Case report of an early diagnosed ameloblastic fibroma

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Abstract

Ameloblastic fibroma is a relatively rare benign odontogenic tumor with two neoplastic components; both epithelial and ectomesenchymal. A 10-year-old girl was referred to a dentist for routine dental treatments for evaluation of the distance between mandibular right first permanent molar and mandibular right second deciduous molar and it was associated with an undesirable backward movement of erupted mandibular right first permanent molar and forward movement of mandibular right second deciduous molar. The lesion was enucleated and the material was sent for histopathologic examination. Microscopically, it was composed of neoplastic proliferation of odontogenic epithelium consisted of cords and islands in a cell-rich mesenchymal stroma, with histopathological diagnosis of ameloblastic fibroma. After 24 months of follow-up, no recurrence was observed and the mandibular right first molar was spontaneously aligned. Early diagnosis and proper management will prevent comprehensive orthodontic treatment.

Keywords: Dental tissue neoplasms, Mandibular neoplasms, Odontogenic tumors

Ameloblastic fibroma (AF) is defined as a benign, rare odontogenic tumor with two histologic components that results in classifying the tumor in true mixed odontogenic tumors.\textsuperscript{[1,2]} It composes 1.5-4.5% of all odontogenic tumors according to the literature.\textsuperscript{[1-6]} AF has been reported in a wide age group from 7 weeks to 57 years\textsuperscript{[7]}, but basically it is considered as a tumor of childhood and adolescence that occurs mostly in the first two decades.\textsuperscript{[6,8,9]} In the majority of cases, the lesion is seen in the mandible with the presentation of a slow-growing, painless swelling and/or failure of tooth eruption.\textsuperscript{[8,9]} However, the tooth impaction and delayed eruption may be due to dental developmental defects or abnormalities such as amelogenesis imperfecta.\textsuperscript{[10]} In some cases; however, the tumors are asymptomatic and are noticed during a routine oral/radiographic examination.\textsuperscript{[9]} Radiographic view of AF is a well-defined unilocular or multilocular radiolucency.\textsuperscript{[5,7]} It has been reported that unilocular radiolucencies usually appear as asymptomatic cases, whereas multilocular cases are associated with jaw swelling.\textsuperscript{[11]} The exact pathogenesis is not clear. The tall columnar ameloblast-like cells in the epithelial component are too primitive to induce the cells of the ectomesenchyme, and only little is known about their interactions. It is also unknown why the induction of odontoblastic differentiation is lacking in AF. Immunohistochemical analysis shows positive staining of odontogenic epithelium for cytokeratin, mesenchymal tissue around the dental lamina-like epithelium for tenascin, focal areas of immature dental papilla-like cells, and basement membrane of the epithelium for vimentin. These findings suggest that AF develops at an early stage of tooth formation.\textsuperscript{[12,13]} Mitotic figures may be a part of the histopathologic view in AF. In the case of presence of a large number of mitosis and especially atypical mitosis, malignant entities, like Ameloblastic fibrosarcoma should be considered as differential diagnosis.\textsuperscript{[1]} The best appropriate treatment for AF still remains uncertain. Several authors have suggested a conservative approach.\textsuperscript{[1]} However, recurrence following surgical removal and progress to malignancy is probable to occur.\textsuperscript{[7,14]} Recurrence is usually thought to be related to incomplete removal and remaining of the residual tumor.\textsuperscript{[6]} Hence, long-term follow-up is recommended.\textsuperscript{[15]} The aim of this study was to report an interesting early diagnosed case of AF which affected mandible of a young girl and was associated with an undesirable movement of erupted mandibular right first permanent molar and mandibular right second deciduous molar. Early diagnosis and proper management will prevent comprehensive orthodontic treatment.
Case report

A 10-year-old Iranian girl was referred to the dentistry department with the chief complaint of failed eruption of the right permanent mandibular first molar. Physical examination showed a generally healthy child and the medical, surgical, familial, and social histories were unremarkable. Intraoral examination revealed no expansion of the buccal or lingual cortical mandibular plate. The overlying mucosa was intact and normal in color and consistency. Radiographic examination by means of a panoramic radiography (Fig. 1-A) indicated a well-defined multilocular radiolucent lesion in the right mandible. The second deciduous molar was displaced forward and the first permanent molar was displaced backward. But the periodontal ligament of both teeth was intact. The periapical radiography of the area showed the same mixed radiolucent lesion (Fig. 1-B).

Based on clinical and radiographic findings, a presumptive preoperative diagnosis of ameloblastoma or odontogenic cyst was made. Under local anesthesia, the lesion was totally removed through an intraoral approach (Fig. 1-C). The enucleated material was sent to the Oral & Maxillofacial Pathology Department of the Dentistry School of Isfahan University of Medical Science for histological diagnosis. Microscopically, neoplastic proliferation of odontogenic epithelium consisted of cords and islands was seen (Fig. 2-A). Histopathologic view revealed double layer of cuboidal or columnar cells in a cell-rich mesenchymal stroma with plump stellate and ovoid cells and little collagen closely resembling the primitive dental papilla (Fig. 2-B). Evidence of hard tissue formation in the sample was not observed, so the histopathological diagnosis was considered as ameloblastic fibroma. After 24-month follow-up, no recurrence was observed. The first molar was spontaneously aligned and the patient won’t need any comprehensive orthodontic treatment (Fig. 3).
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Fig 2-B. double layer of cuboidal or columnar cells 
was seen in a cell-rich mesenchymal stroma with 
plump stellate and ovoid cells and little collagens 
closely resembling the primitive dental papilla. The 
histopathological diagnosis was considered as 
ameloblastic fibroma.

Fig 3. After 24-month follow-up, no recurrence was 
observed. The first molar was spontaneously aligned 
and the patient won’t need any comprehensive 
orthodontic treatment

The recommendations of Helsinki declaration were 
theroughly maintained during this study. Ethical 
approval of this article was confirmed by the ethics 
committee of Isfahan University of Medical sciences.

Discussion

Ameloblastic Fibroma (AF) was first described in 
1891 by Kruse. Up to now, 123 patients have been 
reported in the English literature.[16] It is defined by 
WHO as “consists of odontogenic ectomesenchyme 
resembling the dental papilla and epithelial strands and 
nests resembling dental lamina and enamel organ. No 
dental hard tissues are present.”[6] Tomich was the first 
to classify this tumor as a separate entity.[17] It is a 
mixed tumor with both epithelial and mesenchymal 
components.[1]

In this case, an AF has been reported in a 10-year-
old girl at the posterior of mandible. In line with our 
case, the most common age for AF is the first and 
second decades in the literature although it has the 
potential to be diagnosed in 7-week infants to 57-year-
old patients. Unlike this case which has occurred in a 
girl, a slight higher prevalence has been reported in 
males.[5,9,11] Along with this case, the majority of cases 
has been reported in mandible with a predilection for 
the posterior regions.[6] Still, few cases of the AF in the 
maxilla have been reported.[8]

Swelling of the jaw is the most common finding 
which is occasionally associated with pain, tenderness, 
drainage and ulceration.[6] While none of these common 
features was observed in this patient, the only finding 
was displacement of the teeth which is a rare 
accompanied feature. Since it is benign and slow-
growing, it can also be frequently discovered in a 
routine dental and radiographic examination.[1,8] 
According to the literature review, radiographic view 
might be either unilocular or multilocular with well-
defined borders as in the present case in which a 
multilocular radiolucency was noticed in the panoramic 
view.[6] The differential diagnosis is dentigerous cysts, 
ameloblastoma, odontogenic keratocysts and 
ameloblastic fibrosarcoma according to the 
radiographic view.[8,14] When the internal pattern of the 
lesion is a mixed radiolucent-radiopaque then calcifying 
odontogenic tumor is one of the possible differential 
diagnosis.[12]

Previously, these lesions were categorized as a 
spectrum of a single entity with ameloblastic fibromas, 
the least differentiated of the tumors, maturing and 
developing into ameloblastic fibro-odontomas and later 
odontomas. However, nowadays, it is considered as an 
independent entity with its special characteristics. They 
can be differentiated according to the demographic 
features of the patient.

The least differentiated lesion, ameloblastic fibroma, 
actually occurs, on average, at an older age then the 
more differentiated ameloblastic fibro-odontoma and 
odontoma. In addition, Ameloblastic fibro-odontomas 
and odontomas have a different nature and are 
categorized as hamartomas and unlike the ameloblastic 
fibroma have little chance of recurrence or susceptibility 
to malignant transformation.[18] Therefore, despite the
common apparent features, these lesions should be
differentiated and treated differently. Due to the true
neoplastic nature of AF, the treatment plan differs from
the other lesions which are considered mainly as
hamartomas.[19]

Regarding the treatment procedure of this tumor,
there are some controversies. Philipsen et al.[13]enucleated AF and afterwards careful follow-up of
the lesion was performed. Recurrence of the AF may
not be a true one rather a residual lesion as a result of
incomplete enucleation. Some authors believe that
aggressive treatment is not reasonable.[3,7, 15] However,
Chen et al. [7] reported that recurrence is more common
among patients treated with a more conservative
approach. Those authors found recurrence in 14 out of
41 cases.

Dallera et al. [3], on the other hand, reported no
recurrence for 6 cases with an average follow up of 15
years. Therefore, it is necessary to follow a step-wise
treatment principle when the diagnosis of AF has been
made. There is a great emphasis on the patient’s age
during treatment planning. It is recommended to treat
patients under 22 with conservative surgeries; however,a radical surgery should be considered for
patients older than 22.[6] Consistent with data from other
studies, the treatment plan applied for the patient in this
case was enucleation and no recurrence was observed
after 24 months follow up.

Although it is uncommon, the possibility of
malignant transformation of AF into Ameloblastic
fibrosarcoma has been mentioned by Chen et al. [7] and
Kobayashi et al. [14]. According to their reports, AF
showed no signs of malignancy until the second
recurrence. Chen et al. [7] during two 5-year and 10-year
follow-ups reported transformation to malignancy10.2%
and 22.2%, respectively. However, Kousar et al. have
presented [15] a rapid sarcomatous transformation of one
case of AF within 6 months.

In the case reported here, the main feature was
mandibular first molar failure of eruption. This
disturbance may or may not be associated with
pathology, such as AF. Thus, this case highlights the
importance of careful differential diagnosis and early
detection, while it reports a rare lesion. Tooth
placement and delayed eruption were the only signs of
the lesion. These findings are not common
accompanied features of ameloblastic fibroma. Note the
early diagnosis of the lesion leads to early treatment
with no recurrence.

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