



Ameloblastic Fibrodentinoma: Report of a Rare Case

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ABSTRACT

Aim: This is to report a rare case of ameloblastic fibrodentinoma (AFD), including detailed history, histopathologic as well as radiographic findings. Her condition, a mixed odontogenic tumor, has rarely been reported in publications. We also included the gist of theories put forward regarding the histogenesis of this lesion.

Background: AFD is a rare and controversial entity; it boasts a variety of classifications and terminology based on its unique biological nature as well as histopathological features.

Case description: The patient was a 13-year-old girl with a chief complaint of central unerupted tooth on the left mandible. Cone beam computed tomographic images revealed a mixed pericoronal and multilocular lesion surrounding the crown of the aforementioned tooth, displacing it apically to a large extent.

Conclusion: It is important to differentiate AFD from other benign mixed odontogenic tumors with similar radiographic appearance because of different therapeutic approaches in some of these tumors.

Clinical significance: From the clinical point of view, it is of paramount significance for dental practitioners to assess any delayed tooth eruption (more than 6 months and in comparison with its counterpart on the opposite side) radiographically to ascertain or rule out any such similar lesions.

Keywords: Ameloblastic fibrodentinoma, Mixed odontogenic tumors, Dentinoma, Case report.

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BACKGROUND

Ameloblastic fibrodentinoma (AFD), a rare odontogenic tumor¹ is regarded as a hamartomatous lesion with odontogenic origin and is placed at an intermediate stage between ameloblastic fibroma and ameloblastic fibro-odontoma (AFO).² An unanimously agreed-upon definition states that it is a neoplasm similar to ameloblastic fibroma with inductive changes leading to dentin formation.³ It is

slow-growing, asymptomatic and often associated with unerupted tooth.¹ It tends to occur with incisor primary teeth as well as molar permanent ones.¹ There is a growing predisposition for it among young males below the age of 20.⁴ Radiographic findings tend to mimic AFO, odontoma, ameloblastic fibroma, central odontogenic fibroma,⁵ odontoameloblastoma and calcifying cystic odontogenic tumor.

CASE DESCRIPTION

A 13-year-old girl looking otherwise normal and healthy presented at Dental Faculty of Mashhad University of Medical Sciences (Mashhad, Iran) with a chief complaint of central unerupted tooth on the left mandible. She seemed blissfully unaware of the lesion since there was mention of neither pain nor swelling. Periapical radiographic findings showed a mixed pericoronal lesion with demarcated cortex on the central tooth, left mandible. cone beam computed tomographic (CBCT) images revealed a pericoronal and multilocular radiolucency with small calcified foci surrounding the crown of the aforementioned tooth, displacing it apically to a large extent (Fig. 1). With a

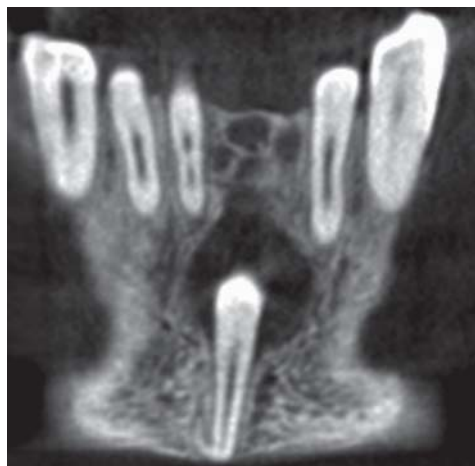


Fig. 1: CBCT, coronal view: a mixed pericoronal lesion affecting mandibular central tooth. Note the clear septa and its cluster view

radiographic differential diagnosis of AFO and calcifying cystic odontogenic tumor, the patient was referred to the Oral Surgery Department where she underwent enucleation.

There were four pieces of the extraction in all, the largest sizing $12 \times 7 \times 3$ mm whereas the smaller ones measuring almost $7 \times 5 \times 3$ mm. On histopathological examination, the hematoxylin and eosin (H&E)-stained section showed a benign neoplastic proliferation of the pulp-like odontogenic mesenchymal cells, odontogenic epithelial cells with enamel organ pattern, dentin, predentin and scattered marginal calcification with no enamel matrix (Figs 2A to C). Those findings were all indicative of AFD.

DISCUSSION

AFD is a rare and controversial entity; it boasts a variety of classifications and terminology based on its unique biological nature as well as histopathological features.⁶

It was first christened 'dentinoma' and depending on its progress, divided into mature and immature categories, which were abolished some time later in 1971.³ AFD and dentinoma have been used synonymously since 1992,³ whereas it is currently a distinct entity in WHO classification system.⁵ There are two main subtypes: peripheral and central,^{1,3} and a pigmented type was reported as well.⁷

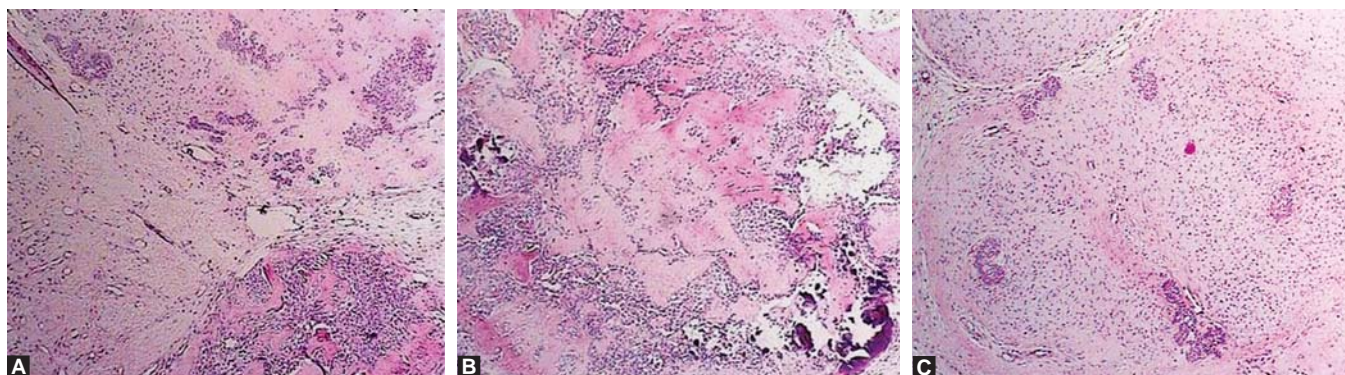
A number of theories have been put forward regarding its histogenesis, which has remained largely unknown to date.⁸ Eversole asserts that it is highly unlikely for an immature type (ameloblastic fibroma) to evolve and grow to a distinct type (complex odontoma), possibly owing to the differentiated factors that are produced by special tumors.⁹ Based on his findings and with regard to the occurrence of AFO in younger patients compared to ameloblastic fibroma, Slootweg concluded that there is a narrow possibility for ameloblastic fibroma to develop into more distinguished odontogenic lesions.¹⁰ Nevertheless, some tend to place AFD in a chronological spectrum of lesions with ameloblastic fibroma on the one end, AFD and

AFO at intermediate stage and odontoma on the other.³ Gardner, for instance, believes that AFD cannot be regarded as a separate category due to the lack of observed different biological behavior and merely based on its inability to create enamel.² Philipsen et al claim that there are two histologically indistinguishable variants for ameloblastic fibroma and AFD. The neoplastic variant, if left *in situ*, fails to develop any further whereas the hamartomatous type is capable of evolving to AFO and subsequently to complex odontoma. However, WHO recently agreed on odontoma as a separate entity, stemming from dental lamina.³

In our case, given the patient's age at the time of referral to our department and the fact that permanent incisors normally erupt at the age of 8, it can be concluded that the lesion had long been in progress, whereas the enamel had not been developed in this period. This, in turn, attests to the claim that AFD, as a separate entity by nature, does not evolve to either AFO or odontoma.

AFD predominantly occurs as a slow-growing asymptomatic swelling, commonly associated with an unerupted tooth.^{5,6} Our patient was, by and large, asymptomatic, since there was mention of neither pain nor swelling and only presented symptom was an unerupted tooth affected by the lesion. However, there are a few articles that have been reported aggressive types of this lesion associated with pain or cortical perforation.⁶ The tumor is reported to occur in childhood and in those below 35, with an average age of around 11 (compared to our case, a 13-year-old girl). There was also reported a male predisposition, with a male to female ratio of 3:1 and also 8:3.^{5,8} Findings also have it that mandibular molars have the most likelihood to be affected, with mandibular incisors involved largely when they are primary.^{8,11} Surprisingly, our case presented with the involvement of mandibular permanent incisors, a very uncommon finding in this respect.^{8,11}

The lesion can present with a disparity of radiographic features, ranging from unilocular radiolucency, multilocular



Figs 2A to C: (A) Ameloblastic fibroma-like appearance plus dentin formation (H&E stain 40× magnification), (B) stellate reticulum and amorphous calcification accommodating dentin (H&E stain at 40× magnification), (C) ameloblastic epithelial islands in dental papilla-like fibrotic tissue (H&E stain at 100× magnification)

radiolucency, mixed (radiolucent and radiopaque) and radiopaque, but commonly observed as a pericoronal radiolucency with varying degrees of opacity owing to unerupted tooth. It makes it radiographically difficult, if not impossible; to differentiate AFD from developing odontoma.⁵ The radiography of presented case showed pericoronal radiolucency with very delicate sparse calcified foci, and severe apical displacement of the affected unerupted tooth as has been reported in other reports.¹²⁻¹⁴ Another interesting radiographic feature in our case concerns obvious septa in the proximity of alveolar crest, creating a grape-like presentation which had also been reported by Lukinmaa et al.¹³ Our initial radiographic differential diagnoses included AFO and calcified cystic odontogenic tumor. The former, although matching age, tends to involve posterior areas and does not usually present with multilocular features on graphy.¹⁵ The latter actually seemed a better match given the patient's age, anterior region involvement, calcification pattern and multilocular appearance.¹⁵

AFD is largely composed of odontogenic epithelium and odontogenic mesenchyme with dentin or dentin-like tissue. Epithelium can be seen in narrow stripes, including two layers of cuboidal cells and papilla-like soft tissue, a common feature with ameloblastic fibroma.⁵ What makes it distinct from ameloblastic fibroma is its exclusive abortive dentin formation in between epithelial and mesenchymal tissue throughout early, formation, growth and maturation stages. Epithelial islands similar to enamel organs are separated from dental papilla-like mesenchyme by a narrow cell-free zone, which are themselves surrounded by round-shaped mesenchymal cells during early stages.⁷ This cell-free layer will subsequently transform to dental matrix with a radial structure surrounded by differentiated mesenchymal cells. Immature dental matrix will then grow to form conglomerates of abortive dentin, which include both epithelial and mesenchymal components. Ameloblastic fibroma sometimes has the narrow cell-free stripe between epithelial and mesenchymal tissue with or without hyalinization but minus the surrounding mesenchymal cells.⁷

It is of paramount significance to differentiate AFD from ameloblastic fibroma and odontoameloblastoma as the latter two are inherently aggressive and thus require different therapeutic approaches. As was mentioned earlier, what makes AFD distinct from ameloblastic fibroma is its dentin formation since both of them share similar epithelial as well as mesenchymal components. Likewise, there can be seen dental papilla or follicle-like odontogenic mesenchymal tissue as well as epithelial stripes along with stellate

reticulum in odontoameloblastoma. However, to differentiate it from AFD, we need to look into calcifications that are similar to the compound or complex odontoma. AFD also resembles AFO clinically, radiographically and pathologically; the distinction though lies in enamel formation in AFO, which is absent in the other one.

The best treatment plan for solitary lesions seems to be enucleation. A conservative modality is recommended in cases of AFO and AFD.⁵

CONCLUSION

It is of paramount significance to differentiate AFD from other benign mixed odontogenic tumors with similar radiographic appearance specially ameloblastic fibroma and odontoameloblastoma as the latter two are inherently aggressive and thus require different therapeutic approaches.

CLINICAL SIGNIFICANCE

As was noted in our case, the only symptom of odontogenic cysts and tumors can be an unerupted tooth affected by the lesion. Hence, it seems prudent on the part of dental practitioners to assess any delayed tooth eruption (more than 6 months and in comparison with its counterpart on the opposite side) radiographically to ascertain or rule out any such similar lesions.

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