

Aneurysmal Bone Cyst of Lower Jaw: A Case Report and Review of Literature

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ABSTRACT

Introduction: Aneurysmal bone cyst (ABC) is an intraosseous expansile osteolytic benign lesion of bone that contains blood-filled cystic spaces. The etiology and etiopathogenesis are unknown. It is less common in the skull bones. The skull bone lesion commonly involves the mandible with three times greater frequency than that of maxilla. The disease is most frequent in adolescents with a slight female predominance. Clinical presentation is of a progressively rapid growth pattern characterized by expansion of involved bone and deformity.

Case report: Herein, we have presented a case of ABC of a 20-year-old Bengali male patient with an ABC located at right angle of the mandible. Cone-beam computed tomography revealed multilocular radiolucency in the right side of the angle and body of the mandible. Incisional biopsy was carried out, which confirmed the diagnosis of ABC.

Conclusion: The purpose of this study was to evaluate the history, etiology, clinical features, and review of literature of this age-old disease entity with very many gray areas, which is being long debated.

Keywords: Aneurysmal bone cyst, Cone-beam computed tomography, Histopathology.

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INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign osteolytic expansile lesion of bones characterized by intraosseous blood-filled spaces with fibroblastic tissue stroma

containing multinucleated giant cells and reactive woven bone. Aneurysmal bone cysts are not frequent in the jaws; more than 50% occur in the long bones and 20% in the vertebrae. Aneurysmal bone cyst is only 1.5% of all the nonodontogenic, nonepithelial cysts of the mandible. It involves the mandible more frequently than the maxilla (3:1). Aneurysmal bone cyst commonly affects young persons, especially adolescents with a slight female predilection. Three main types of ABC described in the literature are the conventional or vascular type, the solid type, and the mixed type. The conventional type represents 95% of all the ABCs, while the solid type comprises the rest 5%. The mixed variant shows characters of both the vascular and the solid types.¹ It is thought by some researchers to be a transitory phase of the former two variants of the lesion caused likely by certain activation of stable solid lesion. The conventional type is a rapidly growing, progressively expansile, destructive lesion causing cortical perforation and soft tissue involvement. The solid type is asymptomatic, may be noticed incidentally as radiolucency on a routine radiograph.²

CASE REPORT

A 20-year-old young male patient reported to the outpatient department of Dr. R. Ahmed Dental College & Hospital with an asymptomatic swelling on the right side of lower jaw over a period of 2 years, with a growth spurt in the last 2 months. He had undergone extraction of the right lower third molar just before the beginning of clinical symptoms. Family history was insignificant, and the onset was insidious without any history of trauma. The lesion caused facial asymmetry. Extraoral examination revealed diffuse swelling on right side of the body and angle of the mandible. The lesion was extending antero-posteriorly from the premolar region to right angle of the mandible measuring approximately 3 × 2 cm. The swelling was firm in consistency and its border was diffuse. On intraoral examination, a diffuse swelling in the buccal vestibule opposite to right molar segment of lower jaw was seen. On intraoral palpation, the lesion extended from concerned premolar region to the retromolar area with expansion of both the cortical plates. The regional second molar was pushed and displaced to the lingual area as shown in the photograph. Panoramic radiograph

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Fig. 1: Panoramic radiograph demonstrating multilocular radiolucent lesion involving body and ramus region of right mandible

revealed a multilocular radiolucency occupying the right side of the ramus of the mandible almost in totality, extending up to permanent lower right second molar in the body of the mandible (Fig. 1). Right molar and premolar teeth tested positive in the vitality test. Cone-beam computed tomography revealed large destructive lesion as radiolucency measuring about 4 × 3 cm involving the right ramus of the mandible almost in its entirety as well as the angle and the body of the mandible extending up to right lower second premolar. The linguoverted right lower permanent second molar was noteworthy. Blood tinged fluid was expressed out on aspiration.

Histopathological examination revealed multiple cyst-like blood-filled spaces separated by connective tissue stroma with trabeculae or osteoid and clusters of osteoclastic giant cell. The blood-filled spaces were devoid of epithelial lining and variable in size. Variable amount of hemosiderin was present in some areas (Fig. 2). The present case was consistent with the classic or vascular form described in the literature. Areas with osteoblastic differentiation and calcifying fibromyxoid were also present.

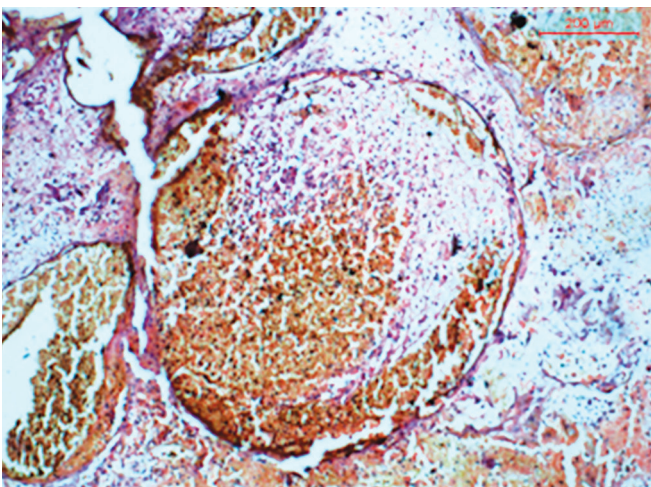


Fig. 2: Tissue section stained with hematoxylin and eosin showing fibrous connective tissue with sinusoidal blood-filled spaces

The etiology of the ABC is obscure. Aneurysm means dilatation of the vessels, but herein expansion of marrow spaces along with that of bone (cortical plates) takes place, which is regarded as the “blow-out effect.” So the nomenclature in usage is sort of a misnomer. There are many theories that attempt to explain its origin and classify these cysts as primary or secondary lesions. Many researchers believe the development of ABC mostly depends on high venous pressure and high marrow content of bone. Because of low venous pressure in the marrow of skull bones, it rarely occurs therein. The lesion is commonly found at the molar-ramus region of the mandible. Many researchers considered these cysts as congenital primary lesions that may coexist with other osseous pathologies. Jaffe and Lichenstein proposed that alteration of local hemodynamics due to increased venous pressure results in resorption of bone; connective tissue replacement and osteoid formation might form these lesions.³ History of trauma and subperiosteal hematoma formation might also be related to the development of ABC, but many cases have been described without any history of trauma,⁴ like ours. Another pathological mechanism could be the bone trauma that may facilitate this hemodynamic alterations.⁵ Even tooth extraction might cause trauma to the existing lesion. Due to the resultant change in hemodynamics, the existing lesion has shown aggressive rapid growth, as in the presented case. Many authors consider that ABCs are secondary lesions related to degeneration of a preexisting bone lesion or coexist with them, such as the central giant cell granuloma, fibrous dysplasia, osteoblastoma, or ossifying and cementifying fibromas. However, this hypothesis has not been in conformity with the histopathological findings.⁶ Clinical presentations are of variable nature. It ranges from a small asymptomatic one to rapidly growing expansile lesion causing pain. Facial deformity, neuralgic pain, pathological fracture, and perforation of cortical bone may also be present.⁷

This presented case showed intraoral lobulated growth with ulceration. The radiological features are also variable with different radiodensities showing multilocular radiolucency with well-corticated border resembling a honeycomb, soap bubble, or even unilocular. Destruction and perforation of the cortex with periosteal reaction have been reported in the literature but was not present in our case. Diagnosis is based mainly on both the radiological and histopathological evaluations. Only radiographic examination is sometimes misleading due to the proximity in radiographic appearances of lesions like ameloblastoma, myxoma, central giant cell granuloma, odontogenic cysts, or central hemangiomas of bone.⁸

Chromosomal anomalies having a role in ABC and cases of familial incidence have been reported in the literature. As the etiology of ABC is obscure, based on the previous reports, the role of genetics in the development of ABC was also palpated.⁹ The association of ABC with t(16;17)(q22;p13) has been reported. Recently, gene rearrangements of the USP6 gene on chromosome 17 and/or the CDH11 gene on chromosome 16 have been shown to be involved in ABC. However, the recent identification of recurrent chromosomal abnormalities indicated that the lesion might result from an arrest of maturation of the osteoblasts caused by USP6 overexpression and dysregulation of autocrine bone morphogenetic protein.¹⁰

MANAGEMENT AND PROGNOSIS

Treatment of the lesion is intralesional curettage or enucleation in usual cases. In the surgical intervention procedure, cryosurgery has also been applied. Rare cases demand more extensive surgical resection or other adjuvant therapy. Reoccurrence rate is 20 to 30% in different studies, and most frequently occur during the first year after surgical exploration, usually when subtotal in nature.

CONCLUSION

Even almost 125 years after the first mention of this kind of lesion and 75 years after referring to it as such, its etiopathology is still obscure and its nomenclature borders misnaming. This is because of the varied etiology, clinical presentation, radiological features, and histopathological presentation. A consensus about confirmatory diagnosis is found wanting most of the times. In the present case, the external appearance does not fully reveal the actual aggression of the lesion, emphasizing

the need for clinical presentation, radiological features, as well as histopathological analysis for confirmatory diagnosis.

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