Ameloblastic Fibroma: A Case Report

Ameloblastik Fibroma: Olgu Sunumu

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ABSTRACT

Ameloblastic fibroma (AF) is a rare benign odontogenic tumor that usually occurs in the first two decades of life. It can occur in the mandible or maxilla but is most commonly found in the posterior region of the mandible.

A 6-year-old female patient applied to our clinic due to swelling in her mouth. The patient had no medical history of previous trauma or pain in the affected area. Enucleation of the tumor was performed under general anesthesia. Following enucleation, surrounding bone curettage was performed and adjacent teeth were removed. As a result of morphological and immunohistochemical examinations, the patient was diagnosed with ameloblastic fibroma.

Although AF is a rare tumor, it is more common in children. Patients with AF need to be followed for a long time due to the ability of AF to turn into ameloblastic fibrosarcoma, which is its malignant form. The patient is followed frequently and has been asymptomatic for 1 year.

ÖZ

Ameloblastik fibroma (AF), genellikle yaşamın ilk yirmi yılında ortaya çıkan nadir görülen benign odontojenik bir tümördür. Mandibula veya maksillada oluşabilir ancak en sık mandibulanın arka bölgesinde bulunur.

6 yaşında kadın hasta ağzındaki şişlik nedeniyle kliniğimize başvurdu. Hastanın tıbbi geçmişi, etkilenen bölgeye daha önce travma veya ağrı öyküsü yoktu. Tümörün enükleasyonu genel anestezi altında gerçekleştirildi Enükleasyonun ardından çevredeki kemik küretajı yapıldı ve komşu dişler çıkarıldı. Morfolojik ve immünhistokimyasal incelemeler sonucunda hastaya ameloblastik fibroma tanısı konuldu.

AF nadir görülen bir tümör olmasına rağmen çocuklarda daha sık görülür. AF'nin, malign formu olan ameloblastik fibrosarkoma dönüşme yeteneği nedeniyle AF'li hastaların uzun süre takip edilmesi gerekmektedir. Hasta sık sık takip edilmekte olup 1 yıldır asemptomatiktir.

Introduction

Ameloblastic fibroma (AF) is included in the benign mixed epithelial & mesenchymal odontogenic tumours according to the WHO 2022 classification of cysts and tumours¹. This group also included dentinogenic ghost cell tumour, odontoma, primordial odontogenic tumour². AF is one of the rare benign odontogenic tumours, most commonly seen in the second decade of life. The incidence is slightly higher in males¹. It can be seen in both jaws. But it is most common in the back of the lower jaw³. An impacted tooth is usually associated with the lesion; the associated tooth is the first or second permanent molar⁴. According to the WHO, AF is defined as a disease consisting of odontogenic ectomesenchyme resembling the dental papilla and epithelial strips and sockets resembling the dental lamina and enamel organ.

Essential diagnostic criteria are a bland, hypercellular, papilla-like mesenchyme, dispersed bilaminar strands of cuboidal or columnar odontogenic epithelium, a well-defined and cortical radiolucency². There is no hard tissue appearance⁵. On routine clinical examination, small cases are usually seen as asymptomatic, unilocular, radiolucent lesions. Larger lesions have a multilocular and radiolucent structure and may cause painless swelling⁶. Due to the rarity of ameloblastic fibromas and the limited literature on treatment, there is controversy about the approach to treatment. Enucleation and curettage are usually sufficient for ameloblastic fibromas. However, extensive and aggressive lesions with a high tendency to recur may require radical surgical treatment⁷.

Case Report

The patient presented in this article is a 6-year-old female. She has no systemic disease and no history of trauma. She was given antibiotics and painkillers due to a misdiagnosis of an odontogenic infection at an outside medical clinic. When her symptoms did not improve, she went to the Oral and Maxillofacial Surgery Clinic of the Faculty of Dentistry

at Selçuk University. Clinical and radiological examination was performed in our clinic. Intraoral examination revealed swelling extending anteroposteriorly from the right mandibular primary canine to the retromolar region. There was no ulceration or inflammation on the mucosa above the swelling and it was the same colour as the surrounding mucosa. The buccal cortical plate was enlarged due to the swelling and there was obliteration in the buccal vestibule. On palpation the swelling was bony and there was mild tenderness over the swelling. Extraoral examination revealed swelling on the right side of the face. Facial asymmetry was associated with this swelling. The swelling is firm and tender to palpation. There was no lymph node involvement. Orthopantomography showed a well-defined multilocular radiolucency with sclerotic margins extending from the unerupted right mandibular first molars to the condylar neck. The lesion was observed to push the unerupted tooth germs mesially (Figure 1.).

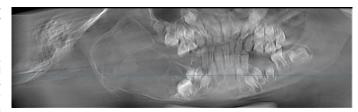


Figure 1. Panoramic view of the patient

Under general anaesthesia, samples were taken by incisional biopsy, including the surrounding intact tissue (Figure 2.).

Gönderilme Tarihi/Received: 11Aralık, 2023
Kabul Tarihi/Accepted: 2 Şubat, 2024
Yayınlanma Tarihi/Published: 19 Ağustos, 2024
Attf Bilgisi/Cite this article as: Aktı A, Çengiz ZO, Erdur Ö, Gürses G. Ameloblastic Fibroma: A Case Report.
Selcuk Dent J 2024;11(2): 227-230 Doi: 10.15311/ selcukdentj.1403551

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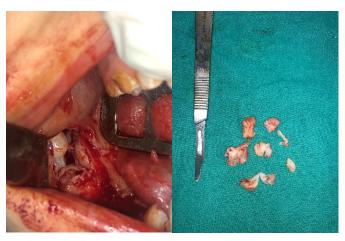


Figure 2. There was made incisional biopsy and were removed some tissues for pathological evaluation

Biopsy tissue samples were fixed in formalin and sent for histopathological analysis. Ameloblastic fibroma was diagnosed following morphological and immunohistochemical studies. Informed consent was obtained from the patient's parents. After local anaesthesia under general anaesthesia, a mucosal incision was made with a scalpel in the region of the buccal sulcus and the lesion was reached by blunt dissection, the lesion was cleaned and enucleated. The adjacent teeth were extracted. After controlling bleeding in the area, the flap was closed with 3-0 silk suture (Figure 3).



Figure 3. The lesion was curratted and enucleated and adjacent teeth were removed

Post-operative follow-up visits were made at 3 days, 1 week and 1 month, and then she came for routine clinical follow-up at regular intervals for 3 months. She was followed up for one year (Figure 4).



Figure 4. 1st year post-operative control radiograph

Discussion

AF belongs to the group of mixed odontogenic tumours and is a rare benign odontogenic tumour. It is so named because histologically it consists of odontogenic tissue of both epithelial and mesenchymal origin⁸. There is controversy in the literature as to whether the tumours included in the mixed odontogenic tumour class are related⁸. Cahn and Blum have theories suggesting that AF develops through continuous differentiation and maturation to become AFO, and then AFO becomes odontoma, a hamartoma⁹. This theory is called the maturation theory. According to this theory, the early stage in the development of an odontoma is AF. However, there are two reasons why this theory is not widely accepted. Firstly, histopathology of almost all residual or recurrent AFs has shown that they do not develop into a hard tissue tumour at any advanced stage. And second, it is known that AFs usually appear after the second decade of life, when odontogenesis is complete^{10,11}. Odontoma, a hamartoma, develops during the period of odontogenesis and therefore it is unlikely that an AF diagnosed after this age represents an early stage of odontoma¹¹.

AF grows slowly and is benign. It often causes no symptoms. It is usually discovered incidentally during routine clinical examinations on radiographs taken to detect unerupted teeth¹². Therefore, regular dental examination is important for early detection of the lesion. Regarding the radiological features of AF, some authors state that it often appears as a unilocular radiolucency^{13,14}, some state that it often appears as a "unilocular radiolucency¹⁵, and some state that it often appears as a "unilocular or multilocular lesion"^{16,17}. Small tumours are usually unilocular and radiolucent lesions^{18,19,20}. In contrast, approximately 75% of larger tumours have a multilocular appearance^{19,20}. These multilocular lesions often cause swelling of the jaw²¹. In our case, although the patient had a large and unilocular lesion, it was not noticed by the patient due to features such as less swelling, slow growth and lack of pain.

If we look at the literature, there are different recurrence rates for atrial fibrillation. Trodahl showed that AF has a high relapse potential with a recurrence rate of 36.4%²². In the study by Buchner et al, the recurrence rate was 16.3% and the malignant transformation rate was 6.4%. While most recurrences were seen in the early decades, malignancy was observed in the advanced decades. One of the most important factors to consider when planning surgery is the age of the patient. The first line of treatment is curettage with enucleation and extraction of the teeth adjacent to the lesion 18 . Some researchers argue that a carefully performed curettage is sufficient to treat the lesion²⁴. This treatment protocol provides continuity of chewing and occlusion and also allows the face and teeth to develop²⁵. In our patient, the lesion was enucleated and the teeth adjacent to the lesion were extracted. In addition, some intact bone tissue around the lesion was removed. Although we felt it was necessary to prevent recurrence, segmental resection was not preferred because it would affect the function, phonation and aesthetics of the jaw and teeth, and the patient's quality of life. After 6 months, it was observed that the surgical area in the patient's mandible had healed with intact bone tissue. In cases of ameloblastic fibroma, it is important to evaluate the size of the lesion with regular clinical examination and radiographic follow-up due to the risk of malignant transformation and the possibility of recurrence. The patient was followed for 1 year and no recurrence was observed.

Değerlendirme / Peer-Review

İki Dış Hakem / Çift Taraflı Körleme

Etik Beyan / Ethical statement

Bu makale, ACBID 15. Uluslararası Kongresi'nde sözlü olarak sunulan ancak tam metni yayınlanmayan "Ameloblastik Fibroma: Olgu Sunumu" isimli sunumun, içeriği iyileştirilerek ve kısmen değiştirilerek yeniden düzenlenmiş halidir.

Bu çalışmanın hazırlanması sürecinde bilimsel ve etik ilkelere uyulduğu beyan edilmiş olup, yararlanılan tüm çalışmalar bibliyografyada belirtilmiştir.

This article is the version of the presentation named "Ameloblastic Fibroma: A Case Report", which was presented orally at the ACBID 15. International Congress, but whose full text was not published, by improving and partially changing the content.

It is declared that during the preparation process of this study, scientific and ethical principles were followed and all the studies benefited are stated in the bibliography.

Benzerlik Taraması / Similarity scan

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Finansman / Grant Support

Yazarlar bu çalışma için finansal destek almadığını beyan etmiştir. | The authors declared that this study has received no financial support.

Çıkar Çatışması / Conflict of Interest

Yazarlar çıkar çatışması bildirmemiştir. | The authors have no conflict of interest to declare.

Yazar Katkıları / Author Contributions

Çalışmanın Tasarlanması | Design of Study: AA (%25), ZOC (%25), ÖE (%25) GG (%25)

Veri Toplanması | Data Acquisition: AA (%25), ZOC (%25), ÖE (%25) GG (%25)

Veri Analizi | Data Analysis: AA (%25), ZOC (%25), ÖE (%25) GG (%25) Makalenin Yazımı | Writing up: AA (%25), ZOC (%25), ÖE (%25) GG (%25) Makale Gönderimi ve Revizyonu | Submission and Revision: AA (%25), ZOC (%25), ÖE (%25) GG (%25)

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