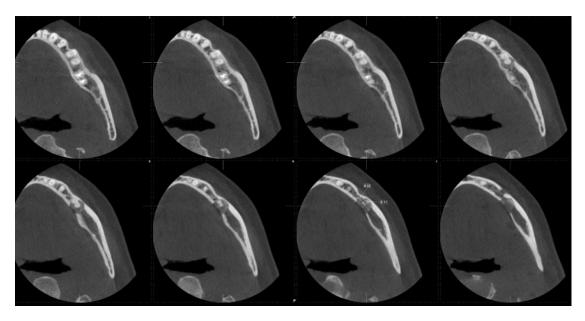


Fig. 2. First evaluation CBCT (Axial view)

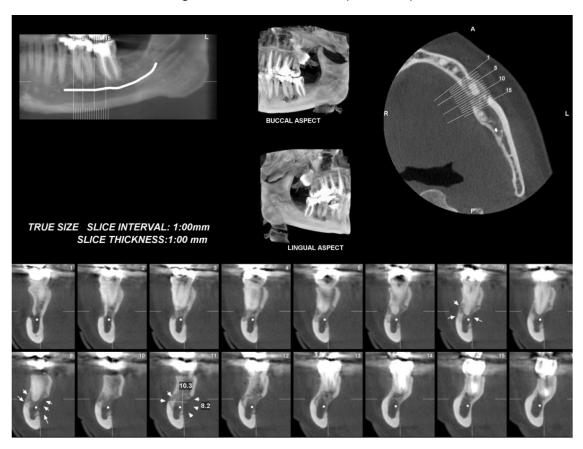


Fig. 3. CBCT view on second evaluation

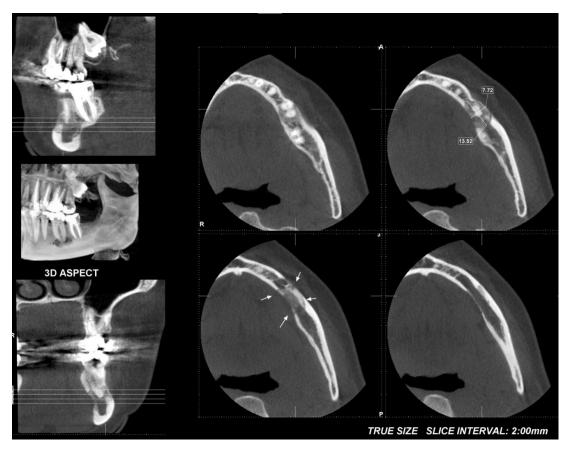


Fig. 4. CBCT view on second evaluation

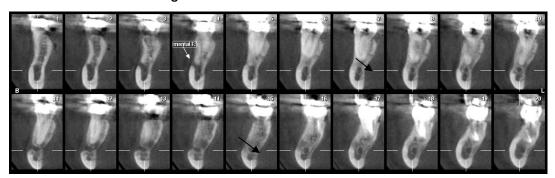


Fig 5. 6 month postoperative CBCT view (cross section), note the healing surgery site with granular bone (black arrows)

As mentioned before, most SBCs are radiolucent lesions without perforation of buccal and lingual cortexes and grow slowly. However, unlike the above typical items, the current case showed a mixed lesion with buccal and lingual cortical perforation and rapid growth. Acording to imaging findings our differential diagnosis included 1) pulpoperiapical lesions that were ruled out due to the tooth vitality, 2) locally aggressive benign tumors such as

ameloblastoma and myxoma, 3) mixed lesions such as COC (Calcifying Odontogenic Cyst) and Calcifying Epithelial Odontogenic Tumor CEOT (Pindborg tumor), 4) fibro-osseous lesions, such as focal cemento-osseous dysplasia FCOD and Cemento-ossifying fibroma COF, cementoblastoma, and finally 6) malignancies. Rapid growth and perforation suggest aggressive destructive behaviour. Moreover, the concomitant pain and numbness,

radiographic appearance (punched out single radiolucency), increased the probability of malignancies such as unifocal multiple myeloma (plasmacytoma). Ameloblastoma, was also taken into account but usually causes roots resorption. Considering that myxoma tends to involve women more than men, the lesion is more likely to be a myxoma. However, myxoma usually involves young people. The mixed radiolucent radiopaque lesions such Calcifying odentogenic cyst and calcifying epithelial odontogenic tumor can be considered as differential diagnosis, but usually they show well defined corticated margins and more expansions in radiographic appearance. The fibro osseous lesions such as Ossifying fibroma and focal osseous displasia can produce such

radiographic internal structures but have more expansion and seldom destruct the cortical plates and inferior alveolar nerve canal cortical boundaries. The radiographic appearance of the lesion is similar to cementoblastoma but cementoblastoma is often seen in young males and provides severe pain, in some cases expansion is seen without cortical plates perforation. As the current study mentioned above, this lesion should be differentiated from malignant lesions because of paresthesia of the lower lip that indicates the neural involvement, rapid growth and destruction of cortical boundaries. In this case paresthesia of lower lip is explained by destruction of cortical border of inferior alveolar nerve canal and compression of inferior alveolar nerve.

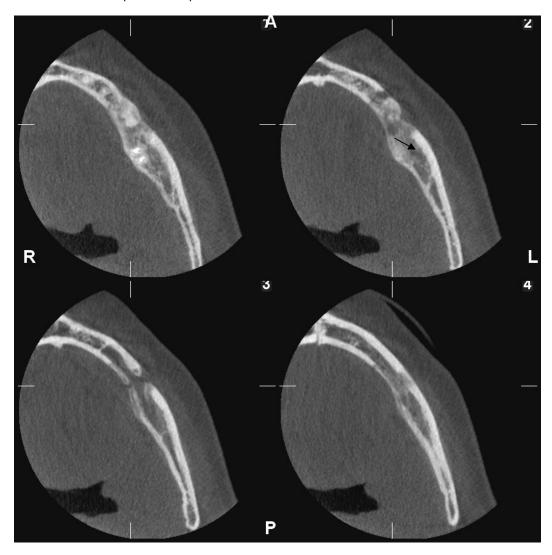


Fig. 6. 12 month postoperative CBCT view (axial view)

4. CONCLUSION

SBC mav have various radiographic presentations, some of which may show an aggressive behaviour: Rapid growth, destruction of bone cortex and involvement of nerve canal. Therefore. SBCs with such imaging presentations should be ruled out from more aggressive and malignant lesions. In such cases, a surgical approach is necessary and samples must be addressed for histopathological analysis.

CONSENT

All authors declare that 'written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

All authors hereby declare that all experiments have been examined and approved by ethics on research committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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