Aneurysmal Bone Cyst: A Case Report with Review of Literature

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Abstract

Aneurysmal bone cyst is an uncommon non-odontogenic benign lesion of specially long bones and vertebral column. However; no part of the skeleton is exempted with the possible exception of skull. These are the rare sites of occurence. Commonly reported in second and third decade, is characterized by a rapid growth with resultant bony expansion. General knowledge of this rare entity is essential since so many radiolucent lesions are similar. Management is either curettage or surgical resection. This paper illustrates a case of Aneurysmal bone cyst in a 30 year old female that presented a rapidly growing, expansile bony growth of mandible.

Key words: Aneurysmal bone cyst, Non-Odontogenic, Radiolucent

Introduction

The World Health Organization defines aneurysmal bone cyst (ABC) as "a benign tumour-like lesion with an expanding osteolytic lesion consisting of blood-filled spaces of variable size separated by connective tissue septa containing trabeculae or osteoid tissue and osteoclast giant cells".[1] Jaffe and Lichtenstein were the first to recognize ABC as an intraosseous, osteolytic lesion chiefly affecting the metaphyseal region of long bones (especially femur and tibia) and between 12 and 30% in the spine.[2] Considering all types of jaw cysts, the ABC is extremely rare with 0.5%.[3] This paper illustrates a rare case of aneurysmal bone cyst in the anterior region extending upto posterior region of mandible in 30-year old female.

Case report

A 30 year old female patient was reported to outpatient department in Shree Bankey Behari Dental College and Research centre, Ghaziabad with a complaint of painful swelling in right lower back tooth region since 1 month. On clinical examination, firm swelling measuring 3 x1inches extending from central incisor to first molar of right side of the body of mandible. Overlying skin was free and normal in color. (Figure 1)

Intra orally the lesion was bluish in color. On palpation it was fixed, rubbery hard, tender and non fluctuant. Submandibular lymph node was just palpable and non tender. The remaining teeth were vital as tested by thermal and electrical vitality test.

On aspiration, blood stained fluid with predominant RBCs was found. On the clinical examination and FNAC it was suspected as Central giant cell granuloma / aneurysmal bone cyst was made. Further the relevant investigations were carried out.

Radiographs

OPG (PAN View) revealed a unilocular radiolucency extending from central incisor to distal surface of first molar. Superiorly involving the roots of remaining teeth without root resorption. Inferiorly compressing the madibular canal with scalloped borders. Few radio opaque foci are visible which are showing honey comb or soap bubble like appearance to some extent. (Figure 2)

After surgical curettage the currated material were sent to the department of Oral Pathology for histopathological examination. Microscopically hematoxylin and eosin stained section revealed connective tissue stroma showed numerous vascular channels with hemosiddern like pigment at some locations. (Figure 3) Mature and immature bony trabeculae showing osteocytes with unevenly distributed multinucleated giant cells were also evident. (Figure 4 & 5) Few vascular channels are not lined with endothelial cells. Based on the above features, the diagnosis was compatible with Aneurysmal bone cyst.

Operative Notes: lesion was curetted surgically by taking trapezoidal flap under general anesthesia. The buccal cortical plate was egg shell thin without perforation and the lingual cortical plate was slight expanded. A bluish brown, soft vascular tissue was found in a multiple loculi after peeling of the buccal cortical plate. The lesion was totally curetted until there was no bleeding, indicating complete removal of lesion. Lastly the incision was sutured back. The patient follow up was uneventful and wound healing was normal.

Discussion

Although the aneurysmal bone cyst is a lesion relatively common in the skeletal structure, is unusual in facial skeleton. In 1893, Van Arsdale [4] called this lesion "homerus ossifying haematoma". In 1940 Ewing used the

term "aneurismal" to describe such lesion [5]. In 1942, Jaffé and Lichtenstein [6] used the term "aneurismal cyst" and in 1950 they coined the term "aneurismal bone cyst" for the first time in literature. In world literature, about 160 cases of aneurysmal bone cyst in the maxillofacial region are reported.7 Bernier and Bhaskar described the first case of ABC in the jaws. [2, 8] Only 1.9% of all ABCs occur in the jaws, representing 1.5% of all nonodontogenic cysts. [9] The average age of occurrence is 13 years and 80% of patients are less than 20 years old with no gender predilection. 10 According, to Marx the peak occurrence is between 5 to 15 years of age, although some cases develop on 20-30 years as well. In facial skeleton, its occurrence is rare with two thirds were located in mandible (the body of the mandible 40%, the ramus 30% and the angle 19%) and one third in the maxilla **(3:1)** It is an expansile osteolytic pseudocyst which can attain great dimensions and may cause symptoms owing to its site and size and rapidity of growth i.e. swelling, deformity, pain, neurologic symptoms, and pathologic fractures**.** [6] In the present case, lesion was found in mandible in 30year old female extending from 41 region to 46 region. Considering the demographic prevalence and the clinical findings our case was compatible with the general agreement.

The etiology of ABC is unclear and controversial. One theory states that trauma causes an inciting injury to periosteal vessels, thus initiating the development of ABC. [6] However Tillman et al, reporting 95 cases, demonstrated no significant history of antecedent trauma. [11]. Jaffé and Lichtenstein refer to alteration in local haemodynamics causing increased venous pressures and engorgement of the vascular bed in the transformed bone, leading to resorption, connective tissue replacement, and osteoid formation. [12] Additional theories about the etiology of this lesion are a subperiosteal intraosseous hematoma. [6] In the present case, trauma could have contributed to the development of the lesion.

.The radiographic features are not pathognomonic and there is no consensus in the literature in this regard. The "blown-out" appearance in radiographs gives the appearance of a "bone cyst". The lesion may appear unicystic or as a unilocular, multilocular (soap-bubble or honeycomb) or moth-eaten radiolucency, causing expansion, perforation or destruction of the bony cortices. There may also be an associated periosteal reaction with reactive new bone formation, resulting in a peripheral sclerotic border in some cases**.** [2, 13] Our case presented as a unilocular radiolucency; however, resorption of roots of the involved teeth was not observed in spite of the close proximity of the radiolucency to the roots.

Histologically the ABC is considered a pseudocyst due to the absence of an epithelial wall. The ABC is an expanding osteolytic lesion containing blood-filled spaces of variable size, separated by connective tissue by bone trabeculae or osteoid tissue and many osteoclastic giant cells.

It is important to differentiate the ABC from other pathologies that occur in the maxillofacial region. These include peripheral and central giant cell reparative granuloma, traumatic bone cyst, brown tumor of hyperparathyroidism, myxoma, fibrous dysplasia, desmoplastic fibroma, fibrous histiocytoma, hemangioma, osteogenic sarcoma, globulomaxillary cyst, hamangioendothelioma, and hemangiopericytoma. [14] The initial diagnosis can be made radiographically. However definitive diagnosis requires histopathological examination of the surgical specimen. In our case the histopathological finding was consistent with above mentioned features.

The treatment of this lesion consist of complete surgical excision, it demonstrates a low recurrence rate. It has been proposed radiation therapy however the risk of subsequent malignant degeneration is present. It has been reported sarcoma arisen within radiated ABC. Even curettage of the cysts has a recurrence rate as high as 50%. Technical difficulties in entirely removing very large lesions can be the explanation for very different recurrence rate within literature. This article suggests how a complete surgical resection can definitively eradicate this aggressive bone lesion with a quite high recurrence rate. [6, 15] In conclusion surgical excision is the treatment of choice and the differential diagnosis of ABC from malignant tumours is the most important clinical aspect. No recurrence was observed at upto 8 moths follow-up.

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Legands

Fig. 1: Swelling extending from central incisor to first molar of right side of mandible (Frontal & Lateral view)

Fig. 2: OPG revealed unilocular radiolucency on the right side of body of mandible

Fig. 3: Numerous vascular channels with hemosiddern like pigment

Fig. 4: Bony trabeculae showing osteocytes

Fig. 5: Stroma showing unevenly distributed multinucleated giant cells

Acknowledgement

I acknowledge with extreme gratitude, the support provided in getting the clinical, radiological, photographs to Dr. Siddharth Gupta, reader, Dept.of Oral Medicine, Diagnosis & Radiology and to Dept. of Oral Surgery.

We acknowledge the contribution of Dr. O.P.Gupta Professor, Ms. Mohita Kapoor in preparation of the manuscript. We thank Mr. Vineet, undergraduate student for the support.

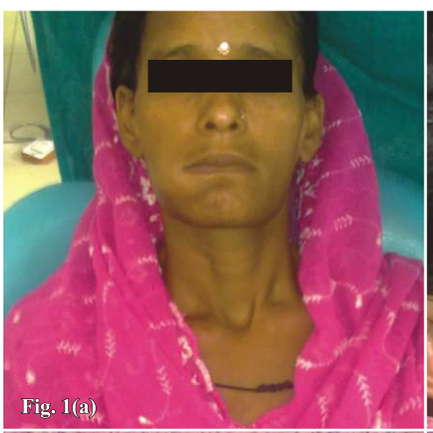
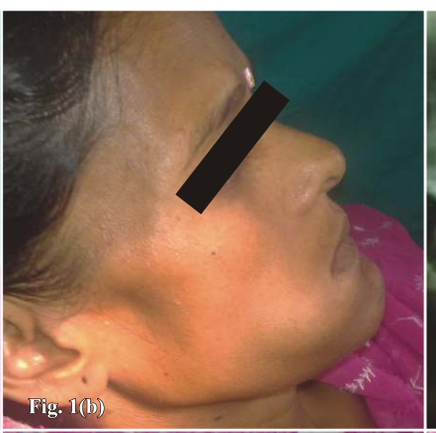


Fig. 1(a)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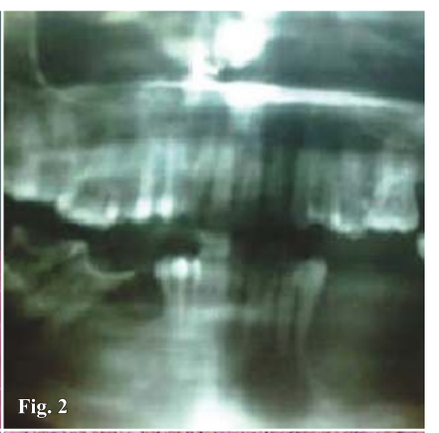
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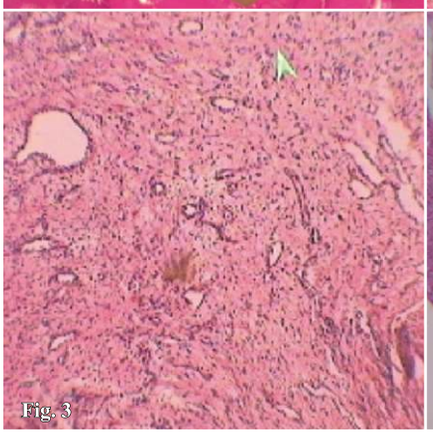
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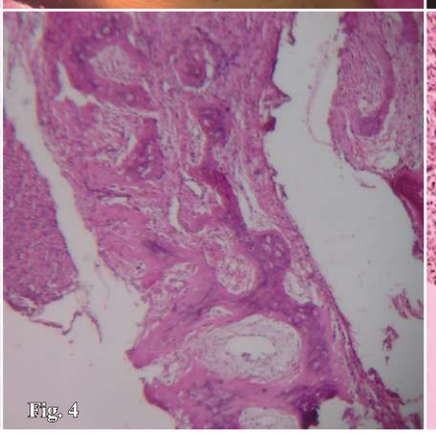
Fig. 4

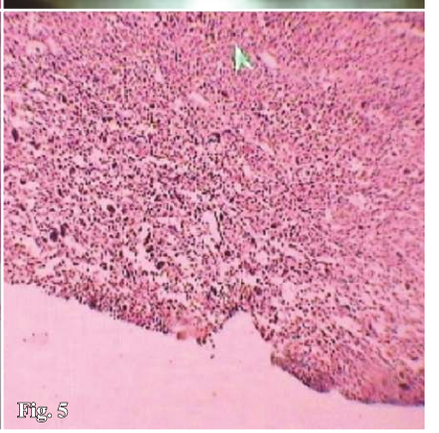
Fig. 5