Aneurysmal Bone Cyst Mimicking Ameloblastoma: A Rare Case Report

Reshna Roy¹, Jahnobi Dutta², Sherin Nair³, Neelutpal Bora⁴

¹PG Resident Surgeon, Department of Oral Pathology and Microbiology, GDC, Dibrugarh, India, ²Senior Lecturer, Department of Oral Pathology and Microbiology, GDC, Dibrugarh, India, ³Reader, Department of Oral Pathology and Microbiology, Kothiwal Dental College, Moradabad, India, ⁴Senior Lecturer, Department of Orthodontics and Dentofacial Orthopaedics, GDC, Dibrugarh, India

Abstract

Aneurysmal bone cyst (ABC) is rare benign lesion of bone which is infrequent in craniofacial skeleton. ABCs are characterized by rapid growth pattern with resultant bony expansion and facial asymmetry. Clinical presentation of the ABC varies from expansile, destructive lesion causing pain, swelling, deformity, and perforation of the cortex to the relatively innocuous type which may not produce any clinically evident bone expansion. As the radiologic and clinical presentation of ABC is extremely variable, histopathologic examination has a great emphasis for the diagnosis. We describe a case of ABC in a 22 years male patient affecting the buccal and lingual cortical plates with expansion on the lower border of mandible. Treatment consisted of hemimandibulectomy of the lesion. A yearlong follow-up revealed complete healing of the involved site with restoration of acceptable esthetics and no recurrence.

Key words: Aneurysmal bone cyst, Pseudocysts, Mandible

INTRODUCTION

Aneurysmal bone cyst (ABC) has been recognized since 1983 when it was described as an ossifying hematoma by van Arsdale. Jaffe and Lichtenstein suggested that the ABC is a bone benign injury, recognized as a solitary clinical-pathological entity. [1-3]

The aneurysmal term is used to describe the balloon-shaped distension of part of the affected bone that results in characteristic radiographic appearance frequently viewed.^[4] Shear and Speight mention a case where an injury was observed near the orbit's floor and another one near the zygomatic arch.^[5] The radiograph presents a uni- or multi-locular radiolucent injury; cortical bone expansion is described as a balloon-shaped stretching of the affected bone. The teeth can be found displaced, and their roots might have resorption. In the computerized

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tomography, an image compatible with uni- or multilocular cyst with cortical expansion is observed, with periosteal reaction or the appearance of moth-eaten shape. [6] There are several pathologic identities that mimic the same image such as the ameloblastoma, myxoma, central giant cells granuloma, the odontogenic cysts, and the central bone hemangioma.

The pathogenesis of the ABC is controversial, and several theories were postulated to explain it. It has been proposed that a trauma, a malformation, or a neoplasia could disorganize the local bone microvasculature resulting in an abnormal vascular condition that is the ABC.^[7]

Macroscopically during the operation, it is common to note an intact periosteum and a very thin layer of bone covering the cyst. When this is removed, several hidden blood vessels can be seen. The bleeding can be profuse and hard to control until the pseudocyst is removed.

CASE REPORT

A 22-year-old male patient had reported to the department of oral and maxillofacial surgery with a chief complaint of pain and swelling in the right side of jaw for 5–6 months.

Corresponding Author: Reshna Roy, Department of Oral Pathology and Microbiology, GDC, Dibrugarh, India. E-mail: reshnaroy6@gmail.com

On Clinical Examination (C/E), there was hard tissue swelling over the buccal and lingual cortical plates with expansion on the lower border of mandible. On palpation, the swelling was firm to hard in consistency with positive crepitation in relation to 43–47 [Figure 1].

Orthopantomogram showed multilocular radiolucency over the right mandible extending from 43 to 47 region with well-defined margins [Figure 2]. A computed tomography scan was requested which showed injury on the right mandible body approximately 4 cm × 5 cm in dimensions [Figure 3].

Based on the clinical findings and radiological findings, a provisional diagnosis of multicystic ameloblastoma of the mandible with resorption of molar roots was given. Differential diagnosis of myxoma, central giant cell granuloma, odontogenic cysts, or central hemangioma of the bone was given.



Figure 1: Pre-operative photograph of patient. Arrow pointing the swelling

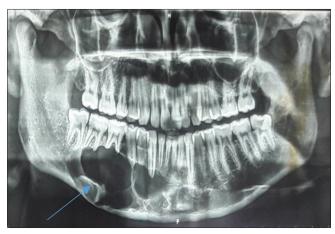


Figure 2: Orthopantomogram showing multilocular radiolucency over the right mandible extending from 43 to 47 region with well-defined margins

The case was referred to the department of oral and maxillofacial surgery. After routine blood examination, the lesion was approached under general anesthesia. The buccal flap was raised, thin cortical border was exposed and the tumor was excised along with the tooth and sent for histopathological examination [Figure.4].

The hematoxylin and eosin stained section (×10) revealed the presence of fibrocellular connective tissue stroma. The stroma shows blood-filled spaces and abundant young fibroblasts [Figure 5]. The stroma contains few inflammatory cell infiltrate, numerous dilated blood vessels, and calcified areas.

DISCUSSION

ABC develops mostly in maxillofacial bones depending on high venous pressure and high marrow content. Therefore, it is rarely seen in the skull bones where there is low venous pressure. The mandible is affected 3 times more when

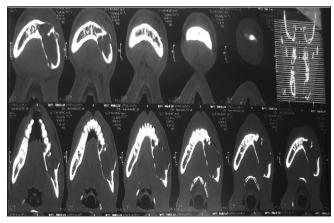


Figure 3: A computed tomography scan showed injury on the right mandible body approximately $4~\rm cm \times 5~cm$ in dimension



Figure 4: The formalin fixed hard tissue specimen with dimension 4 cm \times 5 cm and was brownish-gray in color



Figure 5: The hematoxylin and eosin stained section (x10) revealed the presence of fibrocellular connective tissue stroma. The stroma contains blood-filled spaces and abundant young fibroblasts

compared to the maxilla. It is frequently observed at the molar and ramus regions of the mandible.[1] ABC was first described in the literature by Jaffé and Lichtenstein in 1942. The term "aneurysmatic" emphasizes on expansion of the affected bone, which is called the "blowout effect." The etiology of ABC is controversial. Increased venous pressure and repletion of the vascular bed in the transformed bone caused by the alteration of local hemodynamics were related to resorption, connective tissue replacement, and osteoid formation by Jaffe and Lichenstein. Matsuura et al. had reported that the development of ABC is related to a history of trauma and subperiosteal hematoma formation. [8] In the present case, pain and swelling or facial asymmetry were observed. The radiological features of the ABC in the jaws are variable; the expanded bone appears cystic resembling a honeycomb or soap bubble. Destruction or perforation of the cortex and a periosteal reaction can be also seen. In our case, the ABC represented a multilocular radiolucency causing expansion of the cortical plates. Histologically, the ABC reveals the presence of fibrocellular connective tissue stroma. The stroma contains blood-filled spaces and young fibroblasts abundantly. The stroma contains few inflammatory cell infiltrate, numerous dilated blood vessels, and calcified areas. Recurrence rates range from 20% to 30% in different groups and it occurs most frequently within the 1st year after surgery. Several authors recommend immediate reconstruction of the defect with autogenous grafts in cases of esthetic deformity and in cases with high risk of fractures and loss of mandibular continuity. In the present case, treatment of choice was initiated from curettage of the mass to complete excision.

CONCLUSION

As the radiologic and clinical presentation of ABC is extremely variable, a great emphasis is placed on histopathologic examination for the diagnosis.

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