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Akane Mochizuki
Department of Pathology, Nihon University School of Dentistry, Tokyo, Japan,101-8310,
deak19028@g.nihon-u.ac.jp

Rei Fukui
Department of Pathology, Nihon University School of Dentistry, Tokyo, Japan,101-8310,
tooyama.rei@nihon-u.ac.jp

Toshihiko Amemiya
Department of Oral and Maxillofacial Radiology, Nihon University School of Dentistry, Tokyo, Japan, 101-8310, amemiya.toshihiko@nihon-u.ac.jp

Yoshinori Arai
Department of Oral and Maxillofacial Radiology, Nihon University School of Dentistry, Tokyo, Japan, 101-8310, arai.yoshinori@nihon-u.ac.jp

Masatake Asano
Department of Pathology, Nihon University School of Dentistry, Tokyo, Japan,101-8310,
asano.masatake@nihon-u.ac.jp

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CASE REPORT

A Case of Ameloblastic Fibrodentinoma in the Posterior Maxilla

Akane Mochizuki[1], Rei Fukui[1*], Toshihiko Amemiya[2], Yoshinori Arai[2], Masatake Asano[1]

[1]Department of Pathology, Nihon University School of Dentistry, Tokyo, Japan,101-8310
[2]Department of Oral and Maxillofacial Radiology, Nihon University School of Dentistry, Tokyo, Japan, 101-8310
**Correspondence e-mail to: tooyama.rei@nihon-u.ac.jp*

ABSTRACT

Ameloblastic fibrodentinoma (AFD) is a rare tumor with an incidence rate of less than 1%. When lesion with the histomorphology of ameloblastic fibroma(AF), which is true neoplasms, form dysplastic dentin, and had been referred to as AFD. It histologically consists of odontogenic ectomesenchyme resembling the dental papilla, epithelial strands, and nests resembling dental lamina and enamel organ with dentin formation. Although newly categorized as an odontoma by the WHO in 2017, this lesion was previously referred to as a rare odontogenic tumor by the WHO in 2005. **Objective:** We aim to summarize our case with other previous case reports considered to be equivalent to the conventional WHO classification of AFD. **Case Report:** An 8-year-old girl presented to our hospital complaining of delayed eruption of a tooth. Computed tomography showed an odontoma-like radiopacity in a unilocular radiolucent lesion sized approximately 20 mm. The lesion was extracted under general anesthesia and histopathologically exhibited AFD. Herein, we report a rare case of AFD in the maxilla. **Conclusion:** Although this lesion deviates from the concept of disease as an odontogenic tumor, it is hoped that clinically sufficient follow-up is required and more similar cases will accumulate as independent tumors, rather than simply being recognized as developmental odontomas.

Key words: ameloblastic fibroma, ameloblastic fibro-odontoma, odontoma, odontogenic tumor

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INTRODUCTION

Ameloblastic fibrodentinoma (AFD), classified as an odontoma in the 2017 WHO classification, has been referred to as a rare benign tumor, usually interpreted as a neoplasm similar to ameloblastic fibroma (AF), and characterized by the formation of dysplastic dentin, which shows odontogenic ectomesenchyme resembling the dental papilla, epithelial strands, and nests resembling the dental lamina and enamel organ.[1,2]

The WHO classification in 1971 defined the tumor as a rare odontogenic tumor consisting of odontogenic epithelium and odontogenic connective tissue, characterized by immature dentin formation. Since 1992, the tumor was described as an irregular mixture of odontogenic epithelium and odontogenic ectodermal mesenchyme. The name AFD was used and then classified as an independent tumor. However, for the reason that lesion which exhibited AF with formation of dental hard tissue and reach and exceptional size are most likely developing odontomas, in the 2017 WHO classification revision, on the basis of thistopathological features, AFD and ameloblastic fibro-odontoma (AFO) was categorized as odontoma.[2]

AFD is a rare tumor with an incidence rate of less than 1%, which is less common than AF. The epidemiology is quite similar to odontoma, which are typically diagnosed during the first two decades of life and have most cases of AFD present as painless swelling or are discovered due to disturbances of tooth eruption. Radiographically, it presents as a well demarcated radiolucency with varying levels of radiopacity depending on the extent of mineralization. Thus, lesions sometimes showed an odontoma-like radiographical image. However, since there is also a possibility of malignant transformation, when the lesion reach an exceptional size or recurrent lesions sufficient follow-up is required.

Here, we report a summary of a case considered to be equivalent to the conventional WHO classification (2005) of AFD with a brief literature review.

CASE REPORT

An 8-year-old Japanese girl who visited a local dental clinic for orthodontic treatment and panoramic radiography was found to have an impacted upper right first molar. Thus, the patient presented to our hospital for detailed examination and treatment of delayed eruption of the tooth. There was no swelling, and the surface mucosa was intact. Examination revealed bulging of the bucco-palatal cortical bone with a mild parchment-like feeling in the upper right first molar area. Panoramic radiography revealed a unilocular radiolucent lesion in the region (Figure. 1A). Computed tomography images revealed a spherical mass sized approximately 20 mm bulging on the buccal and palatal sides and loss of continuity of the palatal cortical bone (Figure 1B). The maxillary right first molar with incomplete root formation was present in the maxillary sinus. A spherical mass surrounded the crown, with a small scattering of massive radiopacity. Only the crown of the upper right second molar had formed and was located in the posterosuperior direction of the first molar (Figure. 1C, D). The tumor was removed under general anesthesia based on the diagnosis of an intra-jawbone tumor around the maxillary molar region.

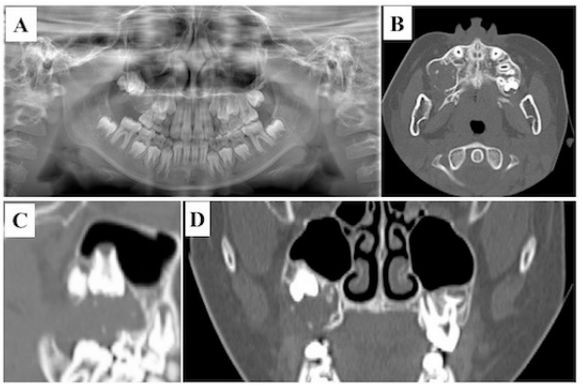

Figure 1. Panoramic radiography (A) shows an impacted upper right first molar, which forms an odontoma-like radiopacity in a unilocular radiolucent lesion. Computed tomography (CT) axial image (B) shows a small scattering massive radiopacity in a unilocular radiolucent lesion near the marginal region. CT sagittal image (C) shows root resorption near the upper right deciduous second molar. CT coronal image (D) shows mild expansion of the bucco-palatal cortical bone.

Histopathological examination showed that the lesion was composed of marked dentinoid material, such as dentin-like hard tissue consisting of spherical osteo-dentin, odontogenic ectomesenchymal tissue, and fibrous tissue. Small epithelial nests were intermingled in the dysplastic dentin (Figure. 2A, B). Small odontogenic epithelial strands and nests were intermingled with mesenchymal cellular tissue, and fibrous tissue included hyalinized changes (Figure. 2C). The odontogenic epithelium with small dark-stained nuclei exhibited small island-like cord-like morphologies showing peripheral palisading, embedded in a cell-rich ectomesenychyme resembling the dental papilla (Figure. 2 D). Enamel organ-like structures were not observed. A diagnosis of odontoma (Ameloblastic fibro-dentinoma [WHO 2005]) was made.

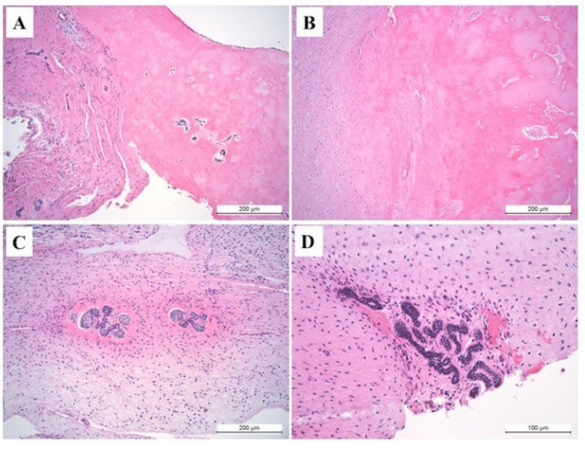

Figure 2. The lesion is composed of mainly dysplastic dentin. The small epithelial islands are intermingled with mesenchymal tissue and fibrous tissue (A). Dysplastic dentin contains dentinoid material, such as dentin-like hard tissue, consisting of spherical osteo-dentin (B). Small odontogenic epithelial islands are intermingled with mesenchymal cellular tissue (C). The epithelial component consists of branching and anastomosing epithelial strands that form knots, with histological features similar to those of the observed AF.

DISCUSSION

AFD was first reported by Straith in 1936 as a rare form of dentinoma.[3] Most of the hard tissue in this type of tumor consists of dentin. The tumor described here was initially defined as an enamel-free odontoma consisting of a small amount of cementitious material, which was later called a dentinoma. In 1970, Gohlin argued that this should be classified as a subtype of dentinoma, considering it as a stage before maturation of the odontogenic epithelial component. Accordingly, the WHO classification in 1971 defined the tumor as a rare odontogenic tumor consisting of odontogenic epithelium and odontogenic connective tissue, characterized by immature dentin formation. In 1992, the tumor was described as an irregular mixture of odontogenic epithelium and odontogenic ectodermal mesenchyme. Hence, the name AFD was used, and this condition was then classified as an independent tumor. However, in the 2017 WHO classification revision,

AFD and ameloblastic fibro-odontoma (AFO) was categorized as odontoma, which is believed to occur during the development of the odontoma.[2]

AFD is pathologically similar to AF with dentin or dentin-like formation. The 2005 World Health Organization classification classified AF, AFD, and AFO as neoplasms composed of proliferating odontogenic epithelium and odontogenic ectomesenchyme. On the basis of the inductive principle, the formation of hard tissue in odontogenic tumors is a result of epithelial-mesenchymal influences, in which the ameloblastic epithelium stimulates the differentiation of odontoblasts from the mesenchyme, and dentin formation subsequently leads to amelogenesis of the enamel matrix.[4,5] Since the mesenchymal tissue is similar to AF in every lesion, it can be considered as an intermediate type of lesion between AF and AFO. It is not yet clear whether there is a transition in these lesions. Takeda et al.[6] suggested that AFD may be involved in the process of tissue differentiation from AF to AFO.

Given this background, it is assumed that its recognition as a developmental odontoma occurring during the developmental process is strongly summed up in the odontoma. However, some lesions diagnosed as AFD or AFO are not usually consistent with the clinical features of an odontoma, such as size and age of onset; therefore, this point remains to be discussed.

From 1936 to 2020, 65 cases of AFD, including the present case, were reported domestically and internationally. [3,7-23] The ages of the patients ranged from 1 to 63 years (1 unknown), with most being relatively young, with an average age of 18.2 years. In terms of gender, there were 39 men (60.0%) and 23 women (35.4%), while 3 (4.6%) were of an unknown gender, indicating that AFD tends to be more common in men. The site of onset was found to be in the maxilla or mandible in 19 (29.2%) and 46 (70.8%) cases, respectively. The tumor can therefore be said to occur more commonly in the mandible. Surgical procedures, such as enucleation or curettage, are generally the treatments of choice, and prognosis is considered to be good. However, malignant transformation to ameloblastic fibrosarcoma has rarely been reported in recurrent lesions. One case recurred 50 months after enucleation of the initial lesion and recurred 92 months later, involving malignant transformation to ameloblastic fibrosarcoma.[19] In another case, the lesion had re-occurred several times before re-occurring as ameloblastic fibrosarcoma 10 years later.[20] As described above, since there is also a possibility of malignant transformation, clinically sufficient follow-up is required.

Although this lesion deviates from the concept of disease as an odontogenic tumor, considering that there have been reports of malignant transformation, it is hoped that more similar cases will accumulate as independent tumors, rather than simply being recognized as developmental odontomas.

CONCLUSION

Herein, we report a case of AFD in the maxilla of an 8-year-old girl with accompanying an overview of our findings. Although these lesions including our present case are clinically most likely developing odontomas, sufficient follow-up is required.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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