Original article

Ameloblastoma: A Clinical and Histopathological Study of 28 Cases Diagnosed in Benghazi

Marwa Elfergani [1], Mansora Abd Alhamid Hamad[2], Rehab El Maddani [3], Huda AL Jilali [4] and Ali Elmurtadi [5]

[1]Demonstrator, Department of Oral Medicine, Oral Pathology, Oral Diagnosis and Radiology, Faculty of Dentistry, University of Benghazi, Benghazi, Libya.
[2]Lecturer, Department of Oral Medicine, Oral Pathology, Oral Diagnosis and Radiology, Faculty of Dentistry, University of Omar Al-Mukhtar, Albayda, Libya.
[3]Assistant Lecturer, Department of Basic Sciences, Faculty of Dentistry, University of Benghazi, Benghazi, Libya.
[4]Lecturer, Department of Oral Biology, Faculty of Dentistry, University of Benghazi, Benghazi, Libya.
[5]Professor, Department of Oral Medicine, Oral Pathology, Oral Diagnosis and Radiology, Faculty of Dentistry, University of Benghazi, Benghazi, Libya

ABSTRACT

Aims:
The study aims to describe the characteristics of of ameloblastoma in a retrospective sample of cases reported at the faculty of dentistry, University of Benghazi, Libya

Methods
A retrospective analysis of cases diagnosed with a ameloblastoma between 2001 and 2016 at faculty of dentistry - Benghazi-libya were reviewed histologically. The cases were then subclassified according to the basic criteria of the World Health Organization. In addition, cases were analysed in terms of clinicopathologic and histopathologic features.

Results:
A total of 28 ameloblastomas types were identified: 8 conventional, 2 peripheral, and 17 unicystic (4 cases ameloblastoma arising in dentigerous cysts) and 1 case is malignant ameloblastoma. Tumours were distributed throughout the jaws with 26 cases in mandibular, 2 cases in maxilla, (2 cases and cross midline, and 2 cases in an anterior location).

Conclusion:
The clinical epidemiological profile of the patients from the present study is very similar to other populations with regards to gender, age and tumor location, with unicystic ameloblastoma being the most common subtype in our population, in contrast to other reports.

Keywords: *Ameloblastoma, retrospective, histopathology, subtypes, Libya.*

Corresponding author:
Rehab El Maddani
Department of Basic Sciences, Faculty of Dentistry, University of Benghazi, Benghazi, Libya.
E-mail: Rehabmadani25@gmail.com

INTRODUCTION

Ameloblastoma is an odontogenic tumor derived from odontogenic epithelium within a mature fibrous stroma [(1)]. Although classified as a benign tumor, ameloblastoma is a locally aggressive odontogenic tumor with severe clinical implications and has the potential for malignant transformation [(2,3)]. It represents approximately 14% of all jaw cysts and tumors and about 9 to 11 % of all odontogenic tumors [(4,5)]. Based on a report of World Health Organization (WHO) on the odontogenic tumor, ameloblastoma classified (i) unicystic, (ii) solid/multicystic, (iii) peripheral or (extraosseous), and (iv) desmoplastic ameloblastoma [(6)]. However, the new classification of WHO in 2017,

ameloblastomas were narrowed to ameloblastoma (conventional), unicystic, extraosseous/peripheral, and metastasizing variants due to the introduction of perspective views based on updates from genetic studies [7]. The most common type of ameloblastoma is the conventional type, accounting for 90% of all ameloblastomas. Within this group, the most frequent pattern is plexiform and follicular histological patterns. The follicular type can show different kinds of cytological differentiation, such as basal cell, granular, and spindle cell types [8].

Clinically, Ameloblastomas usually present as a painless, slow-growing swelling that develops most often in the jaw near the molars, perforation of mandible or maxilla cortical plates, and involves surrounding soft tissue sinonasal structure [9]. Radiographically, ameloblastomas present as unilocular and multilocular radiolucent lesions surrounded by a radiopaque border, primarily located in the posterior mandibular segment [10].

Ameloblastoma has a reportedly varied geographical prevalence ranging from 11-24% of all odontogenic tumors in North America to an estimated world-wide incidence of 0.5 cases per million person-years [11,12]. Ameloblastoma constituted up to 99% of odontogenic tumors in sub-Saharan Africa, with the mandible being more frequently affected [11,13]. Unfortunately, little is known about ameloblastoma in the Libyan population. Therefore, the present study aimed to assess the prevalence of ameloblastoma in a retrospective sample of cases reported at the faculty of dentistry, University of Benghazi.

MATERIALS AND METHODS

This study is a retrospective analysis of 28 cases diagnosed with a ameloblastoma. The data files were retrieved from the data bank of Department of Oral Medicine, Oral Pathology, Oral Diagnosis and Radiology, Benghazi University Faculty of Dentistry. All cases diagnosed with ameloblastoma during the period between 2001 and 2016 were reviewed histologically of haematoxylin and eosin stained section. The revision was performed by two oral pathologists, one of them is a professor of oral pathology. Any disagreement in the diagnosis was resolved by discussions and seeking a third opinion. The cases were then subclassified according to the basic criteria of the world health organization. In addition, cases were analysed in term of clinicopathologic and histopathologic features. Data was summarised using appropriate statistical methods (counts and percentages) to provide a descriptive picture according to subtypes, site within the mouth and gender and age of cases. The statistical analysis was conducted using excel sheets.

RESULTS

Clinical and histolopathologic characteristics of all cases are summarized in Tables 1 and 2. Seventeen patients were males and 12 females. The mean age at time of diagnosis was 30.7 years (range from 13 to 75 years). Table 3 shows the distribution of ameloblastoma cases on the basis of age. We observed that highest number of cases belonged to age group 10-19,20-29,30-39 years (n=7) for each group. Lowest number of cases were observed in the age group 50-59 years (n=0). Tumors were distributed throughout the jaws with 26 cases in mandibular, 2 cases in maxilla, (2 cases and cross midline, and 2 cases in an anterior location).

Twenty-five cases presented as variably sized swellings of the jaws, 2 cases with ulceration, 3cases with pain, 3 cases with tooth mobility and 1 case with paraesthesia. Data on clinical follow-up and radiological assessment were retrieved from the medical record files wherever possible. five cases are associated with impacted tooth, root resorption and bone resorption occurred in 5 and 3 cases respectively, and 1 patient suffered from recurrence. There was insufficient documentation on follow-up in most of cases.

All 28 ameloblastomas were classified according to WHO classification [7] into the following types: 8 conventional, 2 peripheral, and 17 unicystic (4 cases ameloblastoma arising in dentigerous cysts) and 1 case is malignant ameloblastoma.

Table 1. Clinical and histopathological Features of Ameloblastoma

No	Age	Sex	Site	Diagnosis
1	42Y	F	Mandible (premolar area)	Peripheral ameloblastoma
2	49Y	F	Mandible (premolar area)	Conventional ameloblastoma
3	17Y	M	Mandible (Premolar to ramus area)	Unicystic ameloblastoma(mural)
4	20Y	M	Mandible (molar area)	Unicystic ameloblastoma
5	31Y	M	Mandible (molar area)	Unicystic ameloblastoma
6	35Y	F	Mandible (molar area)	Unicystic ameloblastoma
7	28Y	M	Mandible (molar to ramus area)	Ameloblastoma in odontogenic cyst
8	75Y	M	Mandible (premolar to ramus area)	Conventional ameloblastoma
9	31Y	M	Mandible (ramus area)	Ameloblastoma in odontogenic cyst
10	22Y	F	Mandible (molar to ramus area)	Unicystic ameloblastoma
11	36Y	F	Mandible (premolar to ramus area)	Malignant ameloblastoma
12	19Y	M	Mandible (Premolar to ramus area)	Unicystic ameloblastoma
13	60Y	M	Maxilla (Anterior area)	Conventional ameloblastoma
14	41Y	M	Mandible (third molar area)	Ameloblastoma in odontogenic cyst
15	17Y	M	Mandible (premolar to ramus area)	Unicystic ameloblastoma
16	17Y	M	Mandible (premolar to ramus area)	Conventional ameloblastoma
17	21Y	F	Mandible (premolar to ramus area)	Conventional ameloblastoma
18	15Y	F	mandible(molar area)	Unicystic ameloblastoma
19	41Y	M	Mandible(Anterior area)	Unicystic ameloblastoma
20	41Y	M	Mandible (ramus area)	Conventional ameloblastoma
21	27Y	F	Mandible (molar area)	Unicystic ameloblastoma
22	13Y	M	Mandible premolar to molar and cross midline)	Unicystic ameloblastoma
23	14Y	M	Mandible (molar area)	Unicystic ameloblastoma
24	20Y	F	Mandible (molar area)	Conventional ameloblastoma
25	23Y	F	Mandible (Premolar and cross midline)	Conventional ameloblastoma
26	36Y	M	Maxilla (palate right area)	Peripheral ameloblastoma
27	32Y	M	Mandible (premolar area)	Unicystic ameloblastoma(mural)
28	37Y	F	Mandible (molar to ramus)	Ameloblastoma in odontogenic cyst

Table 2: Histopathological Variants of Ameloblastoma

Histopathological subtype	No
Follicular type	2
plexiform type	2
unicystic type	4
Cystic/ Follicular type	5
Cystic/ plexiform type	2
Follicular/ plexiform type	2
Follicular/desmoplastic type	2
plexiform /desmoplastic type	2
Granular /cystic type	2
Cystic/desmoplastic type	1

Table 3: Distribution of ameloblastoma cases on the basis of age

Age (years)	Number of patients
10-19	7
20-29	7
30-39	7
40-49	5
50-59	0
60-69	1
70-79	1

DISCUSSION

Despite the increasing literature about odontogenic tumors, specifically ameloblastoma, little is known about the clinicopathological aspects and frequency of these tumors in Libya.

In this study, there was a slight male predominance in the distribution of ameloblastoma Male: Female (1.3:1). Similar distribution was observed in a study conducted by Verma et al [13] (1.3:1), Montes et al. [14] (1.2:1), Adebiyi et al. [15] (1.5:1), Poon et al. [16] (1.4:1). In addition, the largest study on ameloblastoma published by Reichart et al. [17] reported a male: female ratio of 1.1:1 showing a slight male predominance. However, Similar distribution was observed in other studies [15,18 ,19].

Ameloblastomas were observed in a wide age range (13–75 years) in our study. This finding is in agreement with most reports in the literature [15-18, 20-27]. In the present study, ameloblastoma occurred more often in the first, second, third decades of life. None of the tumour was found below 10 years of age. According to Kim and Jang [28], the frequency of ameloblastoma in young patients (< 19 years) is relatively low, occurring in only 10 to 15% of all reported cases. Our results show a greater frequency, around 25%, for this group of individuals. In addition, our findings were close to those observed by Chidzonga et al. [29], who showed that two thirds of the patients affected by ameloblastomas were less than 40 years of age

Almost 92,8% of ameloblastomas were located in the mandible, with a very high mandible to maxilla ratio (13:1). This is very high compared with the ratios reported by Okada et al [18]. Reichart et al [17] in an extensive review of all of the cases reported in the literature, found the ratio to be around 5:1. The high occurrence in the mandible in the present institutional study may be due to loss of maxillary ameloblastomas to eye-nose throat surgeons.

According to the distribution of ameloblastoma in various anatomical regions of the jaws, the tumours tend to occur commonly in posterior area of the mandible [19, 30-33]. This finding is also compatible with our results.

In relation to the frequency of the different ameloblastoma histological subtypes, the results of the present study differ from previous reports [19,21,34], which demonstrated a greater occurrence of the solid tumor variant. Our findings are similar to those from Montes et al [14]. Pereira Fet al [31], who observed a greater prevalence of the unicystic subtype.

In accordance with the reports of several authors [30,31], the most clinical manifestation of ameloblastoma is swelling in the area affected by the tumor. Our results are in accordance with previous reports.

An interesting finding in the current study was observed 4 cases of ameloblastoma arising from dentigerious cyst. Various studies report that between 15 and 30% of all ameloblastomas form in the wall of a dentigerous cyst [35]. However, it is not known whether they arise from a neoplastic transformation of cells from an otherwise non neoplastic dentigerous epithelium or arise de novo [36]. one case of ameloblastomas have recurred. it is difficult to arrive at a conclusion regarding the relationship between recurrences observed in different subtypes and treatment modalities, as these data were not available for all the ameloblastomas analysed in this study

In our study we found 1of 28 cases amoloblastic carcinoma deposit of ameloblastic carcinomas were considered as extremely rare malignant odontogenic epithelial neoplasm's that may arouse de novo or from a pre-existing odontogenic lesion [37].

CONCLUSIONS

The clinical epidemiological profile of the patients from the present study is very similar to other populations with regards to gender, age and tumor location, with unicystic ameloblastoma being the most common subtype in our population, in contrast to other reports.

REFERENCES

1. Sciubba JJ, Eversole LR, Slootweg PJ (2005). Odontogenic tumours. In: L Barnes, JW Eveson, P Reichart, D Sidransky, eds. *World Health Organization classification head and neck tumours*. IARC Press: Lyon, pp. 283– 328.
2. Bassey GO, Osunde OD, Anyanechi CE (2014). Maxillofacial tumors and tumor-like lesions in a Nigerian teaching hospital: an eleven year retrospective analysis. *Afr Health Sci* **14**: 56– 63.
3. Odukoya O, Effiom OA (2008). Clinicopathological study of 100 Nigerian cases of ameloblastoma. *Niger Postgrad Med J* **15**: 1– 5.
4. Lasisi TJ, Adisa AO, Olusanya AA (2013). Appraisal of jaw swellings in a Nigerian tertiary healthcare facility. *J Clin Exp Dent* **5**: e42– e47.
5. Abayomi OA SA, Oladepo AE. Ameloblastoma: Clinical features and management of 315 cases from Kaduna, Nigeria. J Cranio-Maxillofac Surg. 1993; 21:351–55.

6. Philipsen H. Odontogenic tumours. WHO classification of head and neck tumours. 2005:283-327.
7. Wright JM, Vered M. Update from the 4th edition of the World Health Organization classification of head and neck tumours: odontogenic and maxillofacial bone tumors. Head Neck Pathol 2017 Mar 1;11(1):68-77.
8. Chi AC, Damm DD, Neville BW, Allen CM, Bouquot J. Oral and Maxillofacial Pathology-E-Book. Elsevier Health Sciences; 2008 Jun 11.
9. Ghandhi D AA, Pogrel MA, MacDonald G, Brocklebank LM, Moos KF. Ameloblastoma: A surgeon's dilemma. J Oral Maxillofac Surg. 2006; 64:1010–14.
10. Gupta S, Sexana S, Bhagwat S, Aggarwal P, Gupta PK. Clinicopathological characteristics of ameloblastomas in Western Uttar Pradesh population: An institutional study. Indian J Cancer. 2015 Jan-Mar;52(1):57-60. doi: 10.4103/0019-509X.175557. 6
11. McClary AC, West RB, McClary AC, Pollack JR, Fischbein NJ, Holsinger CF, Sunwoo J, Colevas AD, Sirjani D. Ameloblastoma: a clinical review and trends in management. Eur Arch Otorhinolaryngol. 2016; 273(7): 1649-1661.
12. JingW,XuanM,LinY,WuL,LiuL,ZhengX,TangW, QiaoJ, Tian W. Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. Int J Oral Maxillofac Surg. 2007; 36(1): 20-25
13. Verma S, Khemka A. Clinical features of ameloblastoma cases in local population-A retrospective study. J of Advan Med & Dent Sci Res. 2019 Dec;7(12).
14. Ledesma-Montes C, Mosqueda-Taylor A, Carlos-Bregni R, De León ER, Palma-Guzmán JM, Páez-Valencia C, Meneses-García A. Ameloblastomas: a regional Latin-American multicentric study. Oral diseases. 2007 May;13(3):303-7.
15. Adebiyi KE, Ugboko VI, Omoniyi-Esan GO, Ndukwe KC, Oginni FO. Clinicopathological analysis of histological variants of ameloblastoma in a suburban Nigerian population. Head & Face Med. 2006 Dec 1;2(1):42.
16. Poon CS, Wu PC, So MK. Ameloblastoma in Hong Kong Chinese. HONG KONG MED J. 1996; 2:172-6.
17. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. Eur J Cancer B Oral Oncol. 1995 Mar 1;31(2):86-99.
18. Okada H, Yamamoto H, Tilakaratne WM. Odontogenic tumors in Sri Lanka: analysis of 226 cases. J Oral Maxillofac Surg. 2007 May 1;65(5):875-82.
19. Iyogun CA, Ukegheson GE, Omitola OG. Evaluation of Histopathological Subtypes of Ameloblastoma in a Tertiary Institution in Southern Part of Nigeria. Int J Oral Maxillofac Pathol. 2014 Oct 1;5(4):07-11.
20. Siar CH, Lau SH, Ng KH. Ameloblastoma of the jaws: a retrospective analysis of 340 cases in a Malaysian population. J Oral Maxillofac Surg. 2012 Mar 1;70(3):608-15.
21. Nasrollah Saghravanian, Jahanshah Salehinejad, Narges Ghazi, Mohammad Shirdel, Mahsa Razi. 40-year Retrospective Clinicopathological Study of Ameloblastoma in Iran. Asian Pac J Pre,17 ,619-623.
22. Lu Y, Xuan M, Takata T, Wang C, He Z, Zhou Z, Mock D, Nikai H. Odontogenic tumors: a demographic study of 759 cases in a Chinese population. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998 Dec 1;86(6):707-14.
23. Hong J, Yun PY, Chung IH, Myoung H, Suh JD, Seo BM, Lee JH, Choung PH. Long-term follow up on recurrence of 305 ameloblastoma cases. Int J Oral Maxillofac Surg. 2007 Apr 1;36(4):283-8.
24. Ladeinde AL, Ajayi OF, Ogunlewe MO, Adeyemo WL, Arotiba GT, Bamgbose BO, Akinwande JA. Odontogenic tumors: a review of 319 cases in a Nigerian teaching hospital. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005 Feb 1;99(2):191-5.
25. Tilakaratne WM, Jayasingha RD. Pattern of recurrences of ameloblastoma in Sri Lanka. Sri Lanka Dent J 2004; 34: 20–8.
26. Olgac V, Koseoglu BG, Aksakallı N. Odontogenic tumours in Istanbul: 527 cases. Br J Oral Maxillofac Surg. 2006 Oct 1;44(5):386-8.
27. Avelar RL, Antunes AA, de Santana Santos T, de Souza Andrade ES, Dourado E. Odontogenic tumors: clinical and pathology study of 238 cases. Braz J Otorhinolaryngol. 2008 Sep 1;74(5):668-73.
28. Kim SG, Jang HS. Ameloblastoma: a clinical, radiographic, and histopathologic analysis of 71 cases. Oral Surgery, Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001 Jun 1;91(6):649-53.

29. Chidzonga MM, Perez VL, Alvarez AP. Ameloblastoma: the Zimbabwean experience over 10 years. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1996 Jul 1;82(1):38-41.
30. Karadiguddi P, Mengi R, Melkundi M, BV H, Premnath K. Clinicopathological characteristics of Ameloblastomas in a given population-A retrospective study. J Advan Med & Dent Sci Res. 2019 Jul;7(7):40-43.
31. Pereira F, De Araujo Melo E, Silva Gurgel CA, Cangussu MC, De Azevedo RA, Dos Santos JN. Clinicopathological and demographic characteristics of ameloblastomas in population of Bahia, Brazil. Rev OdontoCienc 2010; 25:2505.
32. Shekhar S, Priya P, Jain S, Verma SK. Histopathological analysis of types of Ameloblastoma: A retrospective study. Pharma Innov J 2019; 8(1): 499-501
33. Naz I, Mahmood MK, Akhtar F, Nagi AH. Clinicopathological evaluation of odontogenic tumours in pakistan-A seven years retrospective study. Asian Pac J Cancer Prev. 2014 Jan 1;15(7):3327-0.
34. El AN, Latif SM. Clinico pathological and immunohistochemical study in odontogenic cysts and ameloblastoma (using laminin-1). Am J Life Sci. 2013;1(3):130-5.
35. Wood NK, Kuc IM. Pericoronal radiolucencies. In: Differential diagnosis of oral and maxillofacial lesion. 5th ed. St.
36. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: a clinicopathological study of 57 cases. J Oral Pathol Med. 1988 Nov;17(9-10):541-6.
37. Avon SL, McComb J, Clokie C. Ameloblastic carcinoma: case report and literature review. J Can Dent Assoc. 2003 Oct;69(9):573-6.