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Case Report

Extensive ameloblastic fibroma of the mandibula in a female adult patient: A case report with a follow-up of 3 years

Sinan Tozoglu[1], Mukerrem Hatipoglu[2], Zeliha Aytekin[2], Elif Inanc Gurer[3]

[1]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Akdeniz University, Antalya, Turkiye,
[2]Department of Periodontology, Faculty of Dentistry, Akdeniz University, Antalya, Turkiye,
[3]Department of Medical Pathology, Faculty of Medicine, Akdeniz University, Antalya, Turkiye

Correspondence: Dr. Mukerrem Hatipoglu
Email: mukerremhatipoglu@hotmail.com

ABSTRACT

Ameloblastic fibroma (AF) is rare benign odontogenic tumour which usually occurs in the first two decades of life. It can occur either the mandible or maxilla but it is most frequently found in the posterior region of the mandible. Treatment of AF in usual is a conservative approach, such as enucleation and curettage but the aggressive lesions require a radical approach. A more radical approach should be considered in older patients who have likely high recurrence tendency. This report describes a case of AF in a 38-year-old female patient identified during a routine radiographic exam. Tomographic examination through three-dimensional reconstruction indicated vestibular fenestration of the cortical bone, with involvement of lingual cortical bone as the lession extended to the posterior region. We removed the tumor under local anesthesia. In this case patient has continued to be followed frequently and has been disease-free for 3 years.

Key words: Ameloblastic fibroma, surgery, vestibular fenestration

INTRODUCTION

Ameloblastic fibroma (AF) is rare, benign neoplasm of odontogenic origin comprising about 1.5–4.5% of all odontogenic tumors.[1] AF is described by the WHO as being "composed of odontogenic ectomesenchyme reminding of the dental papilla and epithelial strands and nests reminding dental lamina and enamel organ. There are no hard tissue appearance."[2]

AF was first described by Kruse 1891.[3] Although reported among a wide age range (0.5–62 years), the tumors are frequently seen in the first and second decades of life 77.7% of cases are diagnosed before the age of 20, and they are primarily considered a tumor of childhood and adolescence.[1,4] Males show slightly higher prevalence than females (1.4/1).[4]

AF has no dental hard tissue.[5] If the lesion has dentinoid tissue without or with enamel formation, it could be ameloblastic fibrodentinoma (AFD), ameloblastic fibroodontoma (AFO), or fully mineralized odontoma (complex and compound) which are members of the group of mixed odontogenic tumors.[6]

There is no consensus among investigators about whether these tumors should be categorized as separate entities or different stages in the maturation of the same lesions.[7] Some authors contend that AF is a distinct, specific neoplastic entity that does not turn into a more differentiated odontogenic tumor,[8] while others contend that if an AF is not extracted, it will eventually mature into an AFO and then continue

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to mature into a completely differentiated odontoma, which is accepted as being a hamartoma.[9]

Several authors documented differences in dental health by socioeconomic status. Bertoldi *et al*. suggested that a lower socioeconomic status class showed many more teeth extracted, decayed, and filled, than did a higher class.[10] This is because they do not have regular dental examinations.

Most cases are discovered during routine oral examination as an asymptomatic, unilocular, small radiolucent lesion. Larger lesions usually have a multilocular and radiolucent pattern, and may cause painless swelling.[11] It appears as a firm, soft tissue mass with a smooth surface. It may or may not be encapsulated.[12]

Microscopically, AFs are composed of both epithelial and connective tissue components, and are characterized by plump fibroblasts and delicate collagen fibrils that closely resemble the dental papilla.[13] The epithelial component which resembles embryonic dental lamina is arranged in various thin branching cords or strands with double or triple layers of cuboidal cells as well as small nests or islands with scanty cytoplasm and basophilic nuclei, while stellate reticulumlike cells are common in larger nests.[11]

Treatment of AF usually involves a conservative approach, such as enucleation with curettage of the surrounding bone along with the removal of the affected tooth, but aggressive lesions require a radical approach.[14] Because of its rarity, there are limited details in the literature regarding its clinical and radiologic appearances. There are also conflicting reports regarding the recurrence and malignant transformation rates of AF. Hence, long-term follow-up is necessary.

CASE REPORT

In November 2012, a 38-year-old female patient presented at the Department of Oral and Maxillofacial Surgery for dental examination of the right mandibular teeth (45–46), which were slightly mobile, with a diffuse, minimal mandibular swelling. A clinical examination revealed smooth swelling obliterating the mucogingival sulcus from the first premolar to the second molar region [Figures 1 and 2].

The growth was insidious and slowly progressing. The patient's medical history revealed no prior trauma

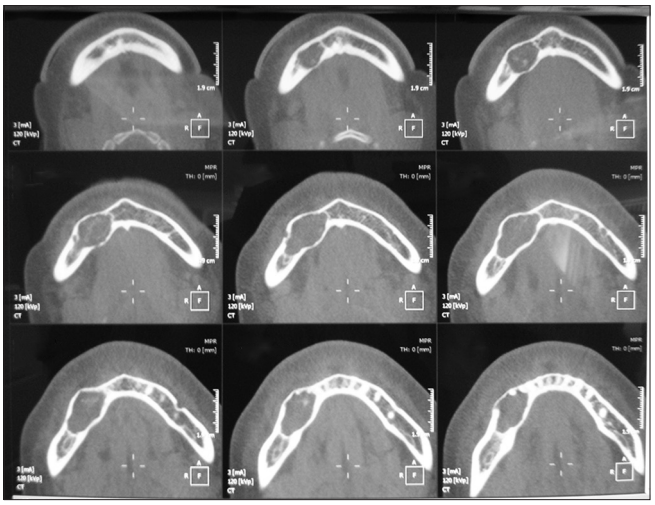

Figure 1: Noncontrast axial computed tomography at bone algorithm showing a tumoral mass occupying the posterior part of the right mandible

or episodes of pain in the affected region. This lesion was associated with a compromised vestibular fornix and severe tooth mobility.

Tomographic examination through three-dimensional reconstruction indicated vestibular fenestration of the cortical bone, with the involvement of the lingual cortical bone as the lesion extended to the posterior region. The lesion measured approximately 22 cm in extension. The approximate size of the lesion was 24 mm × 32 mm. Because aspiration was negative, excisional biopsy was performed [Figures 3 and 4].

As the lesion was well-circumscribed, the patient was submitted to excision of the lesion under local anesthesia. Enucleation of the tumor was performed, followed by curettage of the surrounding bone. The 45 and 46 numbered teeth adjacent to the tumor were extracted. The surgical specimen was fixed in formalin and sent for histopathological analysis. There are various biomaterial combinations used in the regeneration of periodontal bone defects.[15] The tumor cavity was closed without bone graft materials. The immediate postoperative period was uneventful. The bone fully regenerated [Figure 5].

After the wound healing was completed, we started another dental treatment. First, we comprehensively examined all of the patient's teeth and oral tissue. There were deep pockets on the mandibular and maxillary molar areas. We applied the Sc/Rp procedure. Six months after the surgery, a fixed prosthesis was made on the surgical side [Figure 6].

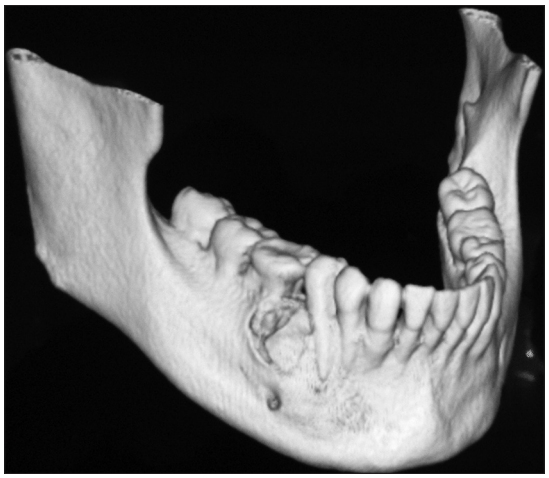

Figure 2: Three-dimensional tomographic reconstruction showing cortical expansion and fenestration

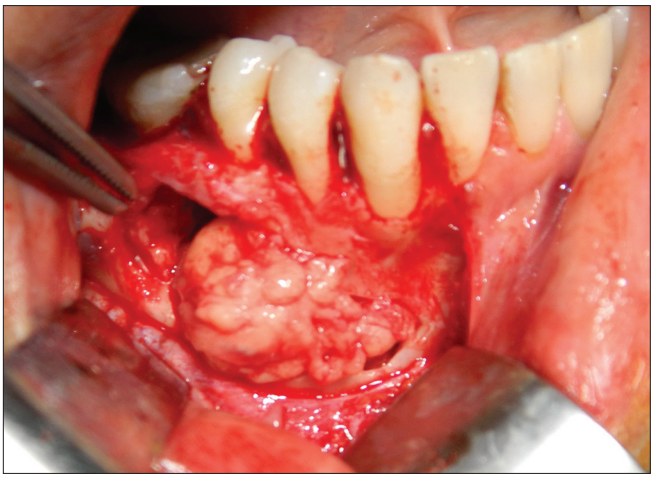

Figure 3: Intraoral image of the tumor during surgery

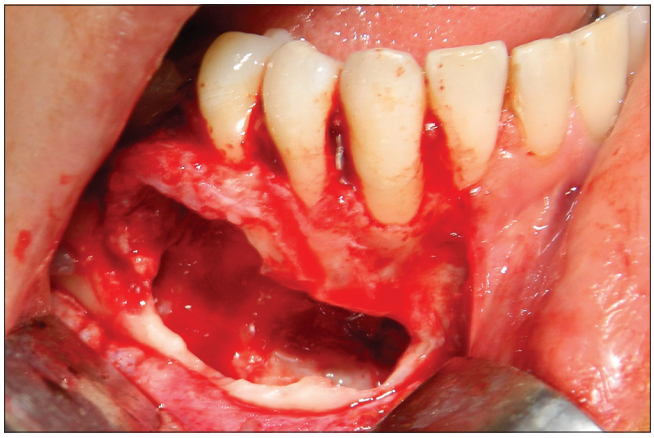

Figure 4: The image of tumor cavity in the posterior region of the mandible after tumor was removed

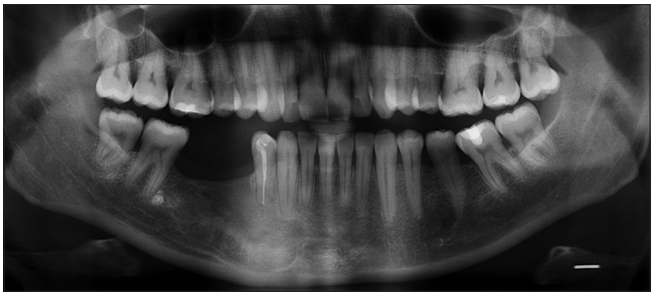

Figure 5: Patient's orthopantograph 6 months after the surgery

DISCUSSION

AF is an uncommon, benign odontogenic tumor with both epithelial and mesenchymal tissues.[7] Cahn and Blum[16] proposed that an AF could eventually develop into an odontoma, a type of hamartoma, if the lesion is allowed to remain. Accordingly, AF, AFD, and AFO would be various stages of the same lesion which are early stages of a developing odontoma.[9]

However, this concept has been challenged for a number of reasons. Several longstanding residual or recurrent AFs have shown no evidence of further maturation into a more differentiated lesion such as AFD, AFO, or odontoma. Moreover, AFs are encountered at ages beyond when odontogenesis is completed, and some AFs occur at advanced age.[1] In our case, there was no odontoma-like development. AF in the posterior mandibular location, is more common than in the maxillary counterpart.[17] Buchner and Vered reported that AF was located in the mandible in 132 (77%) cases and in the maxilla in 40 (23%) cases, with a mandible to maxilla ratio of 3.3:1. A total of 82% were in the posterior region of the jaw, 7.8% were in the anterior region, and 10.2% were in both anterior and posterior regions. In addition, most of the lesions in the anterior region were in the maxilla, and most of the lesions in the posterior region were in the mandible.[18] The presenting lesion was located in the posterior mandible, which is the most common site for this kind of lesion.

Some authors reported that male and female patients are equally affected, whereas others have concluded that males show a slightly higher prediction than females (M: F = 1.26:1[19] M: F 1.42:1).[1] Buchner and Vered reported that AFs were also slightly but not significantly more common in male patients (M: F = 1.36:1).[18] A male to female ratio of 2:1 was reported in a study performed in the Armed Forces Institute of Pathology by Trodahl.[20] Adebayo *et al*.[21] reported that AFs were equally projected for men and women.

AF is a neoplasm of childhood/adolescence with a mean age of 15 years.[19] However, a literature review

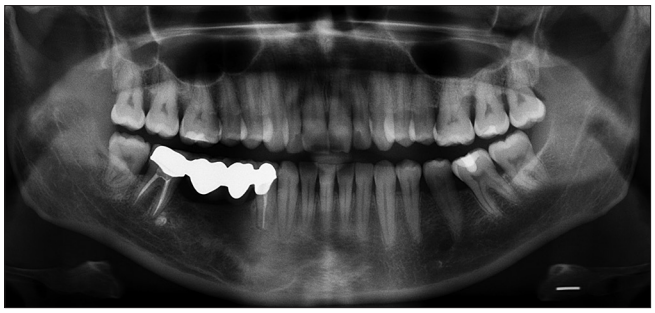

Figure 6: After the surgery fixed prosthesis was made on the surgical side

found that approximately 25% of cases occur in patients over 22 years of age, probably due to lower regular dental attendance.[19] However, Vasconcelos *et al*. reported an AP case in a patient aged 45 years.[22] This age difference may support a step by step proceeding from a benign to a malignant neoplasm in contrast to a *de novo* malignancy.

AF is a benign, slow-growing tumor. Most of the cases are asymptomatic; they are unilocular, radiolucent lesions discovered accidentally on radiographs taken to detect the failure of tooth eruption.[14] By contrast, a multilocular appearance is often seen in larger tumors and is found in about 75% of cases, which are often associated with jaw swelling.[1] In our case, the lesion was unilocular and the patient was unaware of having such a large lesion because of characteristics such as less swelling, slow growth, and a lack of pain and purulent discharge.

A careful treatment planning is necessary as while AF has a little tendency for recurrence, variable recurrence rates have been reported by various authors. Trodahl reported that AF has a high potential for recurrence, with a recurrence rate of 36.4%.[20] The rate of recurrence in Buchner's study was 16.3%, and the rate of malignant transformation to AFS was 6.4%. Most of the recurrences were seen in the younger group, while most malignant transformations were seen in the older group.[18] Therefore the patient's age should be an important consideration when deciding the treatment plan.

The first choice of treatment should be a conservative surgical approach such as enucleation with curettage of the surrounding bone along with the removal of the affected tooth.[3,14] This is especially true in young patient, for whom the priorities are to minimize morbidity and allow mastication and continued facial growth and tooth development. A more radical approach of marginal or segmental resection should be considered in older patients with extensive multilocular lesions and likely high recurrence tendency.[6]

Vasconcelos *et al*. treated a patient who has a large AF on mandibular region. They performed partial mandibular resection with immediate reconstruction by rigid fixation.[22] Pitak-Arnnop *et al.* reported a case report with a follow-up of 4 years. The tumor was enucleated and impacted tooth was extracted. However, the tumor relapsed 2 years after enucleation. Surgery was repeated. The patient has been disease free for 4 years.[17]

In our case, the tumor was enucleated and we extracted the teeth, which were in the lesion. In addition, some healthy bone was removed with bur. Although we think that it is necessary to prevent the recurrence of the lesion, we did not make a wide resection because of its negative effect on quality of life. After 6 months, the surgery cavity of the patient's mandible was fully regenerated. Long-term clinical and radiographic follow-up evaluations are mandatory in all cases. We have still followed our patient for 3 years, and there has been no recurrence.

CONCLUSION

Patients with AF must be followed up for a long period because of AF's ability to transform into ameloblastic fibrosarcoma which is the malignant compartment of AF. In this case, the patient has continued to be followed frequently and has been disease-free for 3 years. Her condition has remained satisfactory, throughout her bone regeneration and follow-up period.

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Conflicts of interest
There are no conflicts of interest.

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