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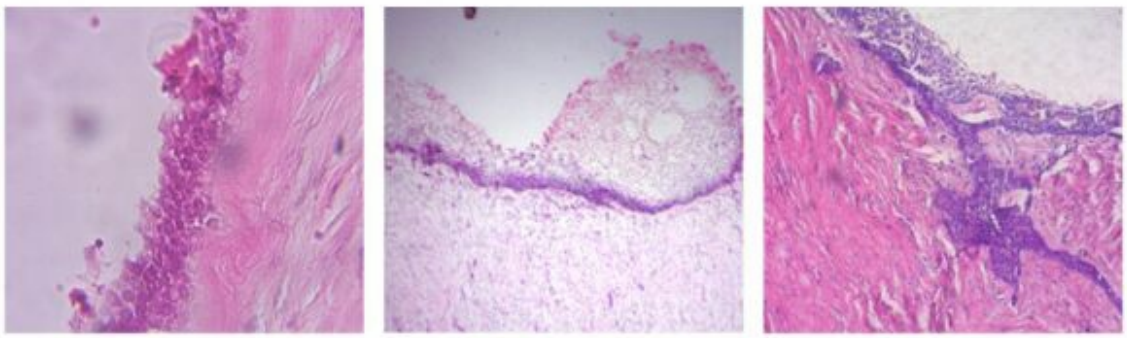

Analysis of Histopathologic Subtypes of Unicystic Ameloblastoma in a Nigerian Population

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ABSTRACT

Objective: Recent studies recommend that the treatment of unicystic ameloblastoma should be varied depending on the histopathologic subtypes of the lesion. This study therefore aims to determine the prevalence, age, gender, jaw-site and histopathologic subtypes of unicystic ameloblastoma in the South-South Nigerian population.

Methods: A 16-year retrospective review of 1473 diagnosed orofacial lesions, including 192 cases of ameloblastoma, among which the unicystic ameloblastomas were selected and their clinicopathological features were analyzed in the Department of Oral Pathology and Medicine, University of Benin Teaching Hospital, Benin City.

Results: There were 81(5.5%) cases of unicystic ameloblastomas, among the diagnosed orofacial lesions, accounting for 42.2% of ameloblastomas in this Centre. The patients' mean age was 31 $\pm$ 1.5 years, with the 2nd and 3rd decades of life as the peak age group (n=46, 56.8%) and a male to female ratio of 1.5:1. The mean duration on presentation was 34.6 months $\pm$ 6.7 S.E and anterior mandible (n=40, 49.4%) was the predominant site of the lesion. The mural unicystic ameloblastoma (n=61, 75.3%) was the most common histopathologic subtype of the lesion, followed by the luminal (n=13, 16.0%) and the intraluminal (n=7, 8.6%) subtypes.

Conclusion: This study observed a higher prevalence of unicystic ameloblastoma compared to previous reports. The lesions were found mostly in young males with predilection for the anterior mandible. The mural unicystic ameloblastoma was the predominant subtype in this population, which may require local surgical resection because of the higher risk of recurrence.

Keywords: Unicystic-ameloblastoma, clinical features, Histopathologic-subtype

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INTRODUCTION

Ameloblastoma is a benign but locally aggressive odontogenic neoplasm of epithelial origin.[1-3] It has been categorized into four distinct clinicoradiological types: solid/multicystic, unicystic, peripheral and desmoplastic.[4] Unicystic ameloblastoma is a type of ameloblastoma that presents clinically and radiologically with features of a jaw cyst.[1,4,5] However, histology of the cystic cavity shows ameloblastomatous epithelial lining with or without luminal and/or mural tumour proliferation.[5,6]

Unicystic ameloblastoma accounts for 10-15% of all intraosseous ameloblastomas.[6,7] It is a less aggressive variant of ameloblastoma[1] that occurs commonly in the second and third decades of life, with equal gender predilection.[1,4] The mandible, especially the posterior part is commonly affected.[1,4-9] Microscopically, there are three variants of unicystic ameloblastoma: luminal, intraluminal and mural.[8] The luminal variant comprises a cystic lining composed focally or uniformly of ameloblastomatous epithelium.[4] In the intraluminal variant, there is proliferation of the ameloblastomatous epithelium into the cystic cavity. The mural variant shows proliferation and invasion of the underlying cystic capsule by the ameloblastomatous epithelium.[4] The different variants of unicystic ameloblastoma show different clinical aggression and recurrence rate.[8] Thus, recent studies recommend different treatment of the lesion depending on the histopathologic variant. Enucleation is believed to be adequate treatment for the luminal and intraluminal variants.[1,4,8] The mural type may require local surgical resection. Recurrence rate for unicystic ameloblastoma is reported to be 10% to 20%, hence post-surgical follow up for possible recurrence is recommended.[4,7]

Some Nigerian studies have reported on the clinical and histopathologic types of

ameloblastoma.[9-13] However, there is paucity of studies reporting specifically on unicystic ameloblastoma in our environment. This study aimed to determine the prevalence, age, gender, jaw-site and the histopathologic variants of unicystic ameloblastoma in a South-South Nigerian population.

MATERIALS AND METHODS

This was a 16-year (January, 2000 - May, 2016) retrospective review of the clinical records, histopathology slides and reports of 1473 diagnosed orofacial lesions in the Department of Oral Pathology and Medicine, University of Benin Teaching Hospital, Benin City. Cases of unicystic ameloblastoma were selected among the 192 diagnosed cases of ameloblastomas and analyzed. The clinical variables of the unicystic ameloblastoma analyzed were the prevalence among diagnosed orofacial lesions and ameloblastomas, age, gender, jaw sites and duration of the lesion. The histopathologic variants of the unicystic ameloblastoma analyzed were Luminal type, Intraluminal type and Mural type. The data were analyzed using the Statistical Package for the Social Sciences (SPSS version 16). Pearson's chi square test was performed. The confidence level was set at 95% and probability (P) values of <0.05 were regarded as significant. Ethical approval was obtained from the Hospital Ethical Committee for the study.

RESULTS

There were 81 (5.5%) cases of unicystic ameloblastoma among the diagnosed orofacial lesions, accounting for 42.2% among the cases of ameloblastoma in this Centre. The patients' age range was from 10 -73 years. The mean age was 31 ± 1.5 years. The 2nd and 3rd decades of life were the peak age groups (n=46, 56.8%). The male (n=49, 60.5%) to female (n=32, 39.5%) ratio was 1.5:1 (Table 1).

Table 1: Age and gender distribution of the patients

Age group	Gender		Total
	Male	Female	
0 – 10	1	0	1
11 – 20	12	11	23
21 – 30	15	8	23
31 – 40	10	7	17
41 – 50	3	3	6
51 – 60	6	2	8
61 – 70	1	1	2
71 – 80	1	0	1
Total	49	32	81

Total493281The mean duration on presentation was 34.6 months (± 6.7 SE). Lesions of 12 and 24 months duration were significantly associated with the 2nd (n=8, 9.9%) and 4th decades (n=6, 7.4%) of life [p=0.001] respectively. The anterior mandible (n=40, 49.4%) was the predominant site of the lesion, followed by the posterior mandible (n=18, 22.2%) (Table 2).

Table 2: Jaw sites of the unicystic ameloblastoma

Site	Frequency	Percent (%)
Anterior mandible	40	49.4
Posterior mandible	18	22.2
Anterio–posterior mandible	14	17.3
Anterior maxilla	3	3.7
Posterior maxilla	4	4.9
Anterior–posterior maxilla	2	2.5
Total	81	100

Table 3: Gender distribution of the histopathologic subtypes of the lesion

Gender	Variants			Total
	Mural	Luminal	Intraluminal	
Male	37	7	5	49
Female	24	6	2	32
Total	61	13	7	81
(%)	75.3	16.1	8.6	100

There was male predilection for all the histopathologic subtypes of the unicystic ameloblastoma (Table 3). The mural unicystic ameloblastoma (n=61, 75.3%) was the most common histopathologic subtype of the lesion. It was followed by the luminal (n=13, 16.1%) and the intraluminal (n=7, 8.6%) subtypes. The mural type occurred mostly in the 3rd decade of life (n=18, 22.2%). The anterior mandible accounted for 31 (38.3%) cases of the mural type (Figure 1). The luminal type occurred predominantly in the 2nd decade of life (n=6, 7.4%), with the anterior mandible accounting for 7(8.6%) cases of the luminal type (Figure 2). The intraluminal type also occurred mostly in the 2nd decade of life (n=3, 3.7%). The sites most commonly affected were the anterior and anterio-posterior mandible,

accounting for 2 (2.5%) cases each of the intraluminal type (Figure 3)

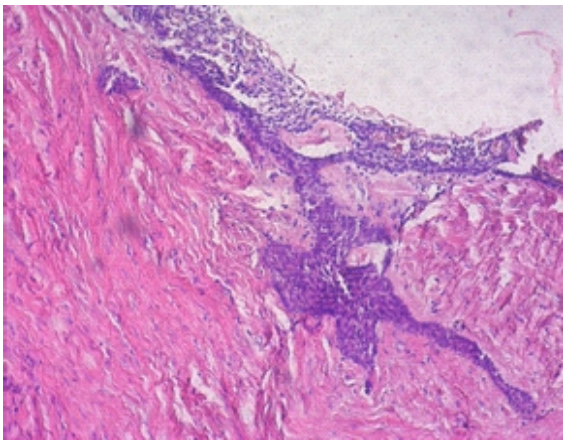

Figure 1: Mural unicystic ameloblastoma showing invasion of the cystic fibrous connective tissue capsule by the ameloblastomatous epithelial lining. (H & E, x 100)

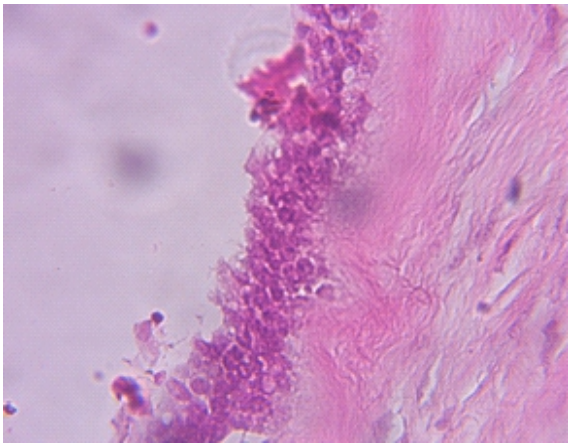

Figure 2: Luminal unicystic ameloblastoma showing cystic cavity lined by inner ameloblastomatous epithelium and outer fibrous connective tissue capsule without tumour invasion. (H & E, x 400)

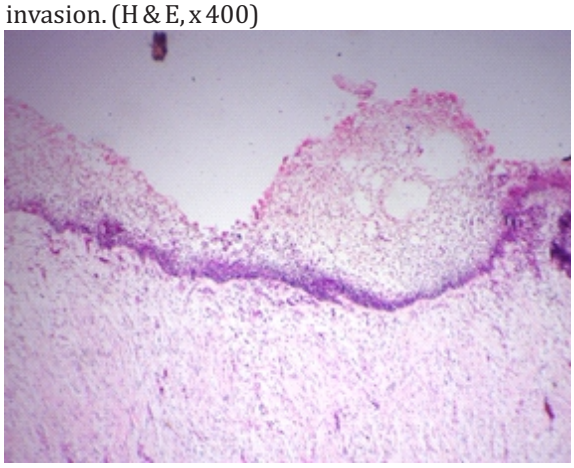

Figure 3: Intraluminal unicystic ameloblastoma showing proliferation of the ameloblastomatous epithelial lining into the cystic cavity without invasion of the underlying fibrous capsule. (H & E,

DISCUSSION

Previous studies have reported a low prevalence of unicystic ameloblastoma among cases of ameloblastoma which ranges from 10-15%.[6,7] However, this study in the South-South Nigerian population observed a higher prevalence (42.2%) of unicystic ameloblastoma among cases of ameloblastoma in this Centre. The lesions were found mostly in young male patients, especially in the 2nd and 3rd decades of life, which is comparable to previous reports.[1,7,8] In contrast to earlier studies that reported equal gender distribution and the posterior mandible as the most common site for unicystic ameloblastomas,[1,4,7] the anterior mandible was the predominant jaw-site for the lesions in this study. These studies support a predilection of unicystic ameloblastoma in young patients, with variable gender and jaw-site distribution of the lesion.

There is agreement in previous studies[1,7,8] that unicystic ameloblastoma is found mostly in the 2nd and 3rd decades of life, with the mural type as the most frequent histopathological variant. Similarly, this study observed that the mural unicystic ameloblastoma was the most common subtype. Furthermore, the mural type was found frequently in the 3rd decade of life, while the luminal and intraluminal subtypes were found commonly in the 2nd decade of life. These findings suggest that patient's age may be useful as clinical indicators for predicting the subtypes of unicystic ameloblastoma. However, the diagnosis of unicystic ameloblastoma and its variants is based only on their histopathological features.

Earlier study[14] of unicystic ameloblastoma regarded the lesions as a rare variant (with a prevalence of 5 to 10%) of intraosseous ameloblastoma, with clinicoradiological features similar to mandibular cyst. Therefore, both lesions were similarly treated by simple enucleation. Recent studies have differentiated primary unicystic ameloblastoma from unicystic ameloblastoma arising from odontogenic cysts, especially radicular cyst / infected radicular cyst and dentigerous cyst. These cysts have ameloblastomatous-like cystic lining that may mimic unicystic ameloblastoma.[15,16] Different treatment options have been recommended depending on the histopathologic variant of unicystic ameloblastoma. Enucleation is thought to be adequate treatment for the luminal and intraluminal variants, while the mural type may require local surgical resection and post-surgical follow up for possible recurrence.[1,4,7,8,14-17]

In a study on the recurrence rate of unicystic ameloblastoma, a higher rate of recurrence was

reported among the mural type (35.7%) compared to the other subtypes (6.7%) after 4 years post-operative follow up.[18] Lau et al.[19] in their study on the treatment modalities of 132 patients with unicystic ameloblastomas found that surgical resection reduced recurrence rate to 3.6%, while recurrence rate was 30.5% for enucleation alone, 16% for enucleation followed by application of Carnoy's solution, and 18% for marsupialization. The mural type was the predominant subtype in the present study; accordingly local surgical resection and long term follow up of the lesion is also recommended in this study population. Further study is also recommended to ascertain the effectiveness of this treatment in reducing recurrence rate of unicystic ameloblastoma in this environment.

CONCLUSION

In conclusion, this study observed a higher prevalence of unicystic ameloblastoma compared to previous reports. The lesions were found mostly in young males with predilection for the anterior mandible. The mural unicystic ameloblastoma was the predominant subtype in this population. Local surgical resection may be required for the mural type due to the higher risk

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