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RESEARCH ARTICLE

AMELOBLASTOMA: A 5 YEAR RETROSPECTIVE ANALYSIS OF CASES IN A TERTIARY HEALTHCARE CENTER IN KERALA

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Abstract

Aim: To supplement the current database of Ameloblastoma by reporting the clinicopathologic features of Ameloblastoma cases from Kozhikode, Kerala, India. Materials and Methods: Archive records of all histologically diagnosed cases of Ameloblastoma from January 2010 through December 2014 in Department of Oral Medicine and Radiology, Government Dental College, Kozhikode, Kerala, India were analyzed with regards to age, gender, clinical manifestation, radiographic aspect, anatomical distribution and histological subtype. Results: Out of the 54 reviewed cases 31 (57.4%) were males and 23 (42.6%) were females. Mostly affected age group was second, third and fourth decades. The patient's age varied from 12 to 65 years, with an average of 37.33 years of age at the time of diagnosis. Posterior mandible was the site of preponderance. The main histological variant seen was solid Ameloblastoma amongst which follicular were most common and none of the case of basal cell type and peripheral variant were seen. Unicystic and desmoplastic variant also found in considerable number. Conclusion: Ameloblastoma mostly affects peoples in the second to fourth decades of life, with slight male preponderance. Posterior mandible is the site of predilection. Commonly appears as multilocular radiolucent lesions but the rare variant i.e. desmoplastic appears as mixed radiopaque-radiolucent lesions. Histopathologically solid variant seen more compare to unicystic variant and desmoplastic variant. Amongst solid type follicular was most prevalent subtype. Relatively higher frequency seen in cases those were in their second decade of life.

Keywords – Ameloblastoma; odontogenic tumours; tumours of jaws

INTRODUCTION

Ameloblastoma is a benign epithelial tumor with no ectomesenchymal component; it has aspects of aggressiveness and local invasion but is also asymptomatic and slow-growing.[1-3] World Health Organization (WHO) defined Ameloblastoma as a "benign, locally invasive epithelial odontogenic neoplasm of putative enamel organ origin".[4] In 1868 Broca first described this tumour and Churchill in 1934 coined the term 'Ameloblastoma'.[5]

There have been several case series of Ameloblastomas from several countries with geographic and/or racial variations. Ameloblastoma ranks as the most common odontogenic tumor in Asia and Africa, whereas odontoma is listed as the most common odontogenic tumor in Europe and America.[6-14] Most of the reported series of Ameloblastoma from Asia are from China and Japan.[6, 14-19] There has been a paucity of information from other countries in Asia such as India.



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The present study aims to analyze a series of Ameloblastoma cases diagnosed in tertiary healthcare center in Kerala, India and to supplement the current Ameloblastoma database by reporting the clinicopathologic features of Ameloblastoma from North Kerala.

MATERIALS AND METHODS:

The surgical biopsy records of all histologically diagnosed cases of Ameloblastoma archived from January 2010 through December 2014 were retrieved from the files of the Department of Oral Medicine and Radiology, Government Dental College, Kozhikode, Kerala, India. Data with regard to age, gender, clinical manifestation, radiographic aspect, anatomical distribution and histological subtype were analyzed. The histopathological diagnosis was based on WHO classification from 2005.

RESULTS:

During a period of five years, fifty four cases were histopathologically diagnosed as Ameloblastoma. Of the 54 cases, 31 (57.4%) were males and 23 (42.6%) were females, making a male/female ratio of 1.35:1. Maximum patients (22.2%) were in second decade of life, 20.4% belonged to the forth decade and 18.5% belonged to the third decade. 61.1% of the patients were below the age of 40 years. The patient's age varied from 12 to 65 years, with an average of 37.33 years of age at the time of diagnosis. (Figure 1).

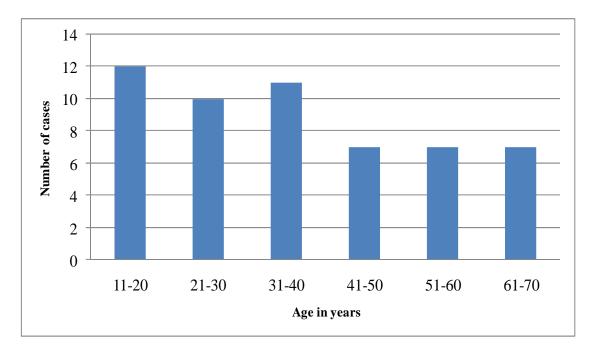


Figure 1: Distribution of cases according to their age



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Of the 54 cases the main clinical feature was a swelling of the affected region (n=50; 92.6%) followed by pain (n=16; 29.6%), tooth displacement (n=13; 24.1%), pus discharge (n=2; 3.7%) and ulceration (n=2; 3.7%). (Table 1)

Table 1: Distribution of cases according to their clinical findings

Symptoms	Number of cases	Relative percentage
Swelling	50	92.6%
Pain	16	29.6%
Tooth displacement	13	24.1%
Pus discharge	2	3.7%
Ulcer	2	3.7%

Mandible was the most common site, corresponding to 46 cases (85.2%). Of these cases, the posterior region (30 cases) was affected more often than the anterior segment. Seven out of 46 mandibular cases were seen crossing the midline.

Maxilla was less frequently affected. Only 8 cases (14.8%) were seen during the study period with maxillary involvement. Ratio between affected cases of mandible and maxilla was found to be 5.75:1. (Table 2)

Table 2: Distribution of cases according to their anatomical location

Anatomic location	Number of cases	Relative percentage	
Posterior mandible	30	55.6%	
Antero-posterior mandible	5	9.2%	
Anterior mandible	4	7.4%	
Crossing the midline in mandible	7	13%	
Posterior maxilla	2	3.7%	
Antero-posterior maxilla	4	7.4%	
Anterior maxilla	2	3.7%	
Total	54	100%	



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In current study 92.6% of the cases showed a pure radiolucent appearance in contrast to mixed radiopaque radiolucent appearance (7.8%). Amongst radiolucent cases, 33 (66%) were multilocular and 17 (34%) were unilocular. Amongst all the cases, resorption of the root (typically knife edge type) was seen in 14 cases (25.9%). (Table 3)

Table 3: Distribution of cases according to their radiographic appearance

Radiographic appearance	Number of sample	Relative percentage	
Multilocular radiolucency	33	61.1%	
Unilocular radiolucency	17	31.5%	
Mixed radiopaque-radiolucent appearance	4	7.4%	
Total	54	100%	

Histologically 64.8% were solid Ameloblastoma (n=35), 27.8% were unicystic and 4 cases (7.4%) were desmoplastic. Not even a single case of peripheral Ameloblastoma was reported. Amongst 35 solid Ameloblastoma cases 18 (51.4%) were follicular, 8 (22.9%) were plexiform, 5 (14.3%) were granular, 3 (8.6%) were acanthomatous and 1 (2.9%) was hybrid lesion (follicular- desmoplastic). Majority of solid Ameloblastomas were found as multilocular radiolucency. Only 2 cases of follicular, 1 case of plexiform and 1 case of granular variant found to be unilocular radiolucency. Most of the unicystic variant seen as unilocular radiolucency but 2 cases also showed multilocularity. All the cases of desmoplastic variant showed mixed radiopaque-radiolucent appearance. (Table 4)

Table 4: Correlation of histological subtype and radiological appearance

Histopathological Subtype	Radiological Ap	Radiological Appearance			
Subtype	Unilocular Radiolucent	Multilocular Radiolucent	Mixed Radiopaque- Radiolucent		
Follicular	2	16	0	18	
Plexiform	1	7	0	8	
Acanthomatous	0	3	0	3	
Granular	1	4	0	5	
Unicystic	13	2	0	15	
Desmoplastic	0	0	4	4	
Hybrid	0	1	0	1	
Total	17	33	4	54	

Patients affected with Ameloblastoma, 88.9% (n=48) were treated with a radical surgery, including recontouring and ample bone resection with a safety margin of healthy bone of about 1 cm. Only 6 (11.1%) young patients (< 25 years) with unicystic Ameloblastomas were treated conservatively with enucleation or curettage.



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CASE-I





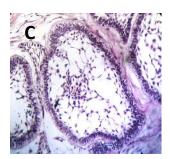
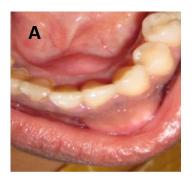


Figure 2: A. Clinical photograph showing slow-growing swelling with expansion of buccal and lingual plate; B. Panoramic radiograph showing multilocular radiolucent lesion with root resorption and displacement of teeth. C: Photomicrograph of follicular pattern of Ameloblastoma (hematoxylin and eosin stain, original magnification X40)

CASE-II





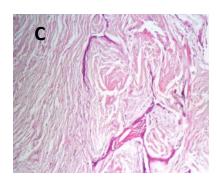


Figure 3: A. Clinical photograph showing slow-growing hard swelling with buccal expansion; B: Intra Oral Peri-apical radiograph showing mixed radiopaque-radiolucent lesion; C: Photomicrograph of desmoplastic pattern of Ameloblastoma (hematoxylin and eosin stain, original magnification X40)

DISCUSSION:

Ameloblastoma is a tumor of worldwide distribution and an analysis of English literature including the case series and meta-analysis originated from different demographic populations revealed striking variability in the clinicopathologic and radiologic features. In this study slight male preponderance was noted. However, some studies from Brazil, [20] Estonia [9] and Srilanka [21] found men and women being affected equally. In contrast other studies from Brazil [22] and Kenya [23] showed a female predilection.

In the present study, Ameloblastoma occurred more often in the second, third and fourth decades of life, although all the age groups were affected. This is similar to findings from other studies reported in India,[13] Brazil [22, 24] and Nigeria [25]. According to Kim and Jang, [26] the frequency of Ameloblastoma in young patients (< 19 years) is relatively low, occurring in

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only 10 to 15% of all reported cases. Results of this study showed a greater frequency, around 18.5%, for this group of individuals. In addition, our findings were very similar to those observed by Chidzonga *et al* [2], who showed that two thirds of the patients affected by Ameloblastomas were less than 40 years of age.

There is a consensus in the literature that Ameloblastoma more frequently affects the mandible, primarily in its posterior region.[1, 2, 9, 11, 22, 24, 26-28] This was also observed in the present study. Some authors states that the lesion site in the mandibular bone is related to the individual's race, with the posterior region more frequently involved in Caucasians and Japanese, while in blacks, especially those of African origin, the anterior/sinfisal bone segment [5, 13, 29] is a more common disease site. Some Indian investigators [30] had observed posterior mandible to be the site of predilection for Ameloblastoma in Dravidian population, similar findings is noted in this study population of Dravidian origin too. In accordance with the reports of several authors [1, 3, 28] the primary clinical manifestation of Ameloblastoma is a slow progressive swelling in the area affected by the tumor, 92.6% of patients presented with complaint of a swelling. Other less frequent symptoms, such as pain, paresthesia, ulceration, and dental mobility were the other presenting complaints.[1, 2, 29]

Multilocular radiolucency surrounded by a radiopaque border is the most common radiographic appearance of Ameloblastoma.[29] In a study conducted in Nigeria by Arotiba *et al*, [28] the authors observed almost 66% of the cases corresponded to multilocular lesions which is in accordance with the findings of the present study where we found that 61% cases had multilocular appearance. Nevertheless, Kim and Jang [26] conducted a study in Korea and reported that almost 60% of cases were composed of unilocular Ameloblastoma, while approximately 20% were multilocular.

In relation to the frequency of the different Ameloblastoma histological subtypes, the results of the present study are similar to previous reports from Estonia [9], Nigeria [25, 28], Zimbabwe [2] and the United States [11]. There is a greater occurrence of the solid tumor variants. Our findings differ from the reports of Ledesma-Mountes *et al* [31], who observed a greater prevalence of the unicystic subtype. Peripheral and desmoplastic Ameloblastomas are considered rare.[5, 32] There were a relatively higher number of cases of desmoplastic variant (7.4%) in this study. Anterior mandible has preponderance for the rare variant, desmoplastic. Sun[33] *et al* also shows anterior mandible predilection for this variant but Ajibola [34] *et al* found posterior mandible predilection for this variant.

CONCLUSION:

Ameloblastoma is a benign, locally aggressive odontogenic tumor. It mostly affects patients in the second to fourth decades of life, with slight male preponderance. Posterior mandible is the site of predilection. Commonly appears as multilocular radiolucent lesions but rare variant i.e. desmoplastic appears as mixed radiopaque-radiolucent lesions. Histopathologically solid variant seen more compared to unicystic variant and desmoplastic variant. Amongst solid type follicular was most prevalent subtype. In comparison to previous studies, minor differences in age, gender and radiographic features were noted.

In contrast to previous studies we had seen relatively higher frequency of cases that was in their second decade of life. Desmoplastic variant had preponderance for anterior mandible region. The differences of clinicopathological data may be influenced by ethnic variation,

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accessibility to medical facilities due to socio-economic status or the availability of documentation.

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