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REVIEW ARTICLE

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Adenomatoid odontogenic tumor: An updated analysis of the cases reported in the literature

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Purpose: To review the clinical and radiographic features of the available data published on adenomatoid odontogenic tumor (AOT) with special emphasis on the comparison of its variants.

Methods: An electronic search was undertaken in July 2018. Eligibility criteria included publications having enough clinical/radiological/histological information to confirm the diagnosis.

Results: A total of 436 publications reporting 1558 cases were included, of which 739 follicular, 247 extrafollicular, and 30 peripheral AOTs. Impacted canine is associated with follicular AOTs in almost 70% of the cases. AOTs were more prevalent in females, in the second decade of life, in maxillae, in anterior region of the jaws, and most are asymptomatic, with a considerable number of lesions presenting cortical bone perforation. Most of the lesions were treated by enucleation. Some cases of recurrence were reported in the literature, but only one was well documented. No difference was found when comparing the clinical/radiological features of the follicular, extrafollicular, and peripheral variants.

Conclusions: Adenomatoid odontogenic tumor variants do not show distinctive clinical radiological features. Recurrence of AOT is very rare, which justify its conservative management.

KEYWORDS
adenomatoid odontogenic tumor, clinical features, odontogenic tumors, recurrence, variants

1 | INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a benign epithelial tumor that shows duct-like structures. AOTs tend to be encapsulated but produce a variety of architectural patterns, most notably multiple, variably sized nodules of polyhedral to spindled epithelial cells with minimal stroma.[1] There is controversy on the literature about its nature, hamartomatous, or neoplastic.[2,3] Cases of recurrence have been a great matter of debate between authors. The aim of the present study was to integrate the available data published in the literature on AOT into an updated comprehensive comparative analysis of their clinical and radiologic features, as well as to try to identify possible cases of recurrence. Moreover, we also compared the clinic-radiographic features of the follicular, extrafollicular, and peripheral variants.

2 | MATERIALS AND METHODS

This study followed the PRISMA Statement guidelines in Appendix S1.[4]

2.1 | Search strategies

An electronic search without time restrictions was undertaken in July 2018 in the following databases: PubMed/Medline, Web of Science,

ScienceDirect, J-Stage, and LILACS. The following terms were used in the search strategies: (adenomatoid odontogenic tumor) OR (adenomatoid odontogenic tumour) OR (adenoameloblastoma) OR (ameloblastic adenomatoid tumor) OR (ameloblastic adenomatoid tumour) OR (adenomatoid ameloblastoma)

Google Scholar was also checked. A manual search of all related oral pathology, maxillofacial and specialist dental and oral journals was performed. The reference list of identified studies and the relevant reviews on the subject were also checked for possible additional studies. Publications with lesions identified by other authors as being AOT, even not having the aforementioned terms in the title of the article, were also re-evaluated by an author (R.S.G.) of the present study.

2.2 | Inclusion and exclusion criteria

Eligibility criteria included publications reporting cases of AOTs, with 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] were used to diagnose a lesion as AOT. The studies could be of any nature (case series, case reports, immunohistochemical studies, histomorphometric studies, radiological studies, etc.), provided that the publication had reported any cases with enough clinical, radiological, and histological information. Except for calcifying epithelial odontogenic tumor-like areas, which are within the spectrum of AOT, hybrid odontogenic tumors containing AOT were excluded.

2.3 | 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 resolved 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 of the present study (R.S.G.), an expert in oral pathology, in order to confirm the diagnosis of AOT.

2.4 | 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, duration of the lesion previously to treatment, lesion location, lesion size, perforation of cortical bone, locularity radiological appearance (unilocular/multilocular), tooth displacement and/or tooth root resorption due to lesion's growth, expansion of osseous region adjacent to the tumor, the presence of clinical symptoms, treatment performed (curettage/excision, enucleation, partial resection, resection with continuity), follow-up period, recurrence, and time to recurrence. The lesion size was determined according to the largest diameter reported in the publications. Contact with authors for possible missing data was performed.

The three clinical and radiographical AOT variants were, according to their intraosseous or extraosseous locations (the intraosseous type varies according to the association or not with the crown of an unerupted tooth), classified as (a) follicular, (b) extrafollicular, and (c) peripheral (or extraosseous) AOT types.

2.5 | Analyses

The mean, standard deviation (SD), and percentage were calculated for several variables. The test performed was the following: Kolmogorov-Smirnov (to evaluate normal distribution), Levene's test (to evaluate homoscedasticity), Student's *t*-test or Mann-Whitney (for two independent groups, continuous variables), Pearson's chi-squared or Fisher's exact test (for categorical variables). The degree of statistical significance was considered $P < 0.05$. All data were statistically analyzed using the SPSS version 25 software (SPSS Inc., Chicago, IL, USA).

3 | RESULTS

3.1 | Literature search

The study selection process is summarized in Figure S1 (Appendix S1). The search strategy in the databases resulted in 2134 papers, 59 additional eligible papers were found in Google Scholar, and 11 papers through hand-searching. At the end, a total of 436 publications were included (Appendix S1).

3.2 | Description of the studies and analyses

A total of 1558 AOTs were identified, of which 739 were follicular, 247 extrafollicular, and 30 peripheral lesions—information was not available for 542 AOTs. Of the 739 follicular AOTs, 507 cases presented one tooth associated with the lesion, 46 cases with 2 teeth (of which in 39 cases one canine was involved), 7 cases with 3 teeth (in all cases one canine was involved), 1 case with 4 teeth (one canine involved), and 2 cases with 6 teeth (both with 2 canines involved). This information was not possible to retrieve for 175 follicular AOTs. When only one tooth was involved by the lesion, the tooth was a canine in 335 out of 493 cases (67.9%) with information available—222 maxillary canines and 113 mandibular canines.

Table 1 presents demographic and clinical features of all cases. AOTs were more prevalent in women than in men, at a nearly 1.9:1 proportion. The mean age of the patients was 19.0 ± 9.0 years (range 1-82). The lesions were noticed by the patient a mean±SD of 13.9 ± 27.5 months (range 0-444) before looking for treatment. Figure 1 shows the distribution of the lesions according to age, separated by the variants. The highest prevalence occurred in the second

TABLE 1 Demographic and clinical features of 1558 adenomatoid odontogenic tumors (AOTs) described in the literature

	Variant			
Variables	**Global**	**Follicular**	**Extrafollicular**	**Peripheral**
n	1558	739	247	30
Age (y), mean ± SD (min–max)	19.0 ± 9.0 (1–82; n = 1480)	16.3 ± 6.0 (1–59; n = 714)	23.9 ± 12.0 (5–82; n = 241)	16.5 ± 7.1 (3–37; n = 30)
Men	18.8 ± 9.3 (5–80; n = 520)	16.2 ± 5.8 (5–56; n = 243)	26.8 ± 14.4 (9–80; n = 85)	16.3 ± 6.6 (9–28; n = 7)
Women	19.2 ± 8.9 (1–82; n = 950)	16.3 ± 6.1 (1–59; n = 469)	22.3 ± 10.1 (5–82; n = 156)	16.5 ± 7.4 (3–37; n = 23)
P value[a]	0.016	0.578	0.094	0.787
Gender, n (%)				
Men	538 (34.8)	246 (33.4)	87 (35.2)	7 (23.3)
Women	1010 (65.2)	491 (66.4)	160 (64.8)	23 (76.7)
Unknown	10	2	0	0
Jaw, n (%)				
Maxilla	940 (61.8)	484 (65.5)	129 (52.4)	27 (93.1)
Mandible	582 (38.2)	255 (34.5)	117 (47.6)	2 (6.9)
Unknown	36	0	1	1
Bone expansion, n (%)				
Yes	724 (89.5)	486 (90.5)	170 (85.4)	–
No	85 (10.5)	51 (9.5)	29 (14.6)	–
Unknown	749	202	48	–
Symptomatic, n (%)				
Yes	97 (11.9)	57 (10.8)	26 (13.3)	0 (0)
No	721 (88.1)	469 (89.2)	170 (86.7)	23 (100)
Unknown	740	213	51	7
Cortical bone perforation, n (%)				
Yes	115 (45.6)	85 (45.2)	30 (47.6)	–
No	137 (54.4)	103 (54.8)	33 (52.4)	–
Unknown	1306	551	184	–
Cortical bone thinning, n (%)				
Yes	381 (96.7)	291 (97.0)	90 (95.7)	–
No	13 (3.3)	9 (3.0)	4 (4.3)	–
Unknown	1164	439	153	–
Bone erosion, n (%) [b]				
Yes	–	–	–	8 (47.1)
No	–	–	–	9 (52.9)
Unknown	–	–	–	13
Locularity, n (%)				
Unilocular	633 (98.9)	461 (98.9)	154 (98.7)	–
Multilocular	7 (1.1)	5 (1.1)	2 (1.3)	–
Unknown	918	273	91	–
Radiological borders, n (%)				
Ill–defined	7 (1.3)	5 (1.2)	2 (1.5)	–
Well–defined	549 (98.7)	412 (98.8)	131 (98.5)	–
Unknown	1002	322	114	–
Tooth root resorption, n (%)				
Yes	86 (17.1)	60 (17.3)	26 (20.3)	–

(Continues)

TABLE 1 (Continued)

	Variant			
Variables	**Global**	**Follicular**	**Extrafollicular**	**Peripheral**
No	418 (82.9)	286 (82.7)	102 (79.7)	–
Unknown	1054	393	119	–
Treatment, n (%)				
None	1 (0.1)	0 (0)	0 (0)	0 (0)
Excision	18 (2.2)	0 (0)	0 (0)	18 (85.7)
Marsupialization	16 (2.0)	13 (2.3)	1 (0.5)	0 (0)
Curettage	32 (4.0)	22 (3.8)	9 (4.6)	1 (4.8)
Enucleation	733 (90.6)	532 (93.0)	180 (92.8)	2 (9.5)
Marginal resection	6 (0.7)	4 (0.7)	2 (1.0)	0 (0)
Segmental resection[c]	3 (0.4)	1 (0.2)	2 (1.0)	0 (0)
Unknown	749	167	52	9
Recurrence, n (%)				
Yes	1 (0.2)	1 (0.2)	0 (0)	0 (0)
No	644 (99.8)	455 (99.8)	155 (100)	13 (100)
Unknown	913	283	92	17
Follow–up time (mo), mean ± SD (min–max)	30.0 ± 36.3 (1–300; n = 352)	28.5 ± 32.9 (1–232; n = 250)	32.2 ± 35.7 (1–174; n = 91)	48.1 ± 89.9 (2–300; n = 10)
Lesion size (cm), mean ± SD (min–max)	3.3 ± 1.7 (0.5–12.0; n = 423)	3.6 ± 1.7 (0.7–12.0; n = 306)	2.5 ± 1.4 (0.5–7.5; n = 102)	1.9 ± 0.9 (0.5–3.0; n = 11)

SD, standard deviation.
[a]Comparison of the mean age between men and women (Mann–Whitney test).
[b]Peripheral lesions only.
[c]Resection with continuity defect.

decade of life. The lesions were more prevalent in the maxilla in comparison with the mandible, and at the anterior region in comparison with the posterior region (Figures 2,34,and). The peripheral variant had a strong prevalence in the anterior maxilla. A considerable percentage of central lesions show signs of cortical bone perforation and only a small amount of AOTs had either multilocular radiological appearance or ill-defined borders. Nearly 17% of the lesions presented root resorption of adjacent teeth. No difference was found comparing the clinical and radiological features of the follicular, extrafollicular, and peripheral variants. Most of the lesions were treated by enucleation. Time of follow-up was informed for 352 lesions, with a mean ± SD of 30.0 ± 36.3 months (min-max, 1-300). An amount of 11.6% of the lesions was followed up for at least 5 years. Some cases of recurrence were reported in the literature, but only one case was identified as possibly presenting strong evidence of an actual recurrence.[5] The lesion recurred twice, 12 years after surgery of the primary lesion, and then again 88 months after the second surgery.

4 | DISCUSSION

The aim of the present study was to integrate the available data published in the literature on AOT. A review of pathological lesions is important as it provides information that can improve diagnostic accuracy, allowing pathologists and surgeons to make informed decisions and refine treatment plans to optimize clinical outcomes.[6-10] The present review observed that AOTs were more prevalent in females, in the second decade of life, in maxillae, in anterior region of the jaws, and most of the lesions are asymptomatic, with a considerable number of lesions presenting cortical bone perforation. Only a small amount of lesions were reported as radiographically presenting ill-defined borders. The evaluation of this criterion by radiographs printed on articles is not always reliable, unless the authors describe it in the text. Thus, the radiological features described in our study may be over or underestimated. Despite this limitation, we could not find any relevant difference between follicular, extrafollicular, and peripheral variants of AOT, which gives additional support to the unifying concept of AOT histogenesis.[11] The clinical and epidemiological profile found in our study confirms results of previous studies.[2,12]

Only one out of more than 1500 AOT cases reported in the literature had substantial evidence of recurrence.[5] However, information on follow-up was available for a merely 22% of the lesions reported in the literature, and about only 11% of these were followed up for at least 5 years. Another case of recurrence was identified by Ide et al[13] being reported in two separated publications, both written in Japanese.[14,15] The authors of the present review were able to retrieve

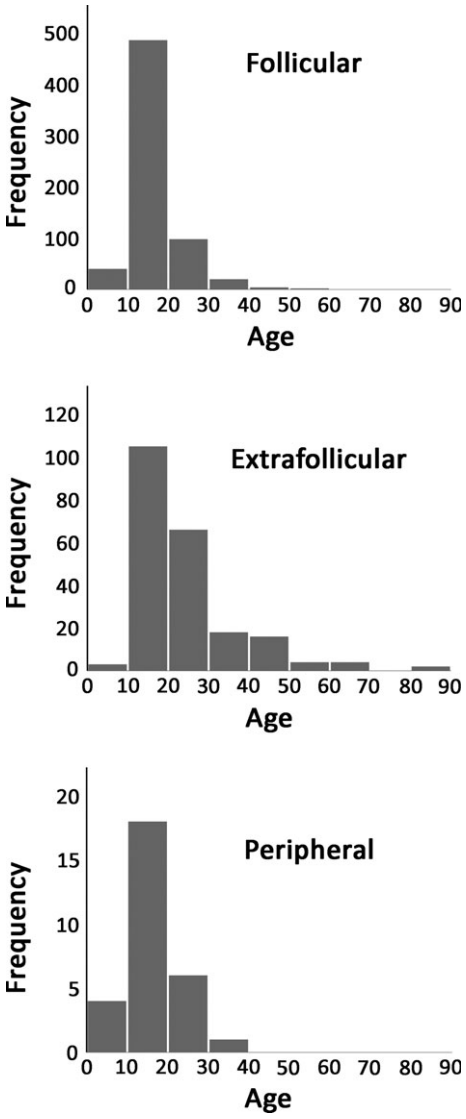

FIGURE 1 Distribution of follicular adenomatoid odontogenic tumors (AOTs), extrafollicular AOTs, and peripheral AOTs according to age (for the cases which the patients' age was informed)

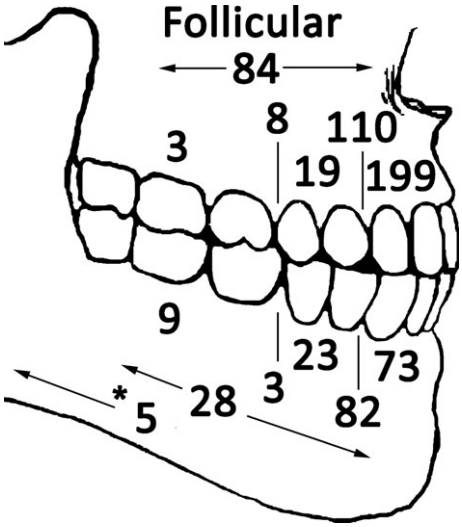

FIGURE 2 Topographical distribution of the known precise locations (n = 646) of follicular adenomatoid odontogenic tumors (AOTs). Cases involving multiple regions (or an entire quadrant) are indicated between arrows. Numbers at the top and bottom of the lines indicate cases involving both adjoining regions: anterior/premolar and premolar/molar. One asterisk (*) indicates the number of lesions from the mandibular body that reached the angle and/or ramus. For the rest of the lesions (n = 93), the location was the "maxilla" (n = 36), "anterior maxilla" (n = 21), "posterior maxilla" (n = 2), "mandible" (n = 10), "anterior mandible" (n = 21), "posterior mandible" (n = 1), and "maxillary sinus" (n = 2). For those cases with available information, 89 out of 650 lesions (13.7%) crossed the midline of the jaws, and the maxillary sinus was significantly affected in 78 out 484 lesions (16.1%) in the maxilla. One lesion reached the coronoid process

the most recent paper[14] but not the first one[15] describing the primary lesion. Moreover, the linguistic barrier would be a limitation in order to properly evaluate the case.

There are some cases in the literature described as recurrent AOTs, but with dubious histopathological results. Takigami et al[16] described a multiple recurrent maxillary AOT with intracranial extension. Rick[3] concluded, however, that this was an ameloblastoma. In the case of Toida et al[17] the radiographic examination showed a small, well-defined radiolucent lesion, measuring approximately 5 mm in diameter 77 months after the enucleation of the primary lesion. Recurrence of the AOT was suspected. However, the patient did not return for further examination. The cases of Yavas et al[18] and Lang et al[19] are other cases described as "recurrent" lesions, but with unconvincing histopathology of AOT. Yoon and Kim[20] described a case of AOT showing a supposed recurrence only 12 months after surgery. However, considering the slow growth potential of AOTs,[3] and the fact that the lesion was initially submitted to marsupialization, this may represent a residual tumor due to inappropriate treatment,[13] even though other cases were submitted to marsupialization followed by enucleation with complete resolution.[21-31] None of these cases were, however, followed up for more than 39 months.

Adenomatoid odontogenic tumor is a successional tooth-associated lesion which develops during the mixed dentition.[11] There has been considerable conjecture as to the nature of this lesion. Recently, KRAS p.G12V mutations were reported in a small cohort of AOT.[32] However, this finding does not prove a neoplastic nature for this tumor because even normal tissues can harbor somatic mutations. On the other hand, a neoplastic biological behavior cannot be discharged only because the tumor is unlikely to recur.

There are some cases described in the literature as multiple AOTs.[32-37] These cases are extremely rare and can be associated with the Schimmelpenning syndrome.[32,38] They were, therefore, not included in the analyses of the present review.

The limitations of the present study include, first, the retrospective nature of the included studies. Because of the retrospective nature of the study, we could not retrieve some relevant information usually associated with typical AOT, such as the presence of enamel hypoplasia and gubernaculum dentis.[11,39] Second, many of the cases have a short follow-up, which could have led to an underestimation

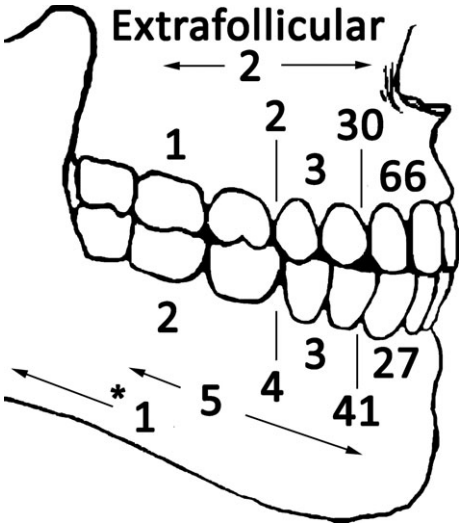

FIGURE 3 Topographical distribution of the known precise locations (n = 187) of extrafollicular adenomatoid odontogenic tumors (AOTs). Cases involving multiple regions (or an entire quadrant) are indicated between arrows. Numbers at the top and bottom of the lines indicate cases involving both adjoining regions: anterior/premolar and premolar/molar. One asterisk (*) indicates the number of lesions from the mandibular body that reached the angle and/or ramus. For the rest of the lesions (n = 60), the location was the "maxilla" (n = 4), "anterior maxilla" (n = 6), "posterior maxilla" (n = 14), "mandible" (n = 9), "anterior mandible" (n = 11), "posterior mandible" (n = 15), and "maxillary sinus" (n = 1). For those cases with available information, 16 out of 201 lesions (8.0%) crossed the midline of the jaws, and the maxillary sinus was significantly affected in 4 out 129 lesions (3.1%) in the maxilla

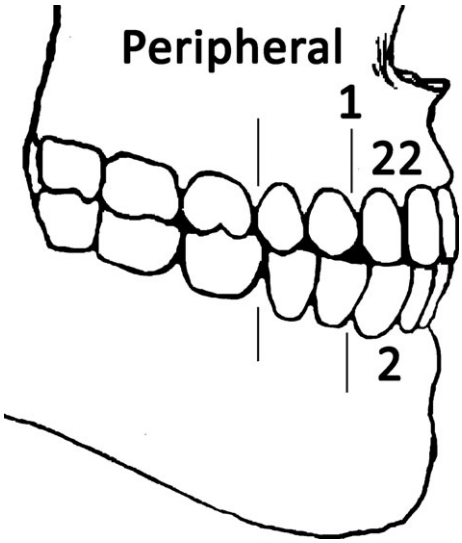

FIGURE 4 Topographical distribution of the known precise locations (n = 25) of peripheral adenomatoid odontogenic tumors (AOTs). Numbers at the top and bottom of the lines indicate cases involving both adjoining regions: anterior/premolar and premolar/molar. For the rest of the lesions (n = 5), the location was the "maxilla" (n = 4), and not available (n = 1). For those cases with available information, 3 out of 27 lesions (11.1%) crossed the midline of the jaws

of the virtually inexistent occurrence of recurrences. Third, the fact that most studies reported only one or a small number of patients followed up for a limited period of time.

5 | CONCLUSIONS

Adenomatoid odontogenic tumor variants do not show distinctive clinical and radiological features. Recurrence of AOT is very rare, which justify its conservative management.

CONFLICT OF INTERESTS

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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