

# A 10-year retrospective study of the clinical presentation, pathology, treatment and outcomes of ameloblastoma and ameloblastic carcinoma in a teaching hospital in Lagos, Nigeria

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## ABSTRACT

**Background.** Ameloblastoma is a common benign, odontogenic tumour with high prevalence in Africans, particularly Nigerians. We describe the epidemiology, clinicopathology, pattern of management and incidence of recurrence of ameloblastoma and ameloblastic carcinoma, in Lagos, Nigeria.

**Methods.** This retrospective study included ameloblastoma cases surgically managed at the Lagos University Teaching Hospital from January 2012 to December 2021. Primary outcome was recurrence; secondary outcomes were length of hospital (LOH) stay and postoperative complications.

**Results.** We included 63 patients with a mean (SD) age of 34.2 (14.8) years, peak incidence (31.7%) in the 3rd decade of life, and male-to-female ratio of 1.03. The most common location, radiological feature and histological type were posterior mandible (77.8%), multilocular radiolucency (90.5%) and follicular ameloblastoma (50.8%), respectively. For surgical intervention, majority of patients had nasotracheal intubation (67.3%) and mandibulectomy (88.9%), and the most common surgical approach was extraoral (67.3%). The mean (SD) LOH stay was 9.4 (2.4) days, and the transoral approach was

associated with shortened LOH. The mean follow-up was 2.7 years, and recurrence was recorded in 2 patients who had conventional ameloblastoma, and one who had ameloblastic carcinoma at 3 years and 3 months postoperatively, respectively. No significant association was noted for recurrence-free survival based on tumour size, tumour diagnosis, histological type and surgical approach ( $p > 0.05$ ).

**Conclusion.** Although the epidemiology, clinicopathology and treatment of ameloblastoma reported were similar to older reports, this study provides more recent information on the persistent public health burden of ameloblastoma, which can be used for comparisons with ameloblastoma in other populations.

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## INTRODUCTION

Ameloblastoma is the most common benign, odontogenic tumour of the head and neck region, comprising about 1% of all cysts and tumours of the jaws and up to 10% of all odontogenic tumours.<sup>1</sup> It arises from epithelial remnants of the developing root sheath.<sup>2,3</sup> It can affect all age groups but is most common in the third to seventh decades, with peak incidence in the third and fourth decades.<sup>4,5</sup> It has a racial preponderance with higher prevalence in Africans and Asians than in Caucasians.<sup>6</sup> The lesion can occur anywhere in the mandible and maxilla but is most commonly found in the posterior mandible.<sup>7</sup> A slight male preponderance has been reported with a male-to-female ratio of 1.17:1.<sup>8</sup>

The tumour is usually asymptomatic, presenting as a painless slow-growing swelling, which may explain the reason for its neglect among patients with low socioeconomic status.<sup>9</sup> However, neglected lesions can grow into large disfiguring sizes (Fig. 1).<sup>9,10</sup> Despite being benign, it is a locally aggressive tumour with a high recurrence rate if not completely resected. Radiological investigations such as orthopantomograms are useful first-line investigations that aid its diagnosis.<sup>11</sup> Advanced imaging such as computed tomography delineates the extent of the tumour and aids treatment planning; however, orthopantomograms can be used for treatment planning if advanced imaging is not available,<sup>11</sup> especially in cases affecting the mandible (Fig. 2). The main goals of surgical treatment of ameloblastoma are to completely remove the tumour and to restore orofacial function and aesthetics.<sup>12</sup> Complete removal of the tumour prevents tumour recurrence and/or transformation to ameloblastic carcinoma.<sup>12</sup> The definitive treatment is surgical resection with wide margins (1–2 cm)<sup>13</sup> with some authors

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recommending immediate bone reconstruction.<sup>14</sup> Conservative management in the form of enucleation and curettage is no longer recommended, even for early lesions, due to the high recurrence rate.<sup>15</sup> In view of the high prevalence of ameloblastoma in Nigeria, and the public health burden it confers on the Nigerian population, it is important to determine its current epidemiological and clinicopathological presentation as well as current treatment modalities and their postoperative outcomes.

We aimed to study the current presentation, treatment and treatment outcomes of ameloblastoma in Lagos, Nigeria, and compare these with what have been previously reported in the population. The specific objectives were to describe the current epidemiology and clinicopathology of ameloblastoma and to describe the current pattern of management and incidence of recurrence of ameloblastoma surgically treated during a 10-year period in a teaching hospital in Lagos, Nigeria.

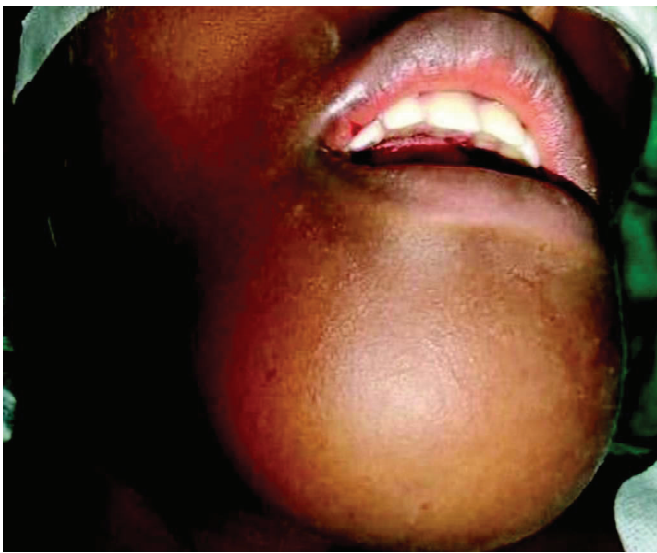


FIG 1. Large expansile lesion of the anterior mandible extending posteriorly

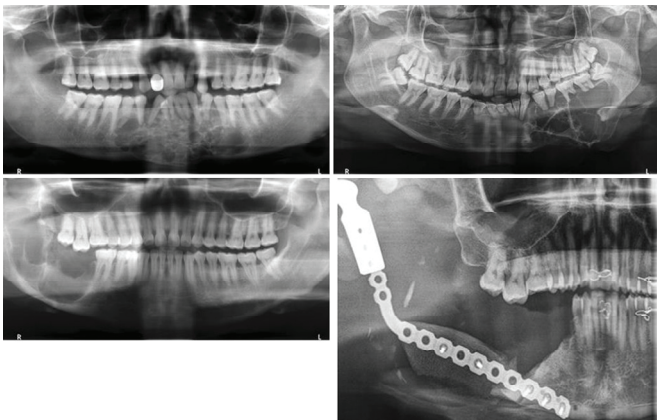


FIG 2. Orthopantomograms showing unicystic ameloblastoma of the anterior mandible (top left), multicystic ameloblastoma of the left posterior mandible (top right), recurrent ameloblastoma of the right posterior mandible (bottom left), and reconstruction of mandibular continuity defect using reconstruction plate, alloplastic condylar prosthesis and iliac crest bone graft secondary to resection of ameloblastoma of the right posterior mandible (bottom right)

**METHODS**

*Study design/sample*

This was a retrospective cohort study of all patients who presented to the Oral and Maxillofacial Surgery clinic, Lagos University Teaching Hospital (LUTH), Lagos, Nigeria for evaluation and management of ameloblastoma and ameloblastic carcinoma between January 2012 and December 2021. The inclusion criteria were (i) a histological diagnosis of ameloblastoma or ameloblastic carcinoma of the mandible or maxilla obtained from a preoperative incisional biopsy, (ii) received surgical intervention for the identified condition, and (iii) had at least 1-year postoperative follow-up.

*Variables*

The predictor variables studied were clinical and radiological characteristics of the lesion, histopathology and surgical intervention. Clinical characteristics were size, location and classification of the lesion. The size of the lesion was its largest diameter. Location of the lesion was divided into 6 groups (modified from the groups proposed by Hong *et al.*<sup>16</sup>): Anterior mandible, left posterior mandible, right posterior mandible, anterior maxilla, left posterior maxilla and right posterior maxilla. The junction of the canine and first premolar served as demarcation between the anterior and posterior region. Classification of the lesion was done in accordance with the WHO classification for odontogenic tumours.<sup>17</sup> Radiological characteristics were described based on reports of orthopantomogram or computed tomography (CT) scans, and histopathology was described based on the histopathological reports. The characteristics of surgical intervention were techniques of anaesthesia, surgical procedure, surgical approach, type of reconstruction and material used for reconstruction.

The primary outcome variable was tumour recurrence. Secondary outcome variables were preoperative and postoperative complications, length of hospital (LOH) stay, length of postoperative follow-up, recurrence-free survival and treatment of recurrence. Postoperative complications were either immediate complications that occurred while the patient was still admitted, or delayed complications that occurred after discharge from hospital. LOH stay was the time from hospital admission before surgery to the time of discharge after surgery when the patient was certified fit to go home, as determined by the managing surgeons. Recurrence-free survival or time to recurrence was the time from surgery to the point of the first noticeable sign of recurrence, or if no recurrence, the most recent disease-free assessment.

Covariates were recorded at first presentation to the Oral and Maxillofacial Surgery Clinic, Lagos University Teaching Hospital and included socio-demographic characteristics such as patients' age, sex at birth and occupation. Occupation was categorized into non-labour (students, unemployed and retirees), unskilled (does not require formal education), semi-skilled (requires formal education but not higher education) and skilled labour (requires higher education).<sup>18</sup> In addition, duration of condition (DOC) and habits of cigarette smoking, alcohol consumption and chewing kola and tobacco were also recorded. DOC was the time from self-reported first symptom onset, to the first presentation at the clinic.

*Data collection methods*

Data were collected from patient records using a proforma. All medical records, radiological, histopathological reports, operative

and postoperative reports within the study period were reviewed for data collection. This study was approved by the Lagos University Teaching Hospital Health Research Ethics Committee, Lagos, Nigeria (ADM/ADM/DSCST/HREC/APP/5786).

### Statistical analysis

Data analysis was performed using the software SAS, version 9.4 (SAS Institute Inc., Cary, NC, USA). Descriptive analysis was done using frequency and proportion for categorical variables and mean and standard deviation (SD) for numeric variables. Data were analysed using Wilcoxon rank sum test, Kruskal–Wallis's test, Pearson's chi-square test and Fisher's exact test wherever applicable, to test for association.  $p < 0.05$  was considered statistically significant. Tests for recurrence-free survival based on prognostic factors for recurrence such as tumour size, tumour diagnosis, histological type and surgical approach were done using Kaplan–Meier analysis and log rank tests.

### RESULTS

Sixty-three patients were diagnosed with and managed for ameloblastoma within the study period. The number of patients treated in a year ranged from 4 (4/63, 6.3%) in 2013 to 10 (10/63, 15.9%) in 2016 with mean (SD) number of patients of 6.3 (1.6) per year (Fig. 3). All 63 patients were followed up for at least 1 year postoperatively with a follow-up ranging from 1 to 10 years and a mean (SD) of 2.7 (2.1) years.

There were 32 (50.8%) males and 31 (49.2%) females with a male-to-female ratio of 1.03 to 1. Their age was between 8 and 67 years with a mean (SD) of 34.2 (14.8) years. Participants' labour categories were unskilled (5/63, 7.9%), semi-skilled (35/63, 55.6%) and skilled (2/63, 3.2%). Twenty-one participants (21/63, 33.3%) were students, retirees or unemployed. Only one patient had a positive family history of tumour (mother had breast cancer), and 14 (14/63, 22.2%) patients had habits of cigarette smoking, alcohol consumption or chewing kola and tobacco.

The DOC ranged from 2 months to 18 years with mean (SD) of 4.6 (3.8) years. There were 9 (9/63, 14.3%) patients with DOC  $< 12$  months, 8 (8/63, 12.7%) with DOC  $\geq 12$  months but  $< 2$  years, 17 (17/63, 27.0%) with DOC  $\geq 2$  years but  $< 5$  years and 29 (29/63, 46.0%) with DOC  $\geq 5$  years. No significant relationship was

noted between age, gender, occupation and DOC ( $p > 0.05$  for all associations, Table 1).

The size of the lesion was recorded for 23 patients, and the largest diameter ranged from 3 cm to 17 cm with a mean (SD) of 8.1 (4.4) cm. The tumour originated more often from the mandible than from the maxilla and, in a decreasing order of frequency, from the right posterior mandible in 27 (27/63, 42.9%) patients, from the left posterior mandible in 22 (22/63, 34.9%) patients, from the anterior mandible in 9 (9/63, 14.3%) patients, from the right posterior maxilla in 3 (3/63, 4.8%) patients and from the left posterior maxilla in 2 (2/63, 3.2%) patients. None of the included tumours originated from the anterior maxilla. About half (50.8%) the lesions at presentation involved 1 (19.1%) or 2 (31.7%) adjacent regions.

The most common radiological features on orthopantomogram or CT scan were multilocular radiolucencies (57/63, 90.5%) and truncation of roots (58/63, 92.1%). Other less common radiological features included unilocular radiolucencies (6/63, 9.5%), cystic necrosis of the jawbone (4/63, 6.3%) and marked destruction of submandibular gland (1/63, 1.6%).

Fifty-three (53/63, 84.1%) patients were diagnosed with conventional ameloblastoma, 4 (6.3%) with unicystic ameloblastoma, 2 (3.2%) with peripheral ameloblastoma and 4 (46.3%) with ameloblastic carcinoma. The most common histological type was follicular ameloblastoma (32/63, 50.8%; Table 2).

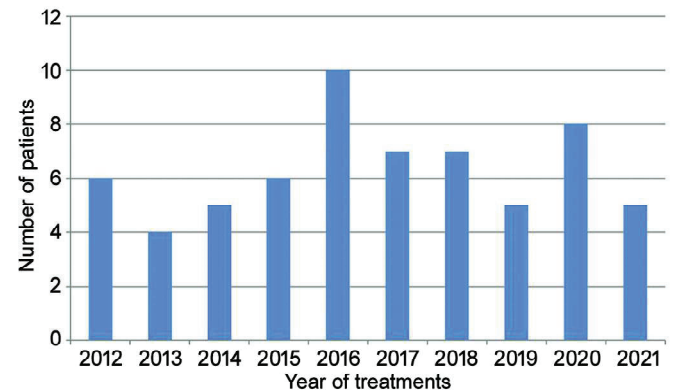


FIG 3. Yearly number of patients treated surgically for ameloblastoma

TABLE 1. Summary of descriptive statistics

Item	Duration of condition (months)				Total	p value
	<12	12 to <24	24 to <60	$\geq 60$		
<i>n (%)</i>						
<i>Age (years)</i>						
1–10	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	1	0.58
11–20	3 (27.3)	2 (18.2)	4 (36.4)	2 (18.2)	11	
21–30	2 (10.0)	1 (5.0)	5 (25.0)	12 (60.0)	20	
31–40	2 (18.2)	3 (27.3)	2 (18.2)	4 (36.4)	11	
41–50	1 (12.5)	0 (0.0)	2 (25.0)	5 (62.5)	8	
51–60	1 (10.0)	1 (10.0)	3 (30.0)	5 (50.0)	10	
61–70	0 (0.0)	0 (0.0)	1 (50.0)	1 (50.0)	2	
<i>Sex</i>						
Female	5 (16.1)	4 (12.9)	10 (32.3)	12 (38.7)	31	0.71
Male	4 (12.5)	4 (12.5)	7 (21.9)	17 (53.1)	32	
<i>Occupation</i>						
Non-labour	3 (14.3)	3 (14.3)	6 (28.6)	9 (42.9)	21	0.89
Unskilled	0 (0.0)	0 (0.0)	1 (20.0)	4 (80.0)	5	
Semi-skilled	6 (17.1)	4 (11.4)	10 (28.6)	15 (42.9)	35	
Skilled	0 (0.0)	1 (50.0)	0 (0.0)	1 (50.0)	2	

\* Fishers exact test

The surgical intervention in 56 (88.9%) patients was mandibulectomy, 5 (7.9%) had a maxillectomy and 2 (3.2%) had surgical excision with peripheral ostectomy (Table 3). Eight (12.7%) patients received additional surgery: 6 (9.5%) had additional surgical excision of soft-tissue lesion, 1 (1.6%) had additional neck dissection and 1 (1.6%) had additional skin graft placement secondary to excision of involved skin.

Complete data on treatment characteristics and complications were available for 52 of 63 patients. Over half the patients had nasotracheal intubation (35/52, 67.3%) and extraoral approach (39/52, 75%) as the anaesthetic technique and surgical approach, respectively. Other less common anaesthetic techniques included elective tracheostomy (11/52, 21.2%), endotracheal intubation (4/52, 7.7%), local anaesthesia plus conscious sedation (1/52, 1.9%) and emergency tracheostomy (1/52, 1.9%; Table 3).

Reconstruction of the jaw defect after surgical resection was either immediate or delayed. Of the 47 patients who had mandibulectomy and complete treatment data, only 4 (48.5%) patients had immediate reconstruction. For maxillectomy patients, only 1 of 3 (33.3%) had immediate reconstruction. For patients who had surgical excision with peripheral ostectomy secondary to peripheral ameloblastoma, reconstruction was not needed.

At the time of data collection, only 19 of 50 (38%) patients with complete data had received reconstruction for jaw defect. In contrast, 31 (62%) patients were either awaiting treatment, refused treatment or were not ready for treatment (financially or psychologically). For mandibulectomy defects, reconstruction was done using non-vascularised iliac crest bone graft for 12 of 16 (75%) patients, vascularised fibular flaps for 3 of 16 (18.8%) patients and clasp retained denture for 1 (6.2%) patient who received marginal resection under local anaesthesia plus conscious sedation; 3 of the iliac crest graft reconstructions were immediate reconstructions and only 1 of the vascularised fibular flap reconstruction was an immediate reconstruction. For maxillectomy defects, immediate reconstruction was done with feeding plate for 1 (33.3%) patient and delayed reconstruction was done with a definitive obturator for 2 (66.7%) patients.

The minimum LOH stay was 1 day (day case) for the patient who received marginal resection under local anaesthesia along with conscious sedation and the maximum LOH stay was 16 days with a mean (SD) of 9.4 (2.4) days. There was a significant difference between surgical approach and LOH stay; transoral approach was associated with shortened LOH stay (Table 4). No preoperative complications were recorded. Immediate postoperative complications were recorded in only 3 of 52 (5.8%) patients: 1 (1.9%) had surgical wound breakdown at postoperative day 14 and 2 (3.8%) developed trismus at postoperative days 7 and 9, respectively. The majority (9/10, 90%) of patients who had extraoral surgical approach developed postoperative complications compared to transoral approach; however, this relationship was not statistically significant (Table 4). No significant relationships were noted between surgical intervention, type of reconstruction, immediate postoperative complications and LOH stay ( $p > 0.05$  for all associations, Table 5). Only 7 patients had delayed postoperative complications (Table 6).

Recurrence was recorded in only 3 (4.8%) patients, 2 with conventional ameloblastoma and 1 with ameloblastic carcinoma. Although recurrence appeared to occur in relationship with larger tumour size ( $> 8$  cm), certain tumour diagnosis and histological type (conventional ameloblastoma [follicular and

granula] and ameloblastic carcinoma) and surgical approach (extraoral), none of these were statistically significant (Table 7).

Time-to-event analysis was done for incidence of recurrence and potential prognostic factors. Kaplan–Meier plot was used

TABLE 2. Histological types of ameloblastoma

Histological type	n (%)
<i>Conventional</i>	
Follicular	31 (49.2)
Cystifying follicular	4 (6.3)
Plexiform	4 (6.3)
Mixed follicular, plexiform	1 (1.6)
Acanthomatous	4 (6.3)
Papillorous keratinizing	1 (1.6)
Desmoplastic	4 (6.3)
Granular	3 (4.8)
Keratoameloblastoma	1 (1.6)
<i>Unicystic</i>	
Luminal	1 (1.6)
Mural	3 (4.8)
<i>Peripheral</i>	
Follicular	1 (1.6)
Plexiform	1 (1.6)
<i>Ameloblastic carcinoma</i>	4 (6.3)
Total	63 (100)

TABLE 3. Details of surgical treatment of ameloblastoma

Characteristic	Mandibulectomy (n=56)	Maxillectomy (n=5)	Surgical excision with peripheral ostectomy (n=2)
<i>Surgery details</i>			
Marginal resection	1	–	–
Segmental	16	–	–
Hemi	15	–	–
Subtotal	23	4	–
Total	1	–	2
With disarticulation	20	–	–
Partial	–	1	–
<i>Anaesthetic technique*</i>			
LA+CS	1	–	–
Nasotracheal	35	–	–
Elective tracheostomy	10	–	1
Emergency tracheostomy	1	–	–
Endotracheal	–	3	1
<i>Surgical approach*</i>			
Transoral	12	–	1
Extraoral	35	3	1
<i>Surgical incision*</i>			
Buccal and lingual gingival sulcus	12	–	1
Submandibula	8	–	–
Bilateral SM plus lower lip split	1	–	–
SM plus lower lip split	6	–	–
SM plus inverted hockey stick	1	–	1
SM with retromandibular extension	3	–	–
SM with anterior midline extension	2	–	–
Modified Schobinger	1	–	–
Visor	12	–	–
Visor plus inverted hockey stick	1	–	–
Trotter Weber Fergusson	–	3	–

\* Complete treatment data is missing for 9 patients who had mandibulectomy and 2 patients who had maxillectomy LA+CS local anaesthesia and conscious sedation SM submandibula

to describe probability of recurrence within 10 years post-operatively for each of the prognostic factors (Figs 4–7). The probability of recurrence was >10%; >20% for tumours >7–11 cm, 20% or more, >20% for ameloblastic carcinoma, about

10% for follicular ameloblastoma, about 50% for granular ameloblastoma and almost 20% for extraoral surgical approach. However, no significant associations were noted between recurrence and potential prognostic factors ( $p>0.05$  for all associations; Table 7).

TABLE 4. Association between reconstruction type, LOH, postoperative complications and surgical approaches

Characteristic	Surgical approach			p value
	Transoral	Extraoral	Total	
Mean (SD) LOH stay (days)	8.0 (2.7)	9.9 (2.2)	9.4 (2.45)	0.015*
<i>Postoperative complications n (%)</i>				
No	12 (71.4)	30 (28.6)	42 (100.0)	0.419†
Yes	1 (10.0)	9 (90.0)	10 (100.0)	

\* Wilcoxon rank sum test † Fishers exact test LOH length of hospitalization

TABLE 5. Association between treatment characteristics, immediate postoperative complications and LOH

Treatment characteristic	Mean (SD) LOH (days)	p value
<i>Surgical intervention</i>		
Mandibulectomy	9.5 (2.4)	0.28*
Maxillectomy	7.7 (1.5)	
Surgical excision with peripheral osteotomy	10.5 (3.5)	
<i>Reconstruction type</i>		
Immediate	9.6 (3.6)	0.48†
Delayed	9.4 (2.3)	
<i>Immediate postoperative complications</i>		
No	9.3 (2.4)	0.53†
Yes	10.0 (2.7)	

\* Kruskal–Wallis test † Wilcoxon rank sum test LOH Length of hospitalization

Table 6. Postoperative complications (n=7)

Complication	Timing of complication	Treatment done
Osteomyelitis of bone graft	1 month	Sequestrectomy of involved bone and osseous curettage 1 month postoperative then secondary iliac crest gbone raft 20 months postoperative
Wound dehiscence and extraoral exposure of reconstruction plate	3 months	Curettage and resuturing for healing by tertiary intention
Recurrence of ameloblastic carcinoma	3 months	Radiotherapy
Broken reconstruction plate	12 months	Replacement of reconstruction plate and reconstruction with iliac crest bone graft
Intraoral exposure of reconstruction plate	36 months	Repositioning of reconstruction plate and reconstruction with iliac crest bone graft
Recurrence of conventional ameloblastoma	36 months	Surgical excision
Recurrence and malignant transformation of conventional ameloblastoma	36 months	Surgical excision

## DISCUSSION

Ameloblastoma is the most common odontogenic tumour of the head and neck region with a high preponderance in the African population.<sup>1,6</sup> Nigeria alone bears over 90% of the total ameloblastoma burden in sub-Saharan Africa; despite an unclear reason, it may be partly due to the large amount of research literature from the region.<sup>19,20</sup> Previous studies<sup>7,20–23</sup> have reported the epidemiological distribution and clinicopathological presentation of ameloblastoma in Nigeria; however, regular review of this information is important for early identification of secular changes due to its high public health impact. In addition, we explored the surgical management of ameloblastoma and outcomes of management after a minimum follow-up of 1 year.

Similar to existing reports,<sup>7,20–23</sup> our study showed a peak incidence of ameloblastoma in the third and fourth decades of life and confirmed previous reports of the rarity of ameloblastoma in the first decade of life.<sup>7,22,23</sup> Conflicting reports exist on the sex distribution of ameloblastoma. Most studies<sup>7,20,23</sup> similar to results of our study, suggest equal sex distribution or a slight male preponderance, while Chukwunke *et al.*<sup>22</sup> reported a female predisposition to ameloblastoma.

Although ameloblastoma is not known to be caused by any occupation, it has been reported to be more common among the low socio-economic class.<sup>24,25</sup> This may be explained by the fact that ameloblastoma sometimes develops from untreated

TABLE 7. Relation of recurrence with prognostic factors

Prognostic factor	Recurrence			p value*	Log rank test p value
	No	Yes	Total		
<i>Size, largest diameter in cm, n†</i>					
≤4	7	0	7	0.08	0.2
>4–7	5	0	5		
>7–11	3	2	5		
>11	6	0	6		
<i>Tumour diagnosis, n</i>					
Conventional	51	2	53	0.41	0.56
Unicystic	4	0	4		
Peripheral	2	0	2		
Ameloblastic carcinoma	3	1	4		
<i>Histologic type, n</i>					
Follicular	32	1	33	0.06	0.28
Plexiform	5	0	5		
Granular	2	1	3		
Ameloblastic carcinoma	3	1	4		
Others	19	0	19		
<i>Surgical approach, n (%)</i>					
Transoral (intraoral)	13	0	13	0.56	0.37
Extraoral	36	3	39		

\* Fishers exact test † Data available for only 23 patients For time-to-event analysis, time at risk began after treatment and ended at the time of recurrence, at the time of other post-treatment complications, at the premature end of follow-up (due to dropout or other reasons) or 10 years after surgery whichever occurred first. Thus, patients with a post-treatment complication other than recurrence were censored at the time of the post-treatment complication and thus left the risk set. Those patients with a premature end of the follow-up that ended before 10 years were censored at the time of the premature follow-up and thus left the risk set as well.

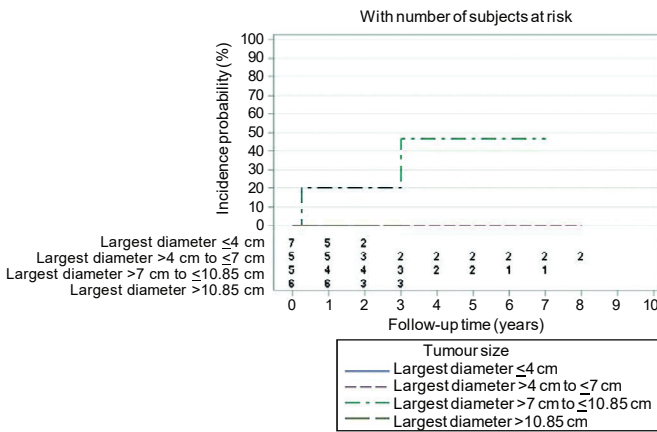


FIG 4. Probability of recurrence within 10-years post-treatment by largest diameter of tumour

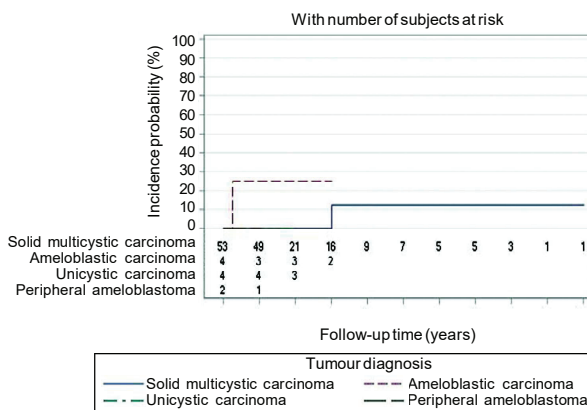


FIG 5. Probability of recurrence within 10-year post-treatment by tumour diagnosis

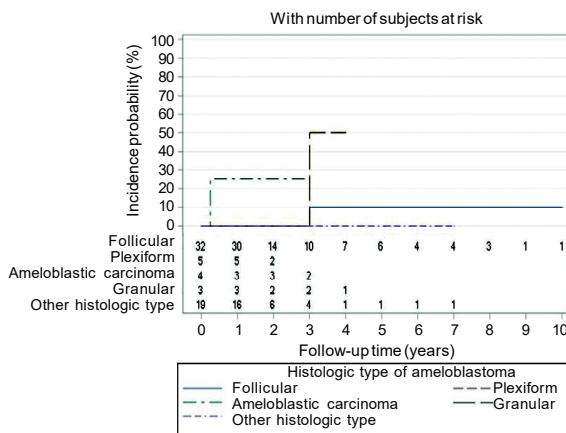


FIG 6. Probability of recurrence within 10-year post-treatment by histological type of ameloblastoma

odontogenic cysts, and people of low socio-economic class are less likely to seek healthcare for financial reasons, resulting in neoplastic transformation to ameloblastoma. In our study only 2 patients were in the skilled labour category while majority were either semi-skilled or unskilled, which supports the current assumptions. The DOC in our study raises public health concerns in that a large proportion of patients lived with their condition despite its grotesque appearance for over 5 years.

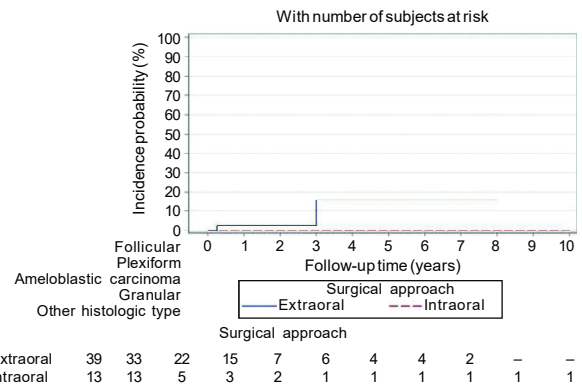


FIG 7. Probability of recurrence within 10-years post-treatment by surgical approach

These are supported by previous reports from within<sup>21,23</sup> and outside Nigeria,<sup>26</sup> pointing to an unaddressed issue of healthcare inequality and inaccessibility.

Ameloblastoma is largely known as a painless, slow-growing, benign lesion. Small lesions are often seen among Europeans and Americans compared to large lesions among Africans and Asians.<sup>6</sup> However, the clinical presentation of the lesion is dependent on the DOC, and large grotesque swellings are seen only with longer DOC.<sup>21,23</sup> Our study, similar to previous Nigerian studies<sup>21,23</sup> reported large lesions with mean tumour diameter of 8.1 cm, and maximum up to 17 cm, with about half involving multiple regions of the mandible or maxilla. The mandible is considered the most common site for ameloblastoma, with some studies<sup>7,19</sup> reporting it predominantly in the posterior mandible, while others<sup>22,23</sup> reported it in the anterior mandible. Similar to previous reports, our study showed that the majority (92.1%) of the lesions occurred in the mandible, and 84.4% of cases involved the left or right posterior mandible. No consensus has been reached on the reason for the predominance in the mandible, but an assumption is that ameloblastoma originates from reduced enamel epithelium and rest cells of Malassez which are predominantly found in the mandible due to its size compared to the maxilla.<sup>21</sup>

In agreement with previous studies from in<sup>21-23</sup> and outside<sup>26,27</sup> Nigeria, the most common radiological features reported were multilocular radiolucency and root truncation representing a multicystic locally invasive odontogenic lesion characteristic of ameloblastoma. Similar to previous reports from Nigeria and/or Africa,<sup>21-23</sup> the most common morphological presentation was conventional ameloblastoma,<sup>21-23</sup> and the most common histological type was follicular ameloblastoma.<sup>7,22</sup> However, Soyele *et al.*<sup>21</sup> reported plexiform type, as the most common type.

A similar study done in the same hospital decades ago by Ladeinde *et al.*<sup>7</sup> reported a prevalence of ameloblastic carcinoma of 4.35%. In our study, the prevalence of ameloblastic carcinoma was 6.3%. Although both reports indicate prevalence <10%, the slight increase in prevalence may suggest an increase in predisposing factors among the Nigerian population and the need for further research into the aetiopathogenesis of ameloblastoma in Nigeria.

Jaw resection and surgical excision with wide margins (1-2 cm) are the recommended treatment for both intraosseous and extraosseous ameloblastoma, respectively.<sup>13,14</sup> Oral and maxillofacial surgeries performed under general anaesthesia require special consideration as routine endotracheal intubation may not be ideal. After non-total mandibular jaw resection,

occlusion must be established between the remaining jawbone and maxillary arch for proper placement and fixation of reconstruction plate and for ideal rehabilitation and functional restoration of the maxillomandibular system after surgery. Endotracheal intubation will prevent this and, hence, the need for alternatives such as nasotracheal intubation, submental intubation and surgical airway or tracheostomy.<sup>28</sup> We used nasotracheal intubation as the most common anaesthetic technique and was only used for mandibulectomies. Endotracheal intubation was used for all maxillectomies as establishment of occlusion was not required. Similar to previous studies<sup>29</sup> this study showed that routine tracheostomy was unnecessary for maxillectomies. No patients in this study had submental intubation; however, it has been shown to be less invasive with minimal intraoperative and postoperative complications compared to tracheostomy and should be considered when endotracheal and nasotracheal approaches are contraindicated.<sup>30</sup>

The more commonly practiced surgical approach for mandibulectomy is extraoral.<sup>31</sup> However, proponents of the transoral approach have suggested its use for resection and immediate reconstruction of benign pathologies when sufficient access can be achieved, with the benefits of limiting postoperative morbidities associated with the extraoral approach.<sup>31</sup> In this study, the transoral approach was used in 12 of 47 mandibulectomy patients and in 1 of 2 patients with surgical excision and peripheral osteotomy. Only 1 of 13 patients in the transoral approach group developed a postoperative complication, although this association was not statistically significant. In addition, statistically significant reduction in LOH was noted with the transoral approach.<sup>31</sup> These issues do not exist for maxillectomy as the universally accepted surgical approach is extraoral using the Weber–Fergusson incision with or without a Trotter or Lynch extension, which is corroborated by the results of our study.

Reconstruction of mandibular jaw resection defects can either be performed immediately at the time of resection or delayed till a later date. Improved infection control and high degree of success following radiotherapy and/or chemotherapy are advantages of the delayed approach, while increased healthcare cost and negative psychological effect from multiple surgeries are disadvantages.<sup>32,33</sup> On the other hand, the immediate approach offers economic advantages and a single surgery experience but at the cost of the increased risk of infection.<sup>32,33</sup> In the immediate approach, non-tension, water-tight seal must be achieved to prevent ingress of saliva into the surgical site, subsequent wound dehiscence, infection and graft failure. In this study, four mandibulectomy patients received immediate reconstruction and one of them developed osteomyelitis 1-month postoperative requiring sequestrectomy of bone graft, curettage of surgical site and delayed secondary reconstruction. Despite the merits of an immediate reconstruction, we recommend it only when non-tension, water-tight seal with adequate infection control can be guaranteed. Reconstruction of maxillary jaw resection defects can be done either by surgical procedure or by prosthetic rehabilitation.<sup>34</sup> Similar to mandibular defects, maxillary defects may be reconstructed using free flaps or grafts from the iliac crest, fibula, radial forearm or scapula.<sup>35</sup> However, the complex anatomy of the maxilla poses a unique challenge in their retention. Like the results of this study, maxillary defects of ameloblastoma are often reconstructed through prosthetic rehabilitation with feeding plates and obturators.

The high rate of recurrence of ameloblastoma following conservative surgical management led to the recommendation of a radical approach for all cases including early stage lesions.<sup>15</sup> In this study, radical resection and excision was done for all patients. Despite this approach, recurrence of ameloblastoma is not uncommon due to its local invasiveness, tissue components and histologic types.<sup>36</sup> Previous studies in Nwoga<sup>37</sup> and outside Nigeria<sup>38</sup> have reported a recurrence rate of 33.3% and 35%, respectively. We report a relatively low recurrence rate of 4.8%, possibly due to the short average follow-up period.

Previous studies by Reichart *et al.* and Ghai<sup>39,40</sup> have reported an increase in recurrence of ameloblastoma when the lesion is multicystic, located in the maxilla and follicular or granular histologic types. The multicystic nature of the lesion points to a more aggressive form of the tumour compared to its unicystic variant. In addition, the lesion spreads more aggressively and recurs more frequently in the maxilla due to the thin maxillary cortical bone that provides a weak barrier for the spread of tumour. Our study corroborates these previous findings. Two-thirds of recurrence reported were seen in multicystic ameloblastoma, and their histologic variants were follicular and granular ameloblastomas. The remaining one-third was seen in ameloblastic carcinoma. Ameloblastic carcinomas are more aggressive and prone to recurrence than conventional ameloblastoma due to their cellular atypia features such as nuclear pleomorphism, hyperchromatism, high mitotic activity, tissue necrosis and vascular invasion.<sup>41</sup> This is supported by the results of this study and found that ameloblastic carcinoma showed a 25% recurrence rate and time-to-recurrence of 3 months, compared to 3.8% and 3 years, respectively, for conventional ameloblastoma. Nonetheless, this and other studies reported that conventional ameloblastoma may undergo malignant transformation during recurrence.<sup>37,38</sup>

We did not find any statistically significant association between recurrence and potential prognostic factors, which may be due to the low recurrence incidence that, in turn, may be due to the short average follow-up period. Ameloblastoma recurrence is usually recorded at 1–10 years postoperative.<sup>36,42</sup> The mean follow-up duration of our study was 2.7 years which may not be adequate for observing recurrence; this risks underestimating the recurrence incidence.

One of the major limitations of this study was the retrospective nature of the data which made it difficult to correlate histopathology and recurrence of the tumours as information such as presence of positive margins after surgery were not recorded. In addition, data on study variables were not evenly available for all included patients. Second, the histopathologic diagnosis reported was based on the acceptable classification of odontogenic tumour at the time of surgical intervention. However, some of these histopathologic diagnoses are no longer recognized by the WHO, due to the ever changing definitions of odontogenic tumours.<sup>43</sup> Third, the study reported a relatively small sample size which was due to the strict inclusion and exclusion criteria. Previous Nigerian studies have reported over 1000 Nigerians living with ameloblastoma.<sup>20</sup> This study, however, did not include all patients with ameloblastoma, but only those who had surgical intervention. The low number of patients who received treatment may be due to the economic challenges and peculiarities of the study location. Finally, the mean follow-up time was not adequate to assess for recurrence following treatment of ameloblastoma. Although, our study included patients treated from 2012 to 2021, and patients treated

during the late study period were not likely to have more than a couple of years of follow-up at the time of data collection, patients treated during the early study period also reported low follow-up time which may be due to patient burnout and economic challenges. We, therefore, recommend that these patients are followed up for a longer period of at least 10 years.

**Conclusion**

Our study showed a similar trend in epidemiological and clinicopathological characteristics of ameloblastoma to those which have been previously reported in the Nigerian and/or other populations. We found a relatively low recurrence incidence of 4.8% and did not find statistically significant associations between recurrence and several prognostic factors. Jaw resection and/or surgical excision plus peripheral ostectomy with wide margins (1–2 cm) represented treatment for intraosseous and extraosseous ameloblastoma, respectively. Special considerations must be made in selecting anaesthetic techniques, surgical approach and reconstruction type to achieve the best possible treatment outcome. Although, the characteristics reported in this study are like previous reports of the Nigerian population, this study provides more recent information on the persistent public health burden that is ameloblastoma, which can be used for comparisons with ameloblastoma in other populations.

**Conflicts of interest.** None declared

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