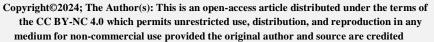
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RESEARCH ARTICLE

AMELOBLASTOMA IN POPULATION OF YEMEN: ANALYZING THE PREVALENCE AND CLINIC PATHOLOGIC FEATURES OF AMELOBLASTOMA IN A YEMENI POPULATION

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Abstract

Background and aims: Ameloblastoma is an uncommon tumor of odontogenic epithelium (ameloblasts, or the outer part of the teeth during development) that can be benign or malignant. It usually appears in the lower jaw rather than the upper jaw. The initial ameloblastoma is often treated with radical tumor resection. Analyzing the prevalence and clinic pathologic features of ameloblastoma in a Yemeni population was the goal.

Material and Methods: This retrospective cross-sectional study was carried out in all patients who were diagnosed with oral tumors at the targeted centers in Sana'a city (Al-Mamoon Center, Central Laboratory, and Al-Awlqy Laboratory Centers) in the period June 2020 to June 2022. This study is based on data taken from patients's files that include age, gender, location, and histopathology. The study used descriptive statistics and SPSS version 26 for data analysis, describing quantitative non-parametric data using median and parametric data using mean and standard deviation.

Results: The ameloblastoma tumor prevalence was 8.8% of all oral tumors (330 cases). The mean age of the ameloblastoma patients was $31.10\pm11.4~(\pm~SD)$ years and ranged from 11 to 80 years, with 1:1.2 males to females. The majority of ameloblastoma cases occurred in the lower jaw (93.1%; N=27), and the posterior part of the lower jaw was the main site for ameloblastoma tumors (79.3%; N=23), and 86.2%; N=23 of ameloblastoma was epithelial origin and 13.8%; N=4 was variegated origin. In terms of histological type, 65.5%; N=19 of the cases were not diagnosed, and the remaining cases showed a rate of 24.1%; N=7 for a multicystic tumor and 10.3%; N=3 for a unicystic tumor. 96.6%; N=28 of ameloblastoma cases were benign tumors, and only 3.4%; N=1 case was a malignant tumor.

Conclusions: The prevalence of ameloblastoma was significant, and these findings increase the awareness level of determent type of lesion for better diagnosis, management, and preventing measures.

 $\textbf{Keywords:} \ \text{Ameloblastoma, odontogenic tumors, or al tumors} \ \ \text{prevalence, Yemen.}$

INTRODUCTION

Odontogenic tumors are defined as unique neoplasms exclusively of the jaw bones and oral mucosa, which are derived from odontogenic epithelial tissue and/or odontogenic ectomesenchymal tissue or both that constitute the tooth-forming apparatus¹.

Ameloblastoma (AB) is one of the most common types of odontogenic tumors^{2,3}. It's distinguished by local invasiveness, tooth displacement, malocclusion, and pathological fracture, with a predisposition for gross

facial deformity, a high rate of recurrence, incidental metastasis, and malignant transformation⁴. It comprised approximately 1% of all jaw tumors and 11% of odontogenic tumors (OT)^{2,5,6}. Manifestation of AB is generally limited, and symptoms are nonspecific. They usually show a painless swelling of the involved region of the jaw. Pain is caused by ulceration in the relative soft tissue⁷. Predominating is detected by chance in radiographs taken for other reasons or during routine examination^{8,9}. While displaying histological characteristics of benignity, this particular condition

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exhibits infiltrative growth patterns that possess significant local destructive capabilities. As a consequence, it frequently obligates radical therapeutic interventions. This tumor can continue if they are not surgically removed to grow to reach massive dimensions¹. The ideal surgical approach for treating AB should aim to minimize the tumor reoccurrence, restore both functional and aesthetic aspects, and result in minimal morbidity in the donor site if any. The planning of the surgical procedure should take into consideration the patient's underlying health conditions, the size and location of the tumor, the available techniques for reconstruction, and the surgeon's level of expertise¹⁰. There is limited knowledge regarding potential etiological factors; occasionally, AB is associated with follicular cysts, impacted teeth, and ectopic teeth⁷. Information derived from every individual particular with regards to their health-related sufferer by AB is required to be reformulated, which should include age, gender, location, size of tumor, histologic type, radiologic form, treatment of AB, reconstruction, and preoperative and post-operative complications were collected from medical reports and reviewed and analyzed retrospectively¹¹. According to the archives of the National Information Center Library in Yemen, many studies have been conducted on cancers in Yemen, including the correlation of the Epstein-Barr virus with breast cancer¹², bladder cancer¹³, renal tumors14, tumors and histological recognition in the operated patients¹⁵, and the prevalence of parotid tumors¹⁶, but there has been no study conducted in Yemen to investigate the prevalence of ameloblastoma (AB) among the Yemeni population.

Therefore, this study aimed to know the prevalence of ameloblastoma (AB) among oral cancer patients and its association with gender and age group, to determine the locations of tumors in the jaws, to study the origin of ameloblastoma, to determine the histological type of ameloblastoma (multicystic tumor/unicystic tumor) and whether it is a benign or malignant tumor.

MATERIALS AND METHODS

Study design: A retrospective study.

Study population: All case reports of patients with oral tumors (reports of biopsies) at the targeted centers in Sana'a city (Al-Mamoon Center, Central Laboratory, and Al-Awlaqy Lab Centers) during the period of the study (June 2020-June 2022). Those laboratory centers were selected as they considered the main laboratory centers in Yemen, where most pathological lesion biopsies were transmitted.

Study site: This study was carried out in Sana'a city. **Sample size:** 330 case reports of oral tumors were found, 29 of which were ameloblastomas.

Inclusion criteria: All biopsy reports of cases diagnosed as oral tumors; all biopsy reports with complete data, including age, gender, anatomical location, and histopathological pattern; all biopsy reports received during the period of the study (June 2020-June 2022).

Exclusion criteria: All biopsy reports with incomplete data of the inclusion criteria, all biopsy reports of cases diagnosed with pathological condition other than oral tumor, all biopsy reports not received during the period of the study (June 2020-June 2022).

Data collection: All biopsy reports that met the inclusion criteria were collected. Data were collected in a data sheet (case sheet), including age, gender, anatomical location, and histopathological pattern. The data collection sheet was designed to have a methodological recording.

Statistical method: Data were described using suitable descriptive statistics. Data analysis was achieved using SPSS version 26. The data were described in as numbers and percents. The quantitative non-parametric data were reported by described in terms of mean and standard deviation (after utilizing Shapiroe-Wilk for testing normality). The threshold of significance was fixed at the 5% level. When $p \le 0.05$, results were considered significant.

Ethical approval: Ethical approval was obtained from the Medical Ethics Committee of the Faculty of Dentistry, Sana'a University, reference number 6/2020, dated June 1, 2020, and all data, including the patient's identity, were kept confidential.

RESULTS

Oral tumor type distribution: The result of oral tumor type distribution showed that the largest number of patients were squamous cell carcinoma (SCC), with a total number of (N=111; 33.6%) of the total patients, followed by lymphoma with a total number of (N=41;12.4%). Ameloblastoma (AB) tumor was in third place with a total number of (N=29; 8.8%) of the total patients. The last group was with (Warthing's tumor), (lymphoma), and (clear cell carcinoma of odontogenic origin), with a total number of (N=1; 0.3%) of the total patients (Table 1).

Prevalence of ameloblastoma: The largest number of non-AB oral tumors was 91.2%; N=301, while the remaining 8.8%; N=29 were AB tumors. The malignant type of ameloblastoma was only (N=1; 0.5%), while N=211; 99.5% had other oral malignancies, of the total (212) patients with oral malignant tumors. The distribution result showed that the benign type of ameloblastoma was (N=28; 23.7%), while other benign tumors were in (N=90; 76.3%) of the total (118) patients with benign tumors (Table 2). **Gender distribution of AB cases:** N=16; 55.2% of the AB cases were male, and the remaining (N=13; 44.8%) were females.

Age distribution of AB cases: The age ranged from 11 to 80 with a mean \pm SD of 31.10 \pm 11.4 years. Nine cases of AB were in the age group 21–30 years (31.0%), followed by the age group 31–40 years with (N=8 cases; 27.6%), and only (N=2 cases; 6.9) in the age group more than 50 years (Table 3).

Site distribution of AB cases: Most of the AB cases were in the mandible (N=27 cases, 93.1%) of the total AB cases, and only (N=2 cases;6.9%) were in the maxilla.

Table 1: Distribution of different oral tumors among Yemeni population attending to the main pathological diagnostic laboratory.

patnological diagnostic laboratory.			
Tumor types	N (%)		
Squamous cell carcinoma (SCC)*	111(33.6)		
Lymphoma	41 (12.4)		
Ameloblastoma (AB)	29 (8.8)		
Odontogenic Fibroma	17 (5.2)		
Pleomorphic adenoma	15 (4.5)		
Spindle cell tumar	11 (3.3)		
Giant cell grannloma	8 (2.4)		
Round cell tumor	8 (2.4)		
Fibro osseons	8 (2.4)		
Malignant undifferentiated tumor	8 (2.4)		
Pyogenic Granuloma	7 (2.1)		
Verrucons carcinoma *	7 (2.1)		
Adenoid cystic carcinoma*	7 (2.1)		
Basal cell carcinoma*	7 (2.1)		
Fibro myxomatous tumor	5 (1.5)		
Lipoma	5 (1.5)		
Papilloma	5 (1.5)		
Capillary hemangioma benigh	5 (1.5)		
Lymphangioma	5 (1.5)		
Cemento-ossifying fibroma	4 (1.2)		
Mucoepidermoid carcinoma*	4 (1.2)		
Odontogenic Myxoma	3 (0.9)		
Wing sarcoma*	3 (0.9)		
Osteosarcoma*	2 (0.6)		
Osteoma	2 (0.6)		
Warthing tumor	1 (0.3)		
Paraganglioma	1 (0.3)		
Clear cell odontogenic carcinoma*	1 (0.3)		
Total	330 (100)		

*Malignant

The origin distribution of AB: Most of the AB tumors originated from the epithelial tissue (N=25 cases, 86.2%), while (N=4 cases; 13.8%) of AB were mixed originated tissue.

AB histological feature: The result of the AB histology feature illustrated in Table 4. The greatest

number of the AB cases was in (Unk) with a total number of (N=19; 65.5%), followed by solid multicystic (N=7 cases; 24.1%) of the total cases, while follicular and plaxiform were less frequent (N=3 cases, 10.3% for each one). Unicystic feature present in (N=3 cases; 10.3%), mural and luminal in (N=1 case; 3.4%) for each feature (Table 4).

Site of AB in mandible: The result of the site of AB in mandible was that most of AB was in the posterior region of the mandible (body-Ramus-Post-Angle) with (N=23; 79.3%), while Ant only represents (N=2; 6.9%) cases. Body site occurred in (N=10 cases; 34.5%). Also, Ramus site of AB occurred in (N=7 cases; 24.1%) and Post site occurred in (N=5 cases; 17.2%), while (N=4 cases; 13.8%) site occurred in unk. The last site was angle, which occurred in (N=1 case; 3.4%) of the total patients (Table 5).

Behavior of AB: The greatest number of AB tumors was benign, counting (N=28 cases; 96.6%); malignant AB tumors occurred only in (N=1 case:3.4%) of the total 29 patients.

DISCUSSION

One of the most prevalent odontogenic epithelial tumors, AB can exhibit a variety of biological characteristics, including simple cystic expansion, malignant transformation, and aggressive solid masses with a high risk of recurrence and local invasiveness. The AB continues to be a hotly debated topic due to its extreme biologic behavior. The purpose of the current investigation was to ascertain the prevalence of AB in Yemeni society. There were two primary factors that went into choosing this topic: First, the National Information Center Library's archive in Yemen indicates that no prior research on the same topic has been done among Yemenis.

Table 2: Prevalence of ameloblastoma among oral tumors and Odontogenic tumors.

Oral tumors	N (%)	Odontogenic tumors	N (%)
AB	29 (8.8)	Ameloblastoma (AB)	25 (42.4)
Others	301 (91.2)	Odontogenic Fibroma	17 (28.8)
Total	330 (100)	Fibro myxomatous tumor	5 (8.5)
Malignant	tumors	Cemento-ossifying fibroma	4 (6.8)
AC	1 (0.5)	Odontogenic Myxoma	3 (5.1)
Others	211 (99.5)	AB Fibroma	3 (5.1)
Total	212 (100)	Class sell adamtassa:	1 (1.7)
Benign tumors		Clear cell odontogenic carcinoma	1 (1.7)
AB	28(23.7)	Carcinoma	
Others	90 (76.3)		
Total	118 (100)	AB fibroma odontoma	1 (1.7)
Total		Total	59 (100)

In addition, the lesion linked to impacted teeth is more common in our social circles. Research on this potentially fatal lesion is necessary to determine its severity and possible asymptomatic characteristics. The study's second goal is to distinguish between the lesions that affect Yemeni patients and the incidence of AB in those patients based on factors such as age, gender, location, and histology. As a result, this study conducted independent research (in Yemen) with the goal of creating a database of oral and maxillofacial

surgery and comparing the information with numerous reports from other parts of the globe.

Table 2 of the current study shows that the prevalence of ameloblastoma (AB), which accounts for 42.4% of odontogenic tumors (OT), was 8.8% of tumors involving the maxillofacial region. 8.8% of AB out of all oral cancers fall into the previously observed range of 1–18% as reported by Siar *et al.*¹⁷, in the Malaysian population and by Valente Pires *et al.*¹⁸, in Barazil. With a frequency of 42.4% of epithelial AB and 49.2%

of all AB, ameloblastoma was found to be the most common odontogenic tumor in the current investigation.

Table 3: Distribution of different age groups of ameloblastoma patients.

differential participation		
Age	N (%)	
1 - 10 years	0 (0.0)	
11 - 20 years	5 (17.2)	
21 - 30 years	9 (31)	
31 - 40 years	8 (27.6)	
41 - 50 years	5 (17.2)	
More than 50 years	2 (6.9)	
Total	29 (100)	

These findings are consistent with previous reports^{19,20}. Nonetheless, Taher *et al.*²¹, found a lower prevalence (34.6%) of AB among odontogenic malignancies. Furthermore, 16.30% of AB to odontogenic tumors were found in the Barazil investigation¹⁸. According to reports, AB is more common in Nigeria and accounts for 63% of all OT in Southwest Nigeria. Adebayo *et al.*²², conducted a five-year retrospective assessment in

North-West Nigeria and discovered that 73% of all odontogenic tumors were AB²². With a little male predilection, the gender distribution of the patients was sixteen men (55.2%) and thirteen girls (44.8%), or a ratio of 1.2:1. These results corroborate those of another investigation^{7,23,24} that discovered a male with modest predominants by ratio 1.2:1. On the other hand, Chae *et al.*²⁵, reported that the female tendency was (2,67:1). But according to most writers^{2,6,9,26-29} there was no sex predominance.

The age range of participants in the current study was 11–80 years. The second decade (31%) and the third decade (27.6%) had the highest prevalence of AB. According to Siar *et al.*¹⁷, the Malaysian patients showed the highest in the second decade, which is comparable to our finding. Numerous research 6,9,23,26,29-31 that documented the peak age of AB incidence in the third decade of life were similar to our study. Zaidi *et al.*², reported a rise in peak incidence to the fourth decade. It is important to note that Hendra *et al.*²⁷, looked into the prevalence of AB worldwide.

Table 4: Distribution of ameloblastoma by histological feature.

			0
	AB histology feature	N (%)	Total N (%)
Multicystic	Follicular	3 (10.3)	7 (24.1)
7(24.1%)	Plexiform	3 (10.3)	7 (24.1)
Unicystic	Mural	1 (3.4)	3 (10.3)
3(10.3%)	Luminal	1 (3.4)	
	Unk	19 (65.5)	19 (65.5)
	Total	29 (100)	29 (100)

According to Hendra et al.27, the incidence peaked in Europe and North America between the fifth and sixth decades, whereas it was lowest in Africa and South America during the third decade. Socioeconomic factors may be the cause of these variations in peak occurrence, as noted by Reichart et al.32, in developing nations, AB tends to occur at a younger age, accelerated aging due to malnutrition, and challenges in accessing the healthcare system, all of which affect peak occurrence. Regarding the anatomical position of AB, where AB in the mandible 93.1% outweighed those in the maxilla 6.9% and posterior of the jaw (body 34.5%, Ramus 24.1%, posterior area 17.2%, and angle 3.4%), this study showed commonalities with most prior investigations. As shown in Table 4^{6,9,29,30}, 33,34, the total (79.3%) is more than the anterior of the mandible (6.9%).

Table 5: Location of AB tumors in mandible.

Location in mandible		N (%)	Total
			N (%)
Ant		2 (6.9)	2 (6.9)
	Body	10 (34.5)	
Dogt	Ramus	7 (24.1)	23
Post	Post	5 (17.2)	(79.3)
	Angle	1 (3.4)	
Unknown		4 (13.8)	4 (13.8)
Total	•	29 (100)	29 (100)

The maxilla is most commonly affected in the mandible, with all cases (6.9%) finding in the posterior region and none in the anterior, which may be due to the previously mentioned small sample size. The primary lesion in the current investigation was epithelial origin, with the majority of research (86.2%) pointing to similar findings³⁵ citing a small number of mixed lesion instances without any meaningful analysis. Regretfully, the majority of biopsy reports (65.5%) did not include information regarding the histological characteristics or the type of AB. Nonetheless, it was discovered that the conventional/multicystic type (24.1%) had the highest number among the remaining reports (34.5%), followed by the unicystic type (10.3%).

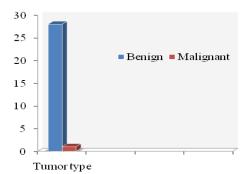


Figure 1: The distribution of AB according to behavior (benign/malignant).

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The most common types were plexiform (10.3%) and follicular (10.3%), followed by mural (3.4%) and luminal (3.4%). This result is consistent with the findings of other investigations^{25-27,30,31,36-38}. While the predominant site of AB was the posterior region, which was thought to be the right or left side, the lesion site in the current study, which was located in the mandible, was localized in the posterior region (body, ramus, and angle), which is identical with most studies^{7,27}. Only one case of amelobastic carcinoma (AC), a malignant kind of lesion, was found in this investigation (3.4%); yet, as previously described²⁷, the benign variety predominates with the greatest number of cases. Remarkable: a male patient's front mandibular region was the site of an exclusive case of malignant AB in the fifth decade. However, this result can be flawed since the MA was misdiagnosed; a CT scan requires additional testing to identify this. Additionally, the patient in the second decade experienced one case recurrence four times over the course of seven years, with the location being in the mandibular, in contrast to Liu et al.³⁹, who reported the existence of recurring 17.2% of the total 1626 cases.

Limitation of the study

The fact that the current study is retrospective in nature and that medical records sometimes, if at all, do not adequately record factors even in situations that could be described is one of its main weaknesses. It is difficult to interpret results using historical data since it is impossible to know for sure if the patients being diagnosed are typical of all (seemingly) comparable individuals. Retrospective studies continue to have a number of significant drawbacks, such as inadequate recording, missing data, and incomplete or absent documentation, but as a methodology, they still have a number of benefits.

CONCLUSIONS

The prevalence of ameloblastoma (AB) in the Yemeni population is as follows: ameloblastoma constitutes 8.8% of all oral tumors, 23.7% of benign tumors, 49.2% of dental tumors, and 0.5% of malignant cancers. Melanoma was found to be most common in the second and third decades of life; the mandible was the most common site of ameloblastoma, accounting for 93.1% of cases. The most common histological types of ameloblastoma were follicular and plexiform variants.

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DATA AVAILABILITY

The accompanying author can provide the empirical data that were utilized to support the study's conclusions upon request.

AUTHOR'S CONTRIBUTIONS

Al-Thobhani SS: Writing the original methodology, investigation. Da'er SAA: Conceptualization. AL-Haddad KA: Writing, review and editing, Al-Moyed KA: Formal analysis, data processing. Al-Kibsi TAM, Al-Shamahy HA: Formal analysis, data processing, conceptualization. Final article was checked and approved by all authors.

CONFLICT OF INTEREST

There are no conflicts of interest in regard to this project.

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