ORIGINAL STUDY

Retrospective Analysis of Ameloblastoma: An Institutional Experience

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Abstract

Introduction: Neoplasms that are derived from odontogenic tissues represent an uncommon and heterogenous group of entities in oral and maxillofacial region comprising about 1% of all jaw tumors. Ameloblastoma received particular attention of oral pathologists due to its local aggressive behavior.

Objective: Indian studies, regarding prevalence of ameloblastoma are very few. The present study was done to analyze and compare clinical, radiographic and histopathologic features of 57 cases of ameloblastomas in India retrospectively.

Materials and methods: 57 cases of histologically diagnosed ameloblastomas were retrieved from the archives over a five year period to analyze the age, sex, site, clinical presentation, radiographic features and histopathology.

Results: In the present study, ameloblastoma accounted for 77% of all odontogenic tumors with a mean age of 37.5 years in males and 42 years in females. About 60% of the cases revealed mandible as most common site predominantly of posterior region with common clinical presentation as painless swelling. Radiographically, multilocular radiolucency was noticed in about 54.3% of cases. Among the histological variants of solid/multicystic ameloblastomas, follicular variant was reported to be the most common (38%) and intraluminal variant (19%) to be the common subtype in unicystic ameloblastomas. Statistical analysis was done among the histological variants of ameloblastomas which showed no statistical significance ($p<0.05$).

Conclusion: The findings in the present study were well in accordance with previous reported studies. However, extensive studies with larger samples need to be carried out in the Indian population to evaluate the incidence of ameloblastoma for more definitive results.

Key words: Ameloblastoma, mandible, odontogenic tumors

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Introduction

Neoplasms that are derived from odontogenic tissues represent an uncommon and heterogenous group of entities in oral and maxillofacial region comprising about 1% of all jaw tumors.[1-3] The odontogenic lesions are classified as benign and malignant tumors although some of them are hamartomas.[4] Ameloblastoma is a benign tumor derived from odontogenic epithelium without involving odontogenic ectomesenchyme. It was first described by Cusack in 1827 as a type of odontogenic cyst. Malassez (1866) differentiated the tumor from other odontogenic cysts. It ranks the most common benign odontogenic tumor in Asian and African countries, where as odontome is the most common odontogenic tumor in Europe and America.[4-6]

Ameloblastoma received particular attention of oral pathologists due to its local aggressive behavior.[4,7] Data on the frequency of this neoplasm among other odontogenic tumors has been reported worldwide. The objective of this study is to do a retrospective analysis of ameloblastoma cases reported in our institution over a period of five years and supplement this data to the English literature.

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Materials and methods

Among 74 cases of odontogenic tumors, 57 cases of clinically and histopathologically diagnosed cases of ameloblastoma which reported to our institution from 2010 -2015 were included in the present study. The following clinical parameters were taken

into consideration such as age, gender, site, symptoms, radiological and histological features. A comparison of these cases was done with other previously reported studies.

Tumors located in the mandible were categorized as anterior for incisor-canine region and as posterior for premolar-molar and ramus-condylar regions. In case of maxilla, the incisor-canine region was considered as anterior, premolar-molar and tuberosity regions were categorized as posterior regions. Lesions crossing midline were considered as bilateral involvement.

Detailed case history of all the cases of ameloblastoma were taken and recorded according to the above parameters. The findings were sent for statistical analysis and results were tabulated.

Results

Descriptive statistical analysis was performed to calculate percentage and frequency of these variants of ameloblastoma. Among 74 cases of benign odontogenic tumors, 57 cases of ameloblastoma were reported. The distribution of these cases in relation to gender and age are presented here. Figure 1 shows male predominance with 56% (32 cases) and females accounted for 44% (25 cases). Figure 2 depicts age wise distribution of cases of which 2% each (1 case) belonged to the age groups of 0-9 and 70 - 79 years, followed by 11% (6 cases) in 10-19 years, 17% each (10 cases) in 20-29 years, 40-49 years, 21% each (12 cases) in 30-39 years and 50-59 years, 9% (5 cases) in 60-69 years age group.

Among 32 male patients, 20 cases (35%) showed solid/multicystic ameloblastoma (SMA), 11 cases (19%) had unicystic ameloblastoma (UA), one case (2%) had desmoplastic Ameloblastoma (DA). Of 25 female patients, 18 (31%) had SMA with six (11%) of UA, one case (2%) had peripheral ameloblastoma (PA) (Table 1). Table 2 shows distribution of histological variants of ameloblastoma according to age. There were a total of 38 cases of SMA the majority of which occurred in 30-39 years of age followed by nine cases (16%) in 49 - 49 years in eight cases (14%) in 50 -59 years. Of the 17 cases of UA, five cases (9%) occurred in 10-19 years of age group with three cases each (5%) in 20 -29 years, 50-59 years, 60-69 years and one case each (2%) in 30-39 years, 40-49 years and 70-79 years. One case each (2%) of PA and DA occurred in 50-59 years and 30-39 years of age.

Of the 57 cases, 36 (63.15%) patients had a clinical presentation of swelling and 21 (36.84%) patients presented with pain and swelling. Radiographic findings of the patients showed 31 cases (54.38%) with multilocular radiolucency, 21 cases (36.84%) with unilocular radiolucency and five cases (8.7%) showed mixed radiolucent radio opacity.

Among 32 males, 21 Ameloblastomas occurred in mandibular posterior region and in 25 female there were 13 (Table 3). There were two cases of ameloblastoma seen in maxillary posterior region in both males and females. The number of cases seen in mandibular anterior region in both males and females were five each whereas two cases involved bilaterally in mandible in males and three cases in maxilla in females. In the maxillary anterior region two cases were seen in males, one case in females and one case showed bilateral involvement in females. The Chi-Square test had a value of 1.9561 for the cases seen occurring in maxilla in males and females with no significant p-value (0.3762). site distribution of ameloblastoma cases was done according together in which Chi-square test value obtained in mandible was 0.7560 with no significant value (p=0.6850).

The various histological subtypes of ameloblastoma are listed in Figure 3. Among the variants of SMA 38% of cases showed follicular type, with 14% acanthomatous type, 9% plexiform type, 3% granular cell type, and 2% each of basal cell, DA and PA. UA cases constituted of about 30% of all the cases, among which 19% showed intraluminal type, 9% of cases were of intramural type and 2% of cases were both intraluminal and intramural type (Figure 4).

The histological variants of ameloblastoma were seen according to gender (Table 4). The follicular variant predominated equally in males and females with 11 cases each, followed by intraluminal variant of UA with 7 cases in males and 4 cases in females. The plexiform variant was seen in 3 males and 2 females, acanthomatous variant 3 cases in males and 5 cases in females. granular cell variant was seen equally in males and females with 1 case each. One case each of the basal cell, desmoplastic and intaluminal-intramural variants were seen in males with one case each in PA and intarmural type of UA were seen in females. The Chi-Square analysis showed 6.6303 with no significance of p value-0.675.

Discussion

The relative frequency of ameloblastoma among all odontogenic tumors ranges from 11-92%.[7] Studies regarding frequency of ameloblastoma done in India are relatively less. Rekha K et al[7] in their study reported 60.3% of ameloblastomas among all odontogenic tumors. The present study is the first study to be done in Khammam district of Telangana state. In the present study, 57 cases (77%) of ameloblastomas were reported among 74 cases of odontogenic tumors over a period of 5 years from January 2010 to June 2015. The results of our study correlated with majority of the previous studies done across the world and also in India.

Among all the odontogenic tumors reported here, ameloblastoma was found to be the most commonly occurring benign odontogenic tumor being in agreement other with studies.[1-3,8-12] This finding supports the fact that ameloblastomas are more commonly seen in developing countries like Asia and Africa compared to developed countries like Europe and America where odontomes are the most commonly reported benign odontogenic tumors.

The reason for high frequency of occurrence of odontomes in developed countries is that they are more likely to have regular dental check-ups and hence are detected during routine radiographic examinations whereas ameloblastomas are the most commonly reported cases in developing countries. The probable explanations for this being the patients usually present to the hospital only when they have symptoms such as swelling or pain/swelling indicating lack of awareness of importance of oral health and differences in socioeconomic status as in agreement with previous studies.[6,13-14] In addition the peripheral population of Khammam district consists of tribes. Also a thorough search of the previous records of our department revealed a majority of the reported cases to be of odontogenic lesions among which ameloblastomas had the highest frequency of occurrence supporting a hypothesis that either the remnants of dental lamina or epithelium of oral mucosa of the people in tribal areas are stimulated by an unknown triggering factor for causation of ameloblastomas.

Clinical features

Age:

The distribution of ameloblastoma showed a wide age range from 9-79 years in our study. This finding is in agreement with many other studies reported.[8,15] The youngest age reported in the present study was of nine years which was in agreement with a study done by Ord et al[16], whereas according to Krishnan et al[17] ameloblastoma was reported in a three year old girl. The oldest age reported was 75 years. Majority of the cases, 12 cases each (21%) occurred in 30-39 and 50-59 years of age group. This was in agreement with previous studies,[3,7-8] whereas it was highest for 15-30 year age group in a previous study.[9] In few other studies, the peak incidence fell in third decade[6,16,18] which was in agreement with studies done by Riechart et al[19] and a multicentric study done in Latin-American population, whereas Pereira et al showed in their study that two thirds of patients had less than 40 years of age.[5]

Gender:

In our study, a slight male predominance was noticed with a male to female ratio as 1.2:1, with 32 cases of males (56%) versus 25 cases of (44%) female patients that correlated well with previous studies.[3,4,9,20-22] The exception to this are selective studies that demonstrated either equal gender distribution[6,8] or female predominance.[17] The mean age of male patients was 39.16 years and females were 39.57 years with a slight female predominance in age. Our finding was in agreement with other studies.[6]

Site:

More number of cases occurred in mandible when compared to maxilla with a ratio of 6.1:1. This was in accordance with previous studies.[1,10,13,19-20] Our study showed 60% of the tumors predominated in posterior region (premolar-molar, ramus) 10% in anterior mandible (incisor-canine) and 5% of crossed the midline. This was in agreement with other studies.[2,5,9,16,18] We found a mean age of occurrence in mandible was 39.7 years where as in maxilla it was 36.1 years.

Table 1: Distribution of amelablastoma cases according to gender

Type	SMA	UA	PA	DA	Total
Males	20 (35%)	11 (19%)	0	1 (2%)	32 (56%)
Females	18 (31%)	6 (11%)	1 (2%)	0	25 (44%)
Total	38 (66%)	17 (30%)	1 (2%)	1(2%)	57 (100%)

SMA – Solid Multicystic Amelablastoma; UA – Unicystic Amelablastoma; PA – Peripheral Amelablastoma; DA – Demoplastic Amelablastoma

Clinical symptoms and radiographic findings:

The primary clinical manifestation of ameloblastoma in the present study was a slow growing painless swelling with few cases accompanied by pain/parestheia. This clinical finding was very much in correlation with several authors.[5,9,22-24] The most common radiographic finding seen in ameloblastoma was a multilocular radiolucency surrounded by a radiopaque border.[25] Some other presentations were unilocular radiolucencies in our study which also correlated with Kim and Jang.[26]

Histological variants:

In the present study solid/multicystic ameloblastoma (SMA) had a highest incidence (66%) when compared to unicystic ameloblastoma (30%), peripheral ameloblastoma (PA) and desmoplastic ameloblastoma (DA) with each of 2% of the reported cases. Among SMA, follicular variant was predominantly seen with 38% of cases followed by acanthomatous (14%), plexiform (9%), granular cell (3%), basal cell (2%) This was in accordance with other studies.[5,6,27] However, these various histopathologic features did not show any behavioural changes and only one tumor demonstrated multiple histopathologic variants. The incidence of PA was about 0.5-9.3% whereas Kittipong D et al[6] reported 4.27% in their study. The plausible explanation for this low incidence is that it does not usually produce gross disfigurement. As a result, patients may not seek medical attention or the attending clinicians may misdiagnose PA as other common benign mucosal lesions and discard the surgical

Table 3: Histological variants of ameloblastoma

Variants	Male	Female	Chi-square (p-value)
Follicular	11	11	$\chi 2=6.6303$ P= 0.675
Plexioform	3	2	
Acanthomatous	3	5	
Granular	1	1	
Basal	1	0	
Desmoplastic	1	0	
Peripheral	0	1	
Uni-IL	7	4	
Uni-IM	4	1	
Uni-IL&IM	1	0	
Total	32	25	

Table 2: Distribution of amelablastoma cases according to age

Type	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79	Total
SMA	1(2%)	1(2%)	7(12%)	10(17%)	9(16%)	8(14%)	2(3%)	0	38(66%)
UA	0	5(9%)	3(5%)	1(2%)	1(2%)	3(5%)	3(5%)	1(2%)	17(30%)
PA	0	0	0	0	0	1(2%)	0	0	1(2%)
DA	0	0	0	1(2%)	0	0	0	0	1(2%)
Total	1(2%)	6(11%)	10(17%)	12(21%)	10(18%)	12(21%)	5(8%)	1(2%)	57(100%)

SMA – Solid Multicystic Amelablastoma; UA – Unicystic Amelablastoma; PA – Peripheral Amelablastoma; DA – Demoplastic Amelablastoma

specimens without submitting them for histopathologic examination.[6]

Among the subtypes of UA, luminal subtype showed 19%, followed by intramural (9%) and intraluminal and intramural (2%) whereas UA with intraluminal and intramural subtype was reported to be maximum in a study conducted by Rekha K et al.[7] The recurrence rate of SMA varies according to many factors such as the type of treatment and histological variant. Recurrence of SMA treated radically is less than curettage. Similarly follicular ameloblastoma shows less recurrence than other variants. The recurrence rate for unicystic ameloblastomas also varies according to the histological subtypes in which intraluminal and intramural subtype is more aggressive and chances of recurrence are more compared to intra luminal variant.

Conclusion

In summary, 57 cases of ameloblastomas, in an Indian population were reviewed to determine their clinicopathologic characteristics. A marked geographic variation in the relative incidences of various ameloblastomas was observed. Results showed that the distribution patterns according to age, gender, site, radiographic and histopathologic features compared favorably to other reported studies with follicular ameloblastomas being the most common histological subtype among solid/multicystic ameloblastomas and intraluminal variant being the commonest one among unicystic ameloblastomas. Since, most data on the frequency of ameloblastomas are derived from hospital based records which may be biased by some socioeconomic factors, the role of genetic and/or environmental factors in modulating geographic variations in the incidence of ameloblastomas requires further investigations.

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