Oral Radiology (2023) 39:220–224
https://doi.org/10.1007/s11282-022-00649-4

CASE REPORT

MR appearance of a rare ameloblastic fibroma with formation of dental hard tissues with histopathologic correlation: a case report

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Received: 21 June 2022 / Accepted: 10 August 2022 / Published online: 24 August 2022

Abstract

An ameloblastic fibroma with formation of dental hard tissues, which the classical name is ameloblastic fibro-odontoma (AFO), is a rare type of mixed odontogenic tumor. An 8-year-old boy was diagnosed with AFO, with an inhomogeneous high signal within the lesion shown by T2-weighted magnetic resonance imaging (MRI). Computed tomography (CT) imaging revealed a unilocular low CT value area of 24 × 19 × 26 mm with buccolingual bony expansion and cortical bone thinning on the left side of the mandible including the crown of the mandibular left second molar. In addition, multiple calcified bodies were detected within the lesion, one of which had a CT value of approximately 2200 HU, equivalent to that of enamel. MRI indicated the lesion to be sized 24 × 19 × 25 mm along with buccolingual bony expansion in the left side of the mandible. Additionally, the lesion showed an internal inhomogeneous high signal, while a portion had an especially high signal in T2-weighted images. That particularly high signal area coincided with the nodular growth area of mucus-rich mesenchymal components without the epithelial component in histopathology findings. The particularly high signal revealed by T2-weighted imaging could be attributed to the mucus-rich component. MRI was found useful for revealing differences in the internal histopathological properties of an AFO in our patient.

Keywords Ameloblastic fibro-odontoma · Ameloblastic fibroma · MRI · Benign tumor · Hamartoma

Introduction

An ameloblastic fibro-odontoma (AFO) is a rare benign odontogenic tumor with enamel and dentin formation and histologic findings of an ameloblastic fibroma (AF)—a mixed tumor consisting of odontogenic ectomesenchyme similar to dental papilla with odontogenic epithelial strands and nests similar to enamel organ and dental lamina [1–3]. AFO accounts for less than 2% of odontogenic tumors. These neoplasms occur predominantly in the mandibular molar region and less frequently in in the maxillary molar region, and are most commonly noted in individuals under

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20 years of age, with a male predilection [4, 5]. These lesions are typically painlessly and slow growing with jawbone expansion [1, 6]. Although AFO was classified as an odontogenic benign tumor in the 3rd edition of the WHO classification, the current WHO classification (4th edition, revised 2017) considers the lesion to be within the spectrum of an odontoma—with the rationale that the hard tissue formation within the AFO lesion are similar to a developing odontoma (hamartoma) [2, 3]. Nevertheless, the concept that AFOs are true neoplasms has not been completely dismissed and it is still listed as rare AFs with formation of dental hard tissues [2].

A typical AFO manifests as a well-defined unilocular or multilocular radiolucency bordering an unerupted tooth, or encompassing its crown. The internal contents have some degree of radiopacity depending on the extent of mineralization that has occurred within the lesion [6, 7]. Most previous reports of AFO have described findings on conventional radiographic images [8–15] and reports detailing the appearances on computed tomography (CT) and magnetic resonance (MR) radiological images are limited. To the best of our knowledge, there have been no previous reports regarding an internal inhomogeneous signal of an AFO shown by T2-weighted MR imaging (MRI). Here, we report a rare case of an AFO that showed a particularly high signal as compared to the surrounding area within an AFO lesion on T2-weighted MR images.

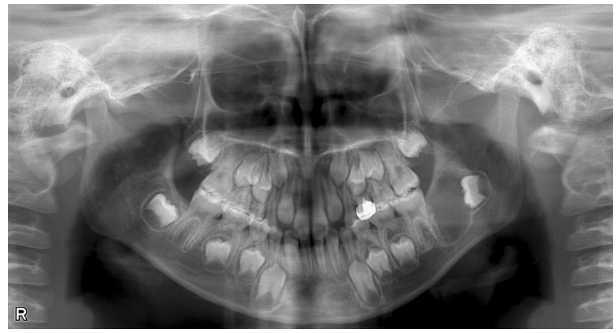

Fig. 1 Panoramic radiograph. A well-defined pericoronal unilocular radiolucency is noted involving the mandibular left first molar, with no root resorption or tooth displacement. Note slight radio-opacity detected on the medial side of the lesion

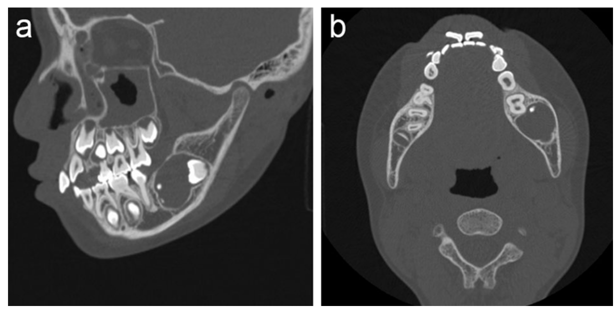

Fig. 2 CT examination. **a** Sagittal section. A unilocular low attenuation area measuring approximately 24 × 19 × 26 mm on the left side of the mandible including the crown of the mandibular left second molar. Multiple calcified bodies are present within the lesion, one of which had a CT number of approximately 2200 HU. **b** Axial section. Buccolingual bony expansion and cortical bone thinning caused by the lesion

Case report

An 8-year-old boy was referred to our hospital for further investigation and treatment of an asymptomatic pericoronal radiolucency involving the mandibular left second molar—an incidental finding on a panoramic radiograph made at a general dental clinic. Extraoral examination showed no facial asymmetry or swelling on the left side of the mandible. However, on intraoral examination, mild swelling was evident on the left side of the mandible. There was no relevant medical or family history.

The panoramic radiograph showed a well-defined unilocular radiolucent area from the distal side of the mandibular left first molar to left mandibular angle, encompassing the crown of the mandibular left second molar (Fig. 1). The mandibular left first molar was not resorbed or displaced. The lesion extended to the inferior alveolar canal and the superior cortex of the canal was not perceptible at the interface. Slight internal radiopacity was detected on the medial side of the lesion. The level of development of the mandibular left second molar was age-appropriate and it was not significantly different from that of the contralateral second molar.

Multi-detector CT images showed a unilocular low attenuation area measuring approximately 24 × 19 × 26 mm, involving the crown of the mandibular second molar. Buccolingual cortical expansion and thinning was detected, extending from the distal side of the mandibular left first molar to the left mandibular angle (Fig. 2). In addition, multiple discrete radiopacities were detected within the lesion, one of which had a CT number of approximately 2200 HU, equivalent to that of enamel.

MRI corroborated the expansile nature of the lesion (Fig. 3). On T1-weighted images, the lesion signal was isointense with muscle, with loss of the normal bone marrow fat signal (Fig. 3a). On T2-weighted images, the lesion signal was inhomogeneous with regions of high signal intensity relative to the surrounding lesion tissue (Fig. 3b). These regions of high signal intensity also appeared as high intensity regions on T2 iterative decomposition of water and fat with echo asymmetry and least-squares estimation (IDEAL) water images (Fig. 3c). The apparent diffusion coefficient

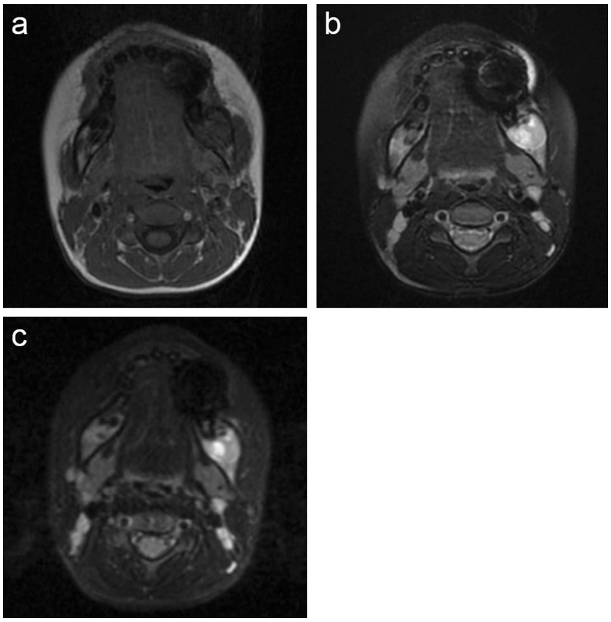

Fig. 3 MR examination. **a** T1-weighted image shows the lesion is isointense with muscle tissue. **b** T2-weighted images reveals an internal inhomogeneous high signal, with a portion of the lesion exhibiting an especially high signal. **c** T2 IDEAL water imaging shows an internal inhomogeneous high signal with a region of especially high signal

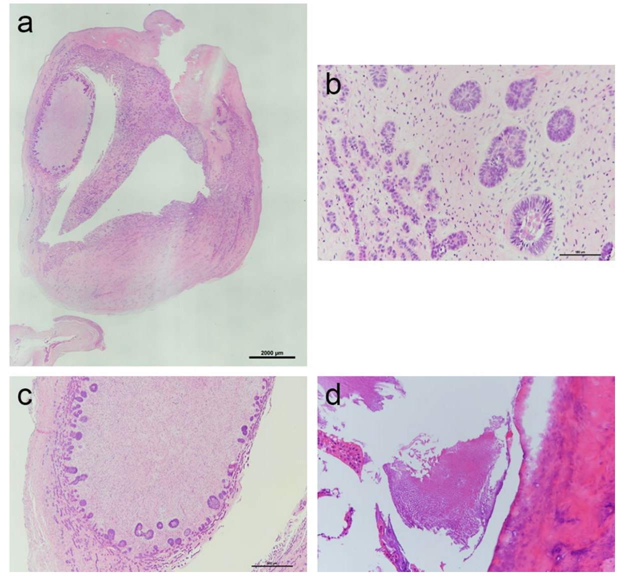

Fig. 4 Histopathologic images (hematoxylin–eosin stain). **a** Low power magnification image of excised lesion (bar = 2000 µm). **b** High magnification of lesion revealed a mixture of odontogenic epithelial nests and odontogenic ectomesenchyme (×20). **c** The portion of nodular growth within the lesion showing a mucus-rich mesenchymal component without an epithelial component, and surrounded by droplet-shaped epithelial nests (×4). **d** One of the multiple hard tissues was a tooth with a diameter of 5 mm, with obvious enamel and dentin formation (×20)

(ADC) value for the entire lesion was 2.45×10^{-3} mm^2/s, while that for the area within the lesion with the particularly high signal in T2-weighted and T2 IDEAL water images was 3.22×10^{-3} mm^2/s. One of the calcified bodies within the lesion identified by CT was depicted as a signal-free area on both T1- and T2-weighted imaging. Based on these findings, we suspected a benign odontogenic tumor associated with dental tissue formation, such as ameloblastic fibroma with formation of dental hard tissues.

Three months after the lesion was noted, a biopsy was performed. Histopathologic examination showed dense odontogenic epithelial nests, though a definitive diagnosis could not be obtained, because an invasive lesion could not be ruled out. With the tentative diagnosis of odontogenic tumor, the lesion was removed by enucleation along with the mandibular left second molar. The lesion easily detached from the surrounding bone and tissue.

Macroscopically, the excised lesion was a substantial mass, with a yellow–white cut surface that resembled an impacted mandibular left second molar and its dental follicle. Histopathological examination revealed cord-like or follicular odontogenic epithelial nests resembling an enamel organ and dental lamina, and odontogenic ectomesenchyme resembling dental papilla (Fig. 4a, b). However, a part of the lesion had grown in a nodular manner from only the mucus-rich mesenchymal component without the epithelial component and was surrounded by droplet-shaped epithelial nests (Fig. 4c). Multiple hard tissues were noted at the margins of the lesion, one of which was a tooth with a diameter of 5 mm with obvious enamel and dentin formation (Fig. 4d). Based on these findings, the final diagnosis of a rare ameloblastic fibroma with formation of dental hard tissues was established. Prior to the current WHO classification, this lesion would have been referred to by its classical name—ameloblastic fibro-odontoma. The postoperative course was uneventful and with no signs of recurrence observed at a post-surgical follow-up examinations up to 3 years.

Discussion

In the 3rd and earlier editions of the WHO classification [3], AFO as well as ameloblastic fibrodentinoma (AFD) were categorized as mixed odontogenic tumors with dental hard tissue formation in an ameloblastic fibroma component, which consists of odontogenic ectomesenchyme similar to dental papilla, and odontogenic epithelium similar to enamel organ and dental lamina. However, in the 4th edition, the current WHO classification, AF is classified as a benign mixed epithelial and mesenchymal odontogenic tumor, and AFO and AFD were integrated into the category of an odontoma, and listed as hamartomas [2]. In this classification, AFO and AFD, which are considered to be neoplastic, are described as "rare AFs with formation of dental hard tissues" [16]. Differentiation between neoplastic and hamartoma disease has important clinical implications, for example, the possibility of recurrence and the potential of malignant transformation, both of which are important considerations for management of AF, AFO, and AFD tumors [17–19]. This emphasizes the need to better characterize the clinical and imaging appearances of AF, AFO and AFD.

The patient demographic in our case matches the tumor's typical distribution. Radiologically, the lesion appeared as a pericoronal radiolucency with small amounts of internal radio-opacity. Although radiopaque components of an AFO are generally located at the center of the lesion [8–13], radio-opacity was found on the medial side in the present case. Although AFOs demonstrate varying calcification patterns that are blended and full or inhomogeneous, including complex odontoma-like calcifications, and have been shown to be associated with impacted teeth on CT images [15], multiple calcified bodies were few in the present case. Previously, Uchiyama et al. reported that the CT value of a calcified body is useful for diagnosis of AFO [14]. In the present case, one of the calcified bodies within the lesion had a CT value of approximately 2200 HU, equivalent to that of enamel. This information allowed exclusion of other lesions that manifest with a mixed radiolucent-radiopaque appearance, such as calcifying odontogenic cyst, calcifying epithelial odontogenic tumor and adenomatoid odontogenic tumor.

The MRI appearance of this lesion was unusual, with an internal inhomogeneous high signal, and a region with a particularly high signal on T2-weighted images. The area with especially high signal coincided with the nodular growth area of the mucus-rich mesenchymal components without the epithelial component, and likely the high signal intensity is due to the presence of a mucus-rich component. The ADC value of the entire lesion was 2.45×10^{-3} mm^2/s, while that of the area with the particularly high signal area in T2-weighted and T2 IDEAL water images was 3.22×10^{-3} mm^2/s. This difference in ADC values might reflect a difference in cell density due to neoplastic growth. To the best of our knowledge, this unusual appearance of an AFO on T2-weighted MR images and the ADC characterization of the neoplasm have not been presented, and the findings in the present study are very rare. The new WHO categorization and change in nomenclature of an ameloblastic fibro-odontoma is controversial [20]. Reports of MR imaging characteristics of these lesions with corresponding histologic evaluation will aid in future analysis of the neoplastic versus hamartomatous nature of these lesions.

In conclusion, we present a rare case of ameloblastic fibroma with formation of dental hard tissues, that showed a particularly high signal within the lesion as compared to the surrounding area in T2-weighted MR images. This difference in signal intensity might reflect different histopathological components within the lesion. This case highlights the clinical value of MR imaging to asses internal composition of odontogenic tumors contributing to assessment of its potential neoplastic features.

Funding Not applicable.

Declarations

Conflict of interest Not applicable.

Ethics approval All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (Osaka University Graduate School of Dentistry) and with the Helsinki Declaration of 1975, as revised in 2008 (5).

Informed consent Informed consent was obtained from the patient for being included in the study.

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