Imaging Science in Dentistry 2024; 54: 207-10
https://doi.org/10.5624/isd.20230247

A rare case report of ameloblastic fibrodentinoma with imaging features in a pediatric patient

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ABSTRACT

Ameloblastic fibrodentinoma (AFD) is a rare benign odontogenic tumor that resembles an ameloblastic fibroma with dysplastic dentin. This report presents a rare case of mandibular AFD with imaging features in a young patient. Panoramic radiography and computed tomography revealed a well-defined lesion with internal septa and calcified foci, causing inferior displacement of the adjacent molars as well as buccolingual cortical thinning and expansion of the posterior mandible. The lesion was surgically removed via mass excision, and the involved tooth was extracted under general anesthesia. During the 5-year follow-up period, no evidence of recurrence was observed. Radiologic features of AFD typically reveal a moderately to well-defined mixed lesion with varying degrees of radiopacity, reflecting the extent of dentin formation. Radiologists should consider AFD in the differential diagnosis when encountering a multilocular lesion with little dense radiopacity, particularly if it is associated with delayed eruption, impaction, or absence of involved teeth, on radiographic images of young patients. ***(Imaging Sci Dent 2024; 54: 207-10)***

KEY WORDS: Odontogenic Tumors; Computed Tomography, X-Ray; Diagnostic Imaging; Fibro-odontoma, Ameloblastic

Ameloblastic fibrodentinoma (AFD) is a rare benign odontogenic tumor, representing only about 1% of all odontogenic tumors reported.[1,2] Typically, this tumor presents as a painless, slow-growing lesion that resembles an ameloblastic fibroma (AF) with internal calcification.[3-6] Histologically, AFD is characterized by odontogenic ectomesenchyme and epithelial strands or nests with dentin formation.[5,7,8] In particular, the lesion is classified as an AFD when its hard tissue can form dysplastic dentin; microscopically, AFD is distinguished by the presence of dentin or dentinoid material, in contrast to an ameloblastic fibro-odontoma (AFO), which contains both dentin and enamel.[1,3,5,6,8]

Radiographically, AFD presents as a relatively well-defined radiolucent lesion containing varying amounts and degrees of internal radiopacity, exhibiting less dense opacity compared to AFO.[3,6] Common features in young patients with AFD include delayed eruption, congenitally missing teeth, and impaction of permanent teeth.[6] Studies indicate that approximately 80% of reported AFD cases are associated with the teeth in the posterior mandible, with the age of onset typically before the second decade of life.[1,6,7,9] To the authors' knowledge, few previous studies have focused on the imaging characteristics of pediatric AFD, particularly with regard to computed tomography (CT).[6,7] Consequently, this report presents a rare case of mandibular AFD in a 6-year-old male patient, highlighting radiological features, including CT images, to aid in the differential diagnosis.

Case Report

A 6-year-old boy was referred to Yonsei University Dental Hospital in June 2018 due to the delayed eruption of a permanent mandibular molar. At the time of the visit, the patient exhibited no clinical signs or symptoms, such as pain or discomfort. No specific previous medical, familial, or psychosocial history was reported, including relevant genetic information. The patient's dental history was unremarkable, with only treatment for dental caries noted; no other specific dental issues were reported. Giv-

Received November 15, 2023; Revised February 29, 2024; Accepted March 8, 2024
Published online April 2, 2024
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Imaging Science in Dentistry · pISSN 2233-7822 eISSN 2233-7830

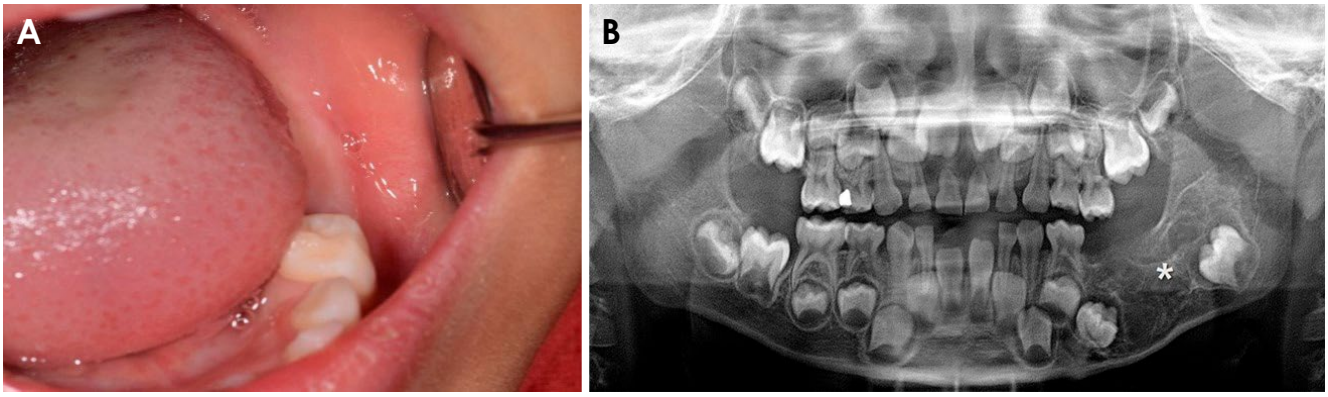

Fig. 1. A. Intraoral photograph reveals missing teeth in the left mandibular posterior region. B. Panoramic radiograph shows a multilocular mixed lesion characterized by internal septa, calcified foci, and the displacement of the left deciduous and permanent molars (asterisk).

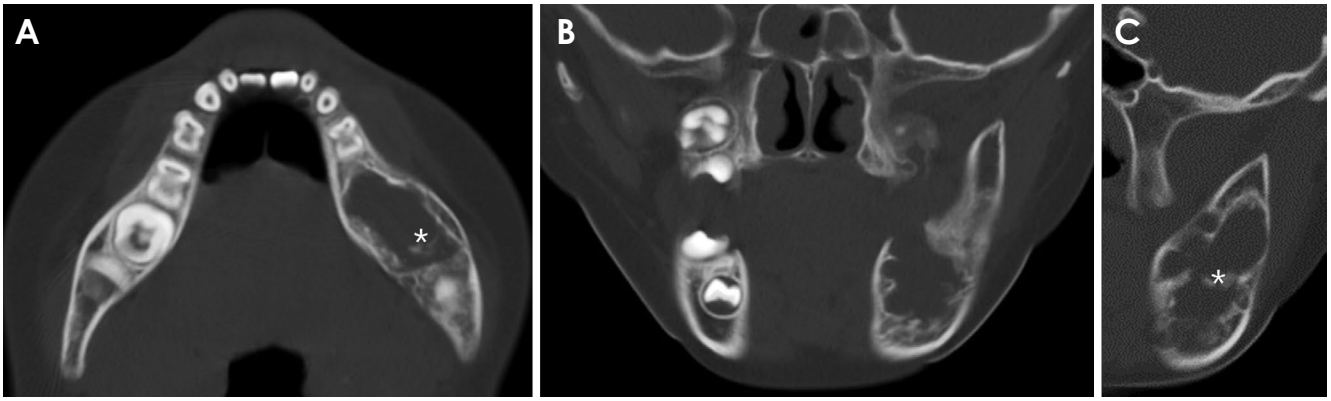

Fig. 2. A. Buccolingual expansion of the left posterior mandible, exhibiting an irregular, diffuse cortical border containing calcified material (asterisk). B. The lesion's border is relatively well-defined, with thin cortication. C. The lesion displays an irregular border with incomplete septa and calcified foci (asterisk).

en the patient's age, absent or delayed eruption of the left mandibular first molar and the left deciduous mandibular second molar was clinically suspected, as illustrated in Fig. 1.

Panoramic radiography (Rayscan Alpha Expert; Ray, Seoul, Korea) revealed a multilocular mixed lesion with internal septa and calcified foci in the left posterior mandible. Inferior displacement of the affected deciduous and permanent molars was observed (Fig. 1B). Initially, an ameloblastic lesion was clinically suspected.

The patient underwent a CT examination (Genesis Hispeed; GE Healthcare, Milwaukee, WI, USA) to aid in differential diagnosis. The images revealed a well-defined, multilocular radiolucent lesion with mild buccolingual expansion and cortical thinning in the left mandibular body and ascending ramus (Fig. 2A). The border of the lesion was relatively distinct, with an irregular, thin, corticated, and scalloped margin (Fig. 2B). Imaging also revealed a partial opening into the oral cavity at the superior aspect of the lesion. The lesion contained incomplete septation and focal areas of calcification (Fig. 2C). Notably, the density of the internal radiopaque foci within the lesion was lower than that of the enamel. Given the patient's young age, AFO was initially suspected; AF was also considered in the differential diagnosis.

The lesion was excised surgically under general anesthesia, and the affected tooth was concurrently removed. Histopathological examination revealed a soft tissue component consistent with AF, alongside calcifying eosinophilic dentinoid material. The soft tissue component exhibited features of mesenchymal tissue similar to the dental papilla, interspersed with small nests of odontogenic epithelium. The final diagnosis was AFD (Fig. 3). The patient experienced uneventful healing with no signs of recurrence

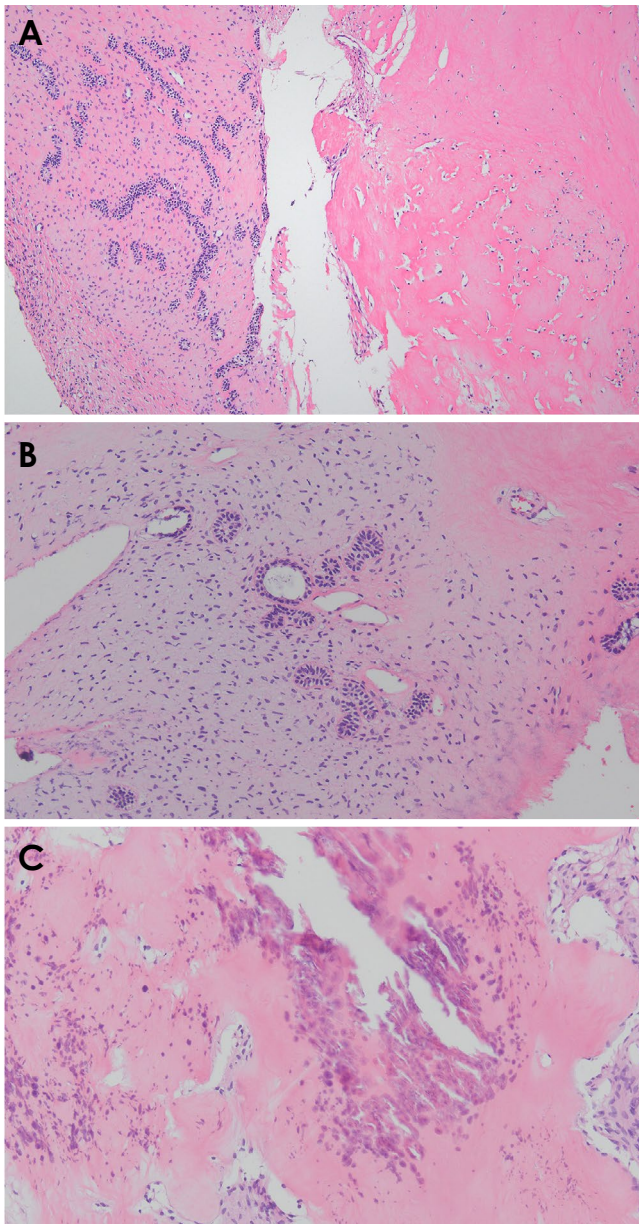

Fig. 3. Histopathological examination using hematoxylin and eosin staining. A. The tumor is composed of a soft tissue component identical to that of an ameloblastic fibroma (left) and adjacent dentinoid material (right) (original magnification ×100). B. The soft tissue component is characterized by mesenchymal tissue that resembles the dental papilla and contains small nests of odontogenic epithelium (original magnification ×200). C. The eosinophilic dentinoid material is in the process of calcification (original magnification ×200).

throughout the 5-year follow-up period. Additionally, orthodontic and prosthetic treatments were administered.

Discussion

AFD is recognized as an independent true tumor, distinct from AFO and AF in terms of its biological characteristics, age of onset, and hard tissue composition.[1,3,10,11] These findings suggest that AFD, as a true neoplasm, possesses the capacity for malignant transformation.[1,12-15] Chrcanovic and Gomez[3] reported that, of 42 cases of recurrent AFD, 2 underwent malignant transformation into ameloblastic fibrosarcoma. In contrast, AFO exhibits a lower propensity for malignant transformation than AFD and generally has a more favorable prognosis.[7] Sanchez-Romero et al.[1] also noted that lesions with a greater amount of hard tissue tend to be less aggressive and have a reduced risk of malignant transformation. Consequently, AFD may exhibit more aggressive growth than AFO, and the potential for malignant transformation should be carefully considered.

Distinguishing between AFD and AFO can be challenging, since both present as mixed lesions with several overlapping clinical features.[3,6] However, previous research has shown that AFO and AFD differ considerably in locularity and radiopacity,[1,3,6] which can assist in the radiological differential diagnosis. Specifically, AFD typically presents as a mixed lesion with relatively well-defined but irregular borders and tends to display more locularity than AFO, with thin, incomplete septa. Additionally, AFD is composed solely of "dentin or dentinoid material" and lacks "enamel matrix or mature enamel."[8] In contrast, AFO is generally unilocular and includes a higher level of radiopacity due to the presence of enamel. Consequently, AFD exhibits less dense opacity compared to AFO and is more multilocular, with varying degrees of radiopacity depending on the amount of internal calcified dentinoid.[1]

If a young patient presents with findings resembling those of AFO but with less internal calcification and a more multilocular appearance, the possibility of AFD should also be considered.[2,16] Since these lesion types can have distinctly different outcomes, radiologists are advised to include the less common AFD in their differential diagnosis rather than quickly attributing the findings to AFO. Furthermore, particular attention should be paid to cases exhibiting an irregular border, bone expansion, and cortical perforation, as these characteristics may indicate malignant transformation.[6] Considering the potential neoplastic features of AFD, it is imperative to maintain long-term follow-up with patients. This enables vigilant monitoring for any signs of recurrence or malignant transformation, in addition to facilitating early diagnosis and treatment.[2,7,9]

With AFD considered a very low-frequency tumor, it is unsurprising that only 70 cases have been reported since the first case of AFD was documented in 1936.[3] Due to the

rarity of AFD, the present report includes a single pediatric case, for which a detailed description of panoramic and CT images was available. Further research focusing on the radiologic aspects of AFD is recommended to better understand the characteristic imaging features of this condition.

In conclusion, AFD typically presents radiologically as a moderately to well-defined multilocular lesion with varying degrees of internal radiopacity, reflecting the extent of dentin formation. This radiopacity is generally lower than that of enamel. Additionally, delayed eruption, displacement, impaction, or absence of teeth may be observed. Radiologists should consider the possibility of AFD, particularly in younger patients, when encountering a mixed radiographic lesion that resembles AFO but exhibits less radiopacity than enamel and a relatively higher degree of locularity.

Conflicts of Interest: None

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