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CASE REPORT

Gingival ameloblastic fibro-dentinoma—Report of a case in a child

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Summary Peripheral ameloblastic fibro-dentinoma (AFD) is an extremely rare benign mixed odontogenic tumor. From a review of the English-language literature, to the best of our knowledge only two gingival cases have previously been reported. Here, we report an interesting case of AFD occurring in the upper labial gingiva between the central incisors of a child, in order to raise awareness of the possible presence of this pathologic entity.

1. Introduction

Ameloblastic fibro-dentinoma (AFD) is a controversial neoplasm with respect to its biological nature and histological diagnosis. Different classifications and terminology have been applied to this tumor, and prior to the 1992-World Health Organization (WHO) classification, the term "ameloblastic fibro-dentinoma" was commonly used [1]. In this 1992-WHO classification of odontogenic tumors, AFD is defined as a neoplasm similar to ameloblastic fibroma that also shows inductive changes leading to the formation of dentin [1]. Furthermore, the terms "AFD" and "dentinoma" are used synonymously in this classification. [1]. However, the existence of the dentinoma as an independent entity has still not been fully accepted [2]. The classical division of dentinoma into mature and immature subtypes has also been discarded.

AFD has predominantly occurred in the posterior region of the jaws, and especially the mandibular posterior region. This is usually in association with unerupted molar teeth in childhood [3]. In addition to the intraosseous lesions, extraosseous AFD has occasionally been reported [4,5]. Here, we report an additional case of AFD occurring in the gingiva of a young child.

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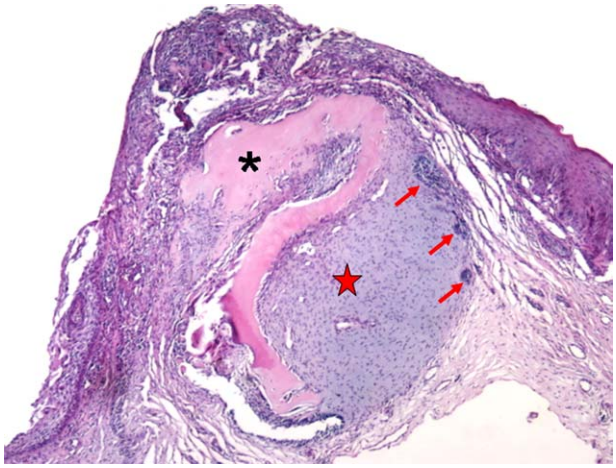

Fig. 1 Low-power magnification photomicrograph showing a primitive dental papilla-like tissue (★) with multiple islands of ameloblast-like cells (arrows) and strands of calcified tissue (*) below the intact gingival (hematoxylin and eosin ×10).

2. Case report

A $2\frac{1}{2}$-year-old girl presented to the Department of Pediatric Dentistry of our institution complaining of a soft tissue swelling in the upper labial gingiva. The mother of the child explained that the swelling had been slowly enlarging for about 1 year and was not associated with any obvious trauma. The patient was otherwise in good health, and her medical and family history was non-contributory.

Examination of the oral cavity revealed the presence of a 0.4 cm soft tissue swelling, located on the labial gingiva between the upper primary central incisors. Upon palpation, this non-ulcerated lesion was found to be firm. Furthermore, the swelling was not tender and, reportedly, painless. Periapical radiography of upper anterior area revealed that bone resorption had not occurred. The clinical impression was of a benign tumor of soft tissue origin, and the entire mass was subsequently excised. At the time of removal, no bony involvement could be visualized. The specimen was then sent for histopathological diagnosis. Low-power examination revealed a neoplastic proliferation of odontogenic epithelial and mesenchymal tissues, beneath an intact labial gingiva (Fig. 1). On higher-power magnification, the odontogenic epithelial component consisted of multiple islands of tall columnar and cubodial ameloblast-like cells with reversed nuclear polarity (Figs. 2 and 3A). The mesenchymal component was primitive connective tissue resembling dental papilla of the tooth germ (Fig. 2). The mineral material consisted of strands of osteodentin (Fig. 2). A fibrillar pattern reminiscent of dentinal tubules is noted in the focal area as marked (Fig. 3B). Enamel formation could not be identified on multiple sections. Ultimately the histological diagnosis was AFD of the gingiva. The excised area healed, and was free of recurrence for 2 years.

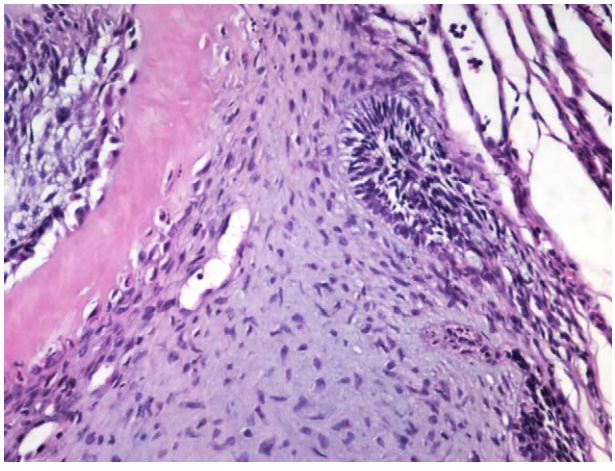

Fig. 2 Higher-power view demonstrating an island of ameloblast-like cells embedded within a primitive dental papilla-like area (hematoxylin and eosin ×100).

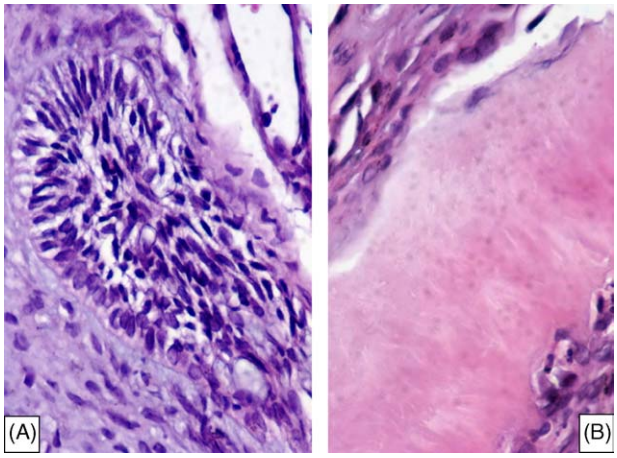

Fig. 3 Columnar and cubodial ameloblastic-like cells with reversed nuclear polarity can be seen (A), as well as a fibrillar pattern reminiscent of dentinal tubules in a focal area of the mineralized tissue (B) (hematoxylin and eosin ×400).

3. Discussion

Peripheral AFD is an extremely rare benign mixed odontogenic tumor consisting of neoplastic odontogenic epithelium and odontogenic mesenchyme with dentin or dentin-like tissues [3]. A review of the English-language medical literature revealed that only two other extraosseous cases have been previously reported with detailed histopathological confirmation [4,5]. The clinical features of these

two previously reported cases, together with the current case, are summarized in Table 1. Characteristically, all three cases were found in the gingiva (buccal/labial or lingual). This concurred with the notion that odontogenic tumors occasionally occur in soft tissue, the gingiva being the most common site [3].

Two cases (cases 2 and 3) occurred in very young children and one occurred in a young adult (case 1) (Table 1). The present case represents the youngest reported peripheral AFD. Two cases (cases 1 and 2) were in white male patients, whereas our case was found in a Chinese female (Table 1). The dimensions of the lesions have been reported in two cases (case 2 and ours); both were small with diameters of approximately 0.5 cm (Table 1). For intraosseous lesion of AFD, it may enlarge to an extreme size [3]. The duration of the two previous gingival lesions was either not stated or uncertain (cases 1 and 2), but the duration (approximately 1 year) in the current case has been noted by the patient's mother. Excision is the treatment of choice and recurrence has not yet been reported.

With respect to histogenesis, some controversy surrounds the relationship between ameloblastic fibroma, AFD, ameloblastic fibro-odontoma and odontoma. Some consider them as separate entities. Others regard them as chronological stages in a continuum beginning from ameloblastic fibroma at one extreme and odontoma at the other extreme with ameloblastic fibro-odontoma as well as AFD in an intermediate stage [2]. In support of this latter proposition, Slootweg [6] analysed 33 mixed odontogenic tumors and found that the mean age of the patients with ameloblastic fibro-odontoma was lower than that of the patients with ameloblastic fibroma. If this finding is correct, the mean age of the patients with ameloblastic fibro-odontoma (which is assumed to differentiate further from ameloblastic fibroma), should have been higher than that of those with ameloblastic fibroma. From this, the author concluded that ameloblastic fibroma represents a separate specific neoplastic entity that does not transit into a more differentiated odontogenic lesion. The age of our patients provides further evidence that contradicts this conclusion as our patient is an extremely young child.

Philipsen et al. [7] have suggested that ameloblastic fibroma and AFD occur in two variants (with indistinguishable histology). The first is a neoplastic lesion, which if left in situ does not appear to mature further. The second variant is a hamartomatous (non-neoplastic) lesion that appears to be able to differentiate into an ameloblastic fibro-odontoma and mature further into a complex odontoma. On the other hand, the compound odontoma is considered as a separate entity resulting from a locally hyperactivity of the dental lamina. This idea has recently been incorporated into the suggested modifications for the 1992-edition of the WHO histological typing of odontogenic tumors [1,8].

On the basis of inductive principle, the development of mineralized components in odontogenic tumors is a sequel of epithelial-mesenchymal interactions in which the ameloblastic epithelium stimulates differentiation of odontoblasts from mesenchyme to deposit dentin. In turn, this induces the formation of enamel matrix. Ameloblastic fibro-odontoma shows a complete interaction between epithelial and mesenchymal components and consequently both enamel and dentin are formed. For lesions of AFD, no enamel has been found and the absence of enamel matrix is the crucial point for histological diagnosis. Therefore, multiple sections should be performed (as done in the present case) to confirm the absolute absence of enamel. Normally, amelogenesis is initiated soon after odontoblasts lay down the dentin. Considering the small amount of dentinoid found in our case, it is possible that the current lesion has already been excised prior to the formation of enamel matrix. If this speculation is true, this may at least partially, explain why the size of the reported cases of peripheral AFD is very small, and confined to the interdental gingiva. Conversely, the lack of enamel in AFD may be because the ability of the deposited denti-

Table 1 Clinical summary of the reported cases of gingival ameloblastic fibrodentinoma

Case no. (authors)	Sex	Age (year-old)	Size (cm)	Race	Location
1. Mckelvy and Cherrick [5]	Male	17	Not-stated	White	Interdental gingiva between lower left 1st and 2nd bicuspids
2. Godjesk et al. [4]	Male	3	0.5	White	Lingual gingiva between lower deciduous left lateral incisor and cuspid
3. Chen et al. [This case]	Female	$2\frac{1}{2}$	0.4	Chinese	Labial gingiva between upper primary central incisors

noid to induce enamel formation has been disrupted through as yet unrecognized etiologies.

In conclusion, a rare case of gingival AFD has been described. This adds to our knowledge of this interesting odontogenic entity, and is of special relevance to pediatric health-care providers since AFD usually occurs in young children.

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