Accepted Manuscript

Title: Ameloblastic fibrodentinosarcoma and ameloblastic fibro-odontosarcoma: a systematic review

Author: Bruno Ramos Chrcanovic Ricardo Santiago Gomez

PII: S2468-7855(18)30074-0

DOI: <https://doi.org/doi:10.1016/j.jormas.2018.03.007>

Reference: JORMAS 166

To appear in:

Received date: 22-10-2017

Accepted date: 18-3-2018

Please cite this article as: Bruno Ramos ChrcanovicRicardo Santiago Gomez Ameloblastic fibrodentinosarcoma and ameloblastic fibro-odontosarcoma: a systematic review (2018), <https://doi.org/10.1016/j.jormas.2018.03.007>

This is a PDF file of an unedited manuscript that has been accepted for publication. As a service to our customers we are providing this early version of the manuscript. The manuscript will undergo copyediting, typesetting, and review of the resulting proof before it is published in its final form. Please note that during the production process errors may be discovered which could affect the content, and all legal disclaimers that apply to the journal pertain.

Ameloblastic fibrodedentinosarcoma and ameloblastic fibro-odontosarcoma: a systematic review

Bruno Ramos CHRCANOVIC ^{1*}

Ricardo Santiago GOMEZ ²

¹ DDS, MSc, PhD; Department of Prosthodontics, Faculty of Odontology, Malmö University, Malmö, Sweden. bruno.chrcanovic@mau.se; brunochrcanovic@hotmail.com

² DDS, MSc, PhD; Department of Oral Surgery and Pathology, School of Dentistry, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil. rsgomez@ufmg.br

* Corresponding author:

Bruno Ramos Chrcanovic. Department of Prosthodontics, Faculty of Odontology, Malmö University, Carl Gustafs väg 34, SE-214 21, Malmö, Sweden. bruno.chrcanovic@mau.se; brunochrcanovic@hotmail.com Mobile: +46 725 541 545 Fax: +46 40 6658503

ABSTRACT

The purpose of the present review was to integrate the available data published on ameloblastic fibrodedentinosarcoma (AFDS) and ameloblastic fibro-odontosarcoma (AFOS) into a comprehensive analysis of their clinical/radiologic features. An electronic search was undertaken in July/2017. Eligibility criteria included publications having enough clinical/radiological/histological information to confirm the diagnosis. 17 publications (8 AFDS, 9 AFOS) were included. The patients with the two different lesions had a similar mean age. The lesions were both more prevalent in mandibles than in maxillae, all showed bone expansion and similar rates of cortical bone perforation, tooth displacement, and locular appearance at radiological exams. Mean follow-up time was higher for AFDS, and the mean size of the lesions were larger for AFDS, although without a statistically significant difference. None of AFDS recurred, while 2 AFOS recurred. As only few cases of AFDS and AFOS have been reported, additional reports are necessary to add evidence to the study of clinical and radiologic features of these lesions.

KEYWORDS

Ameloblastic fibrodedentinosarcoma; ameloblastic fibro-odontosarcoma; odontogenic sarcomas; clinical features; recurrence rate

INTRODUCTION

Odontogenic sarcomas comprise a rare group of odontogenic neoplasms, histologically characterized by a benign ameloblastic epithelium within a sarcomatous mesenchymal component. The mesenchymal component typically comprises of marked cellularity, nuclear pleomorphism and a moderate to high number of mitotic figures. Among the odontogenic sarcomas there are the ameloblastic fibrodentinosarcoma (AFDS) and the ameloblastic fibro-odontosarcoma (AFOS), which are neoplasms similar to the ameloblastic fibrosarcoma (AFS), but in which limited amount of dysplastic dentine (dentinoid) and enamel, respectively, is formed. The differential diagnosis between these lesions depends on histopathological examination [1]. Although the presence of induced dentin and enamel does not appear to alter the basic nature of AFDS/AFOS in relation to AFS, a distinction should be drawn to facilitate correct histologic diagnosis [2].

AFDS and AFOS are considered to be extremely rare lesions. And because of that, there are limited details in the literature regarding their clinical and radiologic features. An epidemiological study of these rare tumors is important because it provides information that can improve diagnostic accuracy, allowing pathologists and surgeons to make informed decisions and refine treatment plans to optimize clinical outcomes [3-5]. Therefore, the aim of the present study was to integrate the available data published in the literature on AFDS and AFOS into an updated comprehensive comparative analysis of their clinical and radiologic features, as well as the frequency of recurrence.

MATERIALS AND METHODS

This study followed the PRISMA Statement guidelines [6].

Search strategies

An electronic search without time restrictions was undertaken in July 2017 in the following databases: PubMed/Medline, Web of Science, and Science Direct. The following terms were used in the search strategies:

(ameloblastic dentinosarcoma) OR (ameloblastic fibrodentinosarcoma) OR (ameloblastic odontosarcoma) OR (ameloblastic fibro-odontosarcoma) OR (ameloblastic fibrosarcoma) OR (ameloblastic sarcoma)

Moreover, Google Scholar was also checked. A manual search of related journals, including *Acta Odontologica Scandinavica*, *Acta Otolaryngologica*, *Annals of Otolaryngology and Rhinology*, *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 Otolaryngology*, *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 AFDS or AFOS, even not having the terms 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. The definitions and criteria of the World Health Classification of Tumors – Head and Neck Tumors book [1] (last updated in 2017), were used in order to diagnose a lesion as AFDS or AFOS. Randomized and controlled clinical trials, cohort studies, case-control studies, cross-sectional studies, case series, and case reports were included. Exclusion criteria were immunohistochemical studies, histomorphometric studies,

radiological studies, genetic expression studies, histopathological studies, cytological studies, cell proliferation/apoptosis studies, *in vitro* studies, and review papers, unless any of these publication categories had reported any cases with enough clinical, radiological and histological information. Hybrid tumors containing parts of AFDS or AFOS were not considered for this study, as they may behave differently from non-hybrid lesions.

Study selection

The titles and abstracts of all reports 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 reported by the publications were thoroughly assessed by one of the authors (R.S.G.) of the present study, expert in oral pathology, in order to confirm the diagnosis of AFDS or AFOS.

Data extraction

The review authors independently extracted data using specially designed data extraction forms. Any disagreements were resolved 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, perforation of cortical bone, locularity appearance in radiological exams (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), tooth displacement and/or tooth root resorption due to lesion's growth, expansion of osseous region adjacent to the tumor, presence of clinical symptoms, and treatment performed (curettage/excision, enucleation, partial resection, resection with continuity). Information on radiotherapy, chemotherapy, the number of recurrences, the occurrence of metastases, and the patient's death were also collected. 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 929 papers. Search in Google Scholar resulted in 1 paper not found in the three main databases. A number of 316 articles were cited in more than one database (duplicates). The reviewers independently screened the abstracts for those articles related to the aim of the present review. Of the resulted 614 studies, 596 were excluded for not being related to the topic or not presenting clinical cases. Additional hand-searching of journals and of the reference lists of selected studies did not yield additional papers. The full-text reports of the remaining 18 articles led to the exclusion of 1 publication because it did not meet the inclusion criteria (same case reported in two different papers). Thus, a total of 17 publications were included in the review.

Description of the Studies and Analyses

Seventeen publications reporting 8 AFDS [2,7-13] and 9 AFOS [14-22] were included in the present review. Table 1 presents the details of each case separately. Table 2 shows a comparison of the demographic and clinical features between AFDS and AFOS. The patients had a similar mean age ($p = 0.815$; Mann-Whitney test). The lesions were both more prevalent in mandibles than in maxillae, mostly in the posterior regions, all showed bone expansion and similar rates of cortical bone perforation, tooth displacement, and locular appearance at radiological exams. One case of AFDS presented tooth root resorption. The mean time of follow-up was higher for AFDS in comparison to AFOS ($p = 0.345$; Mann-Whitney test), and the mean size of the AFDS lesions were larger than of the AFOS ones ($p = 0.556$; Mann-Whitney test), although the differences were not statistically significant. Most cases of both lesions were treated by segmental resection. Two cases of AFDS were submitted to chemotherapy [8,10] and three to radiotherapy [10,11,13]. Only one case of AFOS [16] was submitted to both chemotherapy and radiotherapy. Table 3 shows the recurrence rate according to the treatment performed. None of AFDS recurred, while 2 AFOS recurred, 5.5 [16] and 24 months [20] after surgery.

DISCUSSION

The terms “ameloblastic fibrosarcoma” and “ameloblastic sarcoma” were also used in the search strategies, in order to assess case reports of AFS that could possibly represent cases of AFDS or AFOS due to the presence of dysplastic dentine (dentinoid) and enamel. Through this search method it was possible to find 6 additional cases of AFDS/AFOS in the literature. Even so, also reviewing all cases of AFS in the literature, the authors of the present review cannot figure out how, for example, Chen et al. [14] were able to find 14 cases of AFOS in the literature quoting only 5 references, in which one of them [23] did not show the histological microphotography in order to confirm the diagnosis. In the same way, Reiser et al. [19] stated that 15 cases of AFOS have been reported, and Gatz et al. [16] stated that “seventeen cases (of AFOS) have been reported in the literature to date” quoting only 5 references, not pointing out that these cases consisted of AFOS and of AFDS. More correctly, WHO [1] stated in 2017 that fourteen cases of both AFDS and AFOS were reported in the literature quoting, however, only 3 references. None of these publications declared that the previously published cases in the literature were histologically assessed by the authors in order to confirm the diagnosis, as it was performed in the present review.

About 26% of the AFS cases developed in a recurrent ameloblastic fibroma (AF) [24]. It is thought that ameloblastic fibrodentinoma (AFD) and ameloblastic fibro-odontoma (AFO) to be the precursor for AFDS and AFOS, respectively. Patients diagnosed with AFD and AFO indeed show a lower mean age than AFDS and AFOS [25]. However, none of the cases reviewed here reported to clearly come from a recurrent AFD or AFO. Mainenti et al. [17] reported a case presenting patterns that were recognized as an AFOS arising in a pre-existing AFO. Although it could be possible, one cannot definitively confirm that because there was no previous diagnosis of AFO. And Gatz et al. [16] reported a case of AFOS in patient that presented a hamartomatous complex odontoma, not an AFO, 6 years earlier.

Some cases of AFDS and AFOS showed large areas of the resected tumor with microscopic features of a benign tumor [17,19]. It is therefore important that a histological evaluation of the whole specimen should be carried out, because in AFDS and AFOS the epithelial component is very similar to the one in AFD and AFO, respectively, but the mesenchymal component may show increased cellularity with a sarcomatoid nature in isolated areas.

The use of therapies such as chemotherapy or radiotherapy is not well established for AFDS/AFOS, as there are only few cases submitted to these adjunctive therapies. Segmental resection seems to be the most recommended therapy for AFDS/AFOS, as it is for AFS [24].

The prognosis for the odontogenic sarcomas is apparently better than for other jaw fibrosarcomas, and it is considered a locally aggressive neoplasm with a low incidence of metastasis [8]. One might consider AFDS and AFOS as low-grade tumors, possibly less aggressive than AFS, as none of the cases reviewed here showed clear evidence of metastases in any site (the patient of Gatz et al. [16] developed 3 small pulmonary nodules, presumed to be metastatic lesions from the AFOS. However, a tissue diagnosis was not obtained), and the only death documented [20] was due to a patient with uncontrollable postoperative bleeding on the 4th day of the hospitalization. However, owing to the rarity of cases, it is difficult to accurately estimate long-term prognosis and/or a defined set of criteria for grading. Moreover, the cases of AFDS and AFOS were followed up for a mean of only 64.0 ± 83.9 and 22.5 ± 12.9 months, respectively. This 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/AFOS.

CONCLUSIONS

As only few cases of AFDS and AFOS have been reported, additional reports are necessary to add evidence to the study of clinical and radiologic features of these lesions.

ACKNOWLEDGEMENTS

Declaration of conflicting interests

There are no conflicts of interest to declare.

Funding/grant support

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

REFERENCES

- [1] World Health Organization Classification of Head and Neck Tumours, 4th edition ed. IARC Press, Lyon, 2017.
- [2] Altini M, Smith I. Ameloblastic dentinosarcoma- a case report. *Int J Oral Surg* 1976;5(3):142-7.
- [3] Chrcanovic BR, Gomez RS. Peripheral calcifying cystic odontogenic tumour and peripheral dentinogenic ghost cell tumour: an updated systematic review of 117 cases reported in the literature. *Acta Odontol Scand* 2016;74(8):591-7.
- [4] Chrcanovic BR, Gomez RS. Calcifying epithelial odontogenic tumor: An updated analysis of 339 cases reported in the literature. *J Craniomaxillofac Surg* 2017;45(8):1117-23.
- [5] Chrcanovic BR, Gomez RS. Glandular odontogenic cyst: an updated analysis of 169 cases reported in the literature. *Oral Dis* 2017.
- [6] Moher D, Liberati A, Tetzlaff J, Altman DG, Grp P. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *Ann Intern Med* 2009;151(4):264-9, W64.
- [7] Altini M, Thompson SH, Lownie JF, Berezowski BB. Ameloblastic sarcoma of the mandible. *J Oral Maxillofac Surg* 1985;43(10):789-94.
- [8] Bregni RC, Taylor AM, Garcia AM. Ameloblastic fibrosarcoma of the mandible: report of two cases and review of the literature. *J Oral Pathol Med* 2001;30(5):316-20.

- [9] Corominas-Villafañe O, Cuestas-Carnero R, Corominas O, Jr., Gendelman H. Ameloblastic odontosarcoma: report of a case. *Acta Stomatol Belg* 1993;90(3):149-56.
- [10] da Silva LP, da Rocha Tenorio J, de Melo Junior BC, da Silva Filho JP, do Nascimento GJ, Sobral AP. Ameloblastic fibroodontosarcoma: a rare malignant odontogenic tumor. *Braz J Otorhinolaryngol* 2016;82(5):610-3.
- [11] Delgado Azañero W, Funes Rumiche I, Torres Vega F, Calderón Ubaqui V. Fibrosarcoma ameloblástico encapsulado. *Rev Estomatol Hered* 2008;18(2):128-35.
- [12] Khan M, Ramachandra VK, Kampasi N. Clinical, radiographic, and histologic presentation of fibroodontosarcoma. *Quintessence Int* 2014;45(2):169-72.
- [13] Zabolinejad N, Hiradfar M, Anvari K, Razavi AS. Ameloblastic fibrosarcoma of the maxillary sinus in an infant: a case report with long-term follow-up. *J Pediatr Surg* 2008;43(2):e5-8.
- [14] Chen SJ, Zheng XW, Lin X, Liu H. Ameloblastic fibro-odontosarcoma of the mandible in a pediatric patient. *Eur Ann Otorhinolaryngol Head Neck Dis* 2016;133(6):419-21.
- [15] Forman G, Garrett J. Ameloblastic sarcoma: report of case. *J Oral Surg* 1972;30(1):50-4.
- [16] Gatz SA, Thway K, Mandeville H, Kerawala C, MacVicar D, Chisholm J. Chemotherapy responsiveness in a patient with multiply relapsed ameloblastic fibro-odontosarcoma of the maxilla. *Pediatr Blood Cancer* 2015;62(11):2029-32.
- [17] Mainenti P, Oliveira GS, Valerio JB, Daroda LS, Daroda RF, Brandao G, Rosa LE. Ameloblastic fibro-odontosarcoma: a case report. *Int J Oral Maxillofac Surg* 2009;38(3):289-92.
- [18] Phillips VM, Grotepass FW, Hendricks R. Ameloblastic odontosarcoma with epithelial atypia: a case report. *Br J Oral Maxillofac Surg* 1988;26(1):45-51.
- [19] Reiser V, Alterman M, Shuster A, Kaplan I. Pediatric ameloblastic fibro-odontosarcoma of the mandible: a challenge of diagnosis and treatment. *J Oral Maxillofac Surg* 2013;71(1):e45-57.
- [20] Takeda Y, Kuroda M, Suzuki A. Ameloblastic odontosarcoma (ameloblastic fibro-odontosarcoma) in the mandible. *Acta Pathol Jpn* 1990;40(11):832-7.
- [21] Wang S, Shi H, Wang P, Yu Q. Ameloblastic fibro-odontosarcoma of the mandible: imaging findings. *Dentomaxillofac Radiol* 2011;40(5):324-7.
- [22] Villa VG. Ameioblastic sarcoma in the mandible; report of a case. *Oral Surg Oral Med Oral Pathol* 1955;8(2):123-9.
- [23] Muller S, Parker DC, Kapadia SB, Budnick SD, Barnes EL. Ameloblastic fibrosarcoma of the jaws. A clinicopathologic and DNA analysis of five cases and review of the literature with discussion of its relationship to ameloblastic fibroma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995;79(4):469-77.
- [24] Chrcanovic BR, Brennan PA, Rahimi S, Gomez RS. Ameloblastic fibroma and ameloblastic fibrosarcoma: a systematic review. *J Oral Pathol Med* 2017.
- [25] Chrcanovic BR, Gomez RS. Ameloblastic Fibroodontinoma and Ameloblastic Fibro-Odontoma: An Updated Systematic Review of Cases Reported in the Literature. *J Oral Maxillofac Surg* 2017;75(7):1425-37.

FIGURE LEGENDS

Figure 1. Study screening process.

Figure 2. Distribution of ameloblastic fibroodontosarcomas (AFDS) and ameloblastic fibro-odontosarcomas (AFOS) according to age.

Table 1. Cases of ameloblastic fibroodontosarcoma (AFDS) and ameloblastic fibro-odontosarcoma (AFOS) described in the literature.

Authors	Year	Sex	Age	Location	Bone expansion/symptoms	Lesion size (cm)	Cortical bone perforation / Locularity	Tooth displacement / resorption	Treatment / Recurrence	Chemo / Radiotherapy	Metastasis / Death	Follow-up (months)
AFDS												
Altini and Smith [2]	1976	M	27	34-38	Y / N	NA	Y / U	N / N	Segmental resection / NA	N / N	NA	NA
Altini et al. [7]	1985	M	25	38-41	Y / Y	NA	Y / M	Y / NA	Segmental resection / N	N / N	N / N	18
Corominas-Villafañe et al. [9]	1993	M	12	41-48	Y / N	7 x 5	Y / U	N / N	Enucleation / NA	N / N	N / N	228
Bregni et al. [8]	2001	M	32	31-angle-ramus	Y / N	12 x 10	Y / M	Y / NA	Patient lost to follow-up after biopsy and two	Y / N	N / N	1

Delgado Azañero et al. [11]	2008	F	19	46-48	Y / N	3 x 2	N / M	Y / Y	cycles of chemo Segmental resection / N	N / Y	N / N	65
Zabolinejad et al. [13]	2008	M	4 mo	Left maxillary sinus	Y / N	12	Y / NA	NA	Marginal resection / N	N / Y	N / N	54
Khan et al. [12]	2014	F	17	45-angle	Y / N	4 x 4	Y / U	Y / N	Segmental resection / NA	N / N	N / N	NA
da Silva et al. [10]	2016	M	19	35-angle-ramus	Y / N	NA	Y / M	Y / N	Segmental resection / N	Y / Y	N / N	18
AFOS												
Villa [22]	1955	F	20	Left mandible	Y / N	NA	Y / M	N / N	Segmental resection / N	N / N	N / N	NA
Forman and Garrett [15]	1972	M	17	46-angle-ramus	Y / Y	4 x 4	N / U	Y / N	Enucleation / N	N / N	N / N	14
Phillips et al. [18]	1988	M	29	36-angle-ramus-coronoid	Y / Y	NA	Y / M	Y / N	Segmental resection / N	N / N	N / N	13
Takeda et al. [20]	1990	M	23	34-angle-ramus	Y / Y	NA	Y / M	Y / N	Segmental resection / Y	N / N	N / Y ^a	24
Mainenti et al. [17]	2009	F	12	34-angle-ramus	Y / Y	NA	Y / U	Y / N	Segmental resection / N	N / N	N / N	23
Wang et al. [21]	2011	F	45	47-angle-ramus	Y / N	6 x 4.5	Y / M	N / N	Segmental resection / N	N / N	N / N	12
Reiser et al. [19]	2013	F	6	31-angle-ramus-coronoid-condyle	Y / N	7.8 x 5.5	Y / M	Y / NA	Segmental resection / N	N / N	N / N	12
Gatz et al. [16]	2015	F	14	Right maxillary sinus	Y / N	3.7 x 3.4	N / NA	NA / NA	Debulking + Curettage / Y	Y / Y	N ^b / N	34
Chen et al. [14]	2016	M	4	44-angle-ramus	Y / N	NA	Y / M	Y / N	Segmental resection / N	N / N	N / N	48

NA – not available, Y – yes, N – no, U – unilocular, M – multilocular

^a Patient died from uncontrollable bleeding, on the 4th day of the second hospitalization

^b The patient developed 3 small pulmonary nodules (maximum diameter 12 mm), presumed to be metastatic lesions from the AFOS, which have been treated with radiofrequency ablation. However, a tissue diagnosis was not obtained.

Table 2. Demographic and clinical features of ameloblastic fibrodentinosa (AFDS) and ameloblastic fibro-odontosarcoma (AFOS) described in the literature.

Variables	AFDS	AFOS
n	8	9
Age (years), mean±SD (min-max)	18.9±9.9 (0-32; n=8)	18.9±12.6 (4-45; n=9)
Men	19.2±11.7 (0-32; n=6)	18.3±10.7 (4-29; n=4)
Women	18.0±1.4 (17-19; n=2)	19.4±15.2 (6-45; n=5)
p value ^a	0.643	0.806
Gender, n (%)		
Men	6 (75)	4 (44.4)
Women	2 (25)	5 (55.6)
Unknown	0	0
Jaw, n (%)		
Maxilla	1 (12.5)	1 (11.1)
Mandible	7 (87.5)	8 (88.9)
Unknown	0	0
Bone expansion, n (%)		
Yes	8 (100)	9 (100)
No	0	0
Unknown	0	0
Symptomatic, n (%)		
Yes	1 (12.5)	4 (44.4)
No	7 (87.5)	5 (55.6)
Unknown	0	0
Cortical bone perforation, n (%)		
Yes	7 (87.5)	8 (88.9)
No	1 (12.5)	1 (11.1)
Unknown	0	0
Locularity, n (%)		
Unilocular	2 (28.6)	2 (28.6)
Multilocular	5 (71.4)	5 (71.4)
Unknown	1	2
Tooth displacement/unerupted, n (%)		
Yes	5 (71.4)	5 (71.4)
No	2 (28.4)	2 (28.6)
Unknown	1	2
Tooth root resorption, n (%)		
Yes	1 (20)	0
No	4 (80)	6 (100)
Unknown	3	3

Treatment, n (%)		
Debulking + Curettage	0	1 (11.1)
Enucleation	1 (14.3)	1 (11.1)
Marginal resection	1 (14.3)	0
Segmental resection ^b	5 (71.4)	7 (77.8)
Unknown	1	0
Adjunctive therapy, n (%)		
Chemotherapy		
Yes	2 (25)	1 (12.5)
No	6 (87.5)	7 (87.5)
Unknown	0	1
Radiotherapy		
Yes	3 (37.5)	1 (12.5)
No	5 (62.5)	7 (87.5)
Unknown	0	1
Metastasis, n (%)		
Yes	0	0
No	7 (100)	8 (100)
Unknown	1	1
Death, n (%)		
Yes	0	1 (12.5) ^c
No	6 (100)	7 (87.5)
Unknown	2	1
Recurrence, n (%)		
Yes	0	2 (25)
No	5 (100)	6 (75)
Unknown	3	1
Follow-up time (months), mean±SD (min-max)	64.0±83.9 (1-228; n=6)	22.5±12.9 (12-48; n=8)
Lesion size (cm), mean±SD (min-max)	7.6±4.3 (3.0-12.0; n=5)	5.4±1.9 (3.7-7.8; n=4)

SD – standard deviation,

^a Comparison of the mean age between men and women (Mann-Whitney test)

^b Resection with continuity defect

^c Patient died from uncontrollable bleeding, on the 4th day of the second hospitalization

Table 3. Treatment recurrence – for the lesions with available information about treatment and recurrence.

Treatment	AFDS	AFOS
Recurrence/total (% recurrence)		
Debulking + Curettage	-	1/1 (100)
Enucleation	0/1 (0)	0/1 (0)
Marginal resection	0/1 (0)	-
Segmental resection ^a	0/3 (0)	1/6 (16.7)
Total	0/5 (0)	2/8 (25)
Adjunctive therapy		
Chemotherapy		
Yes	0/1 (0)	1/1 (100)
No	0/4 (0)	1/7 (14.3)
Radiotherapy		
Yes	0/2 (0)	1/1 (100)
No	0/3 (0)	1/7 (14.3)

AFDS - ameloblastic fibro-dentinosarcoma, AFOS - ameloblastic fibro-odontosarcoma

^a Resection with continuity defect