ORIGINAL ARTICLE

Ameloblastic carcinoma of jaw bones: Study of 31 cases from Enugu, Nigeria

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ABSTRACT

Background: Ameloblastic carcinoma is a rare malignant form of ameloblastoma accounting for 0.3–3.5% of all odontogenic tumours. Its management is controversial due to its rarity. This study assessed the pattern, management, and outcome of patients diagnosed with ameloblastic carcinoma at the University of Nigeria Teaching Hospital, Enugu, Nigeria.

Method: This was a retrospective study of all patients diagnosed with ameloblastic carcinoma of the jaw bone at the University of Nigeria Teaching Hospital Enugu, Nigeria between January 2018 and December 2023. The data of interest collected included the age and sex of patients, duration of the lesion (from when the patient first noticed to the time diagnosis was established), site and side of the lesion, treatment, and recurrence on follow-up.

Results: A total of 31 cases of ameloblastic carcinoma were seen during the study period. The patients' ages at diagnosis ranged from 12 to 54 years, with a mean age of 30.58 (SEM = 2.01) years. There was almost an equal distribution of participants by sex (M: F= 1.1:1). The mandible was more affected (93.5%) than the maxilla. All cases that turned out for management (n=24) were managed surgically, with the most frequent (50.0%) procedure being segmental resection of the mandible. Additional surgical procedures included reconstruction of the defect using a metal plate (n= 9) and selective neck dissection (n=3). Adjuvant radiotherapy was given to some (n=13) patients while adjuvant chemotherapy was given to only 3 of patients. During the follow-up, none of the patients had a recurrence of the lesion.

Conclusion: Ameloblastic carcinoma is a rare odontogenic tumour that occurs more in the mandible than the maxilla. It occurs almost throughout all age groups however, those affecting the maxilla appear to occur at an older age group. The mainstay treatment involves resection of the affected jaws with a margin of safety. Radiotherapy and chemotherapy may have a role in its treatment.

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INTRODUCTION

Though ameloblastic carcinoma (AC), a rare malignant form of ameloblastoma accounts for 0.3–3.5% of all odontogenic tumours [1,2] it is considered to be the most common malignant epithelial tumour of odontogenic origin.[1] The age range of patients diagnosed with ameloblastic carcinoma is wide, ranging between 15 to 84 years with a mean age of approximately 30 years,[3] and affects males slightly more compared to females.[4] It tends to affect the mandible more than the maxilla.[3,5] Histologically, ameloblastic carcinoma has some features of ameloblastoma, however, it presents with cytologic atypia, poor differentiation, and a high mitotic index.[6] Despite having no standard guideline for its treatment, surgical resection is the first treatment with or without adjuvant radiotherapy and/or chemotherapy.[2]

Because of the rarity of ameloblastic carcinoma, there have been limited studies[1,4] from Nigeria regarding this disease. However, to the best of our knowledge, there is no study from the southeastern part of Nigeria. This lack of documentation has led to a poor understanding of the proportion, pattern, and treatment protocols used in our locality, and the subsequent outcome of the management offered. Therefore, this study aimed to describe the pattern, management, and outcome of patients diagnosed with ameloblastic carcinoma at the University of Nigeria Teaching Hospital, Enugu, Nigeria.

METHODS

This was a retrospective study of all patients diagnosed with ameloblastic carcinoma of the jaw bone at the University of Nigeria Teaching Hospital Enugu, Nigeria between January 2018 and October 2023. The study was approved by the Health Research Ethics Committee of the University of Nigeria Teaching Hospital Enugu, Nigeria. All cases of jaw tumours were identified and those involving ameloblastoma were isolated. All cases from 2nd January 2018 to 31st December 2023 that had been histologically diagnosed as ameloblastic carcinoma and managed in the centre were included. Reports with inconclusive diagnoses or without a final diagnosis though clinically suspected to be ameloblastic carcinoma were excluded. Postoperative follow-up of the patients was done at 1 month, 3 months, 6 months, 12 months, and 24 months.

The data of interest collected included the age and sex of patients, duration of the lesion (from when the patient first noticed to the time diagnosis was established), site and side of the lesion, treatment, and recurrence on follow-up. Data analysis was done using IBM SPSS Statistics BM for Windows, Version 27.0. Armonk, NY: IBM Corp). Descriptive statistics were applied for proportions, categorical, and continuous data.

Confidentiality of participants' information was ensured throughout the process of data collection, analysis, interpretation, and presentation. Participants were enrolled after obtaining informed consent.

RESULTS

Two hundred and four cases of tumours with histological characteristics of ameloblastoma were seen within the study period, of which 31 cases of ameloblastic carcinoma were recorded. This represents a prevalence of 15.9%. The patients' ages at diagnosis ranged from 12 to 54 years, with a mean age of 30.58 (SEM = 2.01) years. Majority (N=20, 64.5%) were aged 30 years and below [Table 1]. There was almost an equal distribution of participants by sex (M: F= 1.1:1).

The period between the initial presentation of the tumour to the diagnosis ranged between 1 year and 15 years with a mean of 4.92 (SEM = 0.61) years. The mandible was more affected (N=29, 93.5%) than the maxilla at a ratio of 14.5: 1 [Table 1]. Most (N=12, 38.7%) cases had involved the jaws bilaterally, while the left side was affected in 10 patients (32.3%) and the right side was affected in 9 patients (29.0%).

Table 1: Overall distribution of patients according to age groups, sex, and the affected jaw

Age and sex of the participants	The Affected Jaw	
	Mandible (N= 29)	Maxilla (N= 2)
Age Groups (years)		
≤30	20 (69.0%)	-
>30	9 (31.0%)	2 (100.0%)
Mean age (years)	29.5	46.5
Sex		
Female	15 (51.7%)	-
Male	14 (48.3%)	2 (100.0%)

Regarding the treatment offered, 24 patients (77.4%) were managed while 7 (22.6%) did not show up for treatment. The most frequent (N=12, 50.0%) surgical procedure carried out was segmental resection of the mandible (Figure 1). Half of the patients (N=12, 50.0%) had an additional surgical procedure after primary surgery, of which 9 (75.0%) had reconstruction of the defect using a metal plate and 3 (25.0%) had undergone selective neck dissection.

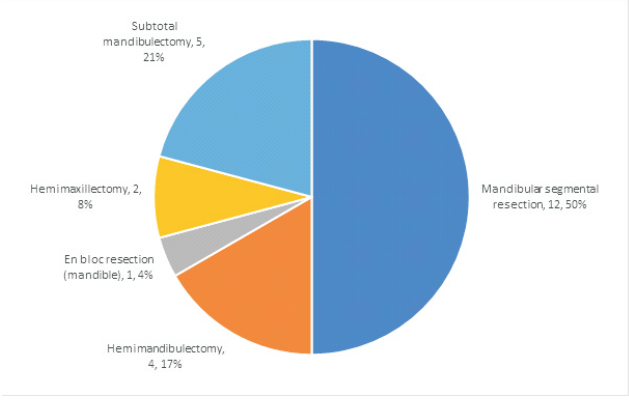

Figure 1: Distribution of patients according to the type of surgical procedure carried out.

Adjuvant radiotherapy after surgery was given to 13 (54.2%) patients, while adjuvant chemotherapy was given to only 3 (12.5%) patients. None of the patients received both chemotherapy and radiotherapy.

All patients who had undergone treatment were followed up for a period ranging from 1 month to 24 months, with a median duration of 8 months (IQR=2,13). During the follow-up, none of the patients had a recurrence of the lesion.

DISCUSSION

The prevalence of Ameloblastic carcinoma (AC) among cases of ameloblastoma has not been previously published however it has been reported to constitute 0.3–3.5% of all odontogenic tumours.[1,2] AC can occur over a wide range of ages.[7] The mean age of the patients in this study is less than other studies from Nigeria. Ndukwe *et al*.[1] reported a mean age of 41.63 years while a more recent study by Soyeye *et al*.[4] reported a mean of 36.8 years. Both studies were carried out in the western part of the country, unlike the present study which was conducted in the eastern part of Nigeria. Our finding is however similar to that reported by Corio *et al*.[7] and Ramesh *et al*.[8] who reported a mean age of 30.1 and 32 years respectively. A much higher mean of 53 years has also been reported previously.[9] This study also showed that the majority of the patients are not more than 30, buttressing the mean age. With regards to the site of presentation, the mean age of those with mandibular AC is less than 30 years, while that of those with maxillary AC is more than 46 years (Table 1). This agrees with other studies that suggest that maxillary AC occurs at an older age.

There is no consensus on sexual predilection in AC, Kruse *et al*.[10] reported a male-to-female ratio of 2.7: 1, while Ramesh *et al*.[8] reported a contrary female-to-male ratio of 3:2. Our study shows a slight male predilection which agrees with other Nigerian studies.[1,4]

Ameloblastic carcinoma is a slow-growing tumour and majorly asymptomatic, this could account for the delay in presentation noticed among patients with this lesion. In this study, the mean duration before presentation for management is similar to that reported by Soyeye *et al*.[4] 5.9 and 7.7 years

respectively for the primary and secondary variants respectively, while it deferred from that by Ndukwe *et al*.[1] and Niu *et al*.[9] who reported a longer duration of presentation of 10.3 years. This long interval before the presentation could be attributed to the asymptomatic nature of the lesion, patient attitude toward their health as most people in the study environment have shown a tendency to delay seeking medical care, additionally, high cost of treatment and out-of-pocket payment for medical services may influence patients' decision to seek traditional option which eventually delays presentation to the hospital and complicates treatment. Dearth of oral and maxillofacial surgeons in the environment may also be a factor.[11] Similar reasons may apply to 26% of the patients who did not present for treatment after having their biopsy done.

Ameloblastic carcinoma was commoner in the mandible in this study. This underscores the finding that it is rare in the maxilla.[1, 4,9,10, 12] Most cases in this present study affected both halves of the mandible with almost equal numbers affecting the right and left sides.

Treatment for AC is still being debated, however, surgical resection with a margin of safety of about 2cm is considered the treatment of choice.[4] Several other treatment options like radiotherapy and chemotherapy have been tried with varying degrees of success.[13, 14] The choice of treatment depends on the clinician and the presentation of the lesion, with most clinicians advocating for surgical resection.[1,4,7] Ramadas *et al*.[14] recommended using chemotherapy in cases of unresectable lesions and as post-surgical adjuvant treatment. These options are in agreement with that of the current study as the commonest treatment carried out was jaw resection (mandibulectomy and maxillectomy) while adjuvant radiotherapy was given in more than half of the patients. The use of adjuvant radiotherapy here agrees with the suggestion by Philips *et al*.[15] that adjuvant should be used for those with positive margins, lymph node involvement, perineural spread, and for those with inoperable lesions. The use of chemotherapy, however, is limited in this study which agrees with the findings reported previously.[16] In this study, the decision regarding adjuvant radiotherapy and or chemotherapy was case to case-based. Generally, the patients we sent for adjuvant therapy included those who had lymph node metastasis, positive tumour margins despite wide resection, and maxillary AC.

We performed neck dissection on a few cases in this study. The role of neck dissection in the treatment of AC is controversial since the route of its spread is still not clear (hematogenous and lymphatics are the suggested possible routes).[17] However, studies indicate its usefulness as there are chances of AC spreading through the lymph nodes to other organs of the body including the, lungs, liver, and brain.[10, 17, 18, 19] Uzawa *et al*.[20] advocates for neck dissection only when there is evidence of regional lymph node involvement, similarly, Giridhar *et al*.[21] recommend prophylactic neck node dissection in AC to be avoided since the progression-free survival and overall survival for patients with or without neck dissection does not differ.

Studies from elsewhere document that the recurrence rate of ameloblastic carcinomas is high[4, 20, 22] and it appears to be higher in the maxilla due to the highly vascular nature of the area, and the presence of many vital structures, which make it difficult to obtain clear margins.[9] On the contrary, in the current study, no case of recurrence was found. Some of the possible explanations for no recurrence in our study could be attributed to a short interval of follow-up in this study (median 8 months), which may not be adequate to conclude that recurrences are rare in AC. Another possibility is because of the protocol we use in our centre. We carry out a wide resection of the tumour beyond 2cm of radiologically apparent margin, as such possibility of cancerous cells being left becomes low hence low recurrence.

CONCLUSION

Ameloblastic carcinoma is a rare odontogenic tumour that occurs in more in the mandible than the maxilla. It occurs almost throughout all age groups however, those affecting the maxilla appear to occur

at an older age group. There is a slight predilection for males in the study environment. The mainstay treatment involves resection of the affected jaws with a margin of safety. Radiotherapy and chemotherapy may have a role in its treatment. A multicentric study involving other Nigerian institutes and other institutes in Africa must be carried out. This will increase our understanding regarding the biological characteristics of AV, and thus develop a better treatment protocol for these tumours

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