

Challenges Associated with the Management of Giant Mandibular Ameloblastoma in a Resource-Poor Environment

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ABSTRACT Ameloblastoma, the most common odontogenic tumor in sub-Saharan Africa, has initial clinical features of painless, slow growth that sometimes results in giant sized, neglected, or gigantic lesions, often called giant ameloblastoma. The definition of giant ameloblastoma is controversial, their full characteristics, especially tumor growth rate, are yet to be fully elucidated, but their large sizes make management quite challenging. This case report presents giant mandibular ameloblastoma that developed over 10 years in a 30-year-old male. The lesion extended from one ramus to the other, measuring 48 cm × 38 cm × 17 cm. The treatment was segmental resection of the lesion after preoperative tracheostomy. Reconstruction was by retaining jaw form using the titanium reconstruction plate due to inability to perform free-bone grafts in this environment. There is need for universal health insurance in Nigeria to improve financial access to health-care services. Improved technical skill of surgeons and other clinical staff will also assist to optimize the treatment outcomes for patients with giant-sized lesions.

KEYWORDS: Ameloblastoma, giant, Nigeria, poverty, tumor growth rate

INTRODUCTION

Ameloblastoma is a rare, benign, locally invasive odontogenic, epithelial tumor that occurs worldwide, comprising about 1%–3% of all jaw tumors.^[1] It is the second most common (after odontomas) in the North American population,^[2] while it is the most common odontogenic tumor among Africans.^[3] The lesion may arise from the enamel organ, remnants of dental lamina, lining of dental cysts, and possibly from the basal cells lining the oral mucosa.^[4] Presentation occurs mostly in the third to fourth decades of life, cases at the extremes of life are rare, it shows no true sexual predilection and more cases are mandibular (80%) than maxillary (20%) especially in the posterior region of the jaws.^[2,3] The tumor is often painless unless secondarily infected, slow growing, causing buccolingual expansion of the jawbone, and sometimes perforating the cortices. It also causes displacement, mobility, and root truncation of teeth.^[3]

Among ameloblastomas, there have been the reports of giant, extensive, extreme, massive, and gigantic lesions due to the sizes attained. While there are no uniform

criteria for such reports, Chaine *et al.*^[5] defined giant mandibular ameloblastoma (GMA) as mandibular ameloblastoma with a large size (>5 cm in length), thinning of the lower and/or posterior mandibular borders and presence of considerable jaw expansion. According to Kanoi *et al.*,^[6] GMA causes a mandibular defect of 7–11.5 cm after segmental resection. These lesions are of peculiar interest for their large sizes and challenges in the management. Furthermore, previous reports of GMA lacked details necessary for calculating the tumor growth rate. The aim of this report is to present patient characteristics and tumor growth rate in a case of GMA with a view to evaluating their roles in the development and management of such patients in a resource-constrained environment. The approval of the institution and informed consent of the patient were

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obtained for inclusion in this report. The patient was assured that all steps would be taken to preserve his identity.

CASE REPORT

A 30-year-old male, resident in an urban center in Nigeria presented with a massive jaw swelling of 10 years' duration. The swelling started from the left posterior region and was progressively increasing till it extended from one ramus to the other. The patient had not sought previous dental treatment due to financial constraints but had gone for prayers and applied "anointing oil" to the lesion. The protrusive mandibular swelling measured about 43 cm × 38 cm, causing Class III skeletal malocclusion and limited mouth opening due to tumor size. The facial appearance showing size of the lesion is shown in Figure 1. The lesion was covered by intact skin and mucosa, bony hard to firm consistency to egg shell crackling in places. All teeth were present but mandibular teeth were mobile.

Aspiration yielded straw-colored cystic fluid. Computed tomography showed multilocular, soap bubble radiolucency extending from right to left ramus with many cortical fenestrations, as shown in Figure 2. Incisional biopsy revealed follicles of odontogenic epithelial cells within the connective tissue. The follicles demonstrated hyperchromatic peripheral cells and stellate reticulum-like central cells, as shown in Figure 3, to give a diagnosis of conventional cystic, follicular ameloblastoma. When the patient was able to obtain financial assistance for treatment, he was subsequently scheduled for surgery. In view of the limited mouth opening caused by the lesion, an elective tracheostomy to facilitate intubation was carried out. Thereafter, segmental resection of the mandible was performed to deliver a lesion with dimensions 48 cm × 38 cm × 17 cm. Figure 4 shows the resected jaw lesion. Using the formula devised by Mehrara *et al.*,^[7] as adapted by Chae *et al.*,^[8] for calculating specific growth rate of tumor (SGR), the SGR for this lesion was 102.3% per year. Reconstruction was carried out with a titanium plate, as shown in Figure 5. Last review was at 6 months postoperatively, and the patient had a fair appearance, as shown in Figure 6. The postoperative radiograph showed the plate is still in place with no evidence of infection or recurrence 10 months after surgery, as shown in Figure 7.

DISCUSSION

Kanoi *et al.*^[6] criticized earlier reports and criteria for the diagnosis of GMA as arbitrary and lacking any benefit in clinical decision making or contributing to



Figure 1: Frontal view preoperative photograph of giant mandibular ameloblastoma of 10 years duration in a 30-year-old male



Figure 2: Computed tomography scan of giant mandibular ameloblastoma showing extensive thinning of bone and multiple fenestrations with invasion of soft tissues

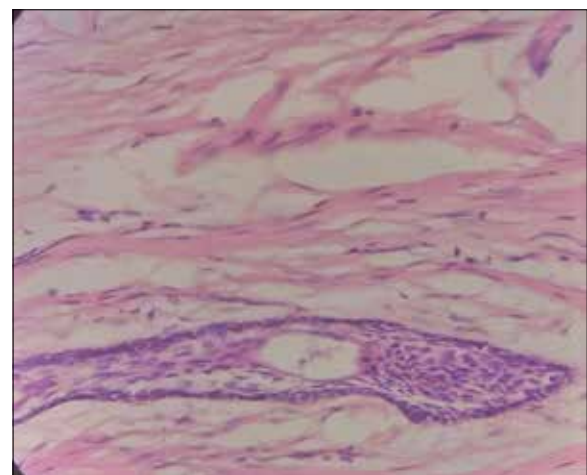


Figure 3: Photomicrograph of section of giant ameloblastoma showing follicular pattern (Hematoxylin and eosin stain, magnification × 100)

management. Therefore, they defined GMA as those causing defects of 7–11.5 cm in the mandible after

segmental resection. The lesion we resected measured 48 cm × 38 cm × 17 cm, total volume was 27,778 cm³ [Figures 1 and 4] is possibly the largest reported case of GMA. Acharya *et al.*^[9] found that patients with GMA often came from the rural parts of India while our patient was an urban resident. Poverty remains an important reason for delays in seeking treatment in resource-constrained settings such as Nigeria^[10] and India.^[6] Other reasons include fear of surgery and ignorance which have resulted in mortality from GMA.^[10] Our patient delayed in presenting for the treatment due to financial handicap. Universal health coverage is not presently available to most Nigerians. Improved financial access to surgical care would assist by reducing delays in treatment, allow for less radical management, and reduce overall cost and postoperative morbidity from surgical care.

Reichert *et al.*^[11] reported the mean age of ameloblastoma as 35.9 years but a recent study by Adebayo *et al.*^[12] found that GMA occur at an older mean age of 44.2 years (range 17–73 years). Our patient was younger than the mean age reported for ameloblastoma; however, GMA can present at any age.^[12] The histology of the GMA in this report was conventional, follicular with multiple cysts in Figure 3. The sexual, site, and histological subtype distribution of our case report was like previous reports of GMA.^[5,6] In view of the rarity and management challenges associated with GMA, its natural history and rate of growth were investigated by Chae *et al.*^[8] and Adebayo *et al.*^[12] From their analyses, the specific tumor growth rate for GMA was between 19.9% and 170% per year.^[8,12] Using the same formula, the SGR for our patient was 102.3% per year, which is within the range of other GMA.

The accepted treatment for GMA is segmental resection of the mandible with immediate reconstruction using free vascularized bone flap preferably from the fibula. This is often followed by rehabilitation with implant-retained over denture.^[5,6] However, in some centers, especially in resource-constrained environments, cost of surgical treatment and inadequate human and technical resources result in deviation from this standard treatment. While some cases of GMA die from uncontrolled disease due to lack of financial access to surgery, others are treated by segmental resection and reconstruction using bone plates [Figure 5] as in this report. Our center presently lacks the capacity to undertake free vascularized bone flap procedures. At 6 months, the postoperative appearance was fair, as shown in Figure 6 while radiograph showed reconstruction plate was still in place [Figure 7]. The patient is currently raising funds for definitive reconstruction. There is need for improved technical expertise especially among nursing and laboratory staff



Figure 4: Photograph of resected specimen of giant mandibular ameloblastoma measuring 48 cm × 38 cm × 17 cm



Figure 5: Intraoperative photograph showing titanium reconstruction plate in place to retain form of resected lower jaw



Figure 6: Frontal clinical photograph showing postoperative appearance 6 months after tumor resection and reconstruction with titanium plate

in Nigeria to support procedures such as free-tissue grafts for improved reconstruction following surgical ablation.



Figure 7: Postoperative posteroanterior radiograph of jaws showing reconstruction plate in place

CONCLUSION

GMA are rare forms of ameloblastoma causing massive jaw swellings that are esthetically unsightly. In addition, these jaw swellings necessitate extensive bone resection with or without reconstruction using free vascularized bone flaps. We present a case that could be the largest GMA ever reported. Tumor characteristics, especially SGR, are similar to previous reports. This study highlights the need to improve financial access to surgical care, reduce ignorance about oral/dental care, and improve the human and surgical technical capacity in developing countries.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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