



A Systematic Review of Adenoid Ameloblastoma: A Newly Recognized Entity

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Abstract

Background Recently, a new odontogenic tumor has been described, the so-called adenoid ameloblastoma (AdAM). The aim of this review was to determine the clinical and imaging features of AdAM and to describe its main histopathological findings.

Methods The systematic review included published cases with a diagnosis of AdAM in the gnathic bones, which had sufficient clinical, imaging, and histopathological data to confirm its diagnosis. The following histopathological diagnostic criteria were adopted: presence of ameloblastoma-like components, duct-like structures, spiral cellular condensations, and a cribriform architecture.

Results Fifteen articles, corresponding to 30 cases of AdAM, were selected. Most cases affected men (63.3%), with a slight preference for the mandible (16:14) and the posterior region of gnathic bones was the most commonly affected site. The mean age at diagnosis was 40.8 years. Clinically, the lesions usually presented as a swelling (53.3%) and, radiographically, as a well-defined radiolucency (33.4%). Surgical resection (40%) was the most frequently adopted treatment and recurrence occurred in 30% of cases. Microscopic examination showed cribriform areas in most AdAM cases (93.3%); duct-like structures and spiral cellular condensations were seen in 100% of the cases.

Conclusion The small number of reported cases, the existence of erroneous diagnoses, and the adoption of initial conservative management make it difficult to determine whether AdAM has a higher risk of recurrence or more aggressive biological behavior than conventional ameloblastomas.

Keywords Adenoid ameloblastoma · Odontogenic tumor · Ameloblastoma · Dentinoid

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Introduction

Ameloblastoma is a benign epithelial odontogenic tumor [1] originally described in 1827 by Cuzack and the publication of further details in 1868 by Broca [2]. It accounts for 13 to 58% of all odontogenic tumors [3], with an incidence rate of 0.92 cases/million people/year. Ameloblastoma shows a peak incidence in the third decade of life and a slight male preference. The mandible is the most affected site. The follicular and plexiform subtypes are the most common histopathological patterns [4, 5].

Clinically, ameloblastoma presents as a painless swelling of slow and expansive growth, which may imply significant morbidity for the patient [4, 6, 7]. Although being considered a benign neoplasm, ameloblastoma tends to be locally aggressive and the recurrence rates are high [8–10]. The adopted treatment depends on the clinicopathological type and the patient's systemic condition, with surgical excision being the mainstay treatment [11].

In 2022, with the update of the World Health Organization (WHO) Classification of Head and Neck Tumors, the term “conventional” was reintroduced and a new entity was recognized, the so-called adenoid ameloblastoma (AdAM) [12, 13]. The latter is characterized histopathologically by a cribriform architecture, the presence of duct-like structures, and the formation of dentinoid material [13]. Concerning their mutational profile, the absence of the BRAF mutation and the scarcity of investigations on other already known mutations in conventional ameloblastomas, such as SMO, represent a research gap in new studies on the molecular profile of these lesions [14, 15].

Several studies have reported cases of AdAM in the gnathic bones. However, further characterization of this tumor is necessary to better understand its etiopathogenesis and biological behavior. Thus, the present systematic review aimed to investigate the clinical and imaging findings of AdAM, as well as to describe its main histopathological findings reported in scientific literature.

Materials and Methods

This systematic review was registered in PROSPERO (Number: CRD42022344636) and was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [16].

Data Sources and Search Strategies

An electronic literature search, with no time or language restrictions, was performed between July and August 2022

in the following databases: PubMed/Medline, Web of Science, Science Direct, Scopus, Embase, and Cochrane Collaboration Library. We also searched the gray literature in OpenGrey, ProQuest, and Google Scholar. In addition, hand searches were performed by crossing the reference lists of the included articles. After identification, duplicate references were removed.

Following the population, intervention, comparison, and outcome (PICO) model, the following research question was formulated for this systematic review: “What are the most common clinical, imaging, and histopathological features of AdAM?”.

The search strategy was based on the combination of the following keywords: (“Adenoid ameloblastoma”[tw] OR “Adenomatoid ameloblastoma”[tw] OR “Dentinoameloblastoma”[tw] OR “Adenoid ameloblastoma with dentinoid”[tw] OR “Adenoid ameloblastoma with dentinoid”[tw] OR “Plexiform ameloblastoma with dentinoid”[tw] OR “Atypical plexiform ameloblastoma with dentinoid”[tw] OR “Dentinoameloblastoma with ghost cells”[tw]).

Eligibility Criteria and Study Selection

All published studies that described cases diagnosed as AdAM in the gnathic bones were eligible. The studies had to provide sufficient clinical, imaging, and histological data so that the diagnosis of AdAM could be confirmed. Randomized controlled clinical trials, cohort studies, case–control studies, cross-sectional studies, case series, and case reports were included. In vitro studies, cytological studies, review articles, and letters to the editor were excluded, unless any of these categories reported cases with sufficient clinical, imaging, and histological information to confirm the diagnosis.

The histopathological diagnostic criteria adopted for inclusion of the articles in the systematic review were the presence of (i) an ameloblastomatous component, (ii) duct-like structures, (iii) spiral cellular condensations, and (iv) cribriform architecture [12].

Four authors (HGFM, RICG, CSOC, and HFP) independently screened the titles and abstracts of all articles identified by the electronic searches. Next, the full texts of the studies that met the inclusion criteria the inclusion criteria were read. Six authors (HGFM, RICG, CSOC, HFP, RPM, and EFM) thoroughly evaluated the clinical, imaging, and histopathological features, as well as molecular findings, in order to confirm the diagnosis of AdAM. In the case of disagreement, the final diagnosis was reached by consensus. If the disagreement persisted, two other experienced pathologists evaluated the cases (MCCM and RAF).

Data Extraction and Analysis

Three of the authors independently assessed the articles eligible for data extraction (HGFM, RIG, and CSOC) and any disagreement was resolved by discussion. The following data were extracted from each article: (i) author(s); (ii) publication year; (iii) country where the study was conducted; (iv) number of reported cases; (v) age and sex of patient(s); (vi) tumor location; (vii) clinical features; (viii) imaging features; (ix) histopathological features; (x) molecular features; (xi) treatment; (xii) recurrence, and (xiii) follow-up period.

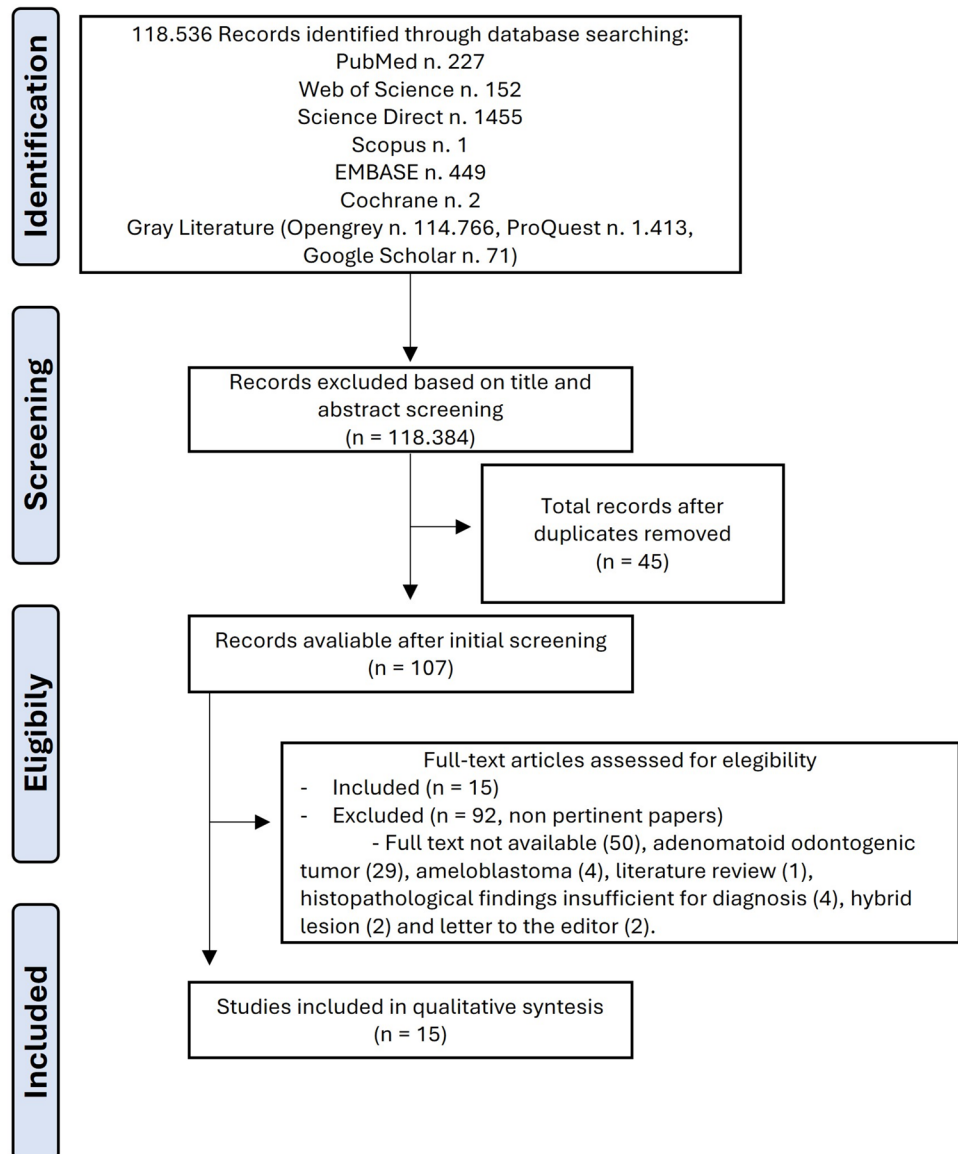
The methodological quality of the articles was evaluated according to the CARE guidelines (for CAse REports), a qualitative checklist for observational studies and case reports [17].

Results

Study Selection

The search strategy retrieved 118,536 articles from the different databases, including those identified by hand searching. After reading the titles and abstracts, 107 articles were potentially eligible and their full texts were assessed by six evaluators (HGFM, RIGG, CSOC, HFP, RPM, and EFM). After reading the full text of the pre-selected articles, 15 studies met all inclusion criteria, and were selected for this systematic review, with a total number of 30 reported cases of AdAM in the gnathic bones. Figure 1 shows the flowchart of the article selection process. All studies were published in English. The 30 reported cases selected for

Fig. 1 Flowchart of the study selection process



this review were from seven countries, covering four of the five continents, except for Oceania. Brazil was the country with the largest number of reported cases (Fig. 2). Detailed information on the studies selected for this systematic review is available in Supplementary Tables 1 and 2.

Clinical and Radiographic Features

Male patients were more commonly affected by AdAM (63.3%). The mean age was 40.8 years, ranging from 15 [18] to 70 years [19]. Only one patient was under 18 years of age [18]. The mandible was the most affected site, with a mandible/maxilla ratio of 16/14, and most tumors involved the posterior region of the mandible (Fig. 3).

Swelling was a frequent clinical finding (53.3%) in the AdAM cases included in this systematic review, whereas pain (13.3%) and paresthesia (10%) were less commonly described. Considering all cases, 30% of the tumors were larger than 3 cm and tooth displacement was reported in only 10% of the cases. Radiographically, AdAM mainly appeared as a well-defined radiolucency (33.4%). There was only one case of peripheral AdAM without bone involvement [20]. In addition, 20% of the cases showed cortical bone destruction.

Surgical resection was the most common treatment (40%) in the included studies. In two of the cases reported by Loyola et al. [21], radiotherapy was used as adjuvant therapy and neck dissection was also performed in one of these cases. Recurrence was a common finding, observed in 30% of cases. Table 1 describes the clinical and imaging data of all AdAM cases included in the present study.

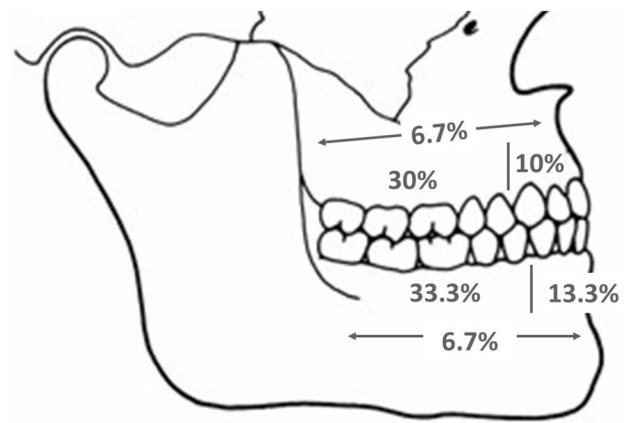


Fig. 3 Anatomical location of the cases diagnosed as adenoid ameloblastoma. The percentage of cases between the arrows indicates tumors located in the anteroposterior region of the mandible or maxilla

Histopathological and Molecular Features

Regarding histopathological features, 50% of the AdAM cases in our review exhibited the plexiform subtype as the predominant morphological pattern, followed by the concomitant presence of the plexiform and follicular patterns (36.7%). Histopathological features such as odontogenic epithelium resembling the stellate reticulum (93.3%) and a cribriform pattern (90%) were present in most of the included cases, while duct-like structures and spiral cellular condensations were found in 100% of the cases. Dentinoid material was present in 70% of the cases. Distinct cell morphologies were also reported, including the presence of clear

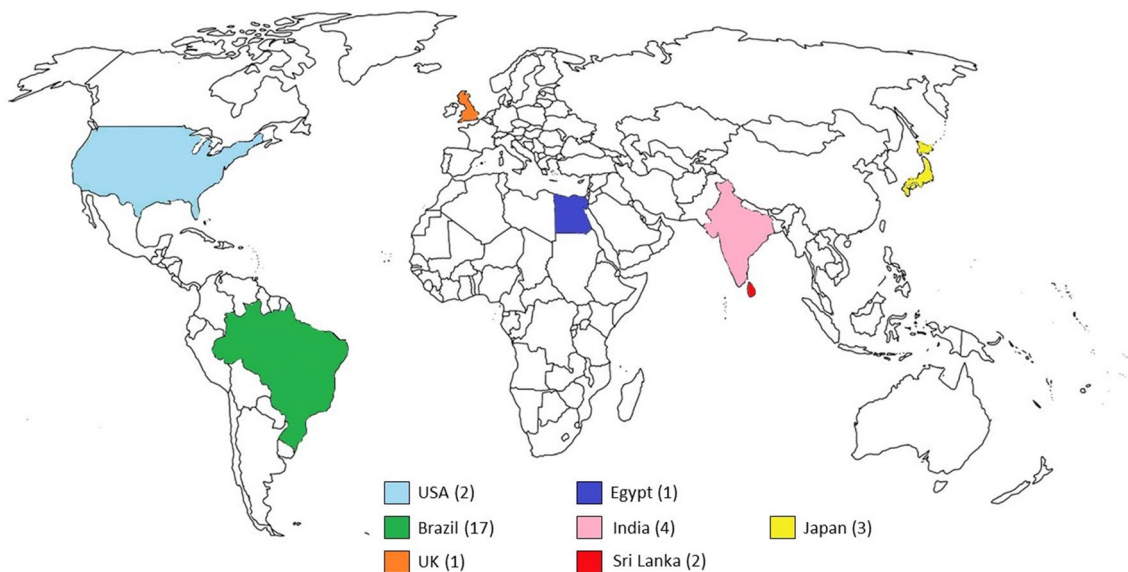


Fig. 2 Geographical distribution of adenoid ameloblastoma cases selected for the present systematic review

Table 1 Clinical–radiographic features of 30 cases of adenoid ameloblastoma reported in the literature included in the systematic review

Clinicopathological variable	<i>n</i>	%
Sex		
Female	11	36.7
Male	19	63.3
Age		
< 20 years old	3	10.0
20–49 years old	19	63.3
> 50 years old	8	26.7
Age mean (SD): 40.8 (± 12.4)		
Evolution time		
≤ 12 months	8	26.7
> 12 months	5	16.7
NI	17	56.7
Location of tumor		
Maxilla	14	46.7
Mandible	16	53.3
Jaw swelling		
Yes	16	53.3
No	1	3.3
NI	13	43.3
Pain		
Yes	4	13.3
No	5	16.7
NI	21	70.0
Numbness/paresthesia		
Yes	3	10.0
No	7	23.3
NI	20	66.7
Size of the tumor		
≤ 3 cm	3	10.0
> 3 cm	9	30.0
NI	18	60.0
Tooth displacement		
Yes	3	10.0
No	5	16.7
NI	22	73.3
Radiographic pattern		
Well-defined radiolucency	10	33.4
Ill-defined radiolucency	3	10.0
Radiolucency with radiopaque foci	4	13.3
No bone involvement	1	3.3
Not informed	12	40.0
Destruction of bone cortex		
Yes	6	20.0
No	4	13.3
NI	20	66.7
Treatment		
Enucleation	6	20.0
Surgical resection	12	40.0

Table 1 (continued)

Clinicopathological variable	<i>n</i>	%
Surgical resection + RT with or without neck dissection	2	6.7
NI	10	33.3
Recurrence		
Yes	9	30.0
No	6	20.0
NI	15	50.0
Follow-up		
≤ 5 years	9	30.0
> 5 years	5	16.7
NI	16	53.3

NI not informed, SD standard deviation, RT radiotherapy

cells (43.4%), ghost cells (23.3%), spindle cells (16.7%), and multinucleated giant cells (20%) (Table 2). Figure 4 illustrates some of the histopathological features described in our systematic review, which refer to a case of AdAM diagnosed at our oral pathology service.

Only 10 cases (33.3%) [19, 22] had their molecular profile investigated. Somatic mutations in FGFR2 and SMO were found in only one case (0.03%) [19] (Supplementary Table 2).

Quality Assessment

The results of the quality assessment of the selected studies are presented in Supplementary Table 3. Most articles adequately described the clinical characteristics (60%), diagnostic techniques used (100%), and therapy modalities (96.7%). However, only 46.7% of the articles reported follow-up data.

Discussion

Odontogenic tumors are an uncommon group of neoplasms that originate from odontogenic tissues and their remnants and that show a heterogeneous biological behavior. The correct diagnosis of these tumors is of the utmost importance for adequate treatment. However, despite advances in the diagnostic methods for odontogenic lesions, controversies regarding their etiopathogenesis, the paucity of information on histopathological and molecular features, and their unclear clinical behavior can make the diagnosis difficult [5, 9].

After several studies have endorsed AdAM to be a distinct entity [11, 22–27], the WHO recognized this tumor in its recent classification [13]. Underreporting of AdAM may exist and many cases may have been diagnosed as either ameloblastoma or adenomatoid odontogenic tumor (AOT),

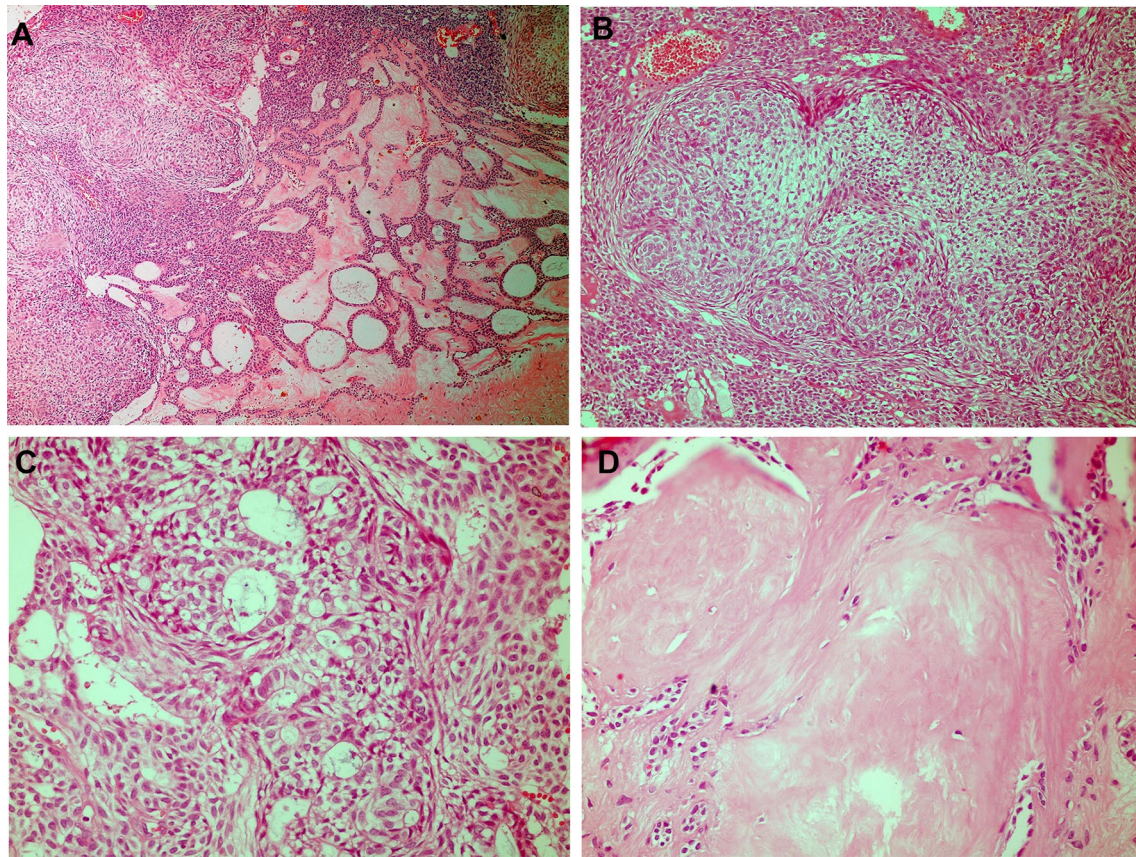


Fig. 4 Histopathological features of adenoid ameloblastoma. **a** Presence of an ameloblastomatous plexiform pattern and cribriform arrangement in solid areas (100×). **b** Spiral epithelial structures and

clear cells (200×). **c** Duct-like structures containing scant eosinophilic material (400×). **d** Dentinoid material near some clear cells (400×)

depending on the predominance of microscopic features of these tumors. The study by Loyola et al. [21] supports this possibility; in that study, 45 cases initially diagnosed as ameloblastomas had their histopathological slides reassessed and four of them were reclassified as AdAM. The current recognition of AdAM as a distinct entity and the better characterization of its histopathological and molecular features are expected to raise the awareness of pathologists and to increase the accurate identification of AdAM. Thus, we believe that more cases of AdAM will be reported over the next years.

This systematic review allowed to identify the clinicopathological and imaging findings of AdAM cases reported in the literature so far, which can be summarized as follows: AdAM was more frequent in male patients, with a mean age of 40.8 years, and the mandible was the most affected anatomical site, with a mandible/maxilla ratio of 16/14. Among cases with mandibular involvement, most occurred in the posterior region. Radiographically, a well-delimited radiolucent appearance predominated, but diffuse and multilocular features were also observed. It is noteworthy that the clinical and imaging features of AdAM are not pathognomonic.

Therefore, thorough histopathological analysis is essential for a correct diagnosis.

The histopathological findings considered to be essential for inclusion of the studies in our systematic review were the presence of an ameloblastomatous component, duct-like structures, spiral epithelial cellular condensations (resembling morulae), and a cribriform architecture. Dentinoid material, clear cells, ghost cells, and areas of calcifications may also be present in AdAM. The formation of dentinoid material in these cases is probably due to an inducing effect of the odontogenic epithelial component. Moreover, a reduction in vascular supply can promote degenerative changes, inducing the formation of a pseudoglandular pattern in large AdAMs [28].

Previous studies have described the molecular profile of AdAM. Immunohistochemical analysis usually shows overexpression of the Ki-67 proliferative marker. Furthermore, mutations in the BRAF and p.V600E genes, commonly identified in ameloblastomas, are absent in AdAM [11, 21]. These findings podem sugerir the view that AdAM is an odontogenic tumor distinct from conventional ameloblastoma. A recent study reported mutations in the SMO and

Table 2 Histopathological features of 30 cases of adenoid ameloblastoma reported in the literature included in the systematic review

Histopathological variable	<i>n</i>	%
Ameloblastomatous pattern		
Solid	1	3.3
Plexiform	15	50.0
Follicular	3	10.0
Plexiform + follicular	11	36.7
Papillary arrangement		
Yes	3	10.0
No	15	50.0
NI	12	40.0
Stellate reticulum epithelium		
Yes	28	93.3
No	0	0.0
NI	2	6.7
Cribriform pattern		
Yes	27	90.0
No	0	0.0
NI	3	10.0
Ductiform structures		
Yes	30	100.0
No	0	0.0
NI	0	0.0
Whorled cellular condensations		
Yes	30	100.0
No	0	0.0
NI	0	0.0
Dentinoid material		
Yes	21	70.0
No	6	20.0
NI	3	10.0
Squamous metaplasia		
Yes	7	23.3
No	4	13.3
NI	19	63.3
Cystic degeneration		
Yes	14	46.7
No	3	10.0
NI	13	43.3
Nuclear hyperchromatism		
Yes	20	66.7
No	0	0.0
NI	10	33.3
Clear cells		
Yes	13	43.4
No	7	23.3
NI	10	33.3
Ghost cells		
Yes	7	23.3
No	12	40.0
NI	11	36.7

Table 2 (continued)

Histopathological variable	<i>n</i>	%
Spindle cells		
Yes	5	16.7
No	7	23.3
NI	18	60.0
Multinucleated giant cells		
Yes	6	20.0
No	3	10.0
NI	21	70.0
Mitosis		
Yes	9	30.0
No	1	3.3
NI	20	66.7
Stroma		
Fibrous/collagenous connective tissue	10	33.3
Hyalinized connective tissue	1	3.3
NI	19	63.4
Inflammatory infiltrate		
Yes	20	66.7
No	0	0.0
NI	10	33.3
Concentric calcifications		
Yes	3	10.0
No	5	16.7
NI	22	73.3

NI not informed

FGFR2 genes in a single case of AdAM [19]. These mutations have been previously detected in some ameloblastomas [29, 30].

Sweeney et al. [14] identified SMO mutations in 39% of ameloblastomas analyzed and 46% harbored BRAF mutations, which tend to be mutually exclusive. The authors suggest that these alterations might define two independent genetic etiologies for ameloblastoma. However, further investigations are needed to conclude whether those mutations influence the development of AdAM and if they are sufficient to justify its recognition as a new entity according to its molecular profile.

Malignant transformation of AdAM has not been reported so far. However, Jayasooriya et al. [11] described a recurrent AdAM case with atypia and hypercellularity, which led to a diagnostic challenge between AdAM or odontogenic carcinoma with dentinoid. However, the mere presence of cellular atypia is not enough to confirm a malignant background [8]. Odontogenic carcinoma with dentinoid is another rare and poorly characterized odontogenic tumor that should be included in the differential diagnosis of AdAM. This low-grade malignant odontogenic neoplasm is histopathologically characterized by the presence of cords and islands of

eosinophilic or clear epithelial cells associated with dentinoid material and, less commonly, duct-like structures [31]. Atypia and perineural invasions are occasionally reported. The absence of epithelium similar to conventional ameloblastoma distinguishes it from AdAM.

Like ameloblastoma itself, AOT is the main differential diagnosis of AdAM. AdAM and AOT share some microscopic features, although the former can be distinguished by the presence of a typical ameloblastomatous component. Furthermore, in contrast to AOT, AdAM does not harbor the KRAS mutation [22, 32]. Thus, the importance of the differential diagnosis between the two tumors must be highlighted, especially because of the aggressiveness of AdAM as opposed to the indolent behavior of AOT.

In our systematic review, 30% of the cases diagnosed as AdAM relapsed. In particular, all cases reported by Loyola et al. [21] underwent surgical resection as primary therapy and the mean time to first recurrence was 9 months. Furthermore, Evans et al. [25] reported a case of recurrence 9 years after initial treatment. However, most recurrences occurred due to misdiagnosis of the lesion as AOT and subsequent conservative treatment. The likelihood of an inaccurate diagnosis can be attributed to the predominance of AOT-like areas upon histopathological analysis, leading to conservative treatment [21]. The biological behavior, prognosis, and appropriate treatment strategies for AdAM remain uncertain and careful and long-term follow-up is therefore essential.

Our results suggest that AdAM is a rare odontogenic tumor, with 30 cases reported to date and included in this study. It mostly affects men in a wide age range, commonly presenting as an asymptomatic and intraosseous growth, with a predilection for the posterior region of the mandible. Radiographically, they predominantly appear as a well-defined radiolucency. Histopathologically, AdAM may present different ameloblastoma morphological patterns with epithelial islands in a spiral arrangement, duct-like structures, and, more rarely, rosette-like structures, associated with dentinoid.

Conclusion

The differential diagnosis, especially with ameloblastoma and AOT, is essential for accurate diagnosis and, consequently, for the correct management of the tumor. Thus, a detailed histopathological analysis of the tumor is imperative considering the findings presented here. Since AdAM is a lesion recently recognized by the WHO (2022) and with few cases reported, this systematic review will help to consolidate information available in the literature. Despite that, it may be early to implement definitive standardized criteria for the diagnosis of AdAM. Eight cases developed multiple recurrences even after wide excision of the tumor, indicating

an aggressive behavior, although this needs further investigation. The few reported cases, erroneous diagnoses, and initial conservative treatment impair a clear conclusion about the actual behavior of AdAM. With the increasing number of cases reported over the last decade and the current recognition of AdAM as a separate entity by the WHO, longitudinal studies on this tumor evaluating its clinical behavior, as well as histopathological and molecular studies elucidating the genetic features of the neoplastic process, will be necessary. Such studies should provide surgeons and pathologists with the data needed for the correct diagnosis, management, and prognosis of this newly recognized tumor.

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Declarations

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Informed Consent For this type of study informed consent is not required.

Consent for Publication For this type of study consent for publication is not required.

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