

Original Article - Evaluative Study

# Large ameloblastic fibro-odontoma in a 7-year-old girl with analysis of 108 cases

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## ABSTRACT

**Background:** Odontogenic tumors such as ameloblastic fibro-odontoma (AFO) are rare conditions in children and are often asymptomatic. AFOs are found by routine clinical and radiological examination or when they cause obvious intra- or extra-oral swelling. **Materials and Methods:** A case of an AFO in a 7-year-old girl is described, and 107 cases from the literature and this report are analyzed. **Results:** The total of 108 cases revealed the average age at presentation of AFO to be 6.3 years in boys and 9.6 years in girls. There was a slight male predilection and AFO lesions most often occurred in the posterior mandible. AFO was almost always associated with an unerupted tooth or teeth. **Conclusions:** While the recurrence rate of AFO was found to be 5.5%, long-term postoperative clinical and radiological follow-up is advised to ensure no future signs of aggressive recurrence.

**Keywords:** Ameloblastic fibro-odontoma, benign tumors, mixed odontogenic tumors

## INTRODUCTION

Ameloblastic fibro-odontoma (AFO) is one of the mixed radiolucent and radiopaque odontogenic tumors. It is relatively rare. Studies indicate that AFO is seen ranging from 0.3% to 1.7% of oral pathology biopsy specimens submitted as possible odontogenic tumors.<sup>[1,2]</sup> AFO was originally termed ameloblastic odontoma before the current nomenclature. Only a few studies exist regarding the characteristics of AFOs as most case reports take the form of single patient publications.<sup>[3]</sup> This study aimed to review cases of AFO reported consecutively in the literature between the years 1967 and 2015 with the addition of a new case.

### Case report

An otherwise healthy 7-year-old girl was referred by her dentist because of swelling of the left side of the mandible. An asymmetric facial swelling was obvious on extraoral examination [Figure 1]. There was no history of trauma or local infection. Intraoral examination revealed normal mucosa overlying the lesion and the absence of the left mandibular first and second molar

teeth [Figure 2]. A distinctive hard bulge was palpable in the vestibule of the left mandible molar region. The mouth opening was normal and there was no pain or other symptoms apart from the swelling. Panoramic radiography [Figure 3] and cone-beam computed tomography scan [Figures 4 and 5] both revealed a large well-defined radiolucent area that contained several radiopaque structures of varying sizes and shapes. The lesion extended from the first molar to the coronoid process and the condylar neck. The borders of the lesion were well circumscribed and the lesion had displaced the developing second molar tooth bud down to the inferior border of the mandible.

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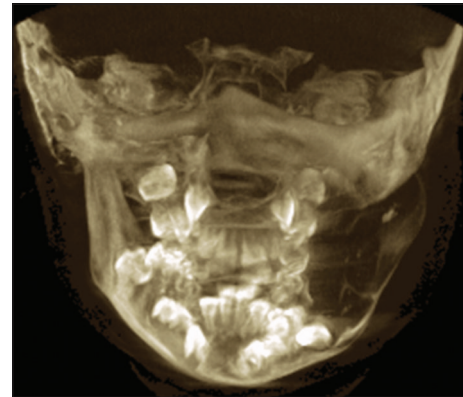
**Figure 1:** Extraoral swelling of the left side of the mandible



**Figure 2:** Missing or absent molars of the left side of the mandible



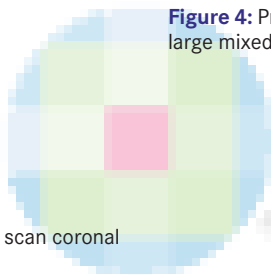
**Figure 3:** Preoperative panoramic radiograph showing large mixed radiolucent and radiopaque lesion of the left ramus and body of mandible



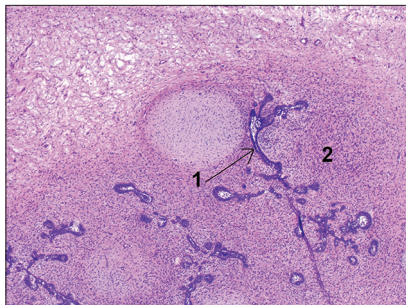
**Figure 4:** Preoperative cone-beam computed tomography scan revealing large mixed lesion of the left ramus and body of mandible



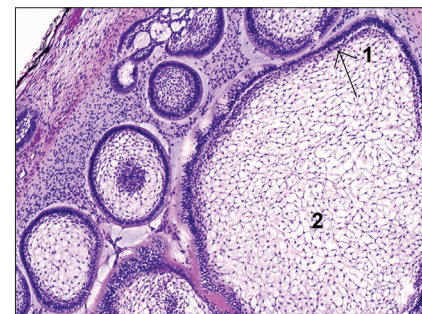
**Figure 5:** Preoperative cone-beam computed tomography scan coronal view of mixed lesion



**Figure 6:** Resected specimen delivered using an intraoral approach



**Figure 7:** Ameloblastic fibro-odontoma with odontogenic epithelium (1) and odontogenic stroma (2) (H and E,  $\times 20$ )



**Figure 8:** Ameloblastic fibro-odontoma exhibiting areas of odontogenic epithelium (1) and (2) (H and E,  $\times 40$ )



**Figure 9:** Panoramic radiograph taken 6 months following surgery with healing of intraosseous lesion of the left ramus and body of mandible

The lesion was enucleated using an intraoral approach under general anesthesia. During the same procedure, tooth numbers 75 and 37 were also extracted. The tumor measured 10 cm  $\times$  4 cm  $\times$  4 cm in size [Figure 6] and was submitted for histopathological examination. The samples were stained with hematoxylin and eosin. A large number of epithelial strands and islands of odontogenic epithelium

**Table 1: Summary of reported cases of ameloblastic fibro-odontoma**

First author	Year of publication	Number of cases	Osseous location	Area	Unerupted teeth (cases)	Sex	Average age	Recurrence reported
Kirjavainen	2015	1	Mand=1	Mol	Yes	Female=1	Female=7	No
Gantala	2015 <sup>[2]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=11	No
Surej Kumar	2014 <sup>[4]</sup>	1	Max=1	Mol	Yes	Female=1	Female=10	No
Banihashem Rad	2014 <sup>[5]</sup>	1	Max=1	Mol	Yes	Female=1	Female=11	No
Lin	2014 <sup>[6]</sup>	1	Max=1	Inc	No	Female=1	Female=3	No
Sreenath	2014 <sup>[7]</sup>	1	Mand=1	Inc	Yes	Female=1	Female=5	No
Nelson	2014 <sup>[8]</sup>	1	Max=1	Mol	Yes	Female=1	Female=15	No
Buchner	2013 <sup>[3]</sup>	10	Max=4, Mand=6	Incisor=1, Premol=1, Mol=5, Multi=3	Yes=9, No=1	Female=4, male=6	Female=14, male=15	U=10
de Souza Tolentino	2010 <sup>[9]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=11	No
Wewel	2010 <sup>[10]</sup>	1	Mand=1	Inc	Yes	Male=1	Male=2	No
Cavalcante	2009 <sup>[11]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=8	No
De Riu	2009 <sup>[12]</sup>	1	Max=1	Mol	Yes	Male=1	Male=26	No
Nascimento	2009 <sup>[13]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=11	No
Zouhary	2008 <sup>[14]</sup>	1	Max=1	Multi	Yes	Female=1	Female=7	No
Hegde	2008 <sup>[15]</sup>	1	Max=1	Multi	Yes	Female=1	Female=12	U
Dolanmaz	2008 <sup>[16]</sup>	1	Max=1	Multi	Yes	Female=1	Female=9	No
Reis	2007 <sup>[17]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=6	No
Gyulai-Gaál	2007 <sup>[18]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=9	No
Chang	2007 <sup>[19]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=16	U
Oghli	2007 <sup>[20]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=3,5	No
Silva	2006 <sup>[21]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=14	No
Hu	2006 <sup>[22]</sup>	1	Max=1	Multi	Yes	Female=1	Female=17	No
Soares	2006 <sup>[23]</sup>	1	Max=1	Multi	Yes	Male=1	Male=4	U
Chen	2005 <sup>[24]</sup>	7	Max=2, Mand=5	Inc=1, Mol=4, Multi=2	Yes	Female=3, male=4	Male=10, female=10	Yes=3, no=2, U=2
Sivapathasundharam	2005 <sup>[25]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=17	U
Reichart	2004 <sup>[26]</sup>	2	Mand=2	Mol=2	Yes, No	Male=2	Male=10	No=1, U=1
Dhanuthai	2004 <sup>[27]</sup>	1	Max=1	Inc	Yes	Female=1	Female=1	No
Alderson	2004 <sup>[28]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=11	U
Chang	2002 <sup>[29]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=26	No
Mosqueda-Taylor	2002 <sup>[30]</sup>	3	Max=2, Mand=1	Multi=3	Yes	Female=1, male=2	Female=25, male=12	U=1, No=2
Friedrich	2001 <sup>[31]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=8	U
Yagishita	2001 <sup>[32]</sup>	1	Mand=1	Multi	No	Male=1	Male=24	U
al-Sebaei	2001 <sup>[33]</sup>	1	Max=1	Multi	Yes	Male=1	Male=11	U
Flaitz	2001 <sup>[34]</sup>	1	Mand=1	Inc	Yes	Male=1	Male=2,5	U
Steinberg	2001 <sup>[35]</sup>	1	Max=1	Multi	Yes	Male=1	Male=6	U
Rao	1999 <sup>[36]</sup>	2	Mand=2	Multi	Yes	Male=2	Male=10,5	U=2
Furst	1999 <sup>[37]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=7	Yes
Savitha	1998 <sup>[38]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=5	No
Favia	1997 <sup>[39]</sup>	2	Max=1, Mand=1	Multi=2	U	Male=2	Male=3,5	No=2
Ozer	1997 <sup>[40]</sup>	1	Max=1	Multi	Yes	Female=1	Female=7	No
Kitano	1994 <sup>[41]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=9	No
Baker	1993 <sup>[42]</sup>	1	Max=1	Inc	Yes	Male=1	Male=0,75	No
Okura	1992 <sup>[43]</sup>	2	Mand=1, Max=1	Mol=2	Yes	Male=1, female=1	Male=7, female=12	No=2
Piette	1990 <sup>[44]</sup>	1	Max=1	Multi	Yes	Male=1	Male=7	U
Glickman	1989 <sup>[45]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=15	U
Warnock	1989 <sup>[46]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=11	U
Hansen	1988 <sup>[47]</sup>	8	Mand=4, Max=4	U=8	U	Male=6, female=2	Male=11,5, female=8	No=4, U=4
Takeda	1988 <sup>[48]</sup>	1	Max=1	Mol	Yes	Female=1	Female=11	No
Hawkins	1986 <sup>[49]</sup>	1	Max=1	Mol	Yes	Female=1	Female=6,5	U
Sole	1986 <sup>[50]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=12	U
Reich	1984 <sup>[51]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=11	No
Anneroth	1982 <sup>[52]</sup>	1	Max=1	Inc	Yes	Male=1	Male=7	No
Hutt	1982 <sup>[53]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=11	No
Daley	1982 <sup>[54]</sup>	1	Max=1	Mol	Yes	Female=1	Female=4	U
Curran	1980 <sup>[55]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=15	No
Bernhoft	1979 <sup>[56]</sup>	1	Max=1	Mol	Yes	Female=1	Female=14	No
Geller	1978 <sup>[57]</sup>	1	Max=1	Mol	Yes	Male=1	Female=15	No
Howell	1977 <sup>[58]</sup>	2	Mand=2	Multi=2	U	Male=1, female=1	Male=36, female=18	Yes=2
Album	1977 <sup>[59]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=5	No

Contd...

**Table 1: Contd...**

First author	Year of publication	Number of cases	Osseous location	Area	Unerupted teeth (cases)	Sex	Average age	Recurrence reported
Herd	1977 <sup>[60]</sup>	1	Mand=1	Mol	Yes	Female=1	Female=11	No
Eda	1977 <sup>[61]</sup>	1	Max=1	Multi	Yes	Female=1	Female=22	No
Cran	1976 <sup>[62]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=9	U
Hanna	1976 <sup>[63]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=11	No
Miller	1976 <sup>[64]</sup>	7	Mand=4, Max=3	Mol=4, Multi=3	Yes=4, U=3	Male=5, female=2	Male=11, female=12	No=7
Meerkotter	1974 <sup>[65]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=9	No
Sanders	1974 <sup>[66]</sup>	1	Mand=1	Mol	Yes	Male=1	Male=19	No
Worley	1972 <sup>[67]</sup>	1	Mand=1	Multi	Yes	Male=1	Male=11	U
O`Brien	1971 <sup>[68]</sup>	1	Max=1	U	U	Female=1	Female=2,5	U
Dutta	1970 <sup>[69]</sup>	1	Max=1	Multi	Yes	Male=1	Male=8	No
Jacobsohn	1968 <sup>[70]</sup>	1	Mand=1	Premol	No	Male=1	Male=15	No
Hammer	1968 <sup>[71]</sup>	2	Mand=2	Mol=2	Yes=2	Male=2	Male=7	No=2
Olech	1967 <sup>[72]</sup>	1	Max=1	Inc	Yes	Female=1	Female=5	No
Total		108	Max=43, Mand=65	Inc=10, Premol=2, Mol=49, Multi=38, U=9	Yes=96, no=5, U=7	Male=66, female=42	Female=9,6, male=6,3	No=62, Yes=6, U=40

Max=Maxilla; Mand=Mandible; Inc=Incisor; Premol=Premolar; Mol=Molar; Multi=Multiple sites; U=Unknown. Numerical superscript next to year of publication indicates citation number of report in references section of manuscript

**Table 2: Characteristics of 108 ameloblastic fibro-odontoma lesions**

Characteristic	Ratio
Female to male ratio	1:1,65
Average age (years)	Female: 9.6 Male: 6.3
Mandible versus maxilla (%)	65 (59.43) versus 43 (40.57)
Molar region (%)	Molar: 47 (44.34) Molar + multiple sites: 85 (80.19)
Unerupted teeth (%)	94 (88.68)
Recurrence (%)	6 (5.57)

the average age at presentation being 6.3 years in males and 9.6 years in females. AFO had a female: male ratio of 1:1.62. Typically, AFO lesions were located in the posterior region of the mandible in 60% of cases. Almost all lesions, 94 of 106 (89%), were associated with the crown of an unerupted tooth or teeth. Clinically, AFO was usually painless and was characterized by asymmetric swelling of the face and delayed tooth eruption. Radiological features of AFO included a cyst-like lesion with radiopaque areas resembling a developing odontoma. The recurrence rate in this review of 108 cases was only 6 recurrent cases or 5.5% [Tables 1 and 2].

were found. The hard tissue component consisted of dentin and enamel. Histopathological examination [Figures 7 and 8] confirmed the clinical and radiological diagnosis of AFO. Clinical and radiological follow-up [Figure 9] was continued over 24 months and the postoperative course was uneventful with no recurrences to date.

## MATERIALS AND METHODS

A search was conducted using Medline and Scopus databases using the search words AFO and odontogenic tumors. This yielded a total of 1254 hits which included many lesions other than AFO, such as ameloblastic fibroma (AF), ameloblastic fibrodentinoma, ameloblastic fibrosarcoma, and odontoma. The search was restricted to AFO and reports including the other diagnoses were removed. The criteria for inclusion in this review were reports describing radiographic features consistent with AFO, histologic features consistent with AFO, and minimum follow-up of 6 months following removal of the lesion. Any report or study failing to mention any of the above inclusion criteria was excluded. This yielded a total of 107 cases which together with the newly reported case of the 7-year-old girl comprising 108 cases forming the database of this review.

## RESULTS

A summary of the findings of AFO reports is presented in Table 1. AFO was found to be a benign tumor of children with

## DISCUSSION

In defining AFO, the World Health Organization lists it as a rare odontogenic tumor with the histologic features of AF in conjunction with the presence of dentin and enamel.<sup>[73]</sup> Histopathologically, the tumor is composed of both soft and hard tissues. The soft tissue component is composed of epithelial strands and small islands of odontogenic epithelium associated with a primitive appearing myxoid connective tissue which resembles the dental papilla [Figures 7 and 8]. The hard tissue component consists of foci of enamel and dentin.<sup>[74]</sup>

Studies have shown that AFO is usually not aggressive and can be treated surgically without removal of the adjacent teeth.<sup>[43,68,71,70,75]</sup> A few studies suggest that AFO can behave aggressively with multiple recurrences.<sup>[24,76]</sup> There had been suggestions that a primarily benign AFO can undergo malignant transformation to ameloblastic fibrosarcoma.<sup>[58,77-79]</sup> Therefore, it is important to continue the follow-up of AFO lesions for several years.

## CONCLUSIONS

AFO is a rare benign mixed odontogenic tumor. Normally, AFO is found at the average age of 9 years in females and 6 years in males. The prognosis is good. The correct diagnosis is based on clinical, radiological, and histopathological findings. AF and complex odontoma must be considered in the differential



diagnosis. The treatment of choice is to remove tumor surgically usually by enucleation.

In this newly reported case, the AFO tumor was enucleated and the area of the left posterior mandible healed uneventfully without complications. Clinical and radiological follow-up has continued for 24 months with no signs of recurrence. Follow-up must be continued on a regular basis for several years. The patient in this case report is now receiving orthodontic treatment and remains under vigilant surgical follow-up.

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#### Conflicts of interest

There are no conflicts of interest.

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