Ameloblastic Fibroma in Mandibular Anterior Tooth Region: A Case Report

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Abstract

Ameloblastic fibroma (AF) is a proliferative mixed tumor which includes components of both odontogenic epithelium and mesenchymal tissue. It is a relatively rare neoplasm, accounting for approximately only 1.5–4.5% of odontogenic tumors. This case report describes an AF that occurred in the mandibular anterior tooth region in a 9-year-old girl who presented with the chief complaint of swelling in the left mandibular anterior tooth region. Intraoral examination revealed a swelling along the labial gingiva, extending from the left mandibular lateral incisor region to the left mandibular canine. Upon palpation, the swelling appeared to comprise a hard tissue. Computed tomography revealed a supernumerary impacted tooth; soft tissue density in the bone surrounding the region extending from the left mandibular lateral incisor to the left mandibular canine; labial bone expansion; and thinning of the labial cortical bone. A biopsy was performed under local anesthesia and the lesion subsequently diagnosed as an AF. Tumor resection and extraction of the supernumerary impacted tooth were carried out under general anesthesia. At 2 years postoperatively the prognosis is good. Although relapse with an AF is rarer than that with an ameloblastoma, strict follow-up is required, as malignant transformation to an ameloblastic fibrosarcoma has been reported in relapsed cases.

Key words: Ameloblastic fibroma — Odontogenic tumor — Benign mixed epithelial and mesenchymal odontogenic tumor — Mandibular tumor — Benign tumor

Introduction

Ameloblastic fibroma (AF) is a benign odontogenic tumor. In the WHO classification of 2017 (4th edition), it is defined as a mixed tumor which includes an odontogenic
mesenchymal component similar to the dental papilla and epithelial component similar to the odontogenic epithelium. It accounts for only 1.5–4.5% of all odontogenic tumors. Herein, we report a case of an AF that occurred in the mandibular anterior tooth region.

Case Presentation

Written informed consent for publication of clinical information pertaining to this case was obtained from the patient and her parents.

The patient was a 9-year-old girl who reported to our clinic with the chief complaint of swelling in the left mandibular anterior tooth region. Her medical history revealed atopic dermatitis. Her family medical history revealed nothing of relevance.

She reported noticing a swelling in the labial gingiva in the left mandibular anterior tooth 1 month earlier. She had visited a dental clinic and undergone panoramic radiography, which revealed a supernumerary impacted tooth and radiolucency extending from the left mandibular lateral incisor region to the left mandibular canine region. She was referred to our clinic for further evaluation.

A full body examination revealed nothing significant, and the face was bilaterally symmetrical. Intraoral examination revealed an indolent labial gingival swelling extending from the left mandibular lateral incisor region to the left mandibular canine region. The overlying mucosa had a healthy color (Fig. 1). Palpation revealed a hard osteoid.

A panoramic radiograph revealed a supernumerary impacted tooth in the mental region and torsion of the lower-left canine (arrowhead). The outline of the lesion was hidden due to shadowing from the cervical vertebrae. Although the outline of the lesion was hidden due to shadowing from the cervical vertebrae, there appeared to be no significant root resorption. The oral and maxillofacial radiologist suggested scanning by computed tomography (CT) to allow a more detailed evaluation (Fig. 2).

The CT images revealed a multi-locular radiolucency lesion in the anterior mandibular area. Axial CT images revealed extension and thinning of the labial cortical bone. The lesion was well-defined, homogeneous, and had smooth borders. Although a supernumerary impacted tooth was found within the...
lesion, there was no obvious root resorption. The density of the lesion was approximately +70 HU, which was comparable to, or slightly lower than, the surrounding muscle. (Fig. 3a–f).

A clinical diagnosis of left mandibular tumor was made.

**Clinical Procedures and Outcomes**

Taking the above clinical diagnosis into account, a biopsy was performed under local anesthesia at the oral surgery out-patient department. Cortical bone tissue was only harvested from the upper area on the labial side of the left mandibular lateral incisor region as the patient was young, indicating the need to keep the procedure as minimally invasive as possible. The biopsy results confirmed an AF. The tumor (enucleation and curettage) was therefore subsequently resected and the supernumerary impacted tooth extracted. A Neumann-Peter incision was performed from the left mandibular first primary molar region to the right mandibular first primary molar region and the mucoperiosteal flap inverted (Fig. 4a). The thin cortical bone in the left mandibular anterior tooth region was removed (Fig. 4b).

The extracted lesion included the supernumerary impacted tooth. There was no adhesion with the surrounding tissue and it was extracted easily. The surrounding bone was removed using a round bur and the surgical incision closed completely (Fig. 4c). Postoperative healing was good and the sutures were removed on the 7th postoperative day. Histopathological examination of the resected tissue confirmed the diagnosis of an AF. Presently, at 2 years postoperatively, progress is good and no recurrence has been observed.

Fig. 3 Axial (a, b, d, and e) and coronal (c and f) CT images revealed multi-locular radiolucent lesion in anterior mandibular tooth region.

Axial CT images revealed expansion and thinning of labial cortical bone (arrowhead). Although supernumerary impacted tooth (arrow) was found within lesion, no obvious root resorption observed in this or surrounding teeth. Density of lesion was comparable to, or slightly lower, than that of surrounding muscle.
According to the WHO classification (3rd edition) of 2005, an AF is defined as a benign odontogenic tumor with odontogenic epithelium and odontogenic ectomesenchyme, but without hard tissue formation\(^\text{11}\). It is different from ameloblastic fibrodentinoma (AFD) and ameloblastic fibro-odontoma (AFO), which fall into the same category, in that it does not form hard tissue. These disparate tumors have been interpreted as a series of tumor types with different epithelial and mesenchymal derivatives. While AF shows the clinical and histopathological characteristics of a neoplasm, the clinical and organizational features of AFD and AFO do not always show such features. The regions in, and age at, which they form also fail to support the hypothesis that AF > AFD > AFO as different stages of the same type of tumor. Arguments have been made for the recategorization of AFD and AFO\(^\text{10}\). Ameloblastic fibroma is classified as an independent true tumor, while the AFD and AFO types have now been deleted from the WHO classification (4th edition, 2017) in favor of treating developmental stage odontomas as hamartomas. However, in rare cases, AFD and AFO have been reported to show considerable development, so many more cases are needed to establish and confirm their categorization\(^\text{3}\).

Yagibashi\(^\text{et al.}\)\(^\text{14}\) noted that, in 18 out of 38 cases (47.4%), the age of occurrence is less than 10 years, and that occurrence is almost

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Fig. 4  Intraoperative photograph.

a: Mandibular bone swelling on labial side with mucoperiosteal flap inverted. b: Cortical bone removed from most swollen part; lesion visible. c: Removal of surrounding bone after extraction. d: Extracted specimen.

Fig. 5  Histopathological image of extraction (H-E staining, magnification 200×).

Mesenchymal component was similar to that of dental papilla. Epithelial component revealed swollen cuboid and columnar cells stretched in 2 layers. Interior resembled enamel pulp, also showing similarity to follicular enamel device.

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**Discussion**

According to the WHO classification (3rd edition) of 2005, an AF is defined as a benign odontogenic tumor with odontogenic epithelium and odontogenic ectomesenchyme, but without hard tissue formation\(^\text{11}\). It is different from ameloblastic fibrodentinoma (AFD)
equal between sexes. The most common area of occurrence was the mandibular molar area (20 of 38 cases [52.6%]), followed by the maxillary molar area (6 cases [15.8%]). In addition, Chen et al.\textsuperscript{12,14} reported that 75 out of 102 cases (73.5%) of AF occurred in the mandibular molar area, and the same tendency is seen in Japan also. In the present patient, the AF occurred in the mandibular anterior tooth region. The prevalence of AF in the mandibular anterior tooth region was reported to be 3 out of 38 cases (7.9%)\textsuperscript{14}, suggesting that the present case was rare.

One feature of AF is that it progresses asymptptomatically and is often found only accidentally on radiographic examination\textsuperscript{9}. In the present case, a painless swelling in the mandibular bone caused the patient to visit a nearby dental clinic for detailed examination. Radiographic examination normally reveals well-defined unilocular and multi-locular radiolucencies. In many cases, radiolucency is accompanied by an impacted tooth\textsuperscript{4,14}, which is usually the first or second molar\textsuperscript{6}. In the present case, however, well-defined multi-locular radiolucencies were seen in the mandibular anterior tooth region, accompanied by a supernumerary impacted tooth. Impactions are often of deciduous teeth or permanent teeth, or of a permanent tooth germ. To our knowledge, no studies to date have reported impacted supernumerary teeth. When the impacted tooth is a permanent tooth or permanent tooth germ, it must be preserved as much as possible and occlusal guidance provided as it affects occlusion\textsuperscript{7}. In the present case, the impacted tooth was extracted as it was supernumerary.

Ameloblastic fibroma is a mixed tumor consisting of epithelium and mesenchymal tissue. The epithelium-rich part shows histopathological similarity to ameloblastoma, while the epithelium-lacking part shows histopathological similarity to odontogenic fibroma. Therefore, it is sometimes difficult to diagnose AF\textsuperscript{9}. In previously reported cases, diagnosis following biopsy was inconsistent with the initial diagnosis\textsuperscript{12,14}. Takano et al.\textsuperscript{12} reported a case wherein they performed biopsy and diagnosed AF by pathological examination. They removed the tumor, determined ameloblastic fibrosarcoma with isolated preparation, and performed an additional extended surgery due to the unclear film structure of the tumor. Therefore, it should be borne in mind that the diagnosis arrived at following a biopsy will sometimes differ from the definitive diagnosis. In the present case, diagnosis at biopsy was consistent with the definitive diagnosis. However, taking the above cited report into consideration, enucleation curettement after first removing surrounding bone was also performed following extraction.

Treatment of AF basically involves complete extraction of the tumor. The prognosis is good, and the relapse rate lower than that seen with ameloblastoma\textsuperscript{4,14}. However, Troedahl et al.\textsuperscript{13} reported that relapse occurred in 10 out of 24 cases (41.7%). Further, Chen et al.\textsuperscript{2} reported that relapse occurred in 41 out of 123 cases (33.3%). In their study, the recurrence rate at 5 years postoperatively was 41.6% and at 10 years 69.2%, with 14 cases (11.4%) showing transformation to ameloblastic fibrosarcoma. One recent study found only a low frequency BRAF V600E mutation in AF\textsuperscript{1}, but mutation of the tumor suppressor gene\textsuperscript{5} has been reported and requires attention. In the present case, at 2 years postoperatively, progress is good and no recurrence has been observed. However, strict follow-up is required, as the possibility of recurrence after a long period of time cannot be ruled out.

**Conclusion**

Here, we have reported a case of AF occurring in mandibular anterior tooth region.

**Conflict of Interest**

The authors wish to report no conflict of interest with regard to this paper.
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