

Original article

A STUDY OF THE HISTOPATHOLOGICAL VARIANTS OF UNICYSTIC AMELOBLASTOMA IN A TERTIARY HEALTH CENTRE IN SOUTH- SOUTH, NIGERIA

Omitola O G, Iyogun CA, Brown A

Department of Oral Pathology and Oral Biology of the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria

ABSTRACT

OBJECTIVE: The aim of this study was to determine the prevalence, clinical profile and histopathological types of all the cases of unicystic ameloblastoma in a Nigerian population.

METHODS: In this retrospective study, case files and biopsy reports of new cases of unicystic ameloblastoma seen at the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria from 2009 to 2017 were retrieved and analyzed for gender, age on presentation, histologic type and site distribution.

RESULTS: A total of 52 cases of ameloblastoma were seen during the period under review among which unicystic ameloblastoma accounted for 19 (36.5%) of the cases. A male to female ratio of 1:1.4 was found. The average age on presentation was 25 years. The lesion was most common in the posterior mandible and the mural variant was the most common histological type.

CONCLUSIONS: The age, gender and site distributions of unicystic ameloblastoma are similar to previous reports but the distribution of the histological types is at variance with previous studies.

KEY WORDS: Unicystic, Ameloblastoma, South-south, Nigeria

Correspondence address:

Dr. C.A. Iyogun Department of Oral Pathology & Oral Biology of the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria
iyoguncornelius@yahoo.com
Tel: +2348033132937

INTRODUCTION

Ameloblastoma is a neoplasm of odontogenic epithelium¹. It is generally a slow-growing but locally invasive tumour. Almost all ameloblastomas are histologically benign. Nevertheless, they may behave in a rather aggressive way by local recurrences when treated by enucleation only.²

Ameloblastoma is the most common odontogenic tumour reported in Nigeria, South Africa and found to be more common among blacks than whites³. According to Arotiba et al, peculiar presentations of this tumour in Black Africans are the late presentation of many patients with massive tumours and the high proportion of patients who are below 20 years at

diagnosis. Other peculiarities include the high predilection for involvement of the symphyseal region of the mandible and the low proportion of the unicystic morphologic type².

It has been postulated that the epithelium of origin is derived from one of the following sources: (1) cell rests of the enamel organ, (2) epithelium of odontogenic cysts, (3) disturbances of the developing enamel organ, (4) basal cells of the surface epithelium, or (5) heterotropic epithelium in other parts of the body⁴. The theory of an odontogenic origin for the ameloblastoma is supported clinically by the tumour's common occurrence in the tooth bearing area and is further reinforced by the finding of Spouge that one in every three such

tumours are mural proliferations in intimate association with the reduced enamel-forming epithelium of dentigerous cysts⁵.

WHO 2017 classification divided ameloblastoma into four categories; conventional, extraosseous / peripheral, unicystic, and metastasizing ameloblastoma⁶. Unicystic ameloblastoma (UA) often presents radiographically as a unilocular, well-demarcated radiolucency that surrounds the crown of an unerupted tooth, resembling a dentigerous cyst. When the tumour grows into the lumen it is called the 'intraluminal type' or when confined to the cyst lining epithelium, it is called the 'luminal type'. If the tumour invades the wall of the cyst, it is called 'mural type'.

Unicystic ameloblastomas have been traditionally treated conservatively, often by "cyst" enucleation, and recurrence has been uncommon. However, there is emerging evidence that unicystic ameloblastomas with mural invasion are known to act as conventional intraosseous ameloblastoma and should be treated as such⁷.

There is dearth of studies of histopathological types of unicystic ameloblastoma among black Africans. This study was therefore designed to determine the prevalence, clinical profile and histopathological types of all the cases of unicystic ameloblastoma in a Nigerian population. This will serve as baseline data for the Centre.

MATERIALS AND METHODS

This was a retrospective review of the clinical case files and biopsy records of all histologically diagnosed cases of unicystic ameloblastoma (UA) from 2009 to 2017 in the Department of Oral Pathology and Oral Biology of the University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria. Nineteen cases of unicystic ameloblastoma were extracted from the records. Hematoxylin and eosin (HE)-stained sections of the 19 cases were retrieved and reviewed to confirm the diagnosis. The 19 confirmed cases were there after categorized into the following 3 variants: luminal, intraluminal and mural based on the 2017 WHO classification of odontogenic tumors. Data on

prevalence age, sex, site and histology of lesions were analyzed descriptively for the various variants of unicystic ameloblastoma. Data analysis was performed with SPSS (version 21, SPSS Inc. Chicago, IL).

RESULTS

During the period of the study, 52 cases of ameloblastoma were histologically diagnosed consisting of 33 (63.5% conventional and 19 (36.5%) unicystic ameloblastoma. (Table1).

Table I: Frequency distribution of histological types of ameloblastoma in the series

Sub type	N	%
Conventional	33	63.5
Unicystic	19	36.5
Peripheral	0	0.0
Malignant	0	0.0
Total	52	100

The patients with UA varied in ages from 10 to 60 years (mean 25 ± 17.08 , median 22.00) with peak prevalence round the 2nd decade of life (Table II). UA was observed to occur slightly more in females (57.9%) than in males (42.1%), with a male: female ratio of 1:1.4.

Table II: Age range in patients with unicystic ameloblastoma

Age range	N	%
0-10	1	5.3
11-20	8	42.1
21-30	2	10.5
31-40	3	15.8
41-50	3	15.8
51-60	2	10.5
Total	19	100

All the tumour cases were exclusively observed in the mandible as no case of UA was seen in the maxilla. The right mandible was the commonest site, accounting for 6 (46.1%) cases of UA tumours (Table III).

The posterior region with 12 (63.2%) patients was affected more often than the anterior segment with 7(36.8%) patients (Figure1).

Table III: Mandibular site distribution of the patients with unicystic ameloblastoma

Location	N	%
Left	5	38.5
Right	6	46.1
Bilateral	2	15.4
Total	13	100

The Mural UA was the most common histologic variant with 11 (57.9%) cases (Figures 2). This was followed by 7 (36.8%) cases of Luminal UA. (Table IV). [Figure 3].

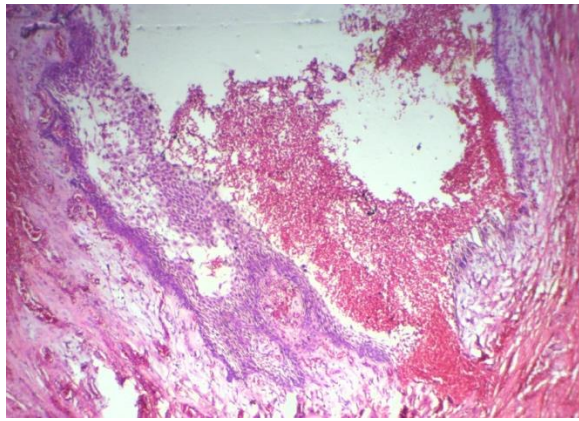


Figure 2: Photomicrograph of mural unicystic ameloblastoma lined by odontogenic epithelium invading the surrounding fibrous capsule (H&E x100)

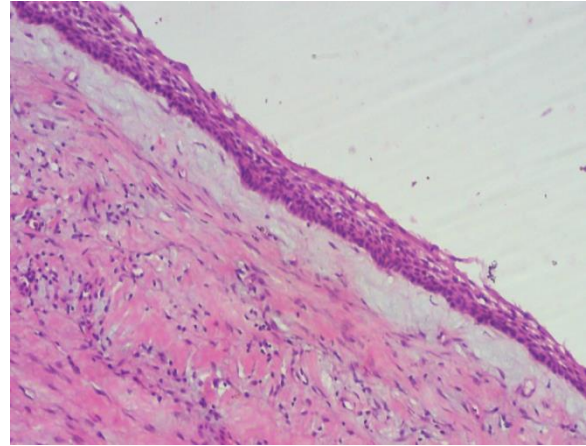
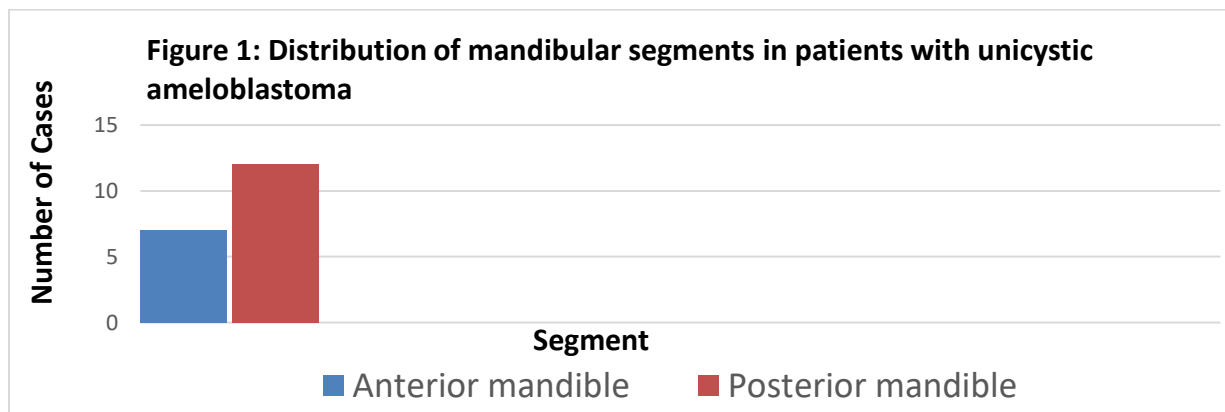


Figure 3: Photomicrograph of luminal unicystic ameloblastoma lined by odontogenic epithelium with prominent hyalinized layer in the outer capsule (H&E x100)

Table IV: Distribution of histologic subtypes in patients with unicystic ameloblastoma

Location	N	%
Intra Luminal	1	5.3
Luminal	7	36.8
Mural	11	57.9
Total	19	100



DISCUSSION

Previous report by Lawal et. al⁸ in Ibadan, Nigeria (2014) suggests a low prevalence (14.3%) of UA among ameloblastoma in Nigerians compared with the 18.9% prevalence of UA report by Tie-Jun Li et al among the

Chinese⁹. However, this study observed a higher prevalence (36.5%) of UA among ameloblastoma. This supports a recent report of a relatively higher prevalence (42.2%) of UA in a South-south Nigerian population by Omoregie et. al (2017)¹⁰.

Participants in this study ranged from 10 years to 60 years, this age range is similar to reports in the literature⁹. The mean age of patient at presentation in this study was 25 years, this is almost equal with a mean age of 23.5 years which was reported by Ackerman et al¹¹(2006) in a study on ameloblastoma. The mean age in this study lies between a value of 18 years reported by Roos et al in 1998¹² and 25.3 years reported by Tie-Jun Li et al in 2000⁹. The peak age group in this study was in the second decade of life, which supports the literature that unicystic ameloblastoma occurs more in younger age group than solid variant of ameloblastoma. Findings in this study are similar to that of Tie-jun Li et al who reported that 70% of their cases occurred in the 2nd and 3rd decades of life⁹.

Unicystic ameloblastoma cases in this study were seen more in females with a male: female ratio of 1:1.4. This is similar to a female gender predilection with M: F= 2:3 reported by Lawal et al.⁸ Findings from this study are in contrast with those observed by Tie-Jun Li et al where there was a male preponderance with M: F =7:4⁹.

Unicystic ameloblastoma was exclusively found in the mandible in this study. This observation is similar to reports in the literature that suggest a predilection for the mandible. Also, it was observed that 63.2% of cases occurred in the molar/premolar region. This again is similar to reports by Kumar et al¹³. Perhaps, the reason for this predilection for posterior mandible is because research has shown that this is also the most common site for impacted teeth which have been associated with several cases of cystic ameloblastoma. Mortazavi et al in 2016 noted that 50-80% of cases of unicystic ameloblastoma are associated with at least one unerupted tooth (mostly the third molar)¹⁴.

Mural variant of unicystic ameloblastoma was the most common histological type representing 53.9% of the cases seen. The observation in this study is similar to reports by Lawal et al⁸, Omoregie et al¹⁰ and Philipsens et al¹⁵ who reported a preponderance of mural variant of UA. The histopathological differentiation of unicystic ameloblastoma to luminal, intraluminal and mural variants is of clinical relevant. This is

because the luminal and intraluminal types are treated conservatively with simple enucleation, while it is now increasingly recognized that the mural type must be treated with surgical excision with safe margin. Treatment of the mural variant with conservative method has been associated with recurrence⁷.

In conclusion, this study observed a relatively higher prevalence of UA over a 9-year period. However, the small sample size makes it difficult to provide a definitive inference. There was predilection of UA for the females, young adults and posterior mandible. The mural UA was the commonest histological type.

Conflict of Interest: None declared

REFERENCES

1. Woo S, Smith- Williams JE, Sciubba JJ, Lippers S. Peripheral Ameloblastoma of the buccal mucosa: Case Report and Review of the English Literature. *Oral Surg Oral Med Oral Pathol* 1987; 67:78-84.
2. Arotiba GT, Hille J, Guthua SW, Adeola H, Odhiambo W, Effiom OA, Osunduwa T, Kirimi J, Dimba E, Olojede ACO, Gbotolorun OM, Adamson O. Ameloblastoma in Black Africans the need for Multi-National collaborative research. *JSM Dent Surg* 2017; 2(2): 1014.
3. 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; 5(4):07-11.
4. Rahman SB, Sadat SMA, Haider IA, Ahmed M. Analysis of histological variants of ameloblastomas of jaws in relation to their clinical presentations. *J Bangladesh Coll Phys Surg* 2017; 35: 61-67.
5. Spouge JD. Odontogenic Tumours: A Unitarian Concept. *Oral Surg Oral Med Oral Pathol* 1967; 24:394-403.
6. Wright JM, Tekkesin MS. Odontogenic tumours: Where are we in 2017? *J Istanbul Univ Fac Dent* 2017; 51:10-30.
7. Philipsen HP, Reichart Pa. Unicystic ameloblastoma. A review of 193 cases from the literature. *Oral Oncol* 1998; 34:317-325.
8. Lawal AO, Adisa AO, Olajide MA. Cystic ameloblastoma: A clinic-pathological review. *Ann Ibd Pg Med* 2014; 12:49-53.

9. Li TJ, Wu YT, Yu SF, YU GY. Unicystic ameloblastoma: a clinicopathological study of 33 chinese patients. *Am J Surg Pathol*. 2000; 24:1385-1392.
10. Omoregie F.O., Orikpete E. Ojo M.A. Analysis of histopathological subtypes of unicystic ameloblastoma in a Nigerian population. *Nig J Dent Research*. 2017; 2(1): 5-9.
11. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: a clinicopathological study of 57 cases. *J Oral Pathol* 1988;17:541–546.
12. Roos RE, Raubenheimer EJ, Van Heerden WF. Clinico-pathological study of 30 unicystic ameloblastomas. *J Dent Assoc S Afr* 1994; 49(11):559-562.
13. Kumar Kumar KR, George GB, Padiyath S, Rupak S. Mural unicystic ameloblastoma crossing the midline: a rare case report. *Int J Odontostomat* 2012; 6(1):97-103.
14. Mortazavi H, Baharvand M. Jaw lesions associated with impacted tooth: A radiographic diagnostic guide. *Imaging Sci Dent* 2016; 46: 147-157.
15. Philipsen HP, Reichart PA. Revision of the 1992- edition of the WHO histological typing of odontogenic tumours. A suggestion. *J Oral Pathol Med* 2002; 31:253-258.