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Review Article

Solitary Bone Cyst and Aneurysmal Bone Cyst: Perplexing Pseudocysts of the Jaws

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ABSTRACT

Pseudocysts of the jaws include two very significant lesions – Solitary bone cyst (SBC) and Aneurysmal bone cyst (ABC). Both these cysts have very similar clinical and radiological findings. Histopathology aids in differentiating the two pathologies, with MRI findings offering confirmatory diagnosis of ABC. Differentiating between the two cysts and establishing a definitive diagnosis cannot be over emphasised as prognosis depends on the correct treatment modality after the diagnosis. A lot of updates with regards to the treatment of these cysts in long bones is available, with the same lacking in the cysts of the jaws. This probably could be due to the relative rarity of these cysts in the jaws. This article aims to address the various aspects of these two enigmatic pseudocysts of the jaws.

Keywords: Pseudocysts; Solitary Bone Cyst; Aneurysmal Bone Cyst; Pathogenesis; Hemorrhage; Head & Neck; Oral

1 INTRODUCTION

A cyst – "is a pathological cavity having fluid, semifluid or gaseous contents and which is not created by the accumulation of pus. It is frequently, but not always, lined by epithelium"[(1)].

By definition, a pseudocyst is a "pathological cavity without lining epithelium and with clinical and radiographic similarities to true cysts, except for histopathological findings"[(2,3)]. Pseudocysts of jaws are – Solitary bone cyst (SBC), Aneurysmal bone cyst (ABC), Static bone cyst and Osteoporotic bone marrow defect[(4)].

2 DISCUSSION

The pseudocysts of jaws are rare, have unclear aetiologies, present similar clinical and radiographic findings and are not easy to identify because of their tricky symptoms. Solitary bone cysts (SBC) and Aneurysmal bone cysts are neither true cysts or tumors (ABC), but can cause severe damage to the jaws.

Rudolph Virchow, a German pathologist first described SBC in 1876[(5)]. SBCs are also known by the names - Traumatic Bone Cyst, Hemorrhagic Bone Cyst and Idiopathic Bone Cavity[(6)]. The name SBC was proposed by Bernier and Johnson[(7)]. It is a rare entity in maxillofacial region, accounting for only 1-2% of all pseudocysts/cysts[(2,3)]. Most of the SBCs are located in long bones (90%), with less than 10% reported in jaw bones and mandible being the most favoured site[(8,9)]. Posterior (premolar-molar) region in either of the jaws is the most common location. Multifocal lesions, although rare are also reported[(8)]. Varying gender predilection has been reported, but males appear to be affected slightly more frequently than females[(10)]. The lesion occurs within 2nd decade of life with stranded cases reported after the third decade[(11)]. The majority of SBCs are detected accidentally by radiographic examinations done for other reasons. Few cases with symptoms of pain, swelling, paraesthesia, displacement and root resorption of the involved teeth, fistula and pathologic fracture have also been reported[(12)].

SBCs can present as solitary or multiple lesions that are usually asymptomatic and self- limiting and show spontaneous healing. Few cases have presented as progressive solitary or multiple lesions with a potential for recurrence. A third type of SBC maybe the ones associated with various non-neoplastic bone lesions [13].

Though the etiopathogenesis of SBC has not been definitively established, three theories - aberrant osseous growth theory, the process of tumour degeneration theory and the trauma-haemorrhage theory have been proposed by Harnet et al. [9] Generally approved theory is the trauma-haemorrhage theory which states that the intramarrow haemorrhage caused by any trauma gets eventually organised into a haematoma. This hematoma will exert pressure and cause venous obstruction and intracapsular exudation. Further, the enzymes in the exudate will promote bone resorption and aseptic necrosis of the bone marrow. This is a plausible explanation for the mandible being involved more than the maxilla, since the maxilla has a loose cortex permitting more vessels and hence more blood supply. The thick cortex of the mandible weakens its self-healing potential [14]. The explanation offered by trauma-haemorrhage theory is sound, yet there is no proof that the etiology is trauma in SBC [15].

Radiographically, solitary bone cysts can present as well defined or poorly defined radiolucent lesions of varying sizes. Though these pseudocysts show scalloping margins around associated roots, associated teeth test vital and root resorption is uncommon. Sometimes the large lesions show vague multilocular appearance and bone expansion. Such expansile multilocular lesions associated with root resorption may resolve slower with standard treatment [16].

SBC is striking in histology for lack of tissue on biopsy. The scanty biopsy tissue makes the diagnosis burdensome. The findings from the scant biopsy tissue are that of fibrovascular connective tissue, extravasated red blood cells and pieces of reactive vital bone without cystic epithelium. The finding of empty or fluid filled space at biopsy is suggestive of SBC. The act of biopsy procedure itself will initiate healing of SBC and it generally shows resolution within 6 months, with larger lesions taking longer time. Prognosis is good, but follow up is indicated [11].

Jaffe and Lichtenstein first described Aneurysmal Bone Cyst (ABC) in 1942 [17]. ABC is a pseudocyst wherein 70% of the cases affect sites other than jaws, the long bones (50%) and spine (20%) and only 2% in jaw bones [18]. They present as an osteolytic lesion similar to SBC, but are more aggressive than SBC [2,3].

Based on the pathogenesis, ABCs can be either primary (70% of cases) or secondary lesions. In primary ABCs CDH11 and USP6 gene (TRE17) rearrangements have been observed. The osteolysis as a result of dilatation and rupture of blood vessels due to intraosseous or subperiosteal haemorrhage or other haemodynamic changes is thought to be the mechanism of primary ABCs [19]. Recurrent alterations in cytogenetics have been observed in primary ABCs. Unlike primary lesions, the secondary ABCs are thought to be a result of its association with various pathologies. In the jaws the accompanying lesions reported are ossifying fibroma, cemento-ossifying fibroma, and malignant or metastatic bone lesions [14].

Patients with SAB generally report with complaint of pain and swelling in the affected region. Not so often they pathological fractures are also noted. Usually, radiograph reveals osteolytic lesion with well-defined margins. Sometimes, when the lesions are expansile involving the periosteum, it may produce soap bubble appearance [20]. The magnetic resonance imaging is useful in diagnosis of ABC as it shows cystic formations with typical fluid-fluid levels due to blood sedimentation [21].

ABC is a rapidly growing, locally destructive lesion. Histologically, numerous blood-filled cavities of different sizes separated by the fibrous septae will be seen. Spindle cells osteoclast-like giant cells, stromal mononuclear cells, and even small strands of osteoid and reactive woven bone will be evident on the fibrous septae [22,23]. Based on the predominance of vascular spaces and fibrous septae, ABCs can be either vascular type or solid type. If both the components are equally prominent then it is called as mixed type. In 95% of cases, the vascular variety of ABCs are present. These are known to cause severe haemorrhage and bone destruction following surgery [22].

A myriad of options is available in the treatment of ABCs. It varies from simple surgical curettage to wide resections. The intralesional curettage may be executed with or without adjuvants. Other modalities like embolization, percutaneous intralesional injections of sclerosing agents such as polidocanol or other agents like doxycycline have been tried. Cryoablation, radiation therapy, radionuclide ablation, or the systemic application of denosumab or bisphosphonates are few other tried methods [21]. Since primary ABCs show altered USP6 genes, targeted therapy options for such cases could be tried. However, no such treatment options are available currently as literature search did not reveal it. A treatment option that is optimal is yet to be ascertained.

3 CONCLUSION

Many cystic bone lesions can present similar symptoms both clinically and radiographically. Establishing definitive diagnosis on histopathology not only helps in identifying the lesion, but also helps the clinician in planning the treatment, as different pathologies require different treatment modalities for best prognosis.

Two such uncommon cystic pathologies with similar clinical and radiographic presentations are SBCs and ABCs. These cystic lesions lack lining epithelium and hence are classified as pseudocysts. Advanced imaging method such as

MRI guides the diagnosis of ABCs. Both these pseudocysts have unclear aetiopathogenesis. These cysts may be harmless that resolve with simple curettage as in most cases of SBCs or may be aggressive and need specific treatment. Generally, ABCs are aggressive lesions that require surgical interventions combined with innovative bone graft materials, autologous red bone marrow, sclerotherapy injections and systemic administration of certain drugs. As a result, a thorough understanding of the aetiology of pseudocysts, as well as quick and accurate diagnosis, is critical in establishing proper treatment modality to increase the cure rate and prognosis of these pseudocysts.

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