Peripheral ameloblastic fibrodentinoma in a 3-year-old boy: Report of an unusual and rare case

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Case Report

INTRODUCTION

Mixed odontogenic tumors are rare in occurrence and are classified based on their differentiation. Most lesions occur in young individuals and are intraosseous. Peripheral odontogenic tumors are relatively rare than their central counterpart. Odontogenic fibroma and ameloblastoma seem to be the most common among peripheral odontogenic tumors.

The first case of ameloblastic fibrodentinoma (AFD) was reported and 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] According to the World Health Organization (WHO) classification, this tumor is termed as “AFD”[2] and considered to be the hard tissue variant of ameloblastic fibroma. The WHO defines ameloblastic fibroma consisting of odontogenic ectomesenchyme resembling the dental papilla and epithelial strands and nests resembling dental lamina and enamel organ. No dental hard tissues are present. AFD represents a variant of ameloblastic fibroma with the presence of dentin.[3]

Besides intraosseous lesions, a peripheral counterpart has been very rarely reported.[4] It represents itself as an asymptomatic slow-growing lesion.

CASE REPORT

A 3-year-old male child reported with a chief complaint of reddened soft tissue growth of the upper labial gingiva of
left central incisor. His medical history was noncontributory and the boy had normal milestones. A history of trauma 5 months ago causing palatal tooth displacement and painful swelling on the labial attached gingiva with pus formation treated by antibiotics was recorded. Two months later, a local firm, tender, nonfluctuant and inflamed soft tissue gingival growth was observed which increased progressively.

Provisional diagnosis of chronic irreversible pulpitis with respect to 61 and gingival fibroma was made [Figure 1a].

Though pulpectomy was initiated considering poor prognosis of the tooth, extraction was planned eventually [Figure 1b]. Periapical radiograph showed no intrabony involvement [Figure 1c]. The tooth bud of the permanent maxillary central incisor was evident and unaffected.

Following 1 month of postextraction follow-up, the patient presented with a mild decrease in inflammation with insignificant reduction in size of mass initially, which increased in size 1 month later. Excision of the mass was planned with parental consent. After hematological examination, excision of the mass was performed under local anesthesia and sent for histopathological evaluation.

Two days postexcision, excised site was asymptomatic with signs of healing [Figure 1d]. Radiographs showed proper alignment of the permanent tooth bud. As loss of space expected was minimal and parents not consenting, space maintainer was not initiated. The patient was kept under regular follow-up. Nine months postexcision, follow-up reveals no recurrence or symptoms clinically and radiographically [Figure 1e].

Histopathological findings
The hematoxylin and eosin-stained section showed surface hyperplastic parakeratinized stratified squamous epithelium with underlying fibrovascular connective tissue stroma [Figure 2a]. The connective tissue stroma showed numerous ameloblastic follicles [Figure 2a]. The follicles were lined by tall columnar ameloblast-like cells at the periphery with apically placed nucleus [Figure 2b]. Stellate reticulum-like cells were also seen adjoining ameloblastic cells at the center of the follicle [Figure 2b]. Abundant cosinophilic calcified areas resembling dentinoid were seen in between the follicles [Figure 2c and d]. The features were indicative of a peripheral ameloblastic fibrodentinoma (PAFD). The tissue section was tested for cytokeratin 19 (CK19) (319M-1 Sigma-Aldrich®) [Figure 3a], following the standard immunohistochemical protocol for confirmation of the odontogenic origin.[9] CK19 was found to be expressed in all the odontogenic epithelial areas [Figure 3b] and especially among those associated with the dysplastic dentinoid matrix [Figure 3c]. The dysplastic dentinoid matrix was not marked by CK19 [Figure 3d]. A final diagnosis of PAFD was returned.

DISCUSSION
The WHO lists AFD as a mixed odontogenic tumor and describes it as a neoplasm similar to the ameloblastic fibroma, showing inductive changes leading to the

Figure 1: Clinical and radiographic images. (a) Preoperative clinical image (before tooth extraction). (b) Preoperative clinical image (after tooth extraction). (c) Preoperative radiographic image. (d) Postoperative clinical image. (e) Postoperative radiographic image.

Figure 2: Composite photomicrographs. (a) Lesion with superficial epithelium and connective tissue stroma with numerous ameloblastic follicles (×4 OM*). (b) Follicle showing ameloblast-like cells, stellate reticulum and dentinoid tissue (×10 OM*). (c) Numerous ameloblastic follicles seen with interspersed dentinoid-like material (×10 OM*). (d) Dentinoid secreted by odontoblast (×40 OM*). *Original magnification
formation of dentine. The occurrence of the latter is indicative of the high rate of differentiation of the lesions.

The present case in discussion had an interesting course. The history of trauma and the displacement of the teeth were most probably the inciting factors. In an area rich with odontogenic tissue (developing tooth follicles), aberrancy in the normal development of tooth structures could be expected as a result of the trauma. Availability of odontogenic tissue in the gingival area for the development of odontogenic lesions is limited. The possibility of odontogenic tissue breach from the central to peripheral of the maxilla can be ruled out due to lack of bony involvement. Unknown causes speeded up by trauma seem to be the likely cause.

The differential diagnoses include peripheral odontogenic fibroma, peripheral ameloblastic fibroma (PAF), PAFD and peripheral ossifying fibroma.

AFD radiographically appears as a well-circumscribed unicocular or rarely multicellular radiolucent defect that contains a variable amount of calcified material with radiodensity of tooth structure. However, in this case, there was no intrabony involvement; hence, differential diagnosis of AFD was ruled out.

Peripheral odontogenic fibroma presenting as a firm, slow-growing and sessile gingival mass covered by normal-appearing mucosa usually occurs on facial gingiva of the mandible causing teeth displacement. Histopathology shows fascicles of cellular fibrous connective tissue interspersed with the areas of less cellular, fibrous, or myxoid connective tissue. However, this lesion does not show odontogenic follicles with dentinoid-like cells similar to the present lesion.

Although PAF has similar histopathological features as of PAFD, there is no enamel or dentin formation.

Some researchers believe that PAFD is only a stage in development of an odontoma while others believe that it is a separate entity exhibiting progressive growth causing bone destruction. Some investigators suggested that ameloblastic fibroma represents immature complex odontoma, and if left undisturbed, it will ultimately differentiate or mature into a lesion known as ameloblastic fibro-odontoma and ultimately continue maturation into completely differentiated odontoma. Eversole et al. proposed that mixed odontogenic tumors are totally dependent upon the presence of differentiation factors.

Peripheral ossifying fibroma can occur at any age although appearing more commonly in children and young adults. Clinically, it represents as well-demarcated focal mass on gingiva, sessile, or pedunculated base having same color as gingiva. Histopathology shows stratified squamous epithelium with bulk made of exceedingly cellular mass of connective tissue comprising large numbers of plump proliferating fibroblasts intermingled throughout a very delicate fibrillar stroma. Several forms of calcifications occur in the form of single or multiple interconnecting trabaculae of bone or osteoid. There may be islands of odontogenic cells but no differentiated ameloblastic tissue.

The present case merits mention because of two interesting features. The peripheral location and the maxillary presentation are relatively rare. A review of literature reveals only five cases hitherto reported until 2015. Table 1 lists the details of the cases reported.

There seems to be no correlating features in the occurrences of these lesions, and there is widespread diversity in the presentations. The ages of occurrence range from 2.5 years to 51 years. There is no sex predilection and lesions seem to occur randomly in both the jaws.

![Figure 3: Composite photomicrograph of immunohistochemistry with cytokeratin 19. (a) Scanner view. (b) Positivity of cytokeratin 19 in epithelial islands (×10 OM*). (c) Cytokeratin 19-positive follicles (×40 OM*). (d) Dentinoid not marked by cytokeratin 19 (×40 OM*). *Original magnification](image)

| Authors          | Age (years) | Sex  | Location                                           |
|------------------|-------------|------|---------------------------------------------------|
| McKelvy and Cherrick | 17         | Male | Interdental gingiva between lower left 1st and 2nd premolars |
| Godjesk et al.   | 3          | Male | Lingual gingival between lower deciduous left lateral incisor and canine |
| Chen et al.      | 2.5        | Female | Labial gingiva between upper primary central incisors |
| Minamizato       | 51         | Female | Interdental papilla of left upper 2nd premolar and 1st molar |
| Priyanka et al.  | 11         | Male | Interdental gingiva between upper central incisors |
| Present case     | 3          | Male | Labial gingiva of maxillary left central incisor |
Studies suggest that conservative excision of the tumor with minimal but adequate margins is the treatment of choice. PAFD is rare, with no published study that estimates its recurrence rate after its surgical excision.\[10\] Hence, conservative surgery with enucleation was performed and patient is kept on regular follow-ups as it appears that the PAFD should be initially treated by conservative surgical therapy.

**Declaration of patient consent**
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**
There are no conflicts of interest.

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