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Olgu Sunumu/ Case Report

ANEURYSMAL BONE CYST OF THE MANDIBLE: REPORT OF A CASE

MANDİBULADA ANEVRİZMAL KEMİK KİSTİ: OLGU SUNUMU

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ABSTRACT

Aneurysmal bone cyst (ABC) is a benign osteolytic lesion. ABC is infrequently encountered in the long bones of the skeleton and its occurrence in the craniomaxillofacial area is a rare condition. Trauma, altered vascular equilibrium and genetic factors have been implicated on the development of ABC. In this report an ABC of the mandible in a 16-year old male patient was presented.

Key words: Aneurysmal bone cyst, jaw lesions, trauma, orthodontics

ÖZET

Anevrizmal kemik kisti (AKK)iyi huylu osteolitik bir lezyondur. AKK iskeletin uzun kemiklerinde seyrek olarak gözlenmekle birlikte çene ve yüz bölgesinde oldukça nadir olarak karşılaşılır. Travma, değişken damar basıncı ve genetic faktörler AKK gelişiminden sorumlu tutulmaktadır. Bu vaka raporunda 16 yaşındaki bir erkek hastanın mandibulasında gözlenen AKK olgusu sunulmuştur.

Anahtar kelimeler: Anevrizmal kemik kisti, çene lezyonları, travma, ortodonti

INTRODUCTION

Aneurysmal bone cyst (ABC) was first described by Jaffe and Lichtenstein in 1942 and later Bernier and Bhaskar reported ABC of the jaws in 1958[1]. The WHO defines ABC as a non-neoplastic intra-osseous lesion, characterized by varying sized blood-filled spaces associated with a fibroblastic stroma containing osteoclast-like multinucleated giant cells, osteoid tissue and woven bone[2]. ABC mainly affects the long bones of the skeleton that most frequently encountered vertebrae and pelvis on the other hand its prevalence in craniomaxillofacial bones is quite rare. ABC is considered as a non-epithelialized cyst that represents about 0.5% of all jaw bone cysts.[3] Mean age at diagnosis is reported as 20 years while it can be seen among children and adults with slight male predilection[4]. Mandibular ramus and corpus are the most frequent sites, more than 75% of the cases involve the mandible and 25% of the cases involve the maxilla[5]. ABC is more commonly observed in the posterior part of the mandible than the anterior part of the mandible. ABC has three distinct variants. Conventional or vascular type ABC presents as an expansile, rapidly growing destructive bone lesion characterized by replacement of the normal bone with fibro-osseous tissue that that causes cortical perforation and soft tissue invasion. The solid type is generally encountered as asymptomatic, solid mass without a cystic compound. The third form is mixed variant that exhibits features of both of previous types. Therefore it is considered as a transitory phase of the ABC between solid and vascular types[6]. Radiologically ABC has variable radiological appearances, it may act as a unilocular or multilocular radiolucent lesion of the jaws as well as mixed radiolucent-radiopaque lesion. Although ABC is a pseudocytic lesion without epithelial involvement, local aggressive behavior causes misdiagnosis of the tumor as a malignant neoplasm, thus histopathological

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evaluation of the biopsied specimen has critical role on the management of ABC of the jaws. In this case report it is aimed to describe the patient with -a rare- solid type mandibular aneurysmal bone cyst and review the literature.

CASE REPORT

A-16-year old male patient referred to the Baskent University Faculty of Dentistry, Department of Orthodontics for his dental crowding. Patient dental and family history was unremarkable. Clinical intraoral and extraoral examination revealed no pathology. Radiological examination revealed 30x35mm radiolus- cent lesion at the midline of the mandible with poor defined borders (Fig 1). Lesion was considered as a traumatic odontogenic cyst but the vitality of the adjacent teeth was normal. An aspiration biopsy was performed but the material was non diagnostic, therefore incisional biopsy procedure was planned. Patient was taken to the operating theatre and following local anesthesia administration a mucope- riosteal flap was raised and bone window was created under copious irrigation. On the other hand no epithelial lining or fibrous material was observed; therefore only curettage of the borders of the lesion was performed. The material, which was a hemorr- hagic mass including tiny bony fragments, was send for histopathological analysis. The microscopic evaluation of the material revealed blood-filled spaces separated by fibrous septae and reactive woven bone fragments (Fig 2). No malignant osteoid formation or stromal atypia was found.

Patient is well healed and radiological analysis showed no further radioluscent lesion therefore patient referred to the department of orthodontics. Patient is still under follow-up for 1 year with no further lesions (Fig 1).

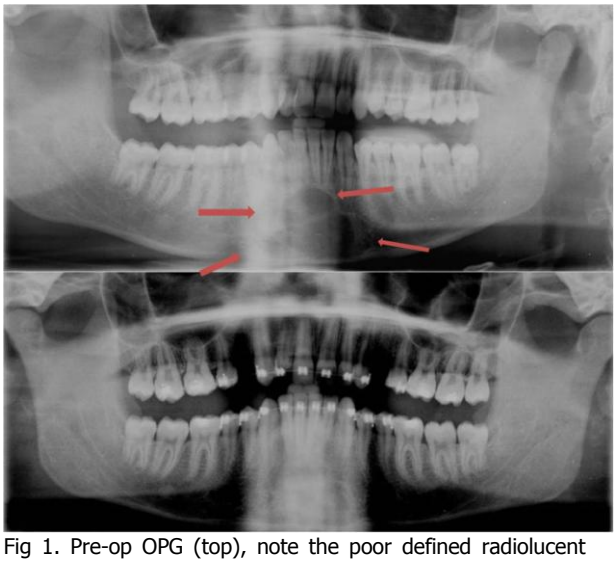

Fig 1. Pre-op OPG (top), note the poor defined radiolucent lesion extending to the inferior border of the mandible and post-op OPG (below), note the well-healed lesion following surgical curettage.

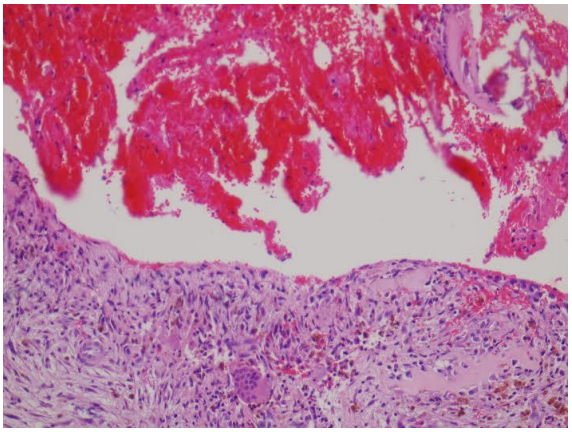

Fig 2. Histopathological view of the specimen. Note the blood-filled cavity that lacks an endothelial lining (above) and the fibroconnective tissue septa containing proliferating osteoblasts and scattered multinucleated giant cells (below). (Hematoxylin-Eosin Stain, 40X)

DISCUSSION

ABC of the bone is an infrequent lesion that consists of 2% of all bone cysts. On the other hand ABC of the jaws is rare with 0.5% prevalence among all cystic lesions of the jaws[7]. The etiology of ABC is unclear. Although history of trauma and subperiosteal hematoma formation are claimed the cause of ABC, Tillman et al. reported that 95 cases without any

history of trauma that was also consistent with our case[8,9]. Jaffe et al. suggested that vascular-haemostatic equilibrium differences have role on formation of ABC, increased venous pressure and repletion the vascular bed in the transformed bone leads to bone resorption with connective tissue and osteoid formation[10]. Bhaskar suggested the characteristic of ABC is similar with giant cell reparative granuloma and claimed if circulation of hematoma had maintained from injured vessel the clinical situation would be ABC[11]. If the injured vessel obliterated and circulation of the hematoma was disrupted the lesion would be a giant cell granuloma. On the other hand mainstay of this theory is debatable, circulation is the cornerstone of bone healing and it is also essential for the resolution of hematoma. Panoutsakopoulos et al. noted chromosomal abnormalities involving band 16q22 was found to be related in 3 patients[12]. In addition Oliveira et al. noted clonal rearrangements of chromosomal bands 16q22 and 17p13 resulting with ABC[13]. On the other hand Leithner et al. investigated 135 patient with ABC and noted 4% heredity[14]. In our case no bone lesion of the patient's relatives was reported that was consistent with the results of Leithner et al. with the low hereditary relation. Given this findings in patients with chromosomal abnormalities that had injury to the vessels may have ABC as a result of the impaired resolution of the hematoma.

Boyd et al suggested a slight female predominance for ABC due to high marrow content[15]. On the other hand Sakuma et al. noted ABC is found in the first two decades of the life with no gender predilection that was consistent with our case[16]. Pathological examination with diagnostic biopsy has critical role on the management of the lesion because there is no consensus for the management of ABC, curettage of the lesion, partial or total resection of the involved segment, embolization or observation had been reported in the literature[1,3]. Recurrence rate of ABC is relatively high, approximately 20% recurrence is observed due to the aggressive nature of the lesion. Kalra et al. noted partial resection might be a suitable option for the management of ABC cases with cortical expansion and facial disfigurement. In addition Kalra et al. suggested radiotherapy may increase the risk for radiation sarcoma, thus radiotherapy is not a suitable management option for ABC[17]. In our case no cortical expansion or facial disfigurement was noted, therefore we preferred only curettage with no further occurrence for 1-year period.

As a conclusion, ABC is benign but locally aggressive in nature, therefore appropriate diagnosis, treatment and follow up have critical role on the management of this lesion. In addition case series with larger samples rather than case reports should be investigated for the determination of the etiology of ABC.

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