



CASE REPORT

Giant Aneurysmal Bone Cyst of the Mandible

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ABSTRACT

An aneurysmal bone cyst (ABC) is a benign osteolytic bony lesion that commonly affects the long bones with rare presentation in the jaws. The etiopathogenesis of ABC is unsure. Several theories have been suggested like trauma, intramedullary hematoma, alterations in local hemodynamics, reactive malformation, and genetic predisposition. Though ABCs are considered as secondary phenomenon in preexisting benign and malignant bony lesions, intermittent reports of ABCs with primary/denovo origin are generating perplexity in the scenario.

Here, we describe a rare case of giant ABC involving mandible extending from right angle of mandible to left canine region which crosses midline, in a 10-year-old female patient, without any evidence of preexisting bony lesion.

Keywords: Aneurysmal bone cyst, Mandible, Primary/denovo, Secondary phenomenon.

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INTRODUCTION

Aneurysmal bone cyst (ABC) was first reported by Van Arsdale in 1893, who described it as ossifying hematoma.¹ Jaffe and Lichtenstein in 1942 were the first to recognize ABC as an intraosseous, osteolytic lesion, chiefly affecting the metaphyseal region of long bones and vertebrae.¹ Bernier and Bhaskar in 1958 described the first case of ABC in the jaws.^{2,3} In 1972 Schajowicz in his Histopathological Classification of Primary Bone Tumors placed ABC in group IX tumor-like lesions, which was latter modified by World Health Organization (WHO) in 1993.⁴

The WHO defined ABC as a benign tumor-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-like giant cells.⁵ Aneurysmal bone cyst is

considered a pseudocyst because it is not lined by epithelium.² Fifty percent of ABCs arise in the long bones and 20% in the vertebral column; and only about 12% affect the head and neck region of which 2% occurs in jaws.^{6,7} Mandible is more frequently affected than the maxilla, the proportions varying from 2:1 to 11:9. The body, ramus, and angle of the mandible are the main locations; rarely it occurs in mandibular condyle and coronoid process.⁷ It generally affects young adults below 20 years of age and there is no definite gender predilection.^{6,8}

Various theories have been proposed for etiopathogenesis of ABCs including post traumatic, reactive malformation, genetic predisposition, and dilatation of local vascular network due to increased venous pressure caused by local circulatory abnormalities.^{9,10} Consistent finding of ABC, like changes in preexisting primary bony lesions, gave rise to the well-accepted theory that it is a secondary phenomenon. Cumulative evidences of ABCs without any association of preexisting lesions are now raising a question on exact etiopathogenesis suggesting a primary/denovo variant.^{8,10}

We hereby report a case of giant ABC without any histological evidence of preexisting bony lesion, involving mandible extending from right angle of mandible to left canine region which crosses midline in 10-year-old female patient.

CASE REPORT

A 10-year-old female patient reported to Department of Oral Pathology and Microbiology, MGM Dental College and Hospital, Navi Mumbai, with a chief complaint of pain and swelling in lower right back region of the jaw since last 8 months. The patient was apparently well 8 months back until she noticed the swelling in the same region, which had suddenly increased to the present size. There was no history of trauma or any other disease affecting the jaw and other bones. Extraoral examination revealed mild swelling was present on the right side of the mandible (Fig. 1). Temperature of the overlying skin was normal and there was no sign of inflammation. An intraoral examination revealed a diffuse bony swelling from right retromolar region to left deciduous canine region, crossing the midline. There was bicortical expansion with vestibular obliteration. On palpation, swelling was firm to hard in consistency and tender.

Orthopantomogram (OPG) and lateral oblique view radiograph revealed a large multilocular radiolucent,

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Fig. 1: Extraoral view showing mild swelling on right side of the mandible

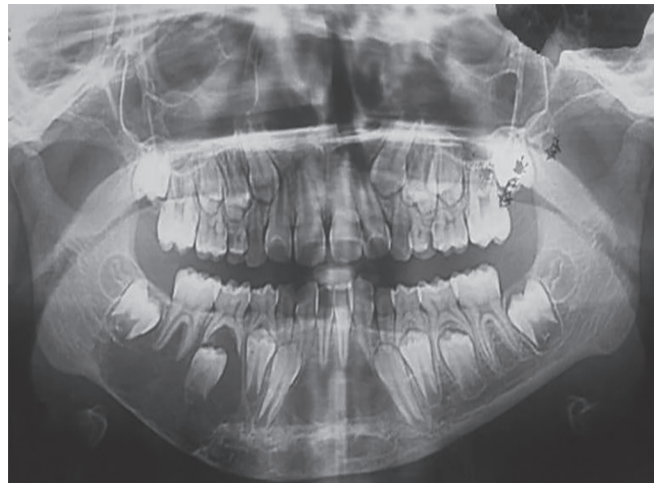


Fig. 2: Preoperative OPG showing a multilocular radiolucency from distal aspect of unerupted 47 to 73 region and thinning of right inferior border of mandible

osteolytic lesion with well-defined border from distal aspect of unerupted 47 to 73 region. No lateral or apical root resorption, displacement, or tipping of the teeth was seen. Thinning of right inferior border of mandible was evident (Fig. 2). Magnetic resonance imaging (MRI) of the lesion showed an expansile multiloculated well-defined lesion extending from right angle of mandible into the right body and across the midline up to the left canine region. The cortex was thinned out but no definite breach was seen. Coronal computed tomography (CT) scan image demonstrated expansile radiolucent lesion of the right mandible. Axial CT image showed circular well-defined radiolucent lesion on the right mandible (Fig. 3). Fine needle aspiration cytology was performed from the buccal aspect of the lesion from 43, 44 region. The aspirate yielded frank blood which showed a large number of erythrocytes predicting a vascular lesion.

Segmental resection of mandible from right angle of mandible to 73 region was done followed by reconstructive treatment protocol (Fig. 4). Gross excised lesion was measuring about $7.5 \times 2 \times 3.5$ cm with buccal and lingual surface showing no perforation (Fig. 5). Radiograph of the excised gross specimen showed multilocular radiolucency and ballooning expansion of buccal and lingual cortex of mandible (Fig. 6). Microscopically, hematoxylin & eosin (H&E)-stained decalcified section showed irregular multiple sinusoidal spaces engorged with blood separated by septa of fibro-collagenous tissue containing few osteoclast-like giant cells and trabeculae of woven bone (Figs 7 and 8). The sinusoidal spaces lacked an endothelial lining (Fig. 9). Meticulous histopathological evaluation of entire specimen confirmed the absence of any preexisting pathology. Thereby, the histopathological diagnosis of ABC was established. The patient is under regular follow-up and there is no clinical evidence of recurrence.



Fig. 3: Axial CT scan image of the right mandible revealing circular radiolucent lesion with bicortical expansion and thinning

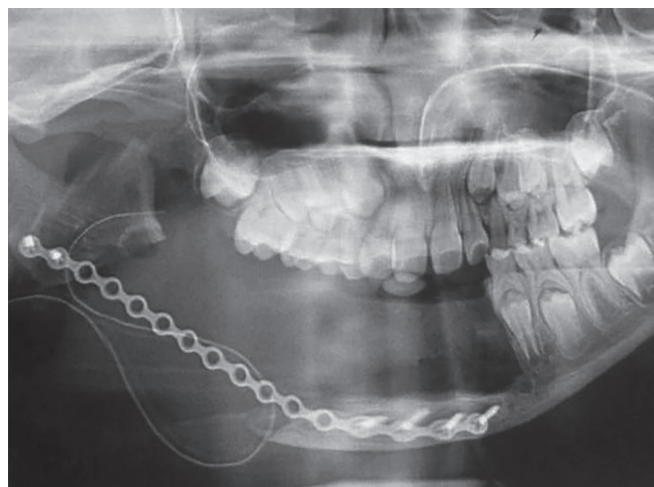


Fig. 4: Postoperative OPG with reconstruction plate



Fig. 5: Gross specimen of mandible showing expansile lesion with a smooth buccal surface

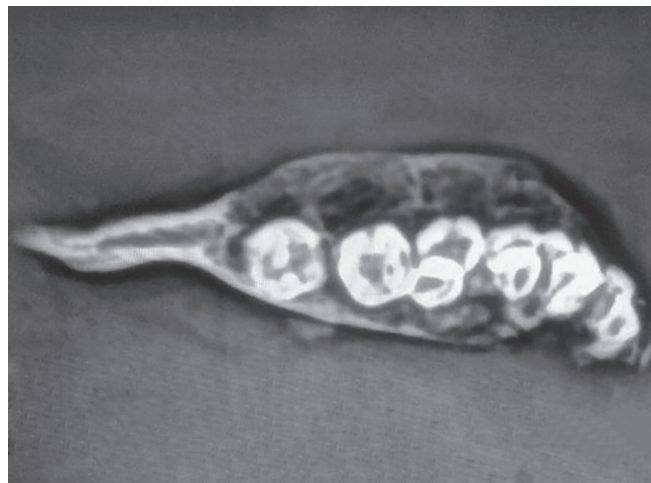


Fig. 6: Radiograph of the excised gross specimen showing multilocular radiolucency and ballooning expansion of buccal and lingual cortex of mandible

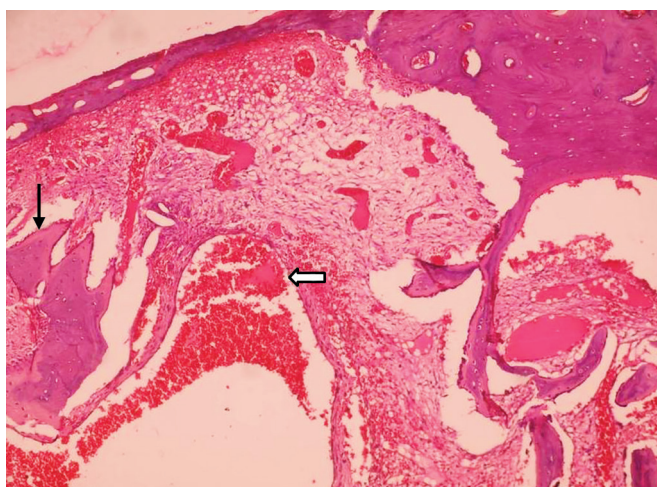


Fig. 7: Sinusoidal spaces engorged with blood (white arrow) separated by septa of fibro-collagenous tissue with trabeculae of woven bone (black arrow) (H&E, 10x)

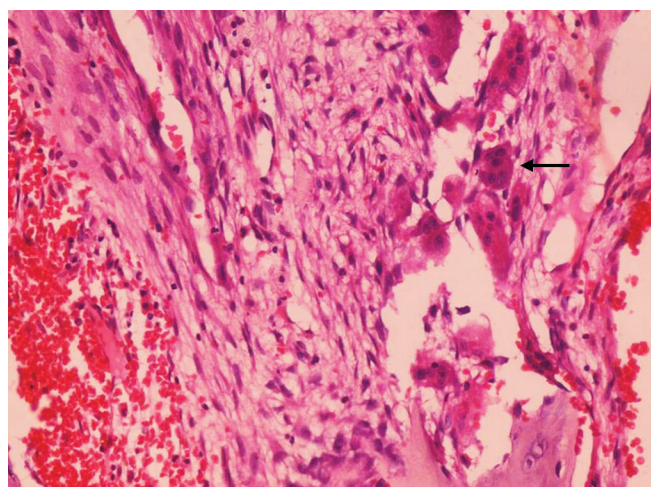


Fig. 8: Multinucleated osteoclast-like giant cells (arrow) in the fibrous tissue (H&E, 40x)

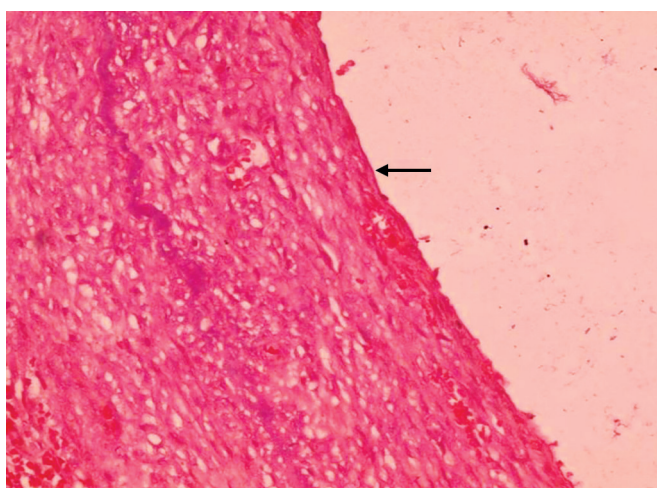


Fig. 9: Sinusoidal space lacked an endothelial lining (arrow) (H&E, 40x)

DISCUSSION

Aneurysmal bone cyst is most commonly found in long bones and in vertebral column. In the jaw, 2% cases are reported.^{1,7} It affects young adults below 20 years of age with no definite gender predilection and, in the jaws, mandible is most common site.^{6,8} In our case, it has affected the mandible of a 10-year-old female patient. Aneurysmal bone cyst is extremely variable in clinical presentation ranging from small, indolent, asymptomatic lesion to rapidly growing, expansile, destructive lesion causing deformity, pain, swelling, and perforation of the cortex.¹¹ In our case, it was a rapidly growing extensive lesion without perforation of the cortex and thus the term giant ABC was suitable.

The radiological features of ABC in the jaws are variable; it may appear as unilocular or multilocular,

expansive, osteolytic radiolucent lesion, with expansion and thinning of surrounding bone.¹² In our case, a multilocular radiolucency causing expansion of the cortical plates and thinning of the inferior border of the mandible was evident. Histopathologically, classic or vascular form of ABC consists of many sinusoidal blood-filled spaces in a fibrous stroma, with multinucleated osteoclast-like giant cells and osteoid. Variable amounts of hemosiderin is also present.^{10,11} Solid form is the other histological type, which consists of a dense stroma, scanty sinusoids, few blood vessels, and caverns. Features of both vascular and solid form are seen in mixed form of ABC.¹⁰ The histological features in our case were consistent with the classical form. However, there was no evidence of preexisting bony lesion in the entire specimen.

Till now cumulative evidences show various possible hypotheses in etiopathogenesis of ABC in regards to its origin (primary/secondary). Aneurysmal bone cyst is a secondary phenomenon arising from preexisting bony lesions, such as ossifying fibroma, giant cell reparative granuloma, chondroblastoma, solitary bone cyst, giant-cell tumor of the bone, osteoblastoma, osteosarcoma, fibrous dysplasia, and fibromyxoma, and is widely accepted.¹³ However, there may be a possibility of extensive degeneration of preexisting lesions in ABCs either by obliteration/reduction of bony pathologies to small undetectable residual portions or by progression of small cystic areas, especially in giant cell lesions to an ABC alone without any identifiable precursor lesions.¹⁴⁻¹⁶ This concept has been challenged by a number of authors leading to another possibility of its primary/denovo origin that is without any association with preexisting bony lesion.^{17,18}

According to Steiner and Kantor,¹⁷ the ABC can develop as either a primary or secondary lesion associated with other bone lesion which was also supported by Harnandez et al.¹⁸ Primary ABCs could be congenital or acquired and could originate from preexisting arteriovenous malformations. The congenital one is seen in children and young adults without any history of trauma, while the acquired one is seen in adults with a history of trauma. The secondary type is suggested to be associated with degeneration of preexisting benign and malignant bony lesions.¹⁸ In the present case, absence of any preexisting bony pathologies through meticulous histopathological evaluation of the entire specimen supports the origin either as primary/denovo or an extensive degeneration of preexisting lesion presenting as ABC alone.

The size, site, and extent of the lesion determine the level of treatment, ranging from simple curettage to extended resection.¹⁹ In the present case, by considering the age of the patient and aggressive nature of the lesion,

a conservative surgical resection of involved portion of the mandible was carried out.¹¹ Aneurysmal bone cysts have a higher rate of recurrence and generally recur within the 1st year after the initial treatment. Reported recurrence rates after surgical curettage and resection of ABCs are 19 to 50 and 11% respectively.²⁰ The patient is under regular follow-up and till date, there is no evidence of recurrence.

CONCLUSION

The lack of knowledge concerning the genesis of ABC and its confusion with similar looking lesions in the bone make exact compilation of data concerning the lesion a difficult task. It is mandatory to correlate the clinical, radiographic, and histopathological examination to arrive at the accurate diagnosis. Aneurysmal bone cyst has a high rate of recurrence, so conservative surgical resection is the treatment of choice.

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