




Original Article

## Clinicopathological and Demographic Characteristics of 171 Cases of Ameloblastoma: A 40-Year Retrospective Institutional Study in Maharashtra

Sarang Gautam Sonawane<sup>1</sup>, MDS, Sanpreet Singh Sachdev<sup>1</sup> , MDS, Manisha Ahire Sardar<sup>1</sup>, MDS, Tabita Joy Chettiankandy<sup>1</sup>, MDS, Jagdish Vishnu Tupkari<sup>1</sup>, MDS, Yogita Baghinath Adhane<sup>1</sup>, MDS

<sup>1</sup>Department of Oral Pathology and Microbiology, Government Dental College and Hospital, Mumbai, Maharashtra, India.



**\*Corresponding author:**

Sanpreet Singh Sachdev,  
Department of Oral Pathology  
and Microbiology, Government  
Dental College and Hospital,  
Mumbai, Maharashtra, India.

[sunpreetss@yahoo.in](mailto:sunpreetss@yahoo.in)

Received: 21 April 2023

Accepted: 08 June 2023

Published: 29 August 2023

**DOI**

10.25259/GJMPBU\_34\_2023

**Quick Response Code:**



### ABSTRACT

**Objectives:** Ameloblastoma (AM) is the most common benign odontogenic neoplasm. Changing trends and geographic variation necessitate the conduction of periodic demographic studies to update the existing demographic data pertaining to AM. The present research aims to further append the existing yet limited demographic data available on AM in India.

**Materials and Method:** One hundred and seventy-one cases of AM were identified out of 7862 departmental archival cases from 1980 to 2020. Demographic variables, clinical and radiographic features, as well as histopathological variants of AM were recorded and compiled. The various histopathological variants of AM were described according to their occurrence in different genders, age groups, and sites.

**Results:** An overall incidence rate of AM among all oral lesions was found to be 2.18% of head and neck lesions. About 61.40% ( $n = 105$ ) of cases comprised conventional AM (CAM), of which plexiform AM (21.64%) was the most common histopathological variant observed, whereas unicystic AM (UAM) constituted 37.43% ( $n = 64$ ) of the cases. About 86% of cases occurred in the mandible, particularly, in the posterior region (83.62%). Multilocular radiolucency was the most frequent radiographic mode of presentation in about 54.97% of the cases while 36.25% of cases presented as unilocular radiolucency.

**Conclusion:** AM constitutes about 2.24% of all head and neck lesions with a mean age of occurrence of 35.84 years. The luminal variant was found to be most common in cases of UAM, whereas plexiform was the most common variant observed in CAM. AM has a predilection to occur in the third decade and in males and exhibits a marked propensity to occur in the mandibular posterior region.

**Keywords:** Odontogenic tumors, Epidemiology, India, Multilocular, Plexiform

### INTRODUCTION

Disturbance in the complex process of interactions between biomolecules and genetic factors that are involved in normal tooth development can lead to the development of a distinctive spectrum of lesions confined to the oral cavity harboring them and are collectively termed odontogenic tumors (OTs).<sup>[1]</sup> Among the numerous entities described and periodically updated as OTs by the World Health Organization (WHO),<sup>[2,3]</sup> approximately 40–50% ( $n = 305^{[4]}$ ,  $n = 250^{[5]}$ ) are found to be AM, making it the most common odontogenic neoplasm in India.

This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-Share Alike 4.0 License, which allows others to remix, transform, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

©2023 Published by Scientific Scholar on behalf of Global Journal of Medical, Pharmaceutical, and Biomedical Update

At present, AM is defined by the WHO (2017) as “a benign intraosseous progressively growing epithelial odontogenic neoplasm characterized by expansion and a tendency for local recurrence if not adequately removed.” The WHO classification (2017) has simplified the classification of AMs into unicystic AM (UAM), conventional AM (CAM) (previously termed as solid/multicystic), and extraosseous/peripheral and metastasizing types.<sup>[3]</sup> The neoplastic odontogenic epithelium may proliferate into the lumen of a UAM (intraluminal) or it may infiltrate the cystic wall which has been described as its mural variant.<sup>[6]</sup>

The proliferating neoplastic odontogenic cells in CAM may exhibit a variety of histopathological patterns such as follicular, plexiform, papilliferous, or adenoid.<sup>[7,8]</sup> In addition, the stellate reticulum-like cells may undergo metaplastic transformation into squamous cells, granular cells, or basaloid cells.<sup>[9]</sup> The connective tissue stroma component may also exhibit various changes such as desmoplastic or hemangiomatic areas, or the formation of dentinoid material [Figure 1].<sup>[8,10]</sup> A single type of pattern predominates although occasionally multiple subtypes may be present in combination, which is termed mixed AM.<sup>[3]</sup>

The trends in the distribution of any lesion show variations regionally as well as temporally and thus periodic and long-term presentation of their demographic data ensures more accurate and updated information that is accessible to clinicians or researchers. The present research aims to further contribute to the existing yet limited demographic data available on AM from the Asian subcontinent, especially in a densely populated country such as India. Data pertaining to the occurrence of AM in different genders, age groups, sites, and histological types have been collected retrospectively from the institutional records of

the previous 40 years and descriptively represented in the subsequent text.

## MATERIAL AND METHODS

Ethical approval for the study was obtained from the Institutional Ethical Review Board. The departmental archival records of 1980–2020 were scanned for possible cases of AM. The respective H- and E-stained histopathological slides were confirmed and classified according to the globally accepted standard 4<sup>th</sup> Edition of the WHO (2017) classification of head and neck tumors.<sup>[3]</sup>

Demographic variables, clinical and radiographic features, as well as histopathological variants of AM were recorded and compiled on a Microsoft Office Excel Sheet (v 2019, Microsoft Redmond Campus, Redmond, Washington, United States). Data were subjected to statistical analysis using the Statistical Package for the Social Sciences (SPSS v 26.0, IBM) for descriptive statistics such as frequencies and percentages for categorical data, mean, and standard deviation (SD) for numerical data.

## RESULTS

### General prevalence and variants

A thorough scan of 7862 cases from archival records yielded 298 cases of OTs (3.79%). Among the OTs, 171 were diagnosed with AM. Thus, the overall incidence rate of AM among all oral lesions was found to be 2.18%. UAM constituted 37.43% ( $n = 64$ ) of the cases, while 61.40% ( $n = 105$ ) of cases comprised CAM. Peripheral AM constituted 1.17% ( $n = 2$ ) of the cases that occurred in the posterior region of the maxilla and mandible in two male patients [Table 1].

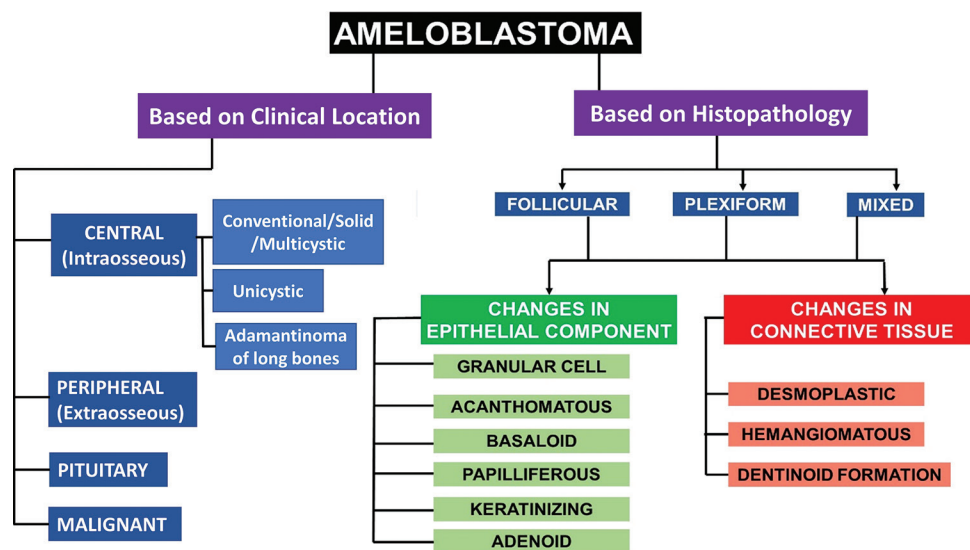


Figure 1: Classification of ameloblastoma based on clinical and histopathological modes of presentation.

**Table 1:** Number of cases of variants of AM based on demographic variables.

Variant of AM	Total number of cases		Gender (n)		Arch (n)			Side (n)		Region (n)			Radiographic features (n)		
	(n)	%	Male	Female	Mandible	Maxilla	Crossing midline	Left	Right	Anterior	Posterior	Unilocular radiolucency	Multilocular radiolucency	Mixed	
UAM															
Luminal UAM	31	18.13	11	20	28	3	11	7	13	12	19	17	14	0	
Intraluminal UAM	14	8.19	3	11	10	4	0	6	8	2	12	7	6	1	
Mural UAM	19	11.11	9	10	17	2	0	10	9	0	19	12	7	0	
Total	64	37.43	41	23	55	9	11	27	26	14	50	36	27	1	
Conventional ameloblastoma															
Plexiform AM	37	21.64	22	15	34	3	5	15	17	5	32	10	22	5	
Follicular AM	20	11.7	8	12	17	3	1	7	12	2	18	8	12	0	
Acanthomatous AM	19	11.11	11	8	15	4	3	8	8	3	16	4	15	0	
Mixed AM	15	8.77	9	6	14	1	1	8	6	0	15	2	12	2	
Desmoplastic AM	7	4.09	6	1	6	1	3	0	4	3	4	0	2	5	
Hemangiomatous AM	4	2.34	4	0	4	0	0	3	1	0	4	1	2	0	
Granular cell AM	2	1.17	0	2	1	1	1	1	0	1	1	0	2	0	
Basal cell AM	1	0.58	1	0	1	0	0	0	1	0	1	1	0	0	
Total	105	61.4	61	44	92	13	14	42	49	14	91	26	67	12	
Peripheral ameloblastoma															
Total	2	1.17	2	0	1	1	0	1	1	0	2	loss of lateral wall/horizontal bone loss			
TOTAL	171	100	104	67	148	23	25	70	76	28	143	62	94	13	

\*AM: Ameloblastoma, UAM: Unicystic ameloblastoma



Cases of UAM comprised 31 luminal, 14 intraluminal, and 19 mural variants. Among the cases of CAM, plexiform AM (21.64%,  $n = 37$ ) was the most common histopathological variant observed, followed by follicular AM (11.7%,  $n = 20$ ), acanthomatous AM (11.11%,  $n = 19$ ), desmoplastic AM (4.09%,  $n = 7$ ), hemangiomatous AM (2.34%,  $n = 4$ ), granular cell AM (1.17%,  $n = 2$ ), and basal cell AM (0.58%,  $n = 1$ ) [Figures 2 and 3]. Fifteen cases (8.52%) of CAM exhibited combined histopathological features of two or more variants and were considered as mixed AM.

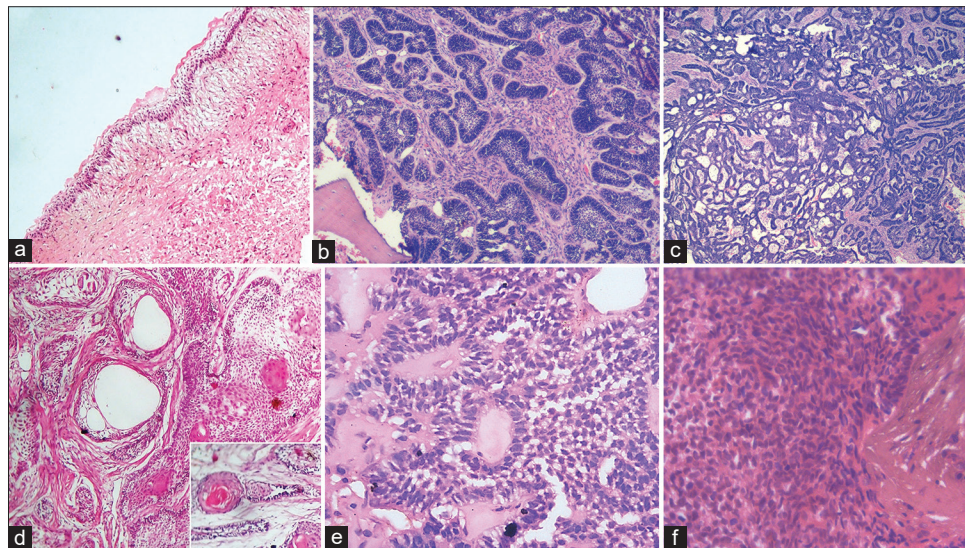
### Demographic distribution

The age of patients diagnosed with AM ranged from 10 to 98 years with a mean age of 35.84 years (SD = 24.929) and a median age of 31 years. The highest frequency of cases was observed in the third decade ( $n = 45$ ), followed by the second

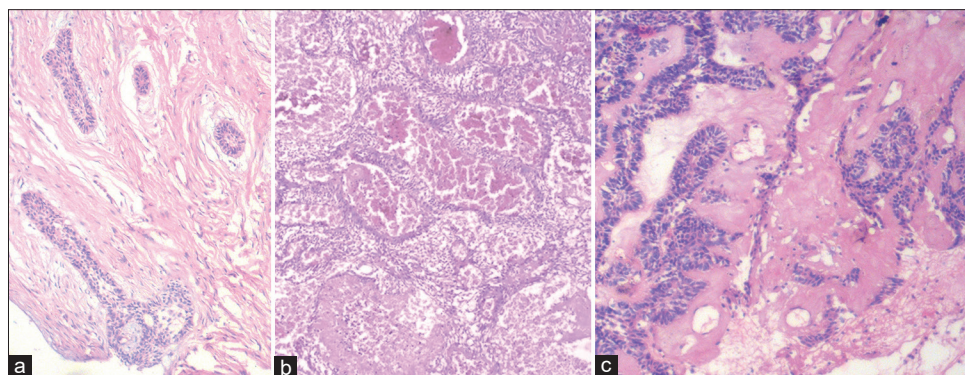
( $n = 34$ ), fifth ( $n = 34$ ), and fourth ( $n = 32$ ) decades. Only three cases were noted that occurred in the first decade, while the least number of cases ( $n = 2$ ) were noted in the tenth decade [Table 2]. A predilection toward the male gender (60.82%) was observed with the male-to-female ratio being 1.55:1 which was even more noticeable in cases of UAM, wherein the ratio was 1.78:1. Except for the follicular variant (M: F = 2:3), all the other subtypes of CAM exhibited slight male predilection. The desmoplastic variant occurred almost invariably in male patients (M: F = 6:1).

### Site of occurrence

The lesions exhibited a marked propensity to occur in the mandible (86.55%,  $n = 148$ ), and particularly in the posterior region (83.62%,  $n = 143$ ). Half of the lesions occurring in the anterior region ( $n = 28$ ) primarily comprised UAM ( $n = 14$ ).



**Figure 2:** H- and E-stained histopathological pictures of- (a). Unicyclic ameloblastoma (AM). ( $\times 10$ ), (b) Follicular AM ( $\times 10$ ), (c) plexiform AM ( $\times 10$ ), and (d) acanthomatous AM ( $\times 10$ ) (Inset: Formation of keratin pearl within ameloblastic follicle), (e) adenoid AM ( $\times 40$ ), and (f) basal cell AM ( $\times 40$ ).



**Figure 3:** H and E-stained histopathological pictures of- (a) desmoplastic ameloblastoma (AM). ( $\times 10$ ), (b) Hemangiomatous AM ( $\times 10$ ) and (c) dentinoid formation ( $\times 10$ ).

**Table 2:** Age-wise distribution of number of cases of AM.

Variant of AM	0–10 years	11–20 years	21–30 years	31–40 years	41–50 years	51–60 years	61–70 years	91–100 years	Total
UAM									
Luminal UAM	1	7	9	3	8	1	2	0	31
Intraluminal UAM	0	4	5	2	2	1	0	0	14
Mural UAM	0	2	7	3	3	1	3	0	19
Total	1	13	21	8	13	3	5	0	64
Conventional ameloblastoma									
Plexiform AM	2	8	12	5	4	3	1	2	37
Follicular AM	0	3	3	3	6	3	1	0	19
Acanthomatous AM	0	4	5	7	2	1	1	0	20
Mixed AM	0	4	1	5	2	1	2	0	15
Desmoplastic AM	0	1	1	3	2	0	0	0	7
Hemangiomas AM	0	0	2	1	1	0	0	0	4
Granular cell AM	0	0	0	0	2	0	0	0	2
Basal cell AM	0	0	0	0	1	0	0	0	1
Total	2	20	24	24	20	8	5	2	105
Peripheral ameloblastoma									
	0	1	0	0	1	0	0	0	2
Total	3	34	45	32	34	11	10	2	171

\*AM: Ameloblastoma, UAM: Unicystic ameloblastoma

Roughly, an equal number of cases occurred on the right (44.44%,  $n = 76$ ) and left (40.94%,  $n = 70$ ) sides of the jaw. Most of the subtypes showed an almost equal propensity for both sides, except for the follicular variant which had a slightly higher predilection to occur on the right side (12/20 cases). About 14.61% ( $n = 25$ ) of the lesions were extensive enough to cross the midline and involve both sides of the jaw.

### Radiographic presentation

Multilocular radiolucency was the most frequent radiographic mode of presentation in about 54.97% ( $n = 94$ ) of the cases. About 36.25% ( $n = 62$ ) cases exhibited a unilocular radiolucent picture, of which UAM accounted for more than half (57.14%). Nearly 40% of the cases of follicular AM and 27.02% of the cases of plexiform AM were unilocular. About 63.80% of the cases of CAM exhibited a multilocular radiolucent radiographic appearance. About 7.39% of the cases exhibited a mixed radiographic picture exhibiting the presence of radiopaque foci/areas, the majority of which were desmoplastic AM and plexiform AM ( $n = 5$  each, respectively). About 71.42% (5/7) of the cases of desmoplastic AM presented as mixed radiographic lesions. Cases of peripheral AM did not exhibit any peculiar radiographic features except for the loss of lateral wall or mild horizontal bone loss involving the associated teeth.

### DISCUSSION

The incidence rate of OTs (3.79%) was found to be slightly higher as compared to the previous extensive demographic

studies conducted in India by Nalabolu *et al.* (2.17%),<sup>[11]</sup> Chawla *et al.* (2.5%),<sup>[12]</sup> and Selvamani *et al.* (3.4%).<sup>[13]</sup> Few demographic studies in India have reported even greater incidence rates of 4.1%<sup>[14]</sup> and 4.29%.<sup>[4]</sup> However, these studies included keratocystic OT (KCOT) which comprised a significant portion of OTs in their results. In the 4<sup>th</sup> Edition of the WHO classification of head and neck tumors (2017), KCOT has been considered as a cystic lesion, termed as odontogenic keratocyst (OKC), and thus, the actual incidence rate of OTs as elicited from these studies may be much lower.<sup>[3]</sup> The incidence of OTs as high as 12.9% in earlier studies that excluded OKC further supports this fact.<sup>[15]</sup>

The observed predominance of AM (57.38%) among other OTs was also slightly higher than previous studies that recorded values of 45.72%<sup>[12]</sup> and 53%.<sup>[13]</sup> The percentage of AM has been reported to be even lower in Western countries (12–14%).<sup>[16]</sup> The lower percentage of AM reported in various studies would actually increase when KCOT is disregarded from their results.<sup>[17]</sup> This fact is reinforced by the results of demographic studies before the classification of OKC as a tumor in 2005 that demonstrated much higher incidence rates of AM (70%).<sup>[15]</sup>

Previous studies have reported CAM to comprise about 55–65% of AMs similar to our results.<sup>[12,13,17,18]</sup> Although studies have found the mural variant to be the most frequent among cases of UAM, our findings suggest that luminal UAM (48.44%, 31/64 cases) was more common.<sup>[12,19,20]</sup> The intraluminal variant of UAM was invariably the least common in all these previous reports from demographic

studies in accordance with our findings (21.88%, 14/64 cases). UAM is generally considered less aggressive, warranting only conservative treatment. On the contrary, due to the more aggressive clinical course and higher chances of recurrence, the WHO, in 2017, recommended that UAM with mural infiltration should be treated equally as CAM.<sup>[3]</sup>

Rarely, AM may occur in soft tissues of the gingiva or edentulous alveolar ridge which has been included as AM, extraosseous/peripheral type in the WHO (2017) Classification. Peripheral AM accounts for 1–10% of all types of AM and may occur at any age.<sup>[21]</sup> It tends to occur in relatively older individuals due to the prevalence of edentulousness in patients in these age groups. Two cases of peripheral AM, occurring in the second and fifth decades, respectively, were noted in our study.

Our findings corroborate global demographic data that have also found a similar mean age of around 30–35 years with the peak incidence of AM in the third decade.<sup>[22]</sup> Similar to findings from previous studies, the majority of cases (65.34%) in our study occurred in the third to fifth decades.<sup>[4,11]</sup> However, UAM had a tendency to occur a decade earlier with the peak incidence in the second decade ( $n = 21$ , 44.68%). Demographic data from the previous studies pertaining to AM have invariably identified a male predilection with M: F ratio ranging from 1.2:1 to 2.1:1.<sup>[12,13,17,18]</sup>

AM has displayed a marked propensity to occur in the mandibular jaw globally across multiple studies with nearly 90% of cases occurring in the mandible.<sup>[13,17,18]</sup> The ratio of cases occurring in mandible to maxilla in our study was found to be 6.43:1, whereas the previous studies have found this ratio similar or even as high as 10:1.<sup>[22,23]</sup> A similarly high propensity was observed in our findings as well as across multiple studies.<sup>[13,24]</sup> There are two plausible explanations for odontogenic lesions occurring in the posterior region of the mandible. First, the developing tooth germs are located at a relatively lower level as compared to those in the anterior region of the jaw. Thus, when the lateral lamina separates the developing dental follicle from the overlying epithelium, there is a greater possibility for remnants that could potentially contribute to the development of odontogenic lesions in this region.<sup>[25]</sup> Second, since odontogenic lesions tend to be associated with an impacted tooth, it would only be rational that the majority would be noted in association with the mandibular third molar, which is the most commonly impacted tooth.<sup>[26]</sup>

All the radiographs in our records were obtained in the form of orthopantomograms, which are crucial when dealing with such lesions that tend to involve significant portions of tooth-bearing and other related areas of the jaw. A greater percentage of UAM presented as a unilocular radiolucent lesion, while multilocularity is typically observed in the radiographic picture of CAM.<sup>[27]</sup> Furthermore, we observed

that lesions of smaller size were unilocular and those of larger size were multilocular. It could, thus, be extrapolated that larger, more aggressive lesions exhibit a compartmented appearance with septae of bone extending into the radiolucent tumor mass.<sup>[27,28]</sup> Additional features such as displacement of teeth and root resorption of adjacent teeth may also be noted.<sup>[27]</sup> At times, long-standing lesions may become extensive enough to cause pathological fractures of the involved jaw [Figure 4].

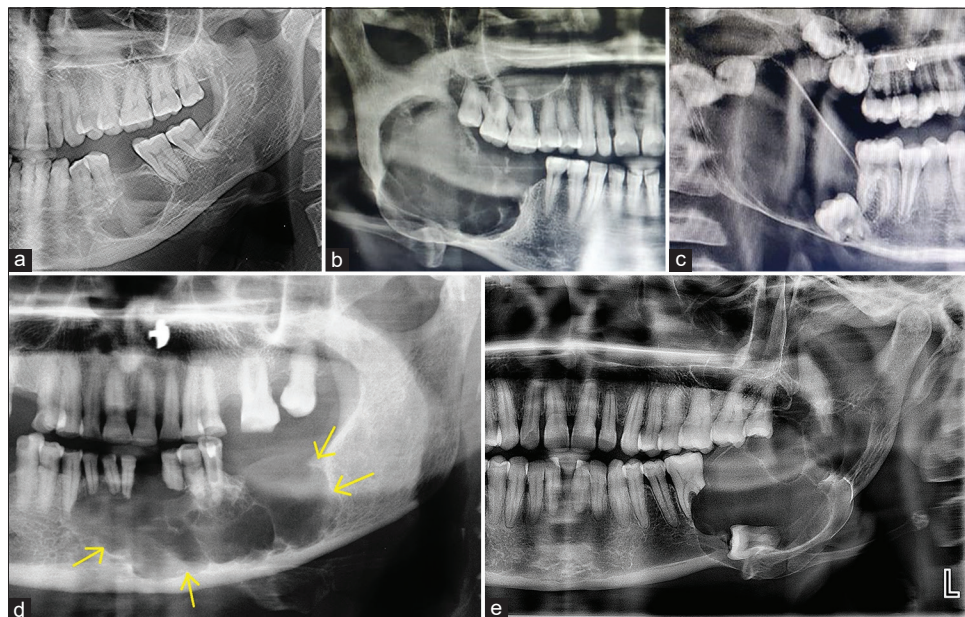
CAM may present itself in several histopathological patterns. However, not much clinical significance was derived depending on the histopathological type of CAM, and thus, these variants were commonly grouped under AM in the 4<sup>th</sup> edition of the WHO Classification of OTs (2017).<sup>[3]</sup> Plexiform and follicular have been variably stated as the most predominant variants across multiple clinicopathological studies similar to our findings with plexiform (21.64%,  $n = 37$ ) being the most frequently observed followed by follicular (11.70%,  $n = 20$ ). Other variants such as acanthomatous, desmoplastic, granular cell, hemangiomas, basal cell, and papilliferous occurred less frequently.<sup>[12,17-19]</sup>

The “potentiality” of a cell is the sum of all its latent capabilities, including every capability that will ever be expressed by the progeny of that cell but that cell itself may not yet have had the occasion to manifest.<sup>[29]</sup> The potentiality of neoplastic odontogenic cells is retained which enables them to differentiate into such varying types of cells having different compositions and functions.

Acanthomatous AM is characterized by squamous metaplasia occurring in the central stellate reticulum-like cells present in the ameloblastic follicles and plexuses. Thus, it should be considered as a product of changes in epithelial components in the primary types of CAM rather than being a distinct variant. Although the previous studies have reported a relatively lower percentage (7–10%) of the acanthomatous variant, we found it (11.11%,  $n = 19$ ) to be almost as common as the follicular variant (11.70%,  $n = 20$ ).

Infrequently, the stellate reticulum-like cells may exhibit transformation into granular cells (3–10%) and basaloid cells (0–2%).<sup>[12,19,30,31]</sup> Their rarity was evident in our study as well, wherein only two cases of granular cell and one case of basal cell variants were observed. Abundant clear cells may be noted among the neoplastic cell population on histopathological examination of AM in some cases. The cell population in a lesion may exhibit clear cell changes due to various reasons such as paucity of organelles, accumulation of substances (e.g., mucin), tumor progression/senescence, or fixation artifacts.<sup>[32]</sup> The term “clear cell AM” is now obsolete and such lesions are regarded as clear cell odontogenic carcinoma according to the 4<sup>th</sup> edition of the WHO (2017) Classification of Head and Neck tumors with nearly 100 well-documented cases to date.<sup>[3]</sup> Therefore, two such





**Figure 4:** Orthopantomograms of cases of ameloblastoma exhibiting (a) unilocular radiolucency, (b) multilocular radiolucency, (c) displacement of mandibular second and third molars with thinning of the inferior border of the mandible, (d) extensive lesion crossing the midline and involving both sides of the mandible, and (e) pathological fracture of the inferior border of mandible with a displacement of the involved mandibular second molar.

cases exhibiting abundant clear cells with AM-like areas in histopathology were excluded from our study.

The derivation of OTs is not always entirely epithelial and may also extend to involve the ectomesenchymal, and/or mesenchymal elements that are or have been a part of the tooth-forming apparatus.<sup>[33]</sup> Various complex epithelial-mesenchymal interactions occur in normal tooth development that is responsible for its morpho-differentiation and histogenesis<sup>[34]</sup> Such interactions inevitably occur in OTs and, thus, account for certain changes noted in the connective tissue surrounding the neoplastic epithelium.

The most common change in the connective tissue is manifested in the form of desmoplasia of collagen fibers that leads to compression of the epithelial component. Such changes, when noted in AM are described as desmoplastic AM which is a rare variant comprising 3–10% of cases. Desmoplastic AM was listed as a separate entity in the 3<sup>rd</sup> edition of the WHO Classification of OTs (2005).<sup>[33]</sup> However, in the recent classification introduced in 2017, it has been regarded as a histopathological variant of AM with no special implications in clinical course or treatment.<sup>[3]</sup> Earlier studies have demonstrated an equal tendency for desmoplastic AM to occur in the anterior region of either of the jaws which was reinforced in our results with only one out of the seven cases occurring in the mandibular posterior region.<sup>[35]</sup>

Hemangiomas AM represents a relatively unusual variant noted in 5–10% of cases of AM. The variant is characterized

by the replacement of stromal components by vascular tissue having numerous endothelial-lined capillaries or spaces filled with blood.<sup>[36,37]</sup> The entity was first described by Kuhn in 1932 as a combination of AM with hemangioma and was later termed adamantinohemangioma by Aisenberg (1950).<sup>[36]</sup> The development of abnormal vascular components may be attributed to the exaggerated granulation tissue response resulting from a disturbance in the repair of neoplastic odontogenic tissue. There were only four cases of this rare variant in our results that occurred invariably in the posterior mandibular region of male patients. The previous studies also support the finding that the hemangiomas variant of AM has the propensity to occur in the mandibular posterior region with a male predilection.<sup>[38]</sup>

Admixed histopathological types and also multiple variations in epithelial and stromal components can be found within the same lesion, and these have been reported as mixed AM.<sup>[3]</sup> Our finding that about 14.28% of cases of CAM exhibited mixed histopathological patterns is in concordance with the previous reports that have suggested that about 7–20% of cases of AM comprise mixed microscopic patterns.<sup>[18,39,40]</sup> However, most of the tissue specimens received in academic institutions comprise incisional biopsies and the patient is referred to specialized multidisciplinary treatment care centers for the excision of tumors. Therefore, the clinicopathologic, demographic, and diagnostic data available in institutional records are majorly constituted from tissues submitted after incisional biopsy procedures, which holds

true for the present study as well. A meticulous observation of an entirely excised specimen may reveal different areas of an AM lesion exhibiting varied histopathological patterns. Thus, the actual percentage of AM comprising admixed histopathological types can be expected to be much higher.

Besides exhibiting a composite histopathological picture of its own variants, AM has also been reported to coexist with other distinct odontogenic cysts/tumors within the same lesion. Such lesions are represented as a spectrum of hybrid OTs. One such peculiar hybrid OT comprising combined histopathological features of adenomatoid odontogenic tumor (AOT) and AM along with the presence of dentinoid material has been termed as “Adenoid AM with Dentinoid” (AAD). The lesion was first described by Slabbert *et al.* in 1992 and termed AAD by AFIP in 1994.<sup>[41,42]</sup>

Data from one of our systematic reviews on AAD have revealed that only 29 cases of AAD have been reported to date. The majority of these have been reported in the past decade due to the increasing awareness amongst pathologists with respect to hybrid lesions.<sup>[43]</sup> The lesion is not recognized as a distinct entity in the WHO classification yet, but with the increasing number of reported cases and subsequent studies demonstrating its clinical significance, it is likely to be included in future classification systems. Since the present study classifies lesions based on the present 2017 WHO classification system, we have included four such lesions into the category of AM which predominantly comprised its histopathological picture (two mixed AM and two plexiform AM, respectively).

Rarely, an AM may metastasize with the most common site being the lungs (70%), lymph nodes (28%), and bone (12%).<sup>[44,45]</sup> Many institutions work majorly on incisional biopsy and operate on minor neoplastic lesions, whereas malignancies of higher grades are referred to oncology centers for further treatment. A complete diagnostic work-up is then carried out in these higher centers and therefore, an accurate estimate of metastasizing AM may not be obtained from institutional records. This constitutes a limitation for such institutional demographic or clinicopathological studies such as the present study which does not have data available pertaining to metastasizing lesions.

Long-term demographic studies conducted in institutions may also present certain other limitations. Various factors such as differences in the request form and changing personnel over time may lead to the inadequacy of certain information in many cases. Due to the lack of data available pertaining to the size and consistency of lesions in many cases, the data were not included in the present study. Standardization of request forms and their uniform incorporation in all geographic areas can ensure completeness and uniformity of data gathered in such studies. In addition, data pertaining to the outcome of the treatment, recurrence, and present status

of the disease in many cases was not sufficiently available owing to lack of adequate follow-up.

Nevertheless, the demographic, radiographic, and histopathological data would contribute to updating the available information on AM on a national level. Emphasis needs to be laid on the creation of multi-stage registries for OTs at a local level which could then be collectively included in national- or global-level databases. Such databases would allow researchers and pathologists to easily access the available data and current trends of various entities for research and clinical purposes.

## CONCLUSION

Although many of our findings overlapped with earlier studies with respect to the parameters in question, certain significant differences were observed which would help pathologists and surgeons to obtain clarity with respect to various subtypes and variants of AM while adding detailed information to the existing data on the subject. AM constitutes about 2.24% of all head and neck lesions with a mean age of occurrence of 35.84 years. The luminal variant was found to be most common in cases of UAM, while plexiform was the most common variant observed in CAM. AM has a predilection to occur in the third decade and in males and exhibits a marked propensity to occur in the mandibular posterior region. Findings from our study would serve to update available information on AM, pertaining to its demographic and clinicopathological characteristics. The data could also contribute to future geography-based research work or that intended to obtain data on a larger scale.

## Declaration of patient consent

Patient's consent not required as there are no patients in this study.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

## REFERENCES

1. Datta A, Goyal P, Maiti S, Ngjelo A, Hiremath A, Kumar K. Odontogenesis, molecular basis of odontogenesis and its relation with common odontogenic cysts and OTs: A review. *J Adv Dent Med Sci Res* 2020;8:5-9.
2. Kramer IR, Pindborg JJ, Shear M. WHO International Histological Classification of Tumours: Histological Typing of Odontogenic Tumours. 2<sup>nd</sup> ed. Heidelberg: SpringerVerlag; 1992.



3. Takata T, Slootweg PJ. Ameloblastoma. In: ElNaggar AK, Chan JK, Grandis JR, editors. WHO Classification of Head and Neck Tumours. 4<sup>th</sup> ed. Lyon: IARC; 2017. p. 215-8.
4. Deepthi PV, Beena VT, Padmakumar SK, Rajeev R, Sivakumar R. A study of 1177 odontogenic lesions in a South Kerala population. *J Oral Maxillofac Pathol* 2016;20:202-7.
5. Sriram G, Shetty RP. OTs: A study of 250 cases in an Indian teaching hospital. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:e14-21.
6. Reichart P, Philipsen H. Odontogenic Tumors and Allied Lesions. London, UK: Quintessence Publishing Co Ltd.; 2004. p. 41-116.
7. Sisto JM, Olsen GG. Keratoameloblastoma: Complex histologic variant of ameloblastoma. *J Oral Maxillofac Surg* 2012;70:860-4.
8. Loyola AM, Cardoso SV, de Faria PR, Servato JP, Eisenberg AL, Dias FL, *et al.* Adenoid ameloblastoma: Clinicopathologic description of five cases and systematic review of the current knowledge. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2015;120:368-77.
9. Sivapathasundharam B, Rajendran R. OTs. In: Shafer's Textbook of Oral Pathology E-book. 8<sup>th</sup> ed. India: Elsevier Health Sciences; 2020. p. 100-15.
10. van Rensburg LJ, Thompson IO, Kruger HE, Norval EJ. Hemangiomatous ameloblastoma: Clinical, radiologic, and pathologic features. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001;91:374-80.
11. Nalabolu GR, Mohiddin A, Hiremath SK, Manyam R, Bharath TS, Raju PR. Epidemiological study of odontogenic tumours: An institutional experience. *J Infect Public Health* 2017;10:324-30.
12. Chawla R, Ramalingam K, Sarkar A, Muddiah S. Ninety-one cases of ameloblastoma in an Indian population: A comprehensive review. *J Nat Sci Biol Med* 2013;4:310-5.
13. Selvamani M, Yamunadevi A, Basandi PS, Madhushankari GS. Analysis of prevalence and clinical features of multicystic ameloblastoma and its histological subtypes in South Indian sample population: A retrospective study over 13 years. *J Pharm Bioallied Sci* 2014;6:S131-4.
14. Lima-Verde-Osterne R, Turatti E, Cordeiro-Teixeira R, Barroso-Cavalcante R. The relative frequency of odontogenic tumors: A study of 376 cases in a Brazilian population. *Med Oral Patol Oral Cir Bucal* 2017;22:e193-200.
15. Simon EN, Stoelinga PJ, Vuhahula E, Ngassapa D. OTs and tumor-like lesions in Tanzania. *East Afr Med J* 2002;79:3-7.
16. Daley TD, Wysocki GP, Pringle GA. Relative incidence of OTs and oral and jaw cysts in a Canadian population. *Oral Surg Oral Med Oral Pathol* 1994;77:276-80.
17. Siar CH, Lau SH, Ng KH. Ameloblastoma of the jaws: A retrospective analysis of 340 cases in a Malaysian population. *J Oral Maxillofac Surg* 2012;70:608-15.
18. Patsa S, Jadv RB, Halder GC, Ray JG, Datta S, Deb T. Demographic and histopathological variation of ameloblastoma: A hospital-based study. *J Oral Maxillofac Pathol* 2016;20:230-3.
19. Intapa C. Analysis of prevalence and clinical features of ameloblastoma and its histopathological subtypes in Southeast Myanmar and Lower Northern Thailand populations: A 13-year retrospective study. *J Clin Diagn Res* 2017;11:C102-6.
20. Tatapudi R, Samad SA, Reddy RS, Boddu NK. Prevalence of ameloblastoma: A three-year retrospective study. *J Indian Acad Oral Med Radiol* 2014;26:145-51.
21. Philipsen HP, Reichart PA, Nikai H, Takata T, Kudo Y. Peripheral ameloblastoma: Biological profile based on 160 cases from the literature. *Oral Oncol* 2001;37:17-27.
22. Avelar RL, Primo BT, Pinheiro-Nogueira CB, Studart-Soares EC, de Oliveira RB, de Medeiros JR, *et al.* Worldwide incidence of OTs. *J Craniofac Surg* 2011;22:2118-23.
23. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, *et al.* Odontogenic tumours: A retrospective study of 1642 cases in a Chinese population. *Int J Oral Maxillofac Surg* 2007;36:20-5.
24. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: Biological profile of 3677 cases. *Eur J Cancer B Oral Oncol* 1995;31B:86-99.
25. Nanci A. Development of the Tooth and its supporting tissues. In: Tenca's Oral Histology. 9<sup>th</sup> ed. China: Elsevier; 2018. p. 68-90.
26. Mortazavi H, Baharvand M. Jaw lesions associated with impacted tooth: A radiographic diagnostic guide. *Imaging Sci Dent* 2016;46:147-57.
27. White S, Pharaoh M. Odontogenic epithelial tumors. In: Oral Radiology- Principles and Interpretation. 6<sup>th</sup> ed. China: Mosby Elsevier; 2009. p. 373-85.
28. Kalkur C, Halim N, Sattur A, Burde K, Naikmasur V. Radiographical approach to multilocular radiolucent lesions of the jaws: A review. *EC Dent Sci* 2019;18:410-20.
29. Cormack DH. Ham's Histology. 9<sup>th</sup> ed. Philadelphia, PA: J. B. Lippincott Co.; 1987. p. 173-4.
30. Taneeru S, Guttikonda VR, Yeluri S, Madala J. Granular cell ameloblastoma of jaw - Report of a case with an emphasis on its characterization. *J Clin Exp Dent* 2013;5:e154-6.
31. Kazakydasan S, Zamhari AK, Achol LT. Basal cell ameloblastoma in a paediatric patient: A case report and review of oral basal cell tumours. *Oral Surg* 2019;12:248-54.
32. Jain A, Shetty DC, Juneja S, Narwal N. Molecular characterization of clear cell lesions of head and neck. *J Clin Diagn Res* 2016;10:E18-23.
33. Barnes L, Everson WJ, Reichart P, Sidradinsky D. Pathology and Genetics. Head and Neck Tumors. 5<sup>th</sup> ed. Lyon: IARC Press; 2005.
34. Ruch JV, Lesot H, Bègue-Kirn C. Odontoblast differentiation. *Int J Dev Biol* 1995;39:51-68.
35. Savithri V, Janardhanan M, Suresh R, Kumar RV. Desmoplastic ameloblastoma with osteoplasia: Review of literature with a case report. *J Oral Maxillofac Pathol* 2013;17:298-301.
36. Kasangari MD, Gundamaraju K, Jyothsna M, Subash AV, Aravind K. Hemangiomatous ameloblastoma- a case report of a very rare variant of ameloblastoma. *J Clin Diagn Res* 2015;9:D08-10.
37. Sharma VK, Verma SK, Goyal L, Chaudhary PK. Hemangiomatous ameloblastoma in maxilla: A report of a very rare case. *Dent Res J (Isfahan)* 2012;9:345-9.
38. Venigalla A, Bojji M, Piniseti S, Babburi S. Hemangiomatous ameloblastoma: Case report with a brief review. *J Oral Maxillofac Pathol* 2018;22:S24-8.
39. Gunawardhana KS, Jayasooriya PR, Rambukewela IK, Tilakaratne WM. A clinico-pathological comparison between mandibular and maxillary ameloblastomas in Sri Lanka. *J Oral*

- Pathol Med 2010;39:236-41.
40. Hertog D, Bloemena E, Aartman IH, van-der-Waal I. Histopathology of ameloblastoma of the jaws; some critical observations based on a 40 years single institution experience. *Med Oral Patol Oral Cir Bucal* 2012;17:e76-82.
  41. Slabbert H, Altini M, Crooks J, Uys P. Ameloblastoma with dentinoid induction: Dentinoameloblastoma. *J Oral Pathol Med* 1992;21:46-8.
  42. Brannon RB. Adenoid Ameloblastoma with Dentinoid. Washington, D.C: Armed Forces Institute of Pathology; 1994. p. 1-94.
  43. Sachdev SS, Chettiankandy TJ, Sardar MA, Adhane Y, Shah AM, Grace AE. Adenoid ameloblastoma with dentinoid: A systematic review. *Sultan Qaboos University Med J* 2022;22:325-38.
  44. Dissanayake RK, Jayasooriya PR, Siriwardena DJ, Tilakaratne WM. Review of metastasizing (malignant) ameloblastoma (METAM): Pattern of metastasis and treatment. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2011;111:734-41.
  45. Kunze E, Donath K, Luhr HG, Engelhardt W, De Vivie R. Biology of metastasizing ameloblastoma. *Pathol Res Pract* 1985;180:526-35.

**How to cite this article:** Sonawane SG, Sachdev SS, Sardar MA, Chettiankandy TJ, Tupkari JV, Adhane YB. Clinicopathological and demographic characteristics of 171 cases of ameloblastoma: A 40-year retrospective institutional study in Maharashtra. *Glob J Med Pharm Biomed Update* 2023;18:18