



## **A Case Report of Peripheral –Type Ameloblastic Fibrodentinoma and a Review**

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### **Authors' contributions**

*This work was carried out in collaboration between all authors and all the authors read and approved the final manuscript.*

### **Article Information**

DOI: 10.9734/BJMMR/2015/17353

#### Editor(s):

(1) Emad Tawfik Mahmoud Daif, Professor of Oral & Maxillofacial Surgery, Cairo University, Egypt.

#### Reviewers:

(1) Pasquale Cianci, Department of Medical and Surgical Sciences, University of Foggia, Italy.

(2) Anonymous, Universidade do Oeste Paulista, Brazil.

(3) Anonymous, Kaohsiung Medical University, Taiwan.

Complete Peer review History: <http://www.sciencedomain.org/review-history.php?iid=1122&id=12&aid=9507>

### **Case Study**

**Received 10<sup>th</sup> March 2015**  
**Accepted 27<sup>th</sup> April 2015**  
**Published 30<sup>th</sup> May 2015**

### **ABSTRACT**

Ameloblastic fibrodentinoma (AFD) is a rare benign tumor usually interpreted as a neoplasm similar to ameloblastic fibroma (AF), characterized by formation of dysplastic dentin on histopathological diagnosis. Peripheral –type ameloblastic fibro-dentinoma (PAFD) is exceedingly rare, with all previously reported cases involving young patients. We here report a case of PAFD occurring in upper gingiva of an 11-year old male child, which was surgically incised.

**Keywords:** Ameloblastic fibro-dentinoma; ameloblastic fibroma; peripheral –type ameloblastic fibrodentinoma.

## 1. INTRODUCTION

Ameloblastic fibro-dentinoma (AFD) was first described by Straith in 1936 as “a very rare neoplasm composed of odontogenic epithelium and immature connective tissue and characterized by the formation of dysplastic dentin” [1]. However in 2005, in WHO’s classification of odontogenic tumors, AFD is defined as a neoplasm similar to ameloblastic fibroma (AF) that also shows inductive changes leading to the formation of dentin [2,3].

Majority of AFD are located in posterior mandibular region in association of interrupted molar teeth, in children [4]. Besides intraosseous lesions, peripheral counterpart has occasionally been reported [5]. Here we report an additional case of PAFD, occurring in the gingiva of a young child.

## 2. CASE REPORT

An 11 year old boy presented to our clinic with a soft tissue growth in upper labial gingiva. The parents of the child explained that the growth had been slowly enlarging for about 6 months and was not associated with any trauma. The patient was otherwise in a good health and his medical and family history was non – contributory.

Examination of the oral cavity revealed the presence of 1.6 cms. Soft tissue growth, located on the labial gingiva between the upper permanent central incisors. Upon palpation, this non-ulcerated lesion was found to be firm. Furthermore, the growth was not tender and reportedly painless. Periapical radiograph of upper incisor area revealed no radiographic changes (Fig. 1).

The clinic-radiological diagnosis a benign tumor of soft tissue origin. The entire mass was then excised. At the time of excision, no bony involvement could be found. The specimen was then sent for histopathological evaluation.

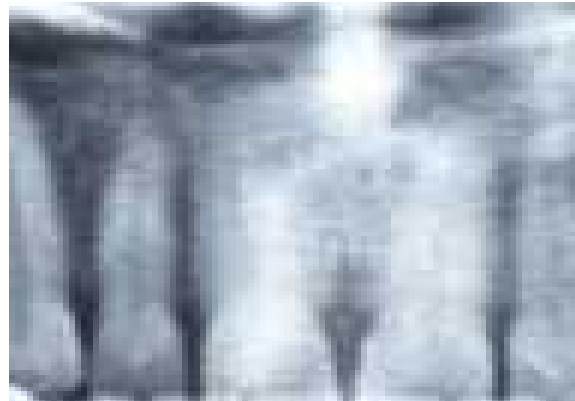
### 2.1 Histopathological Findings

Under, scanner power view a proliferating neoplastic mesenchymal tissue and odontogenic islands were appreciated (Fig. 2a).

Under low power view the odontogenic epithelium component shows strands and islands of odontogenic epithelium showing peripheral palisading appearance. The epithelium strands

lie in myxoid cell- rich stroma with stellate shaped fibroblast (Fig. 2b). Some of the areas were highly cellular showing zone of hyalinization and formation of dentoid at the periphery of odontogenic epithelial islands (Figs. 2c, 2d). Enamel formation could not be identified.

Based on above features and clinical history we reach to a final diagnosis of Peripheral –type ameloblastic fibrodentinoma (PAFD).



**Fig. 1. Periapical radiograph showing no relevant changes**

## 3. DISCUSSION

Mixed odontogenic tumors comprises of a rare group of lesions with diverse histopathological types and clinical behavior. Despite of numerous efforts, there is still considerable confusion concerning the nature and inter-relationship of these mixed odontogenic tumors and related lesions. In general, these lesions comprise AF, AFD, and ameloblastic fibro- odontoma (AFO).

AFD has been considered by some to be an intermediate stage between AF and AFO in terms of histological differentiation [6]. However the histological definition used by Riechart is as follows:

“A hamartomatous lesion similar to the ameloblastic fibroma, but also showing inductive changes that lead to formation of dentoid”.

When the histogenesis of these mixed odontogenic tumors are considered, some controversy surrounds AF, AFD, AFO and odontoma. Some regard them as chronological stages in a continuum beginning from AF at one extreme and odontoma at other extreme with AFO as well as AFD in an intermediate stage [7] Others consider them as different entities.

Clinically and pathologically, AFO and AFD are almost same. Presence of tooth germ elements i.e. enamel and dentin in combination, or only dentin in isolation, helps in differentiation of these two lesions. Because of the presence of dentin only and complete absence of enamel, even in multiple sections, the present case was considered as AFD.

PAFD presents itself as an asymptomatic slow growing lesion and is rare in existence. The

lesion in the present case was on maxillary gingival, as a growth. 2.

AFD histologically is composed of strands and islands of odontogenic epithelium, in a cell rich primitive ecto-mesenchyme resembling the dental papilla (Figs. a, b). Dentinoid or osteodentin is deposited often preceded by a zone of hyalinization.

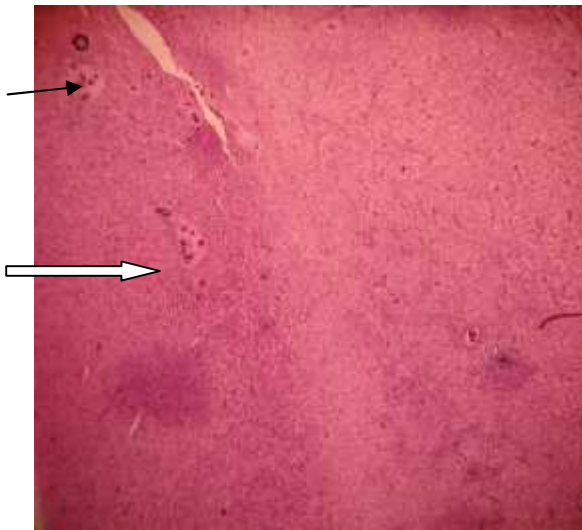


Fig. a. - Scanner view (2X)

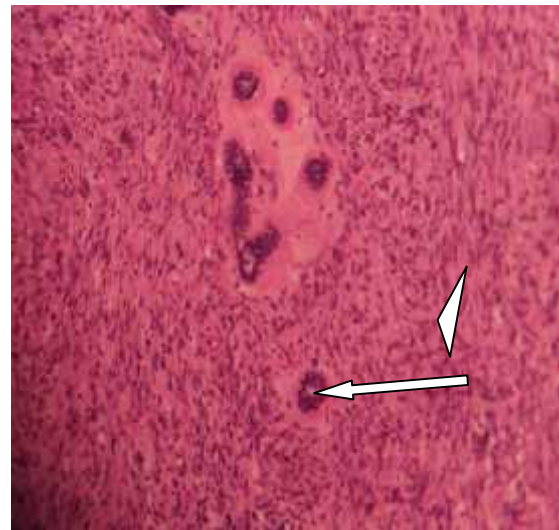


Fig. b. Low power View (10X)

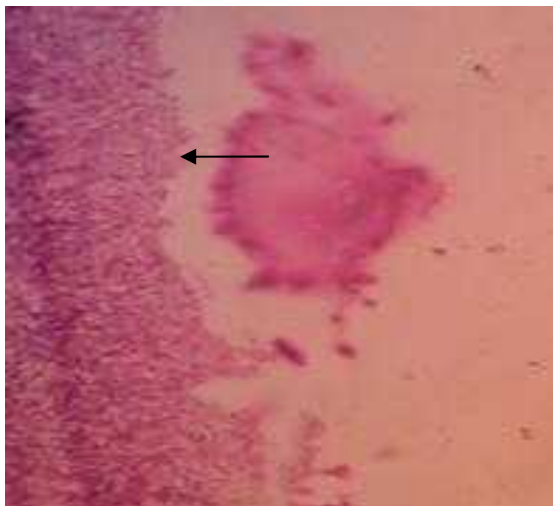


Fig. c. Low power View (10X)

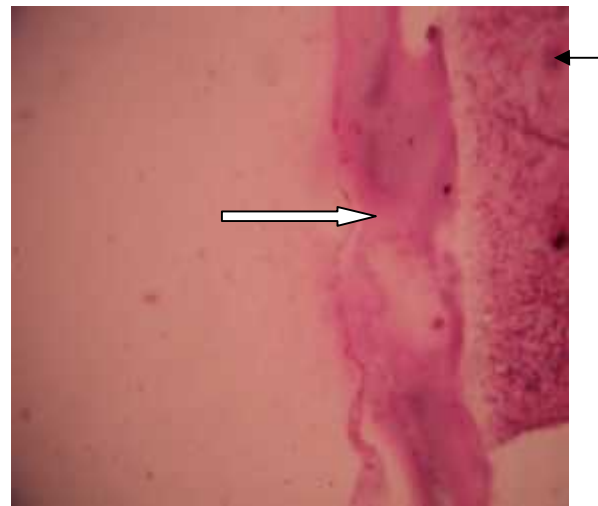


Fig. d. Low power View (10X)

**Fig. 2. (a) Proliferating neoplastic mesenchymal tissue and odontogenic islands (arrows) (2x amplification). (b) Odontogenic epithelium component shows strands (thin arrows) and islands of odontogenic epithelium showing peripheral palisading appearance (largr arrows) (10x amplification). The epithelium strands lie in myxoid cell- rich stroma with stellate shaped fibroblast (arrowheads). (c) Areas highly cellular showing zone of hyalinization (arrow) (10x amplification). (d) Formation of dentinoid at the periphery of odontogenic epithelial islands (arrow) (10x amplification)**

**Table 1. Clinical summary of the reported cases of gingival ameloblastic fibrodentinoma 8, 9, 10, 11**

Case no. (authors)	Sex	Age (year-old)	Size (cm)	Race	Location
1. Mckelvy and Cherrick [10].	Male	17	Not-stated	White	Interdental gingiva between lower left 1 <sup>st</sup> and 2 <sup>nd</sup> bicuspid
2. Godjesk et al. [9].	Male	3	0.5	White	Lingual gingival between lower deciduous left lateral incisor and cuspid
3. Chen et al. [11]	Female	2.5	0.4	Chinese	Labial gingiva between upper primary central incisors
4. Tokutaro Minamizato et al. [8]	Female	51	8 mm × 6 mm	Japan	left upper interdental papilla between the second premolar and first molar
5. Priyanka et al. (present case)	Male	11	1.6 cm	India	Interdental gingiva between upper central incisors

In some cases diagnosed as AFD, dentin matrix or dentoid tissue is an area of hyalinization around epithelial component according to some authors but others have suggested that such hyalinized zone may not represent dentin formation. In other cases, a hyalinized area containing entrapped cells has been considered to be abortive dentin or dentoid tissue [7] because of its proximity to the odontogenic epithelium (Figs. c, d). [12].

In this case definitive odontogenic epithelium was seen in cellular connective tissue stroma. The connective tissue in this case appeared relatively immature and a diagnosis of PAFD was made, as the presence of enamel like material could not be seen.

To the best of our knowledge 4 cases have been reported until 2014 Table 1 above.

Another diagnosis to be considered in our case would be peripheral odontogenic fibroma (POF). Odontogenic fibroma (OF) consists of connective tissue derived from odontogenic ectomesenchymal cells. The connective tissue of OF shows relatively mature fibrous, fibromyxoid and myxoid appearances with low cell density.5 The presence of immature dense cellular fibrous tissue and dysplastic dentin in our case is not consistent with a typical OF.

#### 4. CONCLUSION

We have described a rare case of peripheral-type ameloblastic fibro-dentinoma, in an 11 year

old male child patient, in which histological picture resembles ameloblastic fibroma except for presence of dentin. Thus the identification and careful examination in this lead us to the diagnosis of PAFD which has low recurrence rate when compared to AF and POF.

#### CONSENT

Consent was obtained from patients parents before writing the script.

#### ETHICAL APPROVAL

It is not applicable.

#### COMPETING INTERESTS

Authors have declared that no competing interests exist.

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