

RESEARCH ARTICLE

A 40-year Retrospective Clinicopathological Study of Ameloblastoma in Iran

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Abstract

Ameloblastoma is the most common epithelial odontogenic tumor. It may show locally invasive behavior resulting in recurrence and malignancy. Therefore, appropriate diagnosis of this tumor is necessary. The aim of this study was to evaluate clinicopathological characteristics of ameloblastomas in an Iranian population. We present a 40-year retrospective study of patients diagnosed from 1971 to 2010 in the Department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad, Iran. Information gathered from patient records included age, gender, tumor location and histologic type. The frequency of odontogenic tumors among all lesions was 2.08% and ameloblastoma with 88 samples demonstrated the greatest prevalence (41.5%). Regarding gender, 60% of samples occurred in males. The mean age of studied patients was 33.02 ± 15.74 years with a peak of occurrence in the third decade of life. The most frequent location of tumor was the mandibles (93.2%). Eighty five (96.6%) tumors were recorded as benign and 3 (3.4%) as malignant. Of benign tumors, 62 (72.9%), 20 (23.5%) and 3 (3.6%) cases were of conventional, unicystic and peripheral types, respectively. In contrast to most previous studies, the most common histologic subtype in the present study was plexiform. Knowledge of the incidence of ameloblastoma and its clinicopathologic features including most common location, gender and age distribution in different ethnogeographic backgrounds is necessary for accurate diagnosis and proper treatment.

Keywords: Odontogenic tumor - ameloblastoma - plexiform

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Introduction

Odontogenic tumors are lesions derived from epithelial or mesenchymal remnants. Therefore, these lesions are observed exclusively in the mandible and maxilla. Some of these lesions are real neoplasm which rarely represents behavior of malignant tumors; others may be considered as tumor-like malformation such as hamartoma (Manor et al., 1999; Kumamoto et al., 2000). By considering histological origin, odontogenic tumors are categorized in three groups: 1-Epithelial odontogenic tumors; odontogenic ectomesenchyme doesn't have role in their formation. 2) Mixed odontogenic tumors; these tumors are composed of odontogenic epithelial and ectomesenchymal components. 3) Ectomesenchymal odontogenic tumors; basically formed from ectomesenchyme. Etiology and pathogenesis of related lesions are unclear and there is no evidence for specific cause or motive for them (Neville et al., 2009).

Ameloblastoma is the solid/multicystic odontogenic tumor. It is the most common clinical tumor within all odontogenic tumors if odontoma is considered as hamartoma. According to the latest classification of World Health Organization (WHO), ameloblastoma is known

as the prototype of odontogenic tumors with epithelial origin (Bachmann et al., 2005; Salehinejad et al., 2011). Improper diagnosis and treatment of this tumor may lead to significant morbidity and occasionally death. Its incidence in males and females is equal and the mean age of involvement is approximately 35 years. Ameloblastoma is divided into three types by considering clinical and radiography findings: conventional (86%), unicystic (13%) and peripheral (1%) (Mendenhall et al., 2007).

Surgery is primary treatment but there has been still some debate for the most appropriate method of surgical removal. This debate ranges from conservative to radical modes of treatment (Vohra et al., 2007). The first step of treatment is an appropriate categorization of tumor and gaining knowledge about the incidence of histopathological subtypes as well as age, gender and involved site of lesion.

Due to the lack of information in the English-language literature about the clinicopathologic features of ameloblastoma in Iranian population, we reviewed ameloblastoma tumors over a 40-year period. The purposes of this study were to evaluate the clinicopathologic features of ameloblastoma in Iranian population and to

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provide data for comparison with other Studies.

Materials and Methods

In this cross-sectional study we evaluated all files of the department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad, Iran with diagnosis of odontogenic tumors from 1971 to 2010. All completed and detailed case files were entered the study and incomplete records were excluded from the study procedure.

Then histologic diagnosis records were reevaluated for patients who were treated for ameloblastoma. Then all types of Ameloblastoma were assessed based on histopathological findings and World Health Organization (WHO) classification.

When diagnosis was confirmed, information gathered about age, gender and location of tumor.

Recorded data were analyzed by SPSS version 11, the descriptive data including demographics were expressed as Mean±SD (continuous quantitative variables). Frequency and percentiles were used for qualitative and discrete quantitative variables. And for comparing qualitative variables (gender, tumor type and position), the chi-square test was used. To analyze the age as a quantitative variable, the independent T-test was applied for dichotomous comparisons and Analysis of Variance (ANOVA) for more variables within more than two groups. In all calculations P-value less than 0.05 was considered significant.

To review the literature in term of published case series about Ameloblstoma, we searched Pubmed

with the keywords with the following search strategy: (ameloblastoma AND jaws) AND (“retrospective studies” OR “retrospective analysis” OR “clinicopathological studies” OR “cases-analysis” OR “clinical review” OR “clinical features”) we retrieved multiple papers then the data of relevant studies with retrospective descriptive design were extracted. By considering the archival mode of this study and using paraffin blocks, no ethical problem and privacy safety were assumed to be vital.

Results

We studied all case files of patients who referred to department of Oral and Maxillofacial Pathology, Faculty of Dentistry, Mashhad, Iran with diagnosis of odontogenic tumors during 1971 to 2010. Of all odontogenic tumors 88 (41.5%) were ameloblastoma (Table 1).

Among them, 51(58%) were recorded in men and 37(42%) in women. Chi-square test showed no significant difference for gender distribution in different decades (P=0.068).

Average age of studied individuals was 33.02 ± 15.74 years, (mean=33.02, SD 15.74). In terms of age groups, the highest percentage of patients was from 20-29 years category with 27 cases (30.7%) followed by the age groups of 30-39 years and less than 20 years with 19 (21.6%) and 17(19.3%) patients, respectively.

Regarding location of tumor, 82 (93.2%) cases were in mandible, 3 cases in maxilla (3.4%) and 3 (3.4%) cases were located at soft tissue. From tumors which located in the mandible, 66 (80.5%) cases were in posterior, 5 (6.1%) cases were situated in the anterior of mandible and in 11(13.4%) cases anteroposterior aspect was not recognizable (Table 2).

Among the maxillary tumors, there were two cases in the posterior region and one in the anterior region. One of the soft tissue tumors was in buccal mucosa and others were in mandibular posterior vestibule .

By considering the histological type of tumor, 85 (96.6%) tumors were recorded as benign and 3 (3.4%) as malignant. Among the benign tumors, 62 (72.9%) cases

Table 1. Frequency of Ameloblastoma Tumors among All Samples

Decade	All samples	Ameloblastoma	Percentage
1971-1980	1298	12	0.92
1981-1990	2336	18	0.77
1991-2000	3114	20	0.64
2001-2010	3420	38	1.11
Total	10168	88	0.86

Table 2. Relative Frequency of Ameloblastoma

n	Mean age	Gender			Location				
		Male	Female	Peripheral	Central				
					Maxilla		Mandible		
		3	82		Ant.	Post.	Unknown		
88	33.02	51	37	3	1	2	5	66	11

Table 3. Frequency of Different Clinical Subtypes of Ameloblastoma

Type of tumor	n	Gender		Mean age	Location of tumor		
		Male	Female		85		Peripheral
					Mandible	Maxilla	
Conventional	62	33	29	35.5	60	2	0
Unicystic	20	13	7	24.5	19	1	0
Peripheral	3	3	0	27.7	0	0	3
Carcinoma	3	2	1	42.7	3	0	0

were conventional, 20 (23.5%) unicystic and 3 (3.6%) peripheral type (Table 3).

Our findings indicated that all peripheral ameloblastoma cases were observed within males and incidence of malignancy between males was two times more than females (Table 3). Based on Likelihood Ratio test, there was no significant difference in distribution of tumor type according to gender ($P=0.226$). In spite of an increase in the number of ameloblastoma patients, the percentage of ameloblastoma cases among the whole samples showed a gradual decline between 1971 and 2000, and a gradual rise during 2000s in comparison to the previous decades.

Distribution of tumor types based on age range groups showed that among all types of benign tumors, the group of 20-29 years had the highest prevalence ($P<0.05$).

The comparison of mean age within different type of tumors showed that among benign tumors, the average age of those who had conventional type was higher than other groups. Also the average age of patients with peripheral tumors was more than patients with unicystic tumors.

On the other hand, the average age of patients with malignant tumor was higher than benign group. Peripheral and malignant tumors were mostly seen in 20-29 and 40-49 age groups respectively. According to the ANOVA test, the mean difference between these four mentioned groups was statistically significant ($P = 0.028$).

Histopathologic findings showed that the plexiform ameloblastoma (26 cases, 46.4%) was the most common type in both gender. According to Likelihood Ratio test, there was no significant difference between genders ($P=0.524$). Thereafter, follicular and follicular & acanthomatous had the most frequencies, 15 (26.8%) and 4 (7.1%) patients, respectively (Table 4).

Table 4. Frequency of Conventional Tumors Based on Histopathologic Type

Type of conventional tumors	n	Gender	
		Female	Male
Plexiform	26	10	16
Follicular	15	8	7
Acanthomatous	3	0	3
Follicular & acanthomatous	4	2	2
Plexiform & basal cell	3	1	2
Desmoplastic	2	1	1
Follicular & plexiform	3	2	1
Unknown type	6	-	-

Table 5. Compariative Incidence of Ameloblastoma in Different Published Studies

Author	Country of study	Total No. of cases	Most common Histologic Type	Mean of Age	Sex predilection	Most common involved area
Present study	Iran	88	Conventional	3rd decade	Male	Mandible
Chawla R et al. (2013)	India	91	plexiform and unicystic	32.5	Male	Mandible
Butt F M et al. (2012)	Kenya	127	unknown	2nd decade	Equal	Mandible
Siar C H et al. (2012)	Malaysia	340	solid/multicystic	2nd decade	Male	Mandible
Ruhin-Poncet B et al. (2011)	France	116	Follicular	36	unknown	unknown
DarshaniGunawardhanaks et al. (2010)	Sri Lanka	286	solid/multicystic	33.2	Equal	Mandible
Fregnani E R et al. (2010)	Brazil	121	solid	33.2	Slight female predilection	Mandible
Krishnapillai et al. (2010)	India	73	unicystic	30.2	Slight male predilection	Mandible
Adeline V L et al. (2008)	Kenya	184	solid/multicystic	30.2	Female	Mandible
Odukoya O (2008)	Nigeria	100	solid/multicystic	less than 20 years	Male	Mandible
Adebisi K E et al. (2006)	Nigeria	77	Follicular	3rd decade	Male	Mandible
Ladeinde AL et al. (2006)	Nigeria	207	Follicular	31.67	Male	Mandible

In our findings, the most common site of tumors was the mandible except for peripheral tumor. A significant difference was observed between tumor types and its location, ($P<0.001$).

The findings (the most common histologic types, mean of age and male: female ratio) of 13 published studies reporting 1722 case of ameloblastoma are presented in Table 5.

Discussion

Ameloblastoma is the most important tumor among ectodermal odontogenic tumors. The relative incidence of this tumor is equal to incidence of all odontogenic tumors, except odontoma. Ameloblastoma with invasive behavior and slow development rate is considered as the most common tumor between odontogenic tumors (Tamme et al., 2004).

In the current study, histological types of ameloblastoma are evaluated in an Iranian population for the first time.

Of 212 odontogenic tumors referred to Oral and Maxillofacial Pathology Department of Mashhad Dental School during forty years, 88 cases were ameloblastoma.

Saghravani et al studied the odontogenic tumors in an Iranian population in a 30-year period. Correspondingly, they reported that the ameloblastoma was the most frequent tumor followed by odontoma, odontogenic myxoma, and adenomatoid odontogenic tumor (Saghravani et al., 2010).

In consistent with our findings, several studies reported the ameloblastoma as the most common odontogenic tumor (Lu et al., 1998; Ladeinde et al., 2005; Simon et al., 2005; Jing et al., 2007; Avelar et al, 2011; Osterne et al., 2011). whereas some other studies demonstrated odontoma as the most frequent odontogenic tumor (Tawfik et al., 2010, Taklif et al., 2010); this difference could be due to the racial differences or sample size.

During forty years, the number of patients with ameloblastoma referred to our department was increased; this could be explained as an outcome of increasing the population of the country and worldwide. In terms of gender distribution, about 60% of cases were diagnosed in men. The findings indicated that in all over the study period, superiority of gender distribution was in males. Correspondingly, several studies showed that peripheral ameloblastoma cases were occurred among males (Razavi

et al., 2001; Adebayo et al., 2005; Arotiba et al., 2005). Dhanuthai et al. showed that distribution of females was higher than males in their study (Dhanuthai et al., 2012). They found different results in comparison with ours, while Mosadomi et al observed that gender distribution was equal for males and females in Africa (Mosadomi, 1997).

In our study, ameloblastoma was observed in individuals with younger average age; but in Dhanuthai's study the mean age was 48 years that show older age than our samples (Dhanuthai et al., 2012).

The posterior region of the mandible is the most common location for ameloblastoma in the present study (93.2%) followed by maxilla (3.4%), which agrees with the studies performed in Asia, Europe, Africa and South America (Table 5) (Arotiba et al., 2005). According to the results of mentioned studies, it seems that odontogenic tumors particularly ameloblastoma more likely involve the lower face especially mandible (Fulco et al., 2010). Correspondingly, Kim et al. concluded that plexiform of conventional tumors possessed the most prevalence and the difference between two gender groups wasn't significant (Kim et al., 2001).

In the present study, the most common tumor was the conventional, whereas Ladeinde (Ladeinde et al., 2006) and Adebisi (Adebisi et al., 2006) from Nigeria and Ruhin-Poncet et al (Ruhin-Poncet et al., 2011) from France reported the follicular type as the most frequent; Odukoya (Odukoya et al., 2008) in Nigeria, Adeline (Adeline et al., 2008) in Kenya, Darshani et al (Darshani et al., 2010) in Sri Lanka (Siar et al 2012) in Malaysia declared the solid/multi cystic as the most frequent tumor in their study. Chawla et al (2013) and Krishnapillai et al (Krishnapillai et al., 2010) in India reported the unicystic/plexiform and unicystic as the most common types, respectively. Fregnan et al (Fregnan et al., 2010) showed the solid one was the most frequent in Brazil, while Butt et al (2012) in Kenya couldn't report the most abundant tumor .

In conclusion, according to our findings, ameloblastoma is the most common odontogenic tumor in the mandible and in males with most cases appearing in the third decade of life. Most of the ameloblastic tumors are benign and the conventional type is the most reported one. Our findings confirm worldwide studies results although there are some differences, especially in histological subtypes.

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