ATATÜRK ÜNİVERSİTESİ YAYINLARI
ATATURK UNIVERSITY PUBLICATIONS

Current Research in Dental Sciences

Case Report *Olgu Sunumu* DOI: 10.17567/ataunidfd.950084

Aneurysmal bone cyst of the mandible: A case report of 5 years follow-up

Mandibulada anevrizmal kemik kisti: Beş yıl takipli olgu sunumu

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ABSTRACT

Aneurysmal bone cysts are not real cysts or aneurysms; they are rare non-neoplastic bone lesions. Although aneurysmal bone cysts occur mainly in the long bone metaphyses, they are rarely seen in the facial bones. In the jawbones, the mandible is involved more commonly than the maxilla. It is mainly seen in the first and second decades. Aneurysmal bone cyst affecting the left mandible of a 10-year-old male patient is presented in this case report. The diagnosis of aneurysmal bone cysts is essential and appropriate treatment should be done considering various factors such as age, surgical complications, and the possibility of recurrence.

Keywords: Aneurysmal, cyst, mandible

ÖZ

Anevrizmal kemik kistleri gerçek bir kist veya anevrizma değildir, nadir görülen neoplastik olmayan bir kemik lezyonudur. Anevrizmal kemik kistleri, çoğunlukla uzun kemik metafizlerinde ortaya çıkmasına rağmen nadir olarak yüz kemiklerinde de görülür. Çene kemiklerinde mandibula tutulumu maksilladan daha sıktır. Çoğunlukla birinci ve ikinci dekadda görülür. Bu vaka raporunda 10 yaşında erkek hastanın sol mandibulasını etkileyen anevrizmal kemik kisti sunulmuştur. Anevrizmal kemik kistlerinin tanısı çok önemlidir ve yaş, cerrahi komplikasyon, nüks olasılığı gibi çeşitli faktörler göz önünde bulundurularak uygun tedavi yapılmalıdır.

Anahtar Kelimeler: Anevrizmal, kist, mandibula

INTRODUCTION

An aneurysmal bone cyst (ABC) is not a true cyst or an aneurysm; it is a rare non-neoplastic, enlarged bone lesion. The aneurysmal bone cysts (ABCs) are benign, osteolytic, expansive, and hemorrhagic lesions of bone, which mainly occurred in long bone metaphyses like the tibia, the femur, or the spine. However, only 2% of the lesions arise in the craniofacial skeleton.[1] The World Health Organization defines ABCs as "a destructive, expansive, benign neoplasm of bone composed of multilobulated blood-filled cystic spaces." ABCs usually affects patients under 20 years of age with no sex predilection.[2] Similarly, it occurs in jawbones, mainly in individuals younger than 20 years old. The lesion is prevalent in the mandible than the maxilla contains posterior more often mandible region.[3] In the mandible, ramus and body are generally related. Unusual cases of involvement of condyle and coronoid process have been reported.[4] The aneurysmal bone cyst was first defined in 1893 by Van Arsdale, but Jaffe and Lichtenstein in 1942 were first to use the term of the aneurysmal bone cyst.[5,6] Bernier and Bhasker[7] were the first to define this cyst in the jawbones. This first case in the jaws was in the posterior area of the mandible.

The etiology and pathogenesis of ABCs are still uncertain but are thought to be reactively related to previous trauma or pathology.[8] The leading hypothesis etiology in ABC pathology is vascular and affected by local bone changes[9]. The clinical signs and symptoms of these cysts nonspecific, sometimes making diagnosis difficult. It may be clinically asymptomatic or may arise as a quickly advancing lesion enlarged to cause a pathological fracture.[10] Diagnosis should base on clinical symptoms, radiological, and histopathological examinations.

The purpose of this case report is to emphasize the importance of ABC diagnosis and effective treatment in recurrent cases.

CASE PRESENTATION

A 10-year-old male patient who had previously operated on the left mandible in another center applied to our clinic with complaints of swelling and trismus. Clinically, there was extraoral swelling, and the interincisal mouth opening was 15 mm. A multilocular radiolucent lesion of 2.5 cm in diameter with the sclerotic border was observed in the panoramic radiograph. It was extending from the impacted left second molar to the coronoid process and condyle (Figure 1). The previous histological finding was learned to be a traumatic bone cyst. Considering the patient's age and previous operation, it was decided that the patient should be followed. However, after six months of follow-up, it was observed

Received/Geliş Tarihi: 20.11.2020
Accepted/Kabul Tarihi: 09.06.2021

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Cite this article: Esen A, Baştürk F, Gürses G, Dolanmaz D. Aneurysmal bone cyst of the mandible: A case report of 5 years follow-up. *Curr Res Dent Sci.* 2022; 32(2): 185-188.

that there was no reduction in the size of the lesion. Trismus continued at the same. Since no recovery was observed, the second operation was planned. The patient was operated on under general anesthesia with an intraoral approach. Crestal incision and subperiosteal stripping were made, removed impacted second and third molar germs. The partial osteotomy was performed to reach the lesion, and it was curettage radically. No epithelium was seen in this procedure. However, serious venous bleeding was encountered in the bone cavity. Curettage was completed, the bleeding was brought under control, and the wound was sutured. Intralesional bleeding and the absence of epithelial tissue suggested that the lesion was an aneurysmal bone cyst. The histopathologic result verified our diagnosis. Histological examination revealed fibroblastic connective tissue, cotton-like fibrin, and osteoclastic giant cells around the blood-filled lakes (Figure 2, Figure 3). After six months of follow-up, radiological examination showed that the radiolucency was reduced in the entire lesion. After a 1-year follow-up, the swelling was dissolved, and an adequate bone formation was seen on the panoramic radiograph (Figure 4). The interincisal mouth opening was 28 mm. Trismus completely healed. After five years of follow-up, the patient had no complaints, and no recurrence was observed (Figure 5).

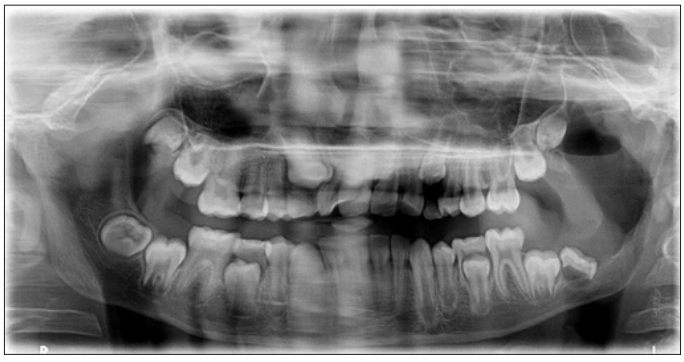

Figure 1. Panoramic radiograph showing a multilocular radiolucent lesion with the sclerotic border extending from impacted left second molar to coronoid process and condyle

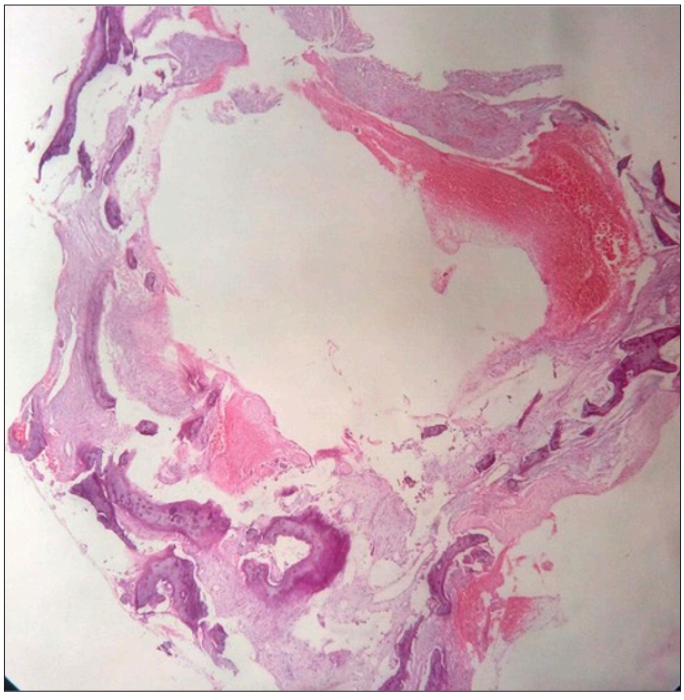

Figure 2. Microphotograph showing a cystic lesion surrounded by bone trabecular (HEx40)

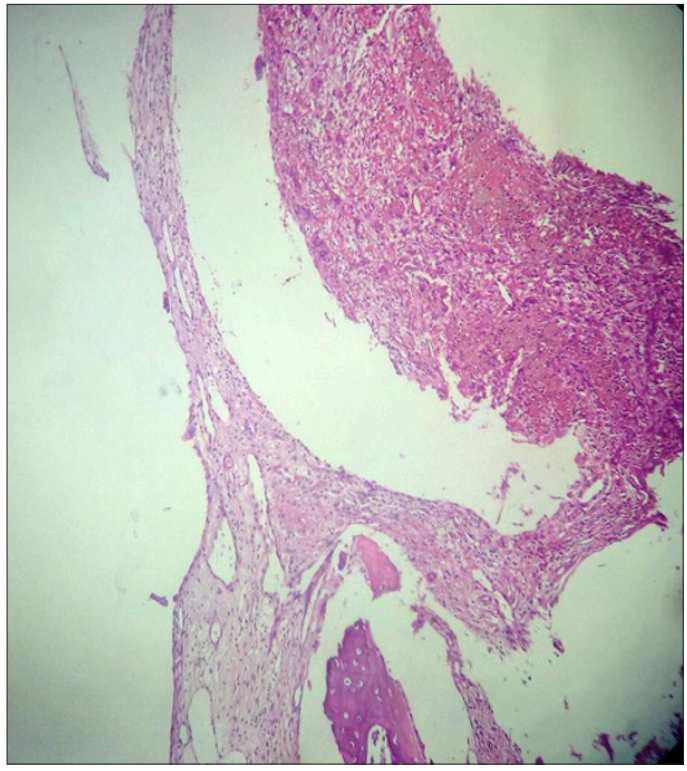

Figure 3. Microphotograph of the lesion showing fibrinoid accumulation, osteoclastic giant cells and bleeding findings (HEx100)

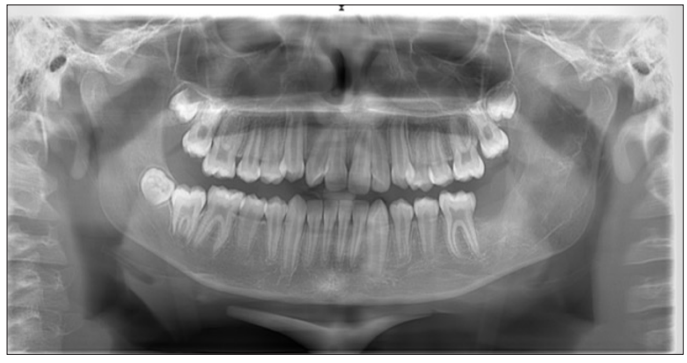

Figure 4. 1-year follow-up radiograph showing healing of the lesion and bone formation

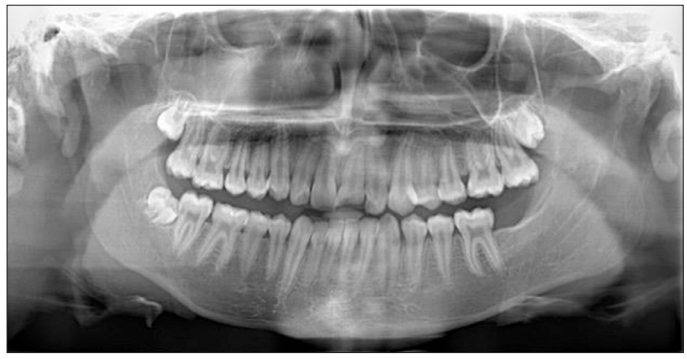

Figure 5. 5-year follow-up panoramic radiograph showing complete bone healing and no sign of recurrence

DISCUSSION

An aneurysmal bone cyst (ABC) is a pseudocyst due to the absence of a cyst epithelial. Jaffe and Lichtenstein defined it in 1942 as blood-filled lesions in which giant cells and irregular bone trabeculae.[6] Bernier and Bhaskar[7] reported the first ABC case containing the maxillofacial skeleton in 1958. ABC was reported in the mandible of an 11-year-old female patient. In the literature, including our case, mandibular involvement more common.[11] Mandibular condyle involvement is extremely rare.[12,13] In our case, the lesion ranged from the ramus to the condylar region. Although there are cases in different age ranges, it is most common under the age of 20.[14] The pathogenesis of ABC is controversial and, it is uncertain whether caused by a previous bone lesion.[6] Its histology consists of fibrous connective tissue stroma with blood-filled vascular spaces, osteoids, and osteoclast-type giant cells.[15] ABC can be seen on the radiograph as unilocular, multilocular, honeycomb, or soap bubbles depends on the types.

Malignant tumors such as Ewing's sarcoma, osteosarcoma, giant cell tumor should be differentiated in the diagnosis of the ABC. Clinical findings may differ depending on the behavior of the lesion. The lesion can keep unchanged for a long period, and it can slowly expanded growth or rapidly expanding swelling with large bone destruction.[16]

There is no consensus on the treatment of ABC, but there are several viable treatment options.[17] Conservative treatments such as calcitonin and methylprednisolone injections, simple curettage, cryotherapy, and complex treatments such as excision, radical resection, and reconstruction with bone grafts can be applied.[18,19] The treatment option should be determined according to the condition of the case. ABC usually responds well to curettage treatment. Motamedi[10] treated 17 mandibular ABC cases with radical curettage and reported no recurrence at 2 to 11 years of follow-up.

ABC occurs in 95% vascular, 5% solid, or mixed form.[16] Bleeding may be a risk, especially in the vascular type. Massive bleeding may be encountered as soon as beginning the surgery. Incomplete curettage can cause bleeding to continue.[14] We encountered major bleeding at the beginning of the operation. After completing the curettage, we finished the operation by controlling bleeding.

The treatment choice will depend on the extent and location of the lesion, the patient's age and symptoms, and the cyst's enlarging to the soft tissues or bone structures (condyle, maxillary sinus) and the feature of the case.[20] Therefore, some lesions can be treated with simple curettage, but active lesions that grow rapidly with the involvement of soft tissues require radical treatment. Khurshida et al.[4] reported an expansive and lytic progressive ABC case in the mandible. The lesion was reported as 7x5 cm in size with destruction of the lingual and buccal cortical bone. Therefore, they resected ABC. They provided reconstruction with a fibula graft.

In cases with mandible posterior region and condyle involvement, facial asymmetry, limitation of mouth opening, pain, and swelling can be seen. In our case, our patient had these symptoms. Various treatment and reconstruction methods for cases involving condyle and coronoid have been reported in the literature. Pelo et al.[13] reported an aneurysmal bone cyst involving the condyle neck in a 10-year-old male patient. They removed the lesion by performing a low condylectomy.

Rapidis et al.[15] reported a case of aneurysmal bone cyst extending from the mandible ramus to the condyle, similar to our case. The authors reported that the lesion recurred after curettage. In the second operation, they performed radical resection and repaired with a costochondral graft.

Ettl et al.[21] reported a case involving the mandibular ramus, condyle, and surrounding soft tissues that recurred six months after surgical curettage. In the second operation, they resected the condyle and part of the ramus. The authors recommended that recurrent cases be treated with more radical surgeries. In our case, the first surgery was probably insufficient. However, we treated it with radical curettage.

In this case, we presented that the radiography of the cyst had a unilocular appearance extending to the coronoid process and condylar neck. However, we encountered bone septa in the cyst during the operation. We entered all parts of the cyst and cleaned the area. After the operation, we found that there was an improvement in a short time radiographically. This dramatic recovery made us think that the curettage performed in the first operation was not enough. Due to the impacted second and third molar teeth may have prevented reaching the lesion, or the lesion around these teeth may not have been adequately cleaned in the previous operation. Therefore, we removed these teeth to provide sufficient vision and to prevent recurrence and made a radical curettage.

Histologically, a cystic structure lined with fibrous connective tissue without a lining epithelium was observed in the sections. Fibrinoid deposition on the lesion wall, osteoclastic giant cells, bleeding findings, and new bone construction were selected. No findings suggesting a neoplastic lesion were detected. These histological findings show that the lesion is an aneurysmal bone cyst, not a simple bone cyst or traumatic bone cyst. In addition, expansion is rare in a simple bone cyst, and serous fluid is present inside the cavity, not blood.[22] This lesion, which was diagnosed as a traumatic bone cyst due to the previous operation, recurred despite curettage. Aneurysmal bone cysts also show persistent recurrences.[21] Therefore, the recurrence of this lesion despite the previous operation shows that it is an aneurysmal bone cyst.

In conclusion, we think that reaching the entire cyst cavity is very important in the surgical treatment of aneurysmal bone cysts. Also, the extraction of the teeth in the cyst cavity may be considered, if necessary, to prevent a recurrence.

Informed Consent: Written informed consent was obtained from patients' parents who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – A.E., D.D.; Design – F.B., A.E.; Supervision – A.E., D.D.; Resources – F.B.; Data Collection and/or Processing – F.B., G.G; Analysis and/or Interpretation – D.D.; Literature Search – F.B., G.G.; Writing Manuscript – F.B., G.G.; Critical Review – D.D.

Declaration of Interests: The authors have no conflicts of interest to declare.

Funding: The authors declared that this study has received no financial support.

Hasta Onamı: Yazılı hasta onamı bu çalışmaya katılan hastanın ailesinden alınmıştır.

Hakem Değerlendirmesi: Dış bağımsız.

Yazar Katkıları: Fikir – A.E., D.D.; Tasarım – F.B, A.E.; Denetleme – A.E., D.D.; Kaynaklar – F.B.; Veri Toplanması ve/veya İşlemesi – F.B., G.G.; Analiz ve/veya Yorum – D.D.; Literatür Taraması – G.G., F.B.; Yazıyı Yazan – F.B., G.G; Eleştirel İnceleme – A.E., D.D.

Çıkar Çatışması: Yazarlar çıkar çatışması bildirmemişlerdir.

Finansal Destek: Yazarlar bu çalışma için finansal destek almadıklarını beyan etmişlerdir.

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