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Ameloblastic fibrodentinoma and ameloblastic fibro-odontoma: an updated systematic review of cases reported in the literature

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ABSTRACT

Purpose: To integrate the available data published on ameloblastic fibrodentinoma (AFD) and ameloblastic fibro-odontoma (AFO) into a comprehensive analysis of its clinical/radiologic features.

Methods: An electronic search was undertaken in August/2016. Eligibility criteria included publications reporting cases of AFD and/or AFO having enough clinical, radiological and histological information to confirm the diagnosis. Demographic data, lesion site and size, treatment approach, and recurrence were analyzed and compared between AFD and AFO.

Results: 55 publications reporting 64 AFDs (60 central, 4 peripheral) and 137 publications reporting 215 AFOs (211 central, 3 peripheral, 1 unknown) were included. The difference in recurrence rate (when the information about recurrence was provided) was not statistically significant. The mean age of the patients affected by AFD was not statistically significantly different from those affected by AFO.

Conclusions: AFD and AFO presented several similarities: higher prevalence in males and in mandibles, similar patients' mean age, rate of cortical bone perforation and of the lesions' association with displaced/unerupted teeth and tooth root resorption, mean lesion size, and recurrence rate. The lesions differed with regard to the presence of radiopacities and locularity. Taken together, our data do not support the concept of progressive maturation of these tumoral conditions.

KEYWORDS

Ameloblastic fibrodentinoma; ameloblastic fibro-odontoma; odontogenic tumors; clinical features; recurrence rate

INTRODUCTION

Ameloblastic fibroma (AF) 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. Rarely, tumors with the histomorphology of AF may form dysplastic dentin, and are called ameloblastic fibrodentinomas (AFD), or dentin plus enamel, and be classified as ameloblastic fibro-odontoma (AFO).¹ There is some controversy in the literature raising doubts whether these lesions represent separate entities or are the same lesion in a continuum representing different stages of evolution.²⁻⁶

The true AFD and AFO, not the developmental odontoma, are considered to be rare lesions. There are limited details in the literature regarding their clinical and radiologic features. The epidemiological study of such rare lesions is of great importance because it provides information that can improve the diagnostic accuracy and will allow pathologists and surgeons to make informed decisions and refine the treatment plan to optimize the clinical outcome. The aim of the present study was to integrate the available data published in the literature on AFD and AFO into an updated comprehensive comparative analysis of their clinical and radiologic features. Moreover, to report the frequency of recurrence of these lesions.

MATERIALS AND METHODS

This study followed the PRISMA Statement,⁷ which is an evidence-based minimum set of items for reporting in systematic reviews. PRISMA focuses on ways in which authors can ensure a transparent and complete reporting of this type of research. A review protocol does not exist.

Search strategies

An electronic search without time or language restrictions was undertaken in August 2016 in the following databases: PubMed/Medline, Web of Science, and ScienceDirect. The following terms were used in the search strategies:

("ameloblastic fibrodentinoma") OR ("ameloblastic fibro-dentinoma") OR ("ameloblastic fibro-odontoma") OR ("ameloblastic fibro-odontome") OR ("ameloblastic fibroodontoma") OR ("mixed odontogenic tumor") OR ("mixed odontogenic tumour") OR ("ameloblastic fibroma")

Moreover, Google Scholar was also checked. A manual search of related journals, including *Acta Odontologica Scandinavica*, *Acta Oto-Laryngologica*, *Annals of Otology Rhinology and Laryngology*, *British Journal of Oral and Maxillofacial Surgery*, *Cancer, Head & Neck*, *Head and Neck Pathology*, *International Journal of Oral and Maxillofacial Surgery*, *Japanese Journal of Oral and Maxillofacial Surgery*, *Journal of Dental Research*, *Journal of Craniofacial Surgery*, *Journal of Cranio-Maxillofacial Surgery*, *Journal of Japanese Society of Oral Oncology*, *Journal of the Japanese Stomatological Society*, *Journal of Laryngology and Otology*, *Journal of Maxillofacial and Oral Surgery*, *Journal of Nihon University School of Dentistry*, *Journal of Oral and Maxillofacial Surgery*, *Journal of Oral Pathology and Medicine*, *Journal of the Stomatological Society*, *Laryngoscope*, *Oral Diseases*, *Oral Oncology*, *Oral Surgery Oral Medicine Oral Pathology Oral Radiology*, *Otolaryngology - Head and Neck Surgery*, and *Quintessence International*, was performed. The reference list of the identified studies and the relevant reviews on the subject were also scanned for possible additional studies. Publications with lesions identified by other authors as being AFD or AFO, even not having the terms "ameloblastic fibrodentinoma" or "ameloblastic fibroodontoma" in the title of the article, were also re-evaluated by an author of the present study.

Inclusion and Exclusion Criteria

Eligibility criteria included publications reporting cases of AFDs or AFOs. The studies needed to have enough clinical, radiological and histological information to confirm the diagnosis of AFD or AFO. Randomized and controlled clinical trials, cohort studies, case-control studies, cross-sectional studies, case series, and case reports were included. Exclusion criteria were review papers and immunohistochemical, histomorphometric, radiological, genetic expression, histopathological, cytological, cell proliferation/apoptosis, and *in vitro* studies, unless any of these publication categories had reported any cases with enough clinical, radiological and histological information. Hybrid tumors containing parts of AFD or AFO were not considered for this study, as they may behave differently from non-hybrid AFD and AFO tumors.

Study selection

The titles and abstracts of all publications identified through the electronic searches were read independently by the authors. For studies appearing to meet the inclusion criteria, or for which there were insufficient data in the title and abstract to make a clear decision, the full report was obtained. Disagreements were solved by discussion between the authors. The clinical and radiological aspects, as well as the histological description of the lesions were thoroughly assessed in order to confirm the diagnosis of AFD or AFO. All included cases of AFD had formation of dentinoid material, mentioned in the text and/or visible at the photomicrographs, besides the presence of ameloblastic fibroma-like areas. All published cases of AFO showing only dentine or dentinoid material deposition, but not enamel, were reclassified as AFD.

Data extraction

The authors independently extracted data using specially designed data extraction forms. Any disagreements were solved by discussion. For each of the identified studies included, the following data were then extracted on a standard form, when available: year of publication, number of patients, patient's sex, age and race, follow-up period, duration of the lesion previously to

treatment, lesion location (maxilla/mandible), anterior/posterior location (three categories: [a] anterior: lesions in the incisors/canine region; [b] premolar region; [c] posterior: lesions in the molars/retromolar region), recurrence, recurrence period, lesion size, presence of erosion of the subjacent cortical bone (for peripheral lesions), perforation of cortical bone, locularity radiological appearance (unilocular/multilocular), presence of radiopacities visible in the radiological exams, association of the lesion with a tooth (the tooth can either be erupted with the entire root(s) encompassed by the lesion or unerupted encompassing the entire tooth), and tooth displacement and tooth root resorption due to lesion's growth. The lesion size was determined according to the largest diameter reported in the publications. Contact with authors for possible missing data was performed.

Analyses

The mean, standard deviation (SD), and percentages were presented as descriptive statistics. Kolmogorov–Smirnov test was performed to evaluate the normal distribution of the variables, and Levene's test evaluated homoscedasticity. The performed tests for two independent groups were Student's t-test or Mann-Whitney test, depending on the normality. Pearson's chi-squared or Fisher's exact tests were used for categorical variables, depending on the expected count of events in a 2x2 contingency table. The degree of statistical significance was considered $p < 0.05$. All data were statistically analyzed using the Statistical Package for the Social Sciences (SPSS) version 23 software (SPSS Inc., Chicago, IL, USA).

RESULTS

Literature search

The study selection process is summarized in Figure 1. The search strategy in the databases resulted in 3610 papers. Search in Google Scholar resulted in 11 eligible papers not found in the

three main databases. A number of 1158 articles were cited in more than one database (duplicates). The reviewers independently screened the abstracts for those articles related to the focus question. Of the resulted 2463 studies, 2218 were excluded for not being related to the topic. Additional hand-searching of journals and of the reference lists of selected studies yielded 22 additional papers. The full-text reports of the remaining 267 articles led to the exclusion of 75 publications because they did not meet the inclusion criteria (see Supplemental Appendix). The excluded studies did not report enough clinical, radiological and histological information to confirm a definite diagnosis of AFD or AFO. Thus, a total of 192 publications were included in the review. Examples of histopathological pictures of AFD and AFO are shown in Figure 2.

Description of the Studies and Analyses

Ameloblastic fibrodentinitomas (AFD)

Fifty-five publications⁸⁻⁶² reporting AFDs were included in the present review, with 64 AFDs, 60 central and 4 peripheral lesions. All included cases of AFD showed formation of dentinoid material, mentioned in the text or visible in the photomicrographs, besides the presence of ameloblastic fibroma-like areas. Table 1 presents demographic and clinical features of all 64 AFDs. AFDs were more prevalent in men than in women, at a nearly 2:1 proportion. The mean age of the patients was higher for the peripheral lesions in relation to the central ones. Figure 3 shows the distribution of all lesions (n=64) according to age, with a higher prevalence in the first two decades of life. The age of the patients with peripheral lesions (n=4) was 2, 3, 11, and 51. The few peripheral lesions were located at/between teeth 11-21 buccal side (n=2), 25-26 buccal side (n=1), and 42-43 lingual side (n=1). The central lesions were more prevalent in the mandible in relation to the maxilla, and at the posterior region in relation to the anterior region (Figure 4). About 30% of the central lesions showed signs of cortical bone perforation and about 60% of the lesions had a radiological unilocular appearance. Radiopacities were observed in 46% of the AFDs, and approximately 80% of the lesions were associated with a tooth. Nearly 20% of the lesions caused root resorption of the

adjacent teeth, most of them deciduous. Treatment of the lesions was known in 58 cases, of which 56 consisted of conservative surgery (6 excisions, 50 enucleations) and 2 cases were treated by segmental resection. The time of follow-up was informed for 37 central lesions, with a mean $\pm$ SD of 50.4 $\pm$ 54.0 months (min-max, 2-258). The race of the patient was reported in 49 cases. Eighteen cases (36.7%) were diagnosed in Caucasians, 11 in Indians, 10 in Asians, 5 in Turks, 2 in blacks, 2 in Hispanics, and 1 in a Persian.

Table 2 shows the recurrence rate according to treatment and radiological locularity. There was information about recurrence for 42 lesions (40 lesions, if the information about the kind of treatment performed was also provided), of which only 2 recurred (4.8%), both central AFDs. One of the cases⁴⁶ was treated by enucleation and recurred twice, with the first recurrence after 50 months, and the second recurrence as ameloblastic fibrosarcoma 92 months after the first one. The second case³¹ had several recurrences within a period of 10 years, until it recurred as an ameloblastic fibrosarcoma. Concerning locularity, the information about recurrence and locularity was provided for 37 lesions. Only one (5.9%) multilocular lesion recurred.

Ameloblastic fibro-odontoma (AFO)

Some studies reporting series of odontogenic studies and including AFOs were found, but their cases were not included here due to lack of enough clinical, radiological and histological information to confirm the diagnosis. These include, for example, Jing et al.⁶³ with 4 cases and Luo and Li⁶⁴ with 12 cases.

One hundred and thirty-seven publications^{2,29,31,32,48,61,65-195} reporting AFOs were included in the present review, reporting 215 AFOs, 211 central and 3 peripheral lesions, and one lesion for which it was not possible to identify whether it was central or peripheral, even though there was enough information to identify it as an AFO. All included cases of AFO showed formation of enamel with or without dentinoid material, mentioned in the text or visible in the photomicrographs, besides the presence of ameloblastic fibroma-like areas. Table 1 presents demographic and clinical features

of the 214 AFOs known to be central or peripheral. AFOs were more prevalent in men than in women, at a 1.3:1 proportion. The mean age of the patients was higher for the central lesions in comparison to the peripheral ones. Figure 5 shows the distribution of the lesions (n=202; all cases for which the patients' age were reported) according to age, with a high prevalence in the first two decades of life. For five lesions were diagnosed in patients in the age range between 20 to 24 years, three lesions were diagnosed in patients between 25 to 29 years old, and four lesions in patients who were 30 years-old or older. The age of the patients with peripheral lesions (n=3) was 3, 8, and 8. The few peripheral lesions were located at/between teeth 11 buccal side (n=1), 11-12 palatal side (n=1), and 13-14 palatal side (n=1). The central lesions were more prevalent in the mandible in comparison to the maxilla, and in the posterior region in comparison to the anterior region (Figure 4). About 27% of the central lesions showed signs of cortical bone perforation and 87% of the lesions had a radiological unilocular appearance. Radiopacities were observed in 86% of the AFOs, and approximately 80% of the lesions were associated with a tooth. Twenty percent of the lesions caused root resorption of adjacent teeth, most of them deciduous. Treatment of the lesions was known in 185 cases, of which 175 consisted of conservative surgery (3 excisions, 172 enucleations) and 10 cases were treated by resection (6 marginal, 4 with continuity). The time of follow-up was informed for 108 central lesions, with a mean $\pm$ SD of 31.6 $\pm$ 33.6 months (min-max, 1-168). The race of the patient was reported in 129 cases. Fifty-five cases (42.6%) were diagnosed in Caucasians, 47 in Asians, 11 in Indians, 6 in blacks, 5 in Hispanics, 2 in Turks, and 2 in Persians, and 1 in other. Table 2 shows the recurrence rate according to treatment and radiological locularity. There was information about recurrence for 146 lesions, with 10 recurrences (6.8%), all central AFOs. Four of these recurrences were diagnosed as complex odontomas,^{2,86,163} one of them as ameloblastic fibrosarcoma after multiple recurrences,³¹ and the other five recurrences as AFOs. Concerning locularity, 119 lesions had information about recurrence and locularity, showing a higher recurrence rate for unilocular lesions in comparison to the multilocular ones ($p=0.658$; Fisher's exact test).

AFD vs. AFO

Only two factors that were statistically significantly different between the two groups of central lesions. The first one was locularity, with AFDs being more frequently multilocular than AFOs ($p < 0.001$). The second factor was the presence of visible radiopacities in the radiological exam, with a higher frequency for AFOs ($p < 0.001$) (Table 1). The difference in recurrence rate (AFD 2/42, 4.8%; AFO 10/146, 6.8%) was not statistically significant ($p = 0.473$; Fisher's exact test).

Table 3 shows details about the cases of recurrence: diagnosis and age of the patients for primary and recurrent lesions.

DISCUSSION

The present study performed an updated comparative analysis of the clinical, radiologic features, and recurrence rate between AFDs and AFOs. The great majority of the cases were reported in isolated case reports or small case series. Peripheral lesions were extremely rare, as only 4 and 3 cases of AFD and AFO, respectively, were identified in the literature. Of the 42 cases of AFD described in the literature with information about recurrence, only 2 recurrences were reported, both diagnosed as ameloblastic fibrosarcoma after multiple recurrences. This suggests that the recurrence of AFDs is unusual. AFOs seem to have a similar recurrence rate to AFDs. Only one case of recurrent AFOs (10% of the recurrences) came back as ameloblastic fibrosarcoma after multiple recurrences.³¹ Forty percent of cases of AFOs recurred as complex odontomas.^{2,86,163} It seems odd that AFOs (a neoplasm) would recur as what is considered a hamartomatous process (complex odontoma), and this raises the possibility of the primary lesions in these particular cases actually being developing odontomas. On the other hand, the finding that six of the recurrent cases (one diagnosed as ameloblastic fibrosarcoma and five AFO) showed no sign of maturation is suggestive that at least some AFOs are not tooth malformations.

The comparisons of several parameters between AFD and AFO were performed taking into consideration the central lesions only, since the peripheral counterparts were so infrequently

reported. AFD and AFO presented several similarities, such as a higher prevalence in males and in mandibles, similar mean age of the patients, rate of cortical bone perforation, rate of association with displaced/unerupted teeth and tooth root resorption, as well as the mean lesion size. The prevalence of AFDs could suggest a bimodal age distribution, even though with a far lower prevalence in the 6th and 7th decades of life than in the 1st and 2nd decades (Figure 3). A possible bimodal age distribution is less clear for AFOs (Figure 5), even having far more cases reported in the literature than AFDs.

AFD and AFO differed with regard to the presence of radiopacities visible in the radiological exams and to the radiographic locularity. It is understandable that AFOs would show a higher prevalence of visible radiopacities in the radiological exams in comparison to AFDs, as enamel is more radiopaque than dentin and dentinoid material. When it comes to locularity, despite the similarities of the mean size of the lesions in both groups, multilocularity was associated with AFD. We do not have any plausible hypothesis to explain such finding, but we have to consider that a statistically significant data may not be biologically relevant.¹⁹⁶ Further studies are necessary to clarify the interpretation of these data.

It is important to mention that the WHO¹ ranked the AFDs as an AF. There are doubts whether the two lesions are similar conditions, but it is of our opinion that AFDs should be considered as a distinct lesion from AFs until new evidence proves otherwise. The discussion about this issue probably began with Cahn and Blum,¹⁹⁷ who postulated that AF represented the least histologically differentiated lesion that evolves from a moderately differentiated form to AFO and ultimately to odontoma. Over the years this concept has not become widely accepted and was put to the test. Eversole et al.³ and Slootweg⁵ argued that, if the AF is the least differentiated tumor and can develop progressively into an AFO and a complex odontoma, then this sequence should be reflected by AFs being found in a younger age group than odontomas, with tumors of intermediate differentiation occurring between the two ages. Yet, all lesions should show the same distribution according to site and the sex predilection should be the same.⁵ However, Eversole et al.'s³ data

indicated that all these tumors occurred within the same age group. Moreover, they found completely different male-to-female ratios for AFs and complex odontomas. These ratios would be expected to be similar if AF routinely developed into odontomas. Slootweg⁵ observed in his review that a comparison of the data on age reveals that the more differentiated AFO occurs at a lower mean age than the histologically primitive AF. Therefore, since it cannot be accepted that a more differentiated lesion should occur at a lower mean age than the lesion from which it develops, the possibility of the AF developing into an AFO and then into an odontoma could be discarded.⁵ Considering the relationship between the AFO and the odontomas, Slootweg⁵ noted that his review's data on age, site, and sex lead to believe that the AFO is an immature complex odontoma. He observed that the distribution according to location is approximately the same for both lesions and the complex odontoma occurs at a larger average age. However, although some "immature odontoma" may be included in every AFO review of cases, the neoplastic nature of this tumor is accepted in the literature.²

The discussion continued with Gardner,⁴ who stated that odontomas, being hamartomas, develop during the period of normal odontogenesis and, therefore, any apparent AF found after that time is unlikely to represent the early stage of a developing odontoma. From the results of the present review, it is interesting to note that twelve cases of AFO and eight cases of AFD affected patients of 20 years of age or older, which is beyond the age of an expected tooth formation. Later, Takeda¹⁹⁸ suggested that an AFD may develop into an AFO. Philipsen et al.⁶ stated that even though there may histologically be a spectrum from the AF to the AFO with the AFD in an intermediate position, this does not necessarily suggest that the AF will differentiate over time into an odontoma. Residual or recurrent cases of AF have shown no evidence of further differentiation and maturation into a more developed odontogenic tumor.⁶ Although the present study observed that 40% of the recurrent cases of AFO were classified as complex odontomas, the other recurrent cases did not show such maturation process. Moreover, the mean age of the patients in AFD was similar to AFO, which do not support the concept of progressive maturation of these tumoral conditions. All these

data support the hypothesis that some AFD and AFO tumors are true neoplastic conditions. We also believe that they should be considered separately from odontoma, unless some contrary evidence is demonstrated.

We agree with the concept of Takeda¹⁹⁸ regarding the differential diagnosis of AFD and immature dentinoma. Microscopically, the epithelial component of 'immature dentinoma' is formed of strands and small islands of odontogenic epithelium without enamel organ-like structures, and the fibrous element varies from cellular to mature collagenous tissue, but the primitive dental papilla-like appearance is not found. The 'immature dentinoma' resembles odontogenic fibroma. On the other hand, the epithelial element in the AFD shows an enamel organ-like structure and the ectomesenchymal component resembles dental papilla. Except for the lack of enamel and the presence of dentinoid structures, AFD shows microscopic similarities to AFO and AF, respectively.

The results of the present study have to be interpreted with caution because of its limitations. First, all included studies were retrospective reports, which inherently results in flaws, manifested by the gaps in information and incomplete records. Second, many of the cases have a short follow-up, which could have led to an underestimation of the actual recurrence rate, because a longer follow-up period can lead to an increase in the recurrence rate. However, it is hard to define what it would be considered a short follow-up period to evaluate the recurrence of AFDs and AFOs. Third, the great majority of the cases described were published as isolated case reports or small case series.

CONCLUSIONS

AFD and AFO presented several similarities: higher prevalence in males and in mandibles, similar patients' mean age, rate of cortical bone perforation and of the lesions' association with displaced/unerupted teeth and tooth root resorption, mean lesion size, and recurrence rate. The lesions differed with regard to the presence of radiopacities and locularity. Taken together, our data do not support the concept of progressive maturation of these tumoral conditions.

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TABLES

Table 1. Demographic and clinical features of ameloblastic fibrodentinomas (AFD) and ameloblastic fibroodontomas (AFO) described in the literature.

	Central lesions		<i>p</i> value (central lesions)	Peripheral lesions	
	A. fibrodentinoma	A. fibroodontoma		A. fibrodentinoma	A. fibroodontoma
Sample size (n)	60	211		4	3
Age (year), mean±SD (min- max)	10.0±9.1 (1-60)	10.1±6.6 (1-55)	0.976 ^a	16.8±23.2 (2-52)	6.3±2.9 (3-8)
Men	10.7±10.1 (1-60)	9.0±5.4 (1-30)	0.318 ^a	7.0±5.7 (3-11)	-
Women	8.7±6.9 (1-24)	11.5±7.7 (1-55)	0.147 ^a	26.7±34.6 (2-51)	6.3±2.9 (3-8)
<i>p</i> value	0.448 ^a	0.007 ^a		1.000 ^b	-
Gender, n (%)					
Men	41 (69.5)	121 (57.9)	0.108 ^c	2 (50)	0 (0)
Women	18 (30.5)	88 (42.1)		2 (50)	3 (100)
Unknown	1	2		0	0
Jaw, n (%)					
Maxilla	17 (28.3)	70 (34.1)	0.399 ^c	3 (75)	3 (100)
Mandible	43 (71.7)	135 (65.9)		1 (25)	0 (0)
Unknown	0	6		0	0
Cortical bone perforation, n (%)					
Yes	13 (28.9)	39 (27.3)	0.833 ^c	0 (0)	0 (0)
No	32 (71.1)	104 (72.7)		4 (100)	3 (100)
Unknown	15	68		0	0
Bone erosion ^e , n (%)					
Yes	-	-	-	0 (0)	0 (0)
No	-	-		4 (100)	1 (100)
Unknown	-	-		0	2
Locularity, n (%)					
Unilocular	31 (63.3)	151 (87.3)	<0.001 ^c	-	-
Multilocular	18 (36.7)	22 (12.7)		-	-
Unknown	11	38		4	3
Radiopacities, n (%)					
Yes	23 (46)	149 (86.6)	<0.001 ^c	1 (33.3)	1 (33.3)
No	27 (54)	23 (13.4)		2 (66.7)	2 (66.7)
Unknown	10	39		1	0
Associated with tooth, n (%)					

Yes ^f	46 (82.1)	124 (79.5)	0.669 ^c	0 (0)	0 (0)
No	10 (17.9)	32 (20.5)		4 (100)	3 (100)
Unknown	4	55		0	0
Tooth displacement/unerupted, n (%)					
Yes	42 (84)	166 (89.2)	0.308 ^c	1 (25)	0 (0)
No	8 (16)	20 (10.8)		3 (75)	3 (100)
Unknown	10	25		0	0
Tooth root resorption, n (%)					
Yes	10 (19.6)	29 (20.1)	0.935 ^c	0 (0)	0 (0)
No	41 (80.4)	115 (79.9)		4 (100)	3 (100)
Unknown	9	67		0	0
Treatment, n (%)					
Excision/curettage	2 (3.7)	0 (0)		4 (100)	3 (100)
Enucleation	50 (92.6)	172 (94.5)		0 (0)	0 (0)
Marginal resection	0 (0)	6 (3.3)	-	0 (0)	0 (0)
Segmental resection ^g	2 (3.7)	4 (2.2)		0 (0)	0 (0)
Unknown	6	29		0	0
Recurrence, n (%)					
Yes	2 (5)	10 (7)	1.000 ^d	0 (0)	0 (0)
No	38 (95)	133 (93)		2 (100)	3 (100)
Unknown	20	68		2	0
Follow-up time (months), mean±SD (min-max)	50.4±54.0 (2-258; n=37)	31.6±33.6 (1-168; n=108)	0.053 ^a	42.0±25.5 (24-60; n=2)	9.0±5.2 (3-12; n=3)
Lesion size (cm), mean±SD (min-max)	3.6±2.3 (0.6-10.0; n=29)	3.6±2.2 (0.5-14.0; n=106)	0.973 ^a	0.8±0.5 (0.4-1.6; n=4)	1.1±0.4 (0.8-1.5; n=3)

For one AFO lesion, there was no information whether it was central or peripheral

SD – standard deviation

^a Student's *t*-test

^b Mann-Whitney test

^c Pearson's chi-squared test

^d Fisher's exact test

^e Applied to peripheral lesions only

^f The tooth(teeth) can either be erupted with the entire root(s) encompassed by the lesion or unerupted encompassing the entire tooth

^g Resection with continuity defect

Table 2. Recurrence rate according to treatment and radiological locularity – for the lesions with available information about both treatment and recurrence, or both locularity and recurrence.

Recurrence/total (% recurrence)	Ameloblastic fibrodentinoma		Ameloblastic fibroodontoma	
	Central	Peripheral	Central	Peripheral
<i>Treatment</i>				
Excision-curettage	1/2 (50)	0/2 (0)	-	0/3 (0)
Enucleation	1/36 (2.8)	-	10/135 (7.4)	-
Marginal resection	-	-	0/6 (0)	-
Segmental resection ^a	0/2 (0)	-	0/2 (0)	-
Total	2/40 (5)	0/2 (0)	10/143 (7)	0/3 (0)
<i>Locularity</i>				
Unilocular	0/20 (0)	-	7/101 (6.9)	-
Multilocular	1/17 (5.9)	-	1/18 (5.6)	-
Total	1/37 (2.7)	-	8/119 (6.7)	-

^a Resection with continuity defect

Table 3. Cases of recurrence: age of the patients for primary and recurrent lesions.

Diagnosis of the primary lesion	Patient's age	Diagnosis of the recurrent lesion	Patient's age at recurrence (months after treatment of the previous lesion)	Reference
AFD	26	AFD, AFD, AFD, AFD, AFD, AFD, AFS	36 (120) ^a	Howell and Burkes ³¹
AFD	23	AFD, AFS	27 (50), 31 (92) ^b	Nagori et al. ⁴⁶
AFO	2	CO	3 (12)	Chen et al. ²
AFO	6	CO	11 (60)	Chen et al. ²
AFO	10	CO	10 (4)	Clausen ⁸⁶
AFO	1	CO	1 (4)	Riddett ¹⁶³
AFO	18	AFO, AFS, AFS, AFS	20 (20), 21 (7 ^b), 21 (2 ^b), 22 (13 ^{b,c})	Howell and Burkes ³¹
AFO	2	AFO	4 (24)	Clausen ⁸⁶
AFO	8	AFO	9 (18)	Friedrich et al. ¹⁰⁶
AFO	7	AFO	9 (24)	Furst et al. ¹⁰⁸
AFO	11	AFO	16 (60)	Wächter et al. ¹⁹³
AFO	11	AFO, AFO	11 (6), (39 ^b)	Frissell and Shafer ¹⁰⁷

AFD - Ameloblastic fibrodentinoma, AFO - Ameloblastic fibroodontoma, AFS - Ameloblastic fibrosarcoma, CO – complex odontoma

^a During 10 years, six attempts of removal of the lesion by curettage were made, and the time period between these treatments was not informed. A new recurrence, which occurred 10 years after the primary lesion, was treated by resection of the mandible, with a diagnosis of ameloblastic fibrosarcoma

^b Time after previous treatment (also in months)

^c Patient died one month after this last procedure

FIGURE LEGENDS

Figure 1. Study screening process.

Figure 2. Histopathological features of an ameloblastic fibrodentinoma (A and B) showing ameloblastic fibroma-like areas with deposition of dentin. Ameloblastic fibroodontoma with enamel formation (C and D).

Figure 3. Distribution of ameloblastic fibrodentinomas (n=64) according to age.

Figure 4. Distribution of the known precise location of central ameloblastic fibrodentinomas (AFD; n=51) and central ameloblastic fibroodontomas (AFO; n=177): AFD/AFO. Peripheral lesions are not included here (see text for the location of the peripheral lesions). Cases involving multiple regions (or an entire quadrant) are indicated between arrows. Numbers at the top and bottom of the lines indicate cases involving both adjoining regions: anterior/premolar, premolar/molar. One asterisk (*) indicates the number of lesions from the mandibular body that reached regions posterior to the 3rd mandibular molar (mandibular angle and/or ramus and/or coronoid process and/or condyle). For the rest of the AFDs, the location was 'posterior' for 1 lesion in the maxilla and 5 in the mandible, 'right mandibular body' for 1 lesion, 'left mandibular body' for 1 lesion, and 'left maxillary sinus' for 1 lesion. For the rest of the AFOs, the location was 'anterior' for 1 lesion in the maxilla and 5 in the mandible, 'posterior' for 9 lesions in the maxilla and 12 in the mandible, 'maxilla' for 1 lesion, 'left maxillary sinus' for 1 lesion, and no information was provided for 6 lesions.

Figure 5. Distribution of ameloblastic fibroodontomas (n=202) according to age.