

# Pattern of Presentation and Management of Ameloblastoma in a Nigerian Regional Reference Hospital

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## ABSTRACT

**Background:** Ameloblastomas are benign but locally aggressive tumor that arises from the odontogenic epithelium. It primarily affects the jaws and is the most common benign odontogenic tumour second only to odontomas. The aim of the study was to review the records of all patients who presented with jaw ameloblastoma cases at the Oral and Maxillofacial surgery clinic of Federal Teaching Hospital (FTH), Gombe, from January 2000 to December 2024 with special attention to their management, complications following treatment as well as the challenges related to the management of these cases.

**Methods:** This was a retrospective study of medical records of all patients with histopathologically diagnosed jaw ameloblastoma at the Oral and Maxillofacial surgery clinic of FTH, Gombe. The data on patient's socio-demographics, tumour location on the jaw, clinical and histopathological parameters, treatments instituted, complications and management challenges were extracted from the patient's folder using data abstraction form. This information was analysed using collection was done using Statistical Package for Social Sciences (SPSS) version 23.

**Results:** 219 cases of ameloblastoma were histologically diagnosed during the period of the study; this amounted to an average of 9 cases per year. The mean age of the patients treated was 33.0 years (SD  $\pm 12$ ) with a male: female ratio of 1.13: 1. All the cases reviewed were located in the mandible with 161(73.4%) of them found in the posterior aspect. The conventional ameloblastoma is the most common clinical presentation 150(69.2%). Follicular variant was the most common histological subtype 123 (56.1%). One hundred and eighty nine (189) patients were treated with resection and non-vascularised iliac crest bone reconstruction. There were 4 recorded cases of tumour recurrence.

**Conclusion:** The result of the study revealed that jaw ameloblastoma cases in this locality shared similar clinico-pathologic presentations as commonly reported in the literature.

**Keywords:** Ameloblastomas, odontogenic tumour, mandibular neoplasms, treatment outcome

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## INTRODUCTION

Ameloblastomas are the most common clinically significant odontogenic tumours.<sup>1</sup> They have a relative frequency that is said to be equal to the combined frequency of all other odontogenic tumours with the exclusion of odontomas.<sup>2</sup> They are tumours of epithelial origin and have been described as usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent.<sup>1</sup>

Ameloblastoma was first recognised by Cusack in 1827 and the term “adamantinoma” was used to describe it by Louis Charles Malassez, a French physician in 1885. It was then renamed ameloblastoma by Ivy and Churchill in 1930.<sup>1, 3</sup> It is currently designated by the World Health Organisation (WHO) as a benign epithelial odontogenic tumour. In 2022, the WHO updated classification describes four clinical sub-types; the conventional, unicystic, extra-osseous and metastasing ameloblastoma.<sup>4</sup> The previous term solid multicystic ameloblastoma was removed as it has no prognostic significance and may lead to confusion with the unicystic ameloblastoma.<sup>5</sup> Desmoplastic ameloblastoma which was initially a clinical sub-type is now considered merely a histopathological variant of the conventional ameloblastoma despite its unique clinical and radiographic features.<sup>1</sup>

Ameloblastoma show a variable geographic prevalence with a global incidence of 0.92 cases per million persons.<sup>6</sup> It is more common in African and Asian populations compared to Caucasians. Several epidemiological reviews considered it as either the most common or second most common benign odontogenic tumour.<sup>7-9</sup> It comprises about 1% of oral tumours and 18% of odontogenic tumours.<sup>10</sup> It has a peak age of incidence of between the 3rd and 5th decade of life but studies in Nigeria and Ghana revealed a high prevalence amongst patients below 19 years of age.<sup>3</sup> In a Chinese study by Lu<sup>11</sup> *et al*, the average age of presentation of ameloblastoma was 31 years with 91% of the tumour occurring in the mandible and a male: female ratio of 1.5: 1. Hatada<sup>12</sup> *et al* reported a mean age of 35 years with a male: female ratio of 1.6: 1 and 93% localisation to the mandible. Mandible to maxilla ratio is higher in Africans (10:1) than in other groups.

In a Nigerian study which assessed prevalence of odontogenic tumours, ameloblastoma accounted for 59-74%; odontogenic myxoma 6-16%; and adenomatoid odontogenic tumour 2-13%. Agbaje<sup>13</sup> *et al* noted that the posterior mandible is the commonest site of occurrence of jaw ameloblastoma with 31% on the right and 26% on the left side. A female preponderance was noted by Chukwuneke<sup>14</sup> *et al* with the anterior mandible being the commonest site. A study by Adebayo<sup>15</sup> *et al* revealed that ameloblastoma is the commonest odontogenic tumour in Nigeria and constituted 73% of all odontogenic tumors in North-West region based on a 5 year retrospective review. Lasisi<sup>16</sup> *et al* reported that ameloblastoma constituted 14% of all jaw tumours and cysts and is the most prevalent odontogenic tumour in developing countries while Adisa<sup>17</sup> *et al* provided a geographic analysis of ethnic distribution of jaw tumours and revealed a hotspot in the North West and South West regions of Nigeria.

Ameloblastoma may arise from several primitive tooth-forming structures like the enamel organ, odontogenic rest cells, and the reduced enamel epithelium. It can also take origin from the epithelial lining of odontogenic cyst, the basal cell layer of the oral mucosa and on rare occasions from heterotrophic epithelium in other parts of the body like the pituitary gland.<sup>18</sup>

Ameloblastomas are slow growing, painless swelling of the mandible or maxilla. Most cases occur in the mandible, predominantly in the posterior molar region. Maxillary ameloblastomas always occur in the posterior molar region.<sup>1, 5</sup> The growth of ameloblastoma occurs in the buccolingual direction, this results in significant bony expansion. Pain is an uncommon symptom of ameloblastoma except where there is infection or haemorrhage inside or adjacent to the tumour. Malocclusion, facial deformity, soft tissue invasion and loosening of teeth are other signs and symptoms of ameloblastoma.<sup>3, 5</sup>

The unicystic variant of ameloblastoma most commonly occur in the paediatric age group and is said to probably arise from a pre-existent dentigerous cyst or from a dental follicle.<sup>19</sup> The extra-osseous sub-type most commonly present as a slow growing, painless, gingival swelling in adults. Metastasing ameloblastoma was re-classified as a benign tumour rather than a malignant tumour

because it shows a benign histopathology in spite of its metastatic potential. It is therefore difficult to differentiate it from conventional ameloblastoma.<sup>20</sup> Histopathologically, ameloblastomas are classically divided into follicular and plexiform variants; however other less common variants have been described like acanthomatous, clear cell, granular cell and the desmoplastic ameloblastoma. There is currently few documented relationship between the histopathological pattern and the behaviour or prognosis of the tumour.<sup>21</sup>

Chemotherapy has no proven role in the treatment of ameloblastoma but may be explored in malignant, recurrent or un-resectable cases. Targeted therapy using BRAF (B-raf serine-threonine kinase) inhibitors e.g. Vemurafenib show promise in case reports. Radiation is rarely used due to risk of malignant transformation hence the mainstay of treatment is conservative or radical surgery. Conservative surgery could be in the form of enucleation or curettage but are associated with high recurrence rates.<sup>22</sup> Radical surgery on the other hand entails total or partial jaw resections with or without disarticulation of the temporo-mandibular joint. Jaw resection may have profound effects on the quality of life of patients because it is often accompanied by complications such as facial deformity, paraesthesia, malocclusion and masticatory deficiencies.<sup>23</sup>

The literatures concerning ameloblastoma continue to evolve and it has been observed that studies conducted in some localities may not be the same when compared to existing findings. There is therefore need for continuous review of cases especially in the context of geographic and environmental pre-disposition. This study sought to review the cases of ameloblastoma seen at FTH, Gombe, Nigeria via her Oral and Maxillofacial surgery clinic from January 2000 to December 2024.

## MATERIALS AND METHODS

This was a retrospective study involving a review of patient's health records from the Oral and Maxillofacial surgery clinic, operating theatres and histopathology laboratory of FTH, Gombe. FTH,

Gombe is a major referral centre for the management of jaw tumours from the North East region which comprises of Bauchi, Adamawa, Taraba, Yobe and Borno states.

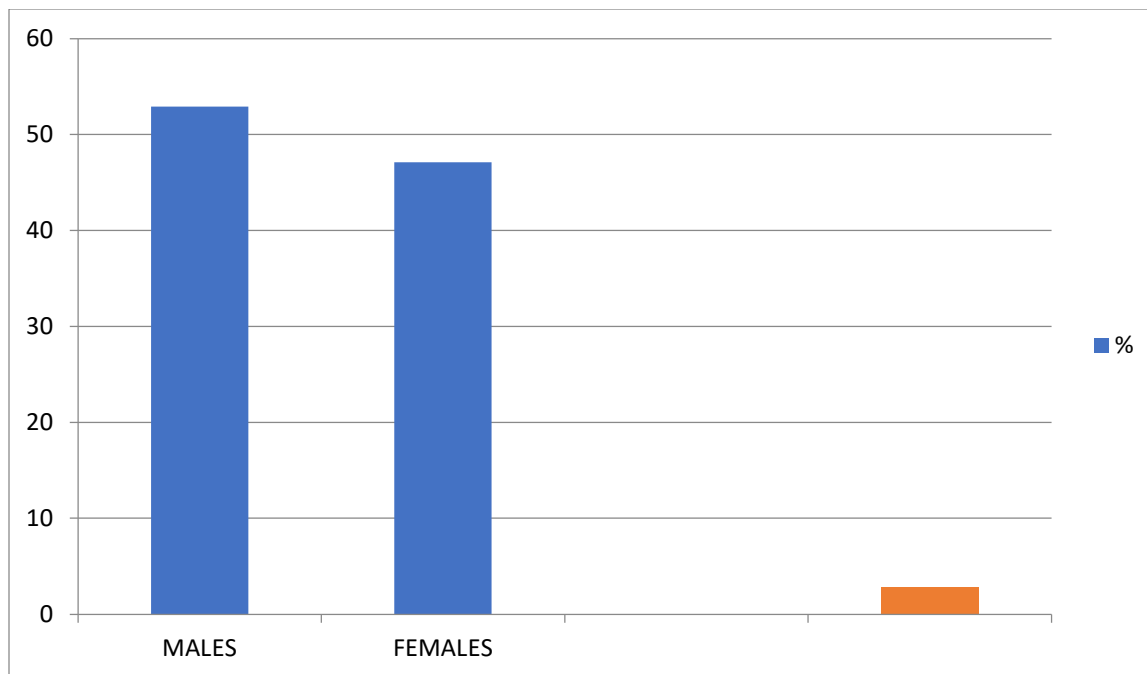
The records of participants considered eligible for the study was retrieved and the variables included were the socio-demographic characteristics, location, clinical sub-type, histopathological variant, the type of treatment provided and complications. All records of patients with a pre-operative or post-operative diagnosis of ameloblastoma from January 2000 to December 2024 were included. Patients with incomplete records were excluded.

Ethical clearance was obtained from the Research and Ethics Committee of the Federal Teaching Hospital, Gombe (NHREC/25/10/2013). The data collected were only used for purpose of the study and the information gathered remained anonymous to protect the identity of participants. Data is presented as charts and graphs and were summarised using descriptive or inferential statistics. Statistical analysis was done using SPSS version 23.

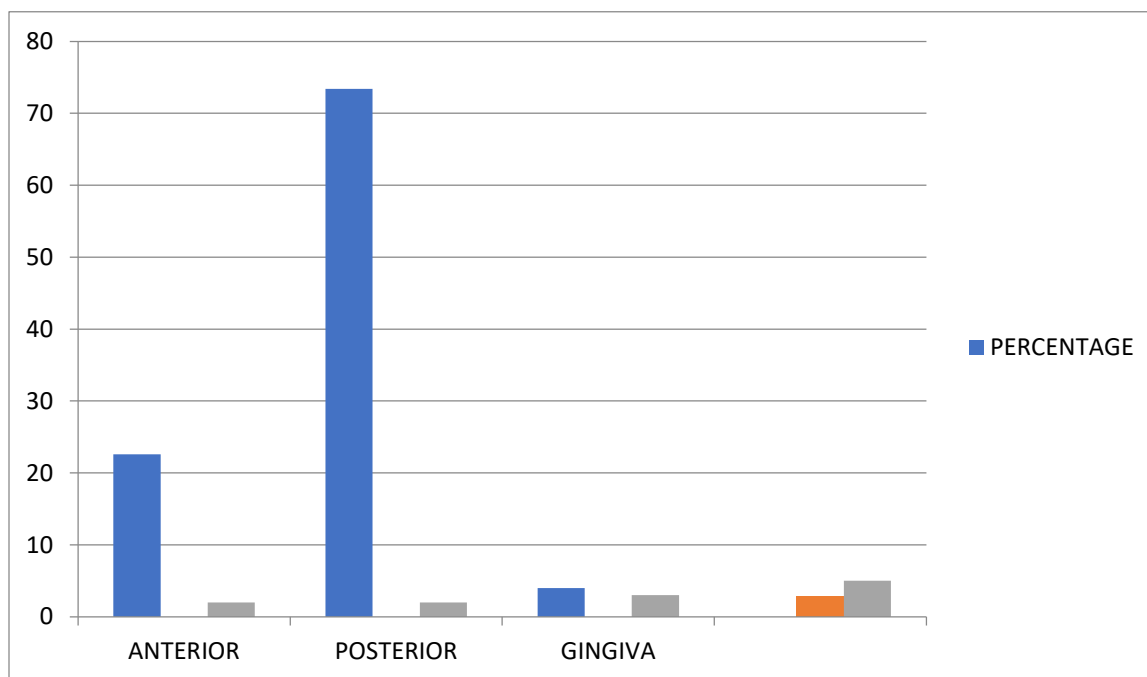
## RESULTS

A total of 219 cases were diagnosed clinically and histo-pathologically during the period of study. The age of the patients ranged from 15 to 55 years, with a mean age of 33 years (SD  $\pm 12$ ). The male: female ratio was 1.13:1 as there were 116 males and 103 female patients. All cases reviewed were located in the mandible with 161 (73.4%) located in the posterior region, 49 (22.6%) in the anterior and 9 (4.0%) where extra-osseous cases located on the gingiva. The conventional ameloblastoma was the most common clinical sub-type 150; (68.75%) and a multilocular radiolucency was the most common radiographic presentation.

Follicular type was the most common histopathologic variant 123; (56.25%) while the granular cell variant was the least common 2; (1.6%). 189 (85.6%) patients' treatment were by surgical resection (radical) and reconstruction was by the use of non-vascularised iliac bone graft. Six patients however, did not report for their surgeries.



**Figure 1:** graph showing the gender distribution



**Figure 2:** chart showing the localisation of the tumour

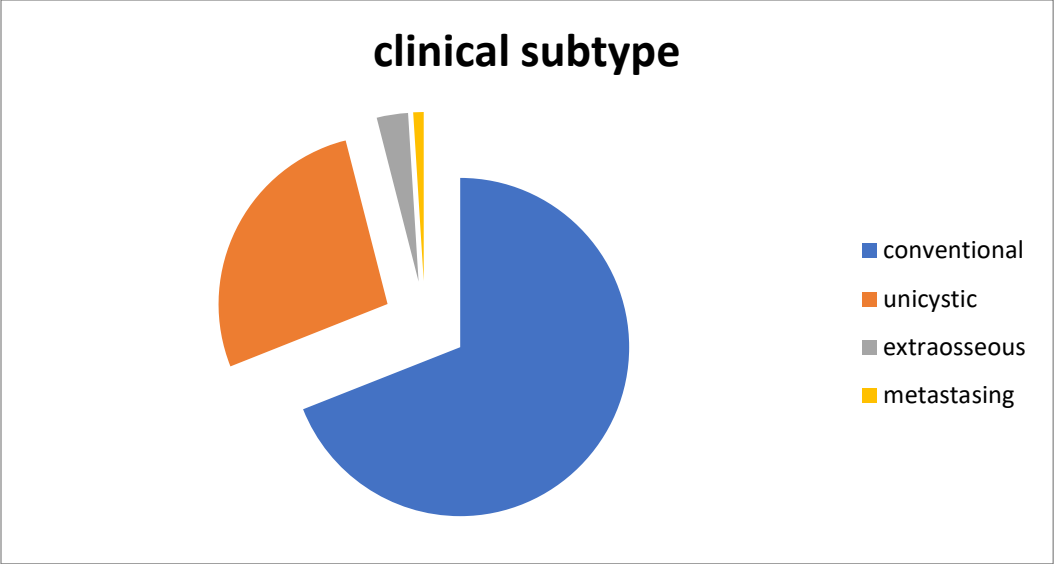


Figure 3: pie chart showing distribution based on clinical presentation

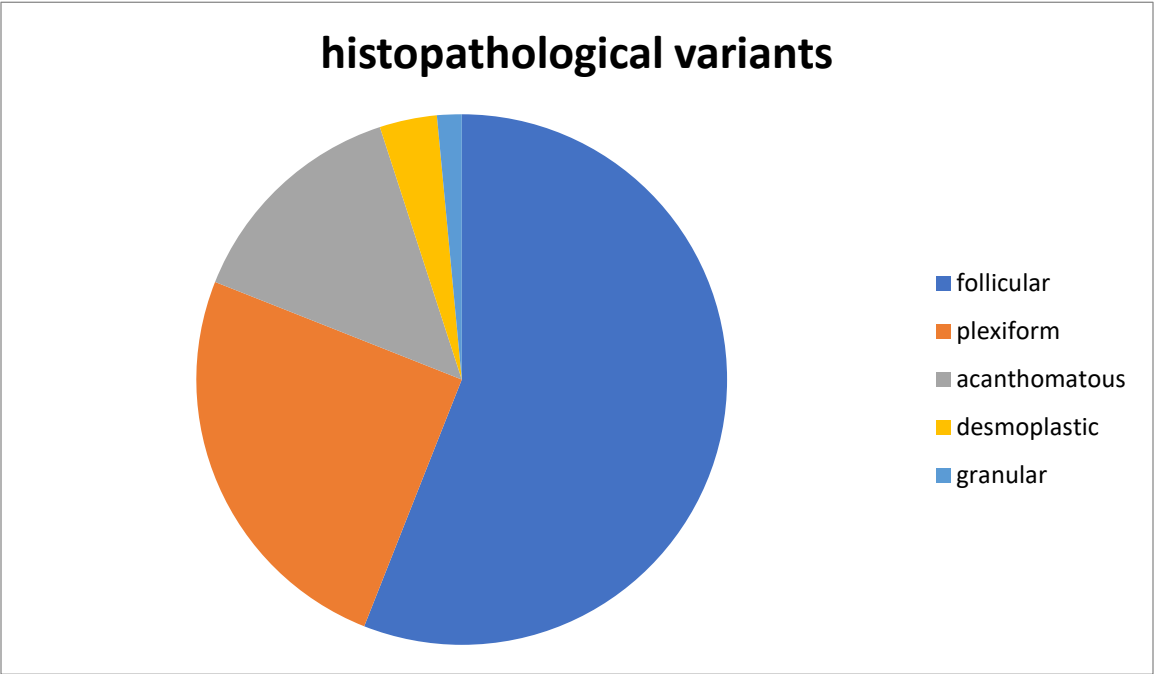


Figure 4: pie chart illustrating the distribution of the histopathological variants

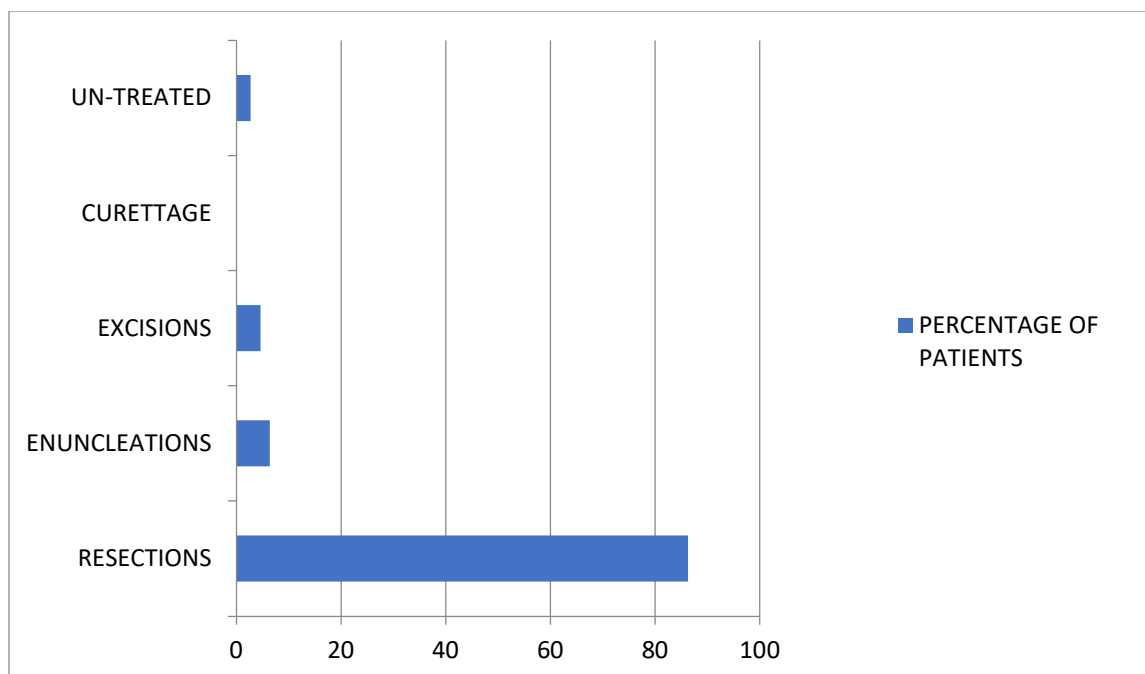


Figure 5: A bar chart showing the distribution of treatment provided

## DISCUSSION

The purpose of this study was to carry out a 25 years review of all ameloblastoma cases that presented at the Federal Teaching Hospital, Gombe. Retrospective studies of this nature have shown varied results. In India, an average of 10 cases annually has been reported while in Ghana between 5 to 7 cases each year was documented. Ladeinde and Adebayo reported a prevalence of between 13 to 15 cases annually in South West Nigeria.<sup>9,15</sup>

The finding of an average of 9 cases of ameloblastoma annually in this present study may suggests a relative decrease in the number of cases when compared with the duration of similar studies conducted in other regions of Nigeria. The decrease may be related to the reduction in hospital attendance occasioned by low socio-economic status and preference for traditional/herbal remedies. The increasing number of maxillofacial surgeons in the North east region of Nigeria may also account for this decrease.

Ameloblastoma presents commonly as a slow-growing and painless tumour which if left un-treated can attain enormous sizes.<sup>5</sup> This is a common finding in our locality where due to low socio-economic status and ignorance patients often seek herbal/traditional remedies before presenting to a hospital.

Ameloblastoma has no gender predilection and is found commonly between the 3<sup>rd</sup> and 5<sup>th</sup> decade of

life. An average age of 31 has been reported among Nigerians.<sup>9</sup> In India and other parts of the Middle East, an average age of 28 years was recorded. A slight female predilection was reported in Saudi Arabia.<sup>6</sup> There are also reports of male predilection in existing literatures. The implication is that ameloblastoma is a non X or Y sex gene-related disease resulting in this varied gender reports.<sup>24</sup>

In this study, there is an almost equal gender ratio with an age range of 15 to 55 years and a mean age of 33 years. This is consistent with the findings in the existing literature and similar also to documented reports.

In terms of clinical presentation, ameloblastoma can be conventional, unicystic, extra-osseous and rarely metastatic. In Africans, the conventional sub-type is the predominant. Olasoji<sup>25</sup> et al and Adeline<sup>26</sup> found the conventional (previously solid/multicystic) as the most common clinical presentation. In this study, 150 (68.5%) of the cases reviewed were found to be conventional while 59 (26.9%) were unicystic and 7 (3.1%) cases were of the extra-osseous type. There were also three cases of metastatic ameloblastoma. Ameloblastoma usually present in the mandible with very minute cases of maxillary presentations being reported. The posterior part of the mandible is the most common location but in India and Nigeria, the anterior part of the mandible is a frequent finding.<sup>10</sup> Maxillary ameloblastomas invariably occurs posteriorly around the maxillary tuberosity. In this



study, there was a relatively higher number of posterior presentations 161 (73.5%) and 54 (24.6%)

anterior cases in the mandible. In the present study, there were no reported cases in the maxilla.



Figure 6: A classical clinical and radiologic presentation of ameloblastoma in the mandible

There are two major histopathological variants of ameloblastoma namely, the follicular and the plexiform. The other less common variants include basal cell, granular cell, clear cell, acanthomatous and desmoplastic ameloblastoma. According to several studies, the follicular variant is the most common. A survey of the relative frequency of different histopathological variants by Gbom and

Maske is thus; follicular 33.9%, plexiform 30.2%, acanthomatous 1.3%, granular cell 3.5%, basal cell 1.4%, desmoplastic 1.4% and keratoameloblastoma 10.1%.<sup>27</sup> This study also shows a high prevalence of the follicular variant with 123 (56.2%), plexiform 65 (25.0%), acanthomatous 31 (14.1%), desmoplastic 8 (3.5%), and granular cell 2 (1.6%).

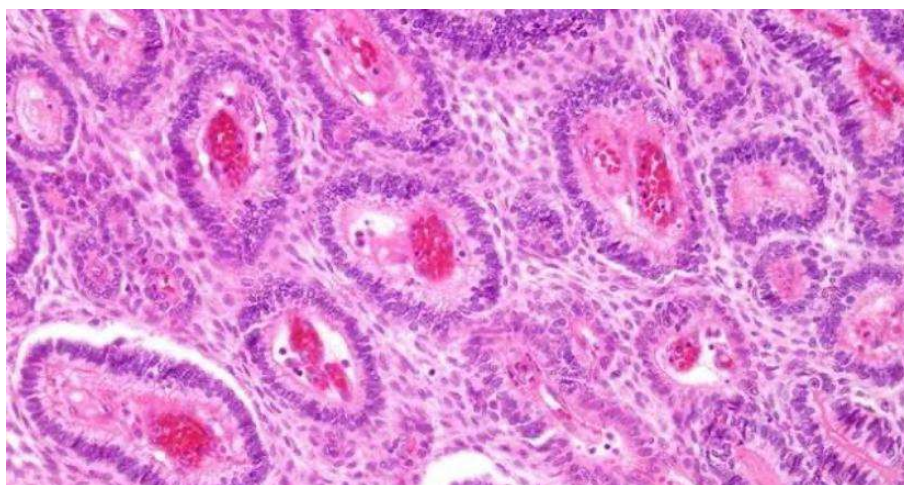


Figure 7: histogram of the patient diagnosed as follicular variant of ameloblastoma; showing islands of cells which resemble the normal dental follicle. The basal cells of these islands are columnar, hyper-chromatic and lined in a palisaded fashion.

Ameloblastoma does not respond to chemotherapy or radiotherapy hence the treatment is surgical. This

can either be conservative (enucleation or curettage) or radical. Conservative surgery is reserved for the

extra-osseous sub-type or smaller lesions because it is associated with a high rate of recurrence.<sup>22, 28</sup> Radical surgeries include various forms of resections like segmental osteotomy, hemi-mandibulectomy, sub-total or total mandibulectomy with or without disarticulation of the temporo-mandibular joint. In this study, most of the patients, 189 (86.3%) were

treated with resections while the 9 extra-osseous cases were managed using excision with wide margins. However, 6 (2.7%) of the patients did not report for surgical treatment and 4 cases of recurrence were recorded during the period of the study.



Figure 8: The same patient treated by surgical resection

A limitation to the study is the possibility of misclassification bias and the fact that the findings may not be generalizable. There is also lack of use of routine radiographic investigations like orthopantomograms which will aid the early detection of ameloblastoma cases, non-documentation of duration of swelling and reason for late presentation. More awareness should be encouraged in this environment so as to discourage the possible use of herbal or traditional interventions.

## CONCLUSION

Ameloblastoma cases at the Federal Teaching Hospital, Gombe are relatively low when compared to other regions of Nigeria but the findings indicate that the patterns are similar to what has been previously reported. The majority of the cases are conventional, located in the mandible and most of them were histopathologically diagnosed as follicular. There were 4 reported cases of recurrence which were initially managed by enucleation. There was no gender predilection and the prevalent age group is similar to what is documented in existing literatures.

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