Demographic Variation of Ameloblastoma: A Retrospective Analysis of 188 Cases in the Indonesian Population

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Abstract

Ameloblastoma is a benign and locally invasive odontogenic tumor originating from remnants of the tooth-forming components. The clinicopathologic features of ameloblastoma in each population in a country can differ with respect to sex, age, and clinical and radiographic features.

To retrospectively evaluate the demographic profiles of patients diagnosed with ameloblastoma in the Indonesian population. A retrospective study of 188 patients with histologically confirmed ameloblastoma was carried out in Dr. Cipto Mangunkusumo National Central Public Hospital, Jakarta, from January 2012 to April 2022. Among the 188 patients, 90 (48.7%) were male and 98 (51.3%) were female, with most of them being in the third to fifth decades of their lives. The majority of tumors measured 4–10 cm (116, 60.7%) and had a dominant growth period of more than 24 months (71, 37.2%). The majority of tumors were located in the mandible (95.3%) and the rest in the maxilla (4.7%), with the ramus and angle of the mandible being the most affected subsite (39.8%).

The most common tumor pattern in our study was the mixed type (57.6%). Distribution patterns according to sex, age, tumor characteristics, tumor subtypes, and surgical treatment in the Indonesian population were in line with other reported series.

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Introduction

Ameloblastoma is a benign odontogenic tumor that usually arises within the jaw.[1] In the recent World Health Organization classification of odontogenic tumors, ameloblastoma was defined as benign and locally invasive epithelial odontogenic neoplasm originating from remnants of the tooth-forming components such as developing enamel, rests of the dental lamina, epithelial linings of dentigerous cysts and basal epithelial cells of the oral mucosa.[2,3] Overall, this tumor accounts for 13%–58% of all odontogenic tumors.[4] This tumor can be classified into microscopic types such as follicular, plexiform, acanthomatous, granular cell, desmoplastic, and basal cell types; however, the follicular type is the most common and recognizable.[5] Surgery is the first choice of therapy considering the high recurrence rate of this tumor.[6,7] Surgical treatment of ameloblastoma can be divided into conservative treatment (enucleation, radical curettage, and cryosurgery) and radical treatment (margin of segmental resection).[6]

Indonesia, a populous country in Southeast Asia with over 270 million people, is a multiracial nation consisting of hundreds of distinct ethnic groups, with the Javanese group being the largest.[8] Many studies have described the demographic profile of ameloblastoma in one or more country[1]; however, there is a paucity of articles on the Indonesian population. However, the clinicopathologic features (sex, age, clinical findings, and radiographic features) of ameloblastoma in some Asian countries differ from those in other continents. The differences may be accounted partially for by ethnic influences and accessibility to medical services.[9,3]

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The paucity of data motivated us to conduct a comprehensive review of the demographic and epidemiologic features of ameloblastoma in the Indonesian population. In particular, data on sex, age, race, risk factor, nutritional status, history of systemic disease, blood type, social & psychological status, local status, surgical treatment, histopathological subtypes, DMFTs score, and OHIs score were evaluated. Data on tumor characteristics, including the size, growing period, site, expansion, and aspiration results were also analyzed. This study aimed to retrospectively evaluate the demographic and epidemiologic profiles of patients diagnosed with ameloblastoma.

Materials and methods

A retrospective study of 188 patients with histologically confirmed ameloblastoma was carried out in Dr. Cipto Mangunkusumo National Central Public Hospital, Jakarta, from January 2012 to April 2022. Institutional review board approval was waived for this study. Each patient's sex, age, race, risk factor, nutritional status, history of systemic disease, blood type, social & psychological status, local status, surgical treatment, and histopathological subtypes were obtained from case summaries accompanying biopsy specimens. Furthermore, other data were also analyzed considering tumor characteristics such as the size, growth period, site, expansion, and aspiration result. We also data on the DMFT score and Oral Hygiene Index (OHI) of each patient. The frequencies and percentages of these variables were calculated. Statistical analyses using SPSS 25 were performed on the collected data and the results were formulated. Correlation analyses, using the chi-square test, were performed between sets of variables; i.e., the site of the tumor and the DMFT score, the blood type and the histopathological subtypes, and the growing period and tooth resorption. The threshold for statistical significance was set at $P < 0.05$.

Results

A total of 188 cases of histologically confirmed ameloblastoma were analyzed in this study. Ninety patients (47.1%) were males and 98 (51.3%) were females [Table 1]. The male-to-female ratio was 1:1.08. The majority of patients in this study 92 (48.2%) were above 34 years, followed by the 13–34 years age group (38.7%) and the below 19 years age group (11.5%). The most abundant ethnic groups were the Javanese 70 (36.6%), followed by the Sundanese 66 (34.6%), and others 51 (26.7%).

Gender distribution n (%)	*Male*		*Female*	
	90 (48.7%)		98 (51.3%)	
Age group distribution n (%)	*<19 years*		*19–34*	*>34 years*
	22 (11.5%)		74 (38.7%)	92 (48.2%)
Race distribution n (%)	*Javanese*		*Sundanese*	*Others*
	70 (36.6%)		66 (34.6%)	51 (26.7%)
Risk factor distribution n (%)	**Alcohol drinking**			
	Yes		*No*	
	4 (2.1%)		184 (96.3%)	
	Smoking habit			
	Yes		*No*	
	53 (27.7%)		135 (70.7%)	
Nutritional status n (%)	*Underweight*		*Normal*	*Overweight*
	27 (14.1%)		69 (36.1%)	92 (48.2%)
Social and psychological status n (%)	**Occupation**			
	Working		*Student*	*Not working*
	87 (45.5%)		33 (17.3%)	68 (35.6%)
	Education			
	High school		*University education*	
	161 (84.3%)		27 (14.1%)	
	Psychological status			
	Calm		*Anxious*	
	137 (71.7%)		48 (25.1%)	
History of systemic diseases n (%)	*Diabetes mellitus*		*Hypertension*	
	10 (5.2%)		30 (15.7%)	
Blood Type n (%)	*A*	*B*	*AB*	*O*
	46 (24.1%)	47 (24.6%)	13 (6.8%)	82 (42.9%)
Local status n (%)	**Association of impacted teeth**			
	Yes		*No*	
	66 (34.6%)		122 (63.9%)	
	Root resorption			
	Yes		*No*	
	130 (68.1%)		58 (30.4%)	
DMFT score n (%)	*Low*		*Moderate*	*High*
	105 (55%)		53 (27.7%)	30 (15.7%)
OHIS score n (%)	*Poor*		*Moderate*	*Good*
	119 (62.3%)		50 (26.2%)	19 (9.9%)

Table 1. Distributions of gender, age group, race, risk factor, nutritional status, social and psychological status, history of systemic diseases, blood type, local status, DMFT score, and OHIS score.

The majority of participants were smokers (53, 27.7%) and 184 (96.3%) of them did not drink alcohol. Most of the patients were overweight (92, 48.2%) followed by normal weight (69, 36.1%) and then underweight (27, 14.1%). Most of the patients were workers (87,

45.5%), (68, 35.6%) were non-workers, and the rest were students (33, 17.3%). The highest level of education attained was high school in 161 (84.3%) patients and university education in 27 (14.1%) patients. At the first meeting, as many as 137 (71.7%) patients looked calm and 48 (25.1%) looked anxious.

Ten (5.2%) patients had a history of type II diabetes mellitus and 30 (15.7%) had a history of hypertension. The most dominant blood type was 'O' (82, 42.9%), followed by 'B' (47, 24.6%), 'A' (46. 24.1%), and 'AB' (13, 6.8%). Impacted teeth were found in 66 (34.6%) patients. A total of 130 (68.1%) cases had root resorption as confirmed by radiographic imaging.

We also performed DMFT and OHIS scoring analyses to track the progression of dental caries and further reflect the deterioration of oral hygiene in patients with ameloblastoma. We found that 105 (55%) patients had low DMFT scores, 53 (27.7%) had moderate scores, and 30 (15.7%) had high scores. A total of 119 (62.3%) patients had poor OHIS scores, 50 (26.2%) had moderate scores, and 19 (9.9%) had good scores.

Tumor characteristics

The majority of tumors (60.7%) measured 4–10 cm, 41 (21.5%) measured <4 cm, and 31 (16.2%) measured >10 cm. Further analyses revealed that the growth periods varied, with most tumors (63, 33%) developed in 12–24 months, 71 (37.2%) developed over 12 months, and 54 (28.3%) developed in less than 12 months. Most of the tumors (76, 39.8%) were located in the angle and ramus of the mandible, 54 (28.3%) in the parasymphysis, 49 (25.7%) in the body and angle of the mandible, and 9 (4.7%) in the maxilla.

Tumor characteristic				
Size	*<4cm*	*4–10cm*	*>10cm*	
	41 (21.5%)	116 (60.7%)	31 (16.2%)	
Growth period	*<12 months*	*12–24 months*	*>24 months*	
	54 (28.3%)	63 (33%)	71 (37.2%)	
Site	*Parasymphysis*	*Body-angle*	*Angle-ramus*	*Maxilla*
	54 (28.3%)	49 (25.7%)	76 (39.8%)	9 (4.7%)
Expansion	*Lingual/palatal-buccal/labial*	*Mesial-distal*	*All direction*	
	57 (29.8%)	25 (13.1%)	106 (55.5%)	
Aspirate	*Yellowish*	*Brownish*	*Purulent*	*Empty*
	28 (14.7%)	131 (68.6%)	6 (3.1%)	23 (12%)

Table 2. Distributions of tumor characteristics, including the size, growth period, site, expansion, and aspirate.

The tumors expanded in all directions in 106 (55.5%) cases, the lingual/palatal-buccal palatal direction in 57 (29.3%) cases, and the mesial-distal direction in 25 (13.1%) cases. Moreover, the aspirate was predominantly brownish 131 (68.6%), followed by yellowish 28 (14.7%), empty 23 (12%), and purulent 6 cases (3.1%).

Histopathologic subtypes

Four clinicopathologic subtypes of ameloblastoma were identified: follicular (31, 16.2%), plexiform (19, 9.9%), mixed (110, 57.6%), and others (13, 6.8%). Mixed subtypes refer to combinations of microscopic patterns of common subtypes. 'Others' refer to less common histopathologic patterns, including the acanthomatous, granular cell, desmoplastic, and basal cell types.

Histopathological subtypes n (%)			
Follicular	*Plexiform*	*Mixed*	*Others*
31 (16.2%)	19 (9.9%)	110 (57.6%)	13 (6.8%)

Table 3. Distributions of histopathological subtypes.

Treatment

All patients underwent surgery. Most of them (149, 78%) underwent radical surgical procedures, including en-bloc resection, segmental resection, hemimandibulectomy, or hemimaxillectomy. The rest of them (22, 11.5%) underwent conservative procedures, including enucleation and radical curettage.

Surgical treatment n (%)	
Conservative	22 (11.5%)
Radical	149 (78%)

Table 4. Distribution of surgical procedures.

Correlation analyses using Pearson's chi-square test between sets of (two) variables; i.e., the tumor site and the DMFT score, the blood type and the histopathological subtype, and the growth period and root resorption, were performed. None of these correlations were statistically significant ($p > 0.05$ for all of them).

Discussion

The present study is a retrospective analysis of the demographic and clinical profiles of ameloblastoma cases registered in the Cipto Mangunkusumo Hospital. This tumor is the most frequent type of odontogenic tumor, which highlights the importance of demographic and epidemiologic studies in this field. In the present study, 51.7% of patients were female and 48.7% were male, which is in line with the findings of Ogunsalu et al. who reported a female predominance with a male-to-female ratio of1:1.14.[10] The peak incidence of ameloblastoma in Asia occurs in the third to fifth decade of life. This is in line with our findings (48.2% of patients were aged more than 34 years).[11]

We collected data on relevant comorbidities and bad habits of patients with ameloblastoma. Regular alcohol consumption and smoking were confirmed in 2.1% and 27.7%, respectively. However, a recent study by Medina et al. reported no correlation between alcohol consumption and smoking habits and the development of ameloblastoma and postoperative complications.[12] In fact, our patients also had comorbidities, of which hypertension (15.7%) and diabetes mellitus (5.2%) were the most frequent pre-existing medical conditions.

The blood type is a genetic characteristic that is usually associated with some diseases and deformities. It can also complicate the prediction of genetic patterns of odontogenic lesions, including ameloblastoma.[13] In 2008, Gheisari et al. also reported the relationship between maxillofacial deformities and the ABO blood type.[14] A study by Rasoul et al. demonstrated that among all blood types, blood type A had the lowest association with maxillofacial deformities and blood type B had the highest association with these deformities.[13] Unfortunately, this finding is contradictory with our results that identify blood type O (42.9%) as the blood type with the highest association with these deformities in our study population.

External root resorption (ERR) is the progressive loss of cementum and dentin due to the continual action of osteoclastic cells.[15] In 1976, Struthers et al. reported that ameloblastoma was associated with higher rates of root resorption than other cystic lesions.[16] Past reports have also suggested that ERR is a result of the pressure caused by a locally invasive tumor-like ameloblastoma.[17] We found that 130 (68.1%) patients had radiologically confirmed root resorption. We also found an association between ameloblastoma and tooth impaction; however, the clear pathogenesis was poorly understood.[18] We found that 66 (34.6%) cases were associated with tooth impaction.

Most of the tumors (95.3%) were located in the mandible and the rest were located in the maxilla (4.7%). The ramus and angle of the mandible were the most affected subsites (39.8%). This finding was similar to those of the study by Singh et al. in 2014, who reported that the mandible, particularly the posterior region, was the most affected site.[19] Up to 60.7% of tumors in this study measured 4–10 cm, a finding similar to those of a study carried out by Ruhin et al., who reported that more than 50% of the lesions measured 5–13 cm.[20]

According to previous studies, the most common patterns are the follicular and plexiform patterns.[14,21] This is in stark contrast to our findings, according to which the mixed type is more predominant (57.6%) than the follicular (16.2%) and plexiform (9.9%) patterns. We also found that the majority of aspirates were brownish (68.6%) and yellowish (14.7%).

The primary treatment of ameloblastoma can be divided into conservative and radical treatments based on the type of tumor and its clinical appearance.[22] Unicystic and peripheral forms of ameloblastoma are usually treated conservatively, while solid and multicystic forms of ameloblastoma are treated radically. Conservative treatment methods such as enucleation and radical curettage are assumed to be associated with high recurrence rates. On the other hand, radical surgery (such as segmental resection) is thought to be associated with a lower recurrence rate; however, it comes with a higher rate of morbidity and requires extensive surgical reconstruction.[23]

The majority of cases in our hospital undergo radical treatment (78%) while the rest are managed conservatively (11.5%). The high recurrence rate was the main reason why most of our cases were managed via resection with a cut margin of 1–1.5mm from the healthy bone. The recurrence rate of ameloblastoma is affected by many factors, including tumor subtypes, surgical treatment, and tumor behavior. Prolonged clinical and radiographic follow-up is necessary to

assess the possibility of recurrence after lengthy treatment.[24]

Conclusions

This study provides comprehensive demographic and epidemiologic data on ameloblastoma in the Indonesian population using different parameters. Distribution patterns according to sex, age, tumor characteristics, tumor subtypes, and surgical treatment were in line with other reported series.

Declaration of Interest

The authors report no conflict of interest.

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