Aneurysmal Bone Cyst Of The Mandible With Unusual Presentation - A Case Report

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Date of Receiving : 15/Sep/2012
Date of Acceptance : 25/Oct/2012

ABSTRACT: Aneurysmal bone cyst (ABC) is an uncommon lesion which has been found in most bones of skeleton, although the majority occurs in the long bones and in the spine. Among all the cystic lesions that can be found at the mandible or the maxilla it is very rare. This lesion represents less than 1% of all the bone cysts biopsied. The rare jaw lesions are encountered in the body and ramus of the mandible. Commonly reported in the second and third decade of life, ABC's are characterized by a rapid growth pattern with resultant bony expansion and facial asymmetry. Although ABC is a benign lesion, it can behave locally in an aggressive manner because of its rapid growth and osteolytic capacity. The literature contains conflicting reports on the clinical and radiological features of ABC of the jaws. The radiographic appearance of ABC varies from a unicystic radiolucency or moth-eaten radiolucency to an extensive multilocular lesion. This paper describes a case of ABC in a 20 years old male patient affecting the body of the mandible. Treatment consisted of surgical curettage of the lesion. A brief review of existing literature on ABC is also made.

KEY WORDS: Aneurysmal Bone Cyst, Pseudocyst, Reactive Lesion, Mandible, Multinucleated

INTRODUCTION

Aneurysmal bone cyst (ABC), is a rare non epithelized pseudocyst of jaws. They represent about 1.5% of all nonodontogenic and nonepithelial cystic of the jaws. Considering all types of jaw cysts the ABC is extremely rare with 0.5%. It was first described by Jaffe and Lichtenstein in 1942. They can usually be found in the metaphysis of the long bones, mainly in the “tibia and the femur”. The average age of occurrence is 13 years and 80% of patients are less than 20 years old with no gender predilection. The WHO defines ABC as a benign intra-osseous lesion, characterized by blood- filled spaces of varying size associated with a fibroblastic stroma containing multinucleated giant cells, osteoid and woven bone. The characteristic radiological features of ABC in the long bones are well documented as a well-defined expansile radiolucent lesion surrounded by a thin overlying cortex. In contrast the descriptions of ABC in the jaws are conflicting and vary from mainly a unilocular radiolucency to a ‘ballooned out' multilocular radiolucency with a honeycomb or soap-bubble appearance.

CASE REPORT

A 20 years old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling and pain on left side of face since 1 month. The patient was asymptomatic 1 month back after which swelling occurred. Initially the swelling was small in size and had gradually increased to the present size. It was also associated with altered sensation. The patient also had difficulty in eating food. The patient's medical and family history was unremarkable and there was no history of trauma. On extra-oral examination, facial asymmetry was apparent with a solitary diffuse swelling on the left side of mandible, measuring approximately \(3\times2\) cm in its greatest dimensions. It extends anteroposteriorly, 2 cm away from tragus of ear to angle of mouth and superoinferiorly from a line joining tragus of ear to angle of mouth to lower border of mandible. On palpation it was tender and firm with normal overlying skin. (Fig. 1)

On intraoral examination, generalised grade I stains and calculus were present. An ill-defined growth was present on the buccal aspect of 35, 36, 37 and 38 with vestibular obliteration, roughly oval in shape, measuring approximately \(3\times1\) cm in its greatest dimensions, extending from mesial aspect of 35 to the retromolar region. The overlying mucosa appears pink with purulent discharge.(Fig.2) On palpation, it was tender, soft in consistency and was associated with pus discharge. Grade II mobility was present w.r.t 36, 37, 38

Considering the history and the clinical examination, provisional diagnosis of chronic suppurative osteomyelitis was made.

In investigations, IOPAR and panoramic view were taken. (Fig. 3,4) The intra oral periapical radiograph shows ill defined radiolucency on the disto-proximal surface of 36 suggestive of grossly carious tooth. Loss of lamina dura was seen at the apex of the mesial and distal roots. An ill defined radiolucency was seen at the apex suggestive of periapical pathology. Horizontal bone loss was seen on the distal aspect of tooth. W.r.t 37, crown appears normal, loss of lamina dura was seen along the mesial and distal aspects and apex of mesial and distal roots. An ill-defined radiolucency was seen in the apical region suggestive of periapical pathology. Horizontal bone loss was seen on the mesial and distal aspect of tooth. W.r.t 38, the crown appears normal with loss of lamina dura on mesial aspect of mesial root. Bone loss was seen on the mesial aspect of tooth. Panoramic radiograph revealed a single unilocular lesion with ill defined borders, roughly oval in shape, approximately \(2.5\times1\) cm.
in its greatest dimensions, extending from distal aspect of 34 to the mesial aspect of 38. No root resorption was evident. The internal structure comprises of mixed radiolucent and radiopaque lesion. On the basis of the radiographic features a radiographic diagnosis of chronic osteomyelitis was made. The lesion was surgically excised and was sent for histopathologic examination. The microscopic features show predominant bony trabeculae with osteocytes within it and osteoblastic rimming. Few areas show fibrocellular stroma with predominant cystic spaces filled with extravasated RBCs. Multinucleated giant cells were evident with inflammatory component. These microscopic features were suggestive of ossifying fibroma with secondary changes of Aneurysmal bone cyst.

**DISCUSSION**

Aneurysmal bone cyst is usually considered to be a reactive lesion of the bone rather than a cyst or true neoplasm. Most believe that ABC is the result of a vascular malformation within the bone. The cause of the malformation is however a topic of controversy. It is not clear whether the lesion is primary or occurs in a preexisting bone lesion. Eighty percent of the ABCs occur in patients below 20 years of age with no gender predilection. However, studies have claimed a slight female preponderance. ABC is most common in those regions of the skeleton where there is a relatively high venous pressure and high marrow content. This explains its rare occurrence in the skull bones in which there is low venous pressure. However, when present, the mandible is most commonly affected (mandible-maxilla ratio 3:1), with a higher predilection for molar and ramus region.

ABC is extremely variable in clinical presentation, ranging from a small, indolent, asymptomatic lesion to rapidly growing, expansive, destructive lesion causing pain, swelling, deformity, neurologic symptoms, pathologic fracture and perforation of the cortex. The radiological features of ABC in the jaws are quite conflicting; may appear as a unilocular or multilocular radiolucency with expansion and thinning of the surrounding cortical bone, appears cystic resembling a honeycomb or soap bubble appearance since it may be traversed by thin bony septa. The periphery is usually well defined and the shape is circular or “hydraulic”. After an ABC becomes large, there is propensity for extreme expansion of the cortical plates and it can displace or resorb teeth. A characteristic radiographic feature of ABC is the “ballooning” distention of periosteum with a thin outline of reactive, subperiosteal bone.

Histologically, ABC consists of a fibrous connective tissue stroma with numerous blood filled caverns of sinusoids, multinucleated giant cells and osteoid. Hemosiderin is present in variable amounts and osteoid and bone formations are variable. Since a normal epithelial lining is lacking, the lesion is also referred to as pseudocyst.

The treatment of the ABC is determined by the nature of any associated lesion. There is no uniform treatment and management of ABC due to its varied nature. The usual treatment of choice is curettage as it is a benign lesion. The lesion regressed spontaneously. Some authors have also recommended supplementing curettage with cryotherapy. The defect can be filled up with bone chips prior to cryosurgery. Segmental resections are performed with immediate bone grafting if the lesions have been found to be extensive and cause functional and cosmetic deformities. Radiation is not recommended as sarcomatous change has been reported in these lesions after irradiation. A high recurrence rate of 53-66% has been reported for ABC in the jaws. Therefore, a close follow-up of the cases is recommended.

**CONCLUSION**

Aneurysmal bone cyst of the jaws represents an enigmatic pseudocyst with variable clinicopathological, histological and radiographic presentations, therefore, often posing a diagnostic dilemma. As the radiological features of ABC are varied, resembling many lesions, histopathologic analysis is a must for the diagnosis.

**REFERENCES**

LIST OF PHOTOGRAPHS

Fig 1: Extraoral swelling on left side of face

Fig 2: Intraoral Photograph

Fig 3: Intra Oral periapical Radiograph irt 36, 37, 38

Fig 4: Panoramic view