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Gross surgical features and treatment outcome of ameloblastoma at a Nigerian tertiary hospital.

AA Olusanya[1], AO Adisa[2], AO Lawal[2] and JT Arotiba[1]
Departments of Oral and Maxillofacial Surgery[1] and Oral Pathology[2], College of Medicine, University of Ibadan, Ibadan, Nigeria.

Summary

Background: Ameloblastoma is a benign odontogenic tumour which is locally infiltrative and may cause severe craniofacial deformities. Its epidemiology, clinical and histologic configurations are replete in local and international literature, but data about its gross surgical patterns and treatment outcome in Nigeria is sparse. We therefore describe the clinical, gross surgical configuration, histopathologic features and the outcome of management seen at a Nigerian tertiary hospital.

Methods: Records of all histologically diagnosed Ameloblastoma from January 2000 to December 2011 at the University College Hospital, Ibadan, Nigeria, were retrieved from the departments of Oral and Maxillofacial Surgery and Oral Pathology. Patients' biodata, clinical, radiographical, gross surgical and histological features of the tumours, type of treatment, mode of jaw reconstruction and post-surgical follow up period data were documented.

Results: One hundred and sixty-three ameloblastomas were diagnosed during the study period, only 92 had complete records and were therefore included in the study. The mean age was 34.2 years (+/-14.25) and the male to female ratio was 1:1.1. Majority of the patients were in the low socioeconomic class (67.4%). Majority of the patients (76.5%) had segmental jaw resection and reconstruction was done mainly with non-vascularised iliac crest grafts. The gross surgical configurations were described as solid, cavitated and cystic.

Conclusion: Despite the locally aggressive nature of ameloblastoma, patients presented late for treatment and majority of them had segmental jaw resection as a mode of treatment. Three gross surgical configurations of the specimens were described.

Keywords; *Ameloblastoma, gross configuration, treatment outcome.*

Correspondence: Dr. A.A. Olusanya, Department of Oral and Maxillofacial Surgery, College of Medicine, University of Ibadan, Ibadan, Nigeria. E-mail: defamyl@yahoo.com

Résumé

Introduction : L'améloblastomie est une tumeur bénigne odontogénique d'infiltrative locale et peut causer des malformations cranio-faciales sévères. Son épidémiologie, ses configurations cliniques et histologiques sont citées dans la littérature locale et internationale, mais les données sur ses caractéristiques chirurgicales brutes et les résultats du traitement au Nigeria sont rares.

Méthodologie : Nous décrivons donc les configurations chirurgicales, caractéristiques histopathologiques brutes cliniques et les résultats de soins au vu d'un hôpital de soins tertiaires du Nigeria. Des relevés de toute améloblastomie histologiquement diagnostiqué de Janvier 2000 à Décembre 2011, au centre Universitaire Hospitalier, UCH, Ibadan, au Nigeria, ont été récupérés par les services de chirurgie buccale, de maxillo-faciale et de pathologie buccale. Les données biographiques des patients, les caractéristiques cliniques, radiographiques, chirurgicales et histologiques des tumeurs, le type de traitement, le mode de reconstruction de la mâchoire et le suivi postopératoire périodique ont été documentés.

Résultats : Cent soixante-trois améloblastomies ont été diagnostiqués au cours de la période de l'étude, seulement 92 avaient des dossiers complets et ont donc été inclus dans l'étude. L'âge moyen était de 34,2±14.25 ans et le ratio hommes-femmes était de 1 : 1.1. La majorité des patients était de la classe socio-économique faible (67,4%). Egalement, la majorité des patients (76,5%) a eu une résection segmentaire de la mâchoire et la reconstruction a été réalisée essentiellement avec les greffes non-vascularisées de la crête iliaque. Les configurations chirurgicales chroniques ont été décrites dans les cavités cystiques.

Conclusion : Malgré la nature agressive de l'améloblastomie locale, les patients se présentaient pour le traitement avec un retard et la majorité d'entre eux ont subi une résection segmentaire de la mâchoire comme mode de traitement. Trois configurations chirurgicales brutes des spécimens ont été décrites.

Introduction

Ameloblastoma is a benign tumour classified by the World Health Organization as an epithelial neoplasm of odontogenic origin [1]. It is the most common benign odontogenic tumour in African and Asians [2-5] and second only to odontomas in the European and American literature [6-7]. Ameloblastoma usually presents as a slow growing, painless swelling which

causes expansion of the bucco-lingual plates and sometimes infiltration of soft tissue. The growth of ameloblastoma may sometimes result in severe deformities of the craniofacial complex [8]. Although it is considered benign, the biologic aggression of ameloblastoma results in significant therapeutic challenges because it is locally invasive and this accounts for recurrence when treated conservatively [9]. Few authors have described the gross appearances of ameloblastoma in literature.

Ameloblastoma's established epidemiology, clinical and histologic configuration is replete in local and international literature, but documented data about its gross surgical appearances and the outcome of management in Nigeria is sparse. This study was therefore designed to describe the clinical, gross and histopathologic features as well as the outcome of management of Ameloblastoma cases seen at a Nigerian tertiary hospital.

Materials and methods

The records of all cases of histologically diagnosed Ameloblastoma seen from January 2000 to December 2011 at the University College Hospital, Ibadan, Nigeria, were retrieved from the archives of the departments of Oral and Maxillofacial Surgery and Oral Pathology. Information was obtained on patients' biodata, clinical, radiographical, surgical and histological features of the tumour, type of treatment, mode of jaw reconstruction and follow up period after treatment. The socioeconomic classes were categorised as Class 1 (high social class), 2 - 3 (middle class) and 4 - 5 (low social classes) [10]. For the cases which had more than one configuration co-existing the specimens were categorised as the type that was mainly represented.

Results

Two hundred and sixty six of the 1,595 lesions diagnosed during the 11 year study period were odontogenic tumours. Out of these, 163 were Ameloblastoma but only 92 had complete records and were therefore included in the study.

Age and gender distribution

- The age ranged from 12 to 80 years with a mean age of 34.2 years (+/-14.25).
- There were 43 Males and 49 female with a male female ratio of 1:1.1

Socioeconomic status

- Majority of the patients were in the low socioeconomic class (67.4%) while only 1.1% of the patients were in the high socioeconomic class (Table 1).

Clinical features

- The duration of swelling ranged from 0.8 to 240.0 months with a mean of 42.9 months (+/-41.8), and a median of 36.0 months.
- Pain was recorded in 54.3% of the cases.
- Two (2.2%) of the cases seen in this study did not present with the typical appearance of ameloblastoma and demonstrated unusual jaw expansion and intra-oral exophytic growth like the patient in Figure 1.
- Multi-locular radiolucency was the commonest radiological pattern seen in this study (Table 1).

Treatment

- Sixty-eight patients were treated surgically. See Table 1.

Reconstruction

- Reconstruction was done mainly with non-vascularised iliac crest grafts (Table 1)

Gross Surgical Appearance

- The mean dimensions of the specimens were 8.4 (+/- 3.198) cm for length, 5.2 (+/-2.068) cm for breadth and 3.4 (+/-1.367) cm for height (Table 2).
- Three main gross configurations of the tumours were observed in this study (Table 1 and Figure 1). These were described as solid, cavitated and cystic. The solid type appeared fleshy, pinkish grey in colour, firm to friable in consistency and had minimal necrotic areas. The cavitated type had multiple cavities with intervening areas of fleshy tumour mass while the cystic type was essential one large cystic space which sometimes was continuous with 2 or 3 other cystic spaces. The cystic types tended to have a thickened continuous lining that appeared yellowish or pinkish and some were cobbled on their inner surface. The lining had the tendency to peel off fairly readily from the underlying bone (Figure 1d). The Cystic type was the commonest while the solid and the cavitated types were of almost equal proportions (Table 1). In 27 (29.3%) of the cases more than one configuration was seen. The combination configurations seen were 15 (16.3%) cases of cystic/solid, 11(12.0%) cases of cavitated/solid and a single (1.1%) case of cystic/solid/cavitated.

Histology

- The cystic histological type was the most common in this study (Table 1).

Follow up and Recurrence

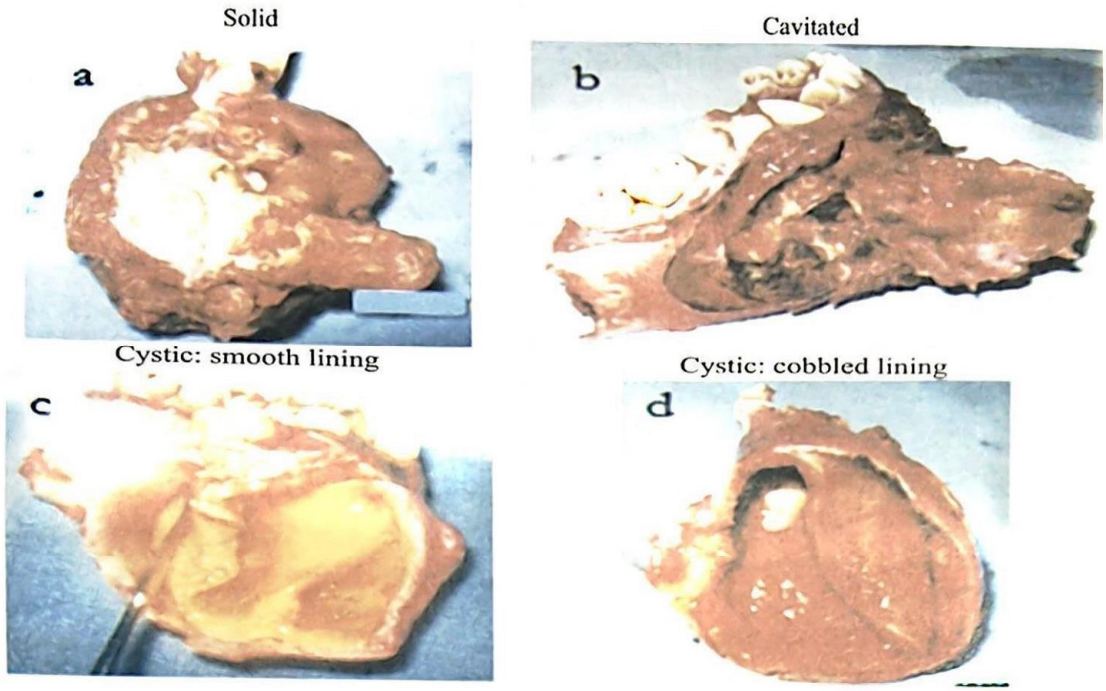

Fig. 1: Gross surgical appearance

Table 1: Table of results

Socioeconomic classes (%)	Class I 1.1	Class II 7.6	Class III 23.9	Class IV 32.6	Class V 34.8	
Site	Mandible 94.6% (87)	Maxilla 5.4% (5)				
Position	Anterior 27.2% (25)	Posterior 72.8% (67)				
Side	Right 33.7% (31)	Left 34.8% (32)	Bilateral 31.5% (29)			
Radiology	Unilocular radiolucency 25.0% (23)	Multilocular radiolucency 57.6% (53)	Mixed opacity 17.4% (16)			
Histology	Cystic 27.2% (25)	Plexiform 25.0% (23)	Follicular 22.8% (21)	Acanthomatous 6.5% (6)	Telangiectatic 3.3% (3)	Others 3.3% (3)
Gross appearance	Solid 26 (28.26%)	Cavitated 24 (26.09%)	Cystic 42 (45.65%)			
Treatment	Segmental resection	Enucleation	Hemi-jaw resection	Subtotal jaw resection		
	76.5%	13.2%	8.8%	1.5%		
Reconstruction	Iliac crest graft	Steinman's pin	Reconstruction plate	No reconstruction		
	66.2%	10.3%	1.5%	22.0%		
Recurrence	*Type of Treatment*	*Recurrence*	*Mean period of recurrence post surgery (months)*	*Site*		
	Enucleation	22.2% (2/9)	28	Mandible		
	Segmental resection	3.8% (2/52)	85.5	Mandible		
	Hemi-jaw resection	16.7% (1/6)	7	Maxilla		

Mean follow up period was 18.51 months (+/-22.7) and ranged from 0.3 to 156.0 months with a median of 11.5 months. During this period, five cases (7.4%) had recurrence. The mean period of recurrence was earlier in the more conservative type of treatment while the recurrence that followed the radical treatment of a hemi-jaw resection occurred in the maxilla. The recurrent cases are as illustrated in Table 1.

Discussion

Mandibular swellings accounted for the majority of tumours in this study which is in accordance to the previous documentations on ameloblastoma both locally and internationally [3,11-13]. The reason for the higher incidence of this tumour in the mandible is still obscure. A few authors have suggested some theories based on infection and chronic irritation but these are yet to be substantiated [11]. The posteriorly located tumours were commoner and almost equal distribution of left sided, right sided and bilateral

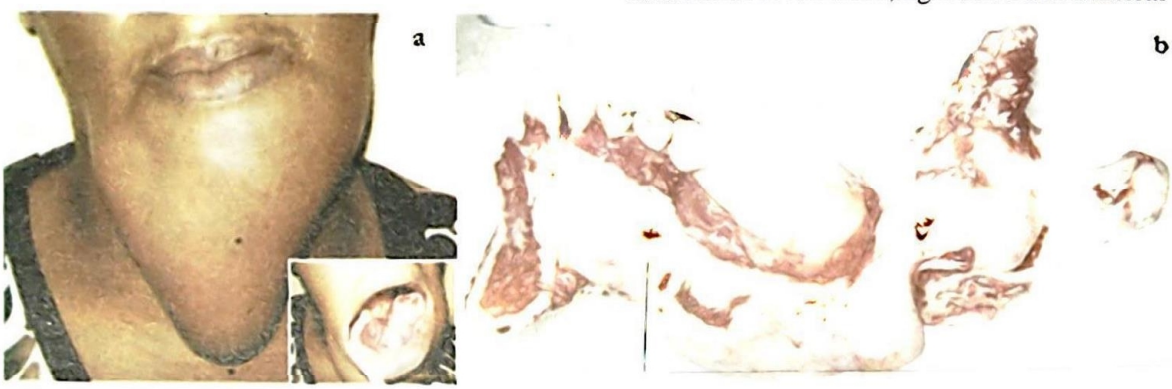

Fig. 2: Atypical presentations of ameloblastoma

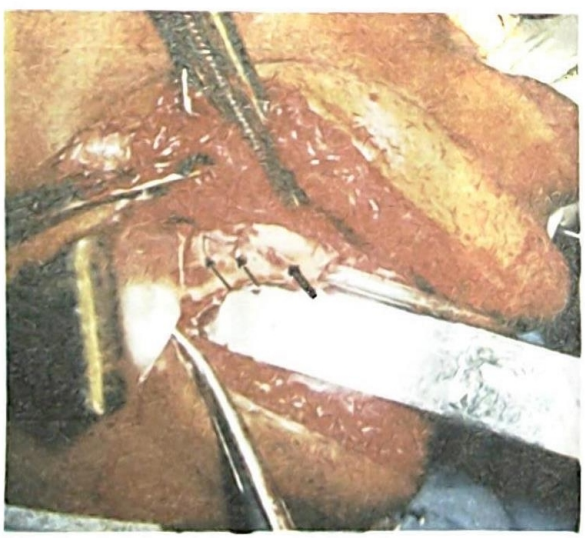

Fig.3: Bone regeneration can be seen engulfing the anchoring wires (thin arrows) and along the length of the Steinman's pin (block arrow).

occurrence was observed. Pain was reported in about half of the patients which was unusual as this symptom is not considered an intrinsic component of the lesion [1]. The pain therefore may have been due to trauma or superimposing infection which can occur secondarily. Indeed Yaacob reported that all Ameloblastoma patients who complained of pain in his study had perforations of their cortical plates [14]. He suggested that this perforation could readily allow tumour infection following any breach of oral soft tissues [14].

Typically ameloblastoma presents as an intra-bony lesion with bucco-lingual expansion. Generally, the covering mucosa, though may be thinned out, usually remains intact with or without imprints of the opposing teeth. On palpation the lesion may be bony hard through depressible and firm to fluctuant where cortical perforation has occurred. Some of the cases seen in this study showed deviation from the typical presentation described above. These atypical presentations are noteworthy as such may mimic the clinical appearance of a malignancy by presenting

Table 2: Dimensions of the gross surgical specimens

	Mean (+/- SD)	Maximum	Minimum	Median
Length (cm)	8.4 (+/- 3.198)	20.0	4.0	8.0
Breadth (cm)	5.2 (+/- 2.068)	12.5	2.0	4.2
Height (cm)	3.4 (+/- 1.367)	8.0	1.5	3.0

as an ulcerative exophytic lesion (Fig. 2a) or some other type of tumour like the intra-modullary progression of Keratocystic odontogenic tumour (Fig. 2b).

Ameloblastomas are commonly radioluscent lesions usually multilocular in pattern especially when large [14]. However, radiographic patterns showing combination of both radiolucency and radiopacity have been previously documented [11]. This was observed in almost a quarter of the patients in this study. This combination pattern may not be unassociated with longstanding lesions as the mean duration of lesion for the lesions with combination radiographic patterns was 5.5 years as compared with the 3.8 years for the radiolucent lesions. Radiographic appearance may thus be an index for approximating the duration of ameloblastoma. This may be significant in our environment as it is common practice for patients to intentionally shorten time intervals of tumours for fear of being blamed for negligent attitude, even though the delay was most likely due to ignorance or poverty [15]. A study in Malaysia inferred that late presentation was due to the absence of pain from the tumour [14].

The commonest modality of treatment for ameloblastoma in our centre is jaw resection with 1.5 to 2.0 cm of clinically free margin. Majority of the jaw reconstructions in this study were done using nonvascularised iliac crest grafts which is the usual practice in our centre because of lack of facilities for microvascular tissue transfer, its ease of harvest, and minimal complications encountered with this type of grafts. Steinman's pin reconstruction was used in cases where the residual normal intra-oral soft is considered insufficient for adequate graft coverage or when the extended surgical period required for jaw resection as well as graft harvest is contra-indicated in patients with compromising, preoperative systemic conditions. Steinman's pin placement is considered as an alternative temporary measure which is removed at a later date via an extra-oral approach and the defect reconstructed with autogenous bone graft. The Steinman's pin, though not osteogenic, has been known to osteoconduct the path of new bone formation along its length which was observed in one of the patients in this study (Figure 3). A number of the patients have also been known to carry these pins for as long as nine years without complications.

Some authors have described the gross appearance of ameloblastoma as solid areas with white to gray color and little haemorrhage or necrosis [16]. Others described the gross appearance as cystic, solid or cystic-solid, however, almost all ameloblastomas show cystic degeneration [17, 18]. This cystic degeneration is believed to be a function of age [17]. The need to distinguish ameloblastoma grossly is necessary because of the better prognosis for the unicystic type after limited surgical treatment than it is for the solid type [19].

Review and follow up of patients after treatment in this part of the world still remains a major challenge to assessing the outcome of treatment interventions [11]. Few studies from Nigeria on odontogenic tumours in general and ameloblastoma in particular have been able to document their outcome of management after varying periods of follow up [3,20,21] while other studies were mainly on clinicopathological patterns without treatment or short follow up periods of less than 12 months without specific figures [4,11,12,22,23]. In this series the follow up period ranged from 0.3 months to 156 months with a mean of 18.7 months.

Olaitan *et al.* [20] reported a recurrence rate of 7.4% which was similarly observed in the present study but, Arotiba *et al.* [3] reported higher recurrence rates of 10% and 40% for tumours treated with wide excision and conservative approaches respectively. Of the five recurrent cases in this study, one recurred in the maxilla (25% of the treated maxillary cases) as compared with 4/64 (6.3%) that recurred in the mandible. The single maxillary lesion recurred much earlier than the mandibular lesions despite the radical treatment undertaken for the case (Table 1). The maxilla is a site that poses a significant challenge to the treatment of ameloblastoma due to its having a lower amount of compact bone; a situation which provides little resistance to the spread of the tumour and may allow spread of daughter cells 1 to 2 cm beyond the apparently free surgical margin.

Conclusion

The pattern of presentation and outcome of management for ameloblastoma in the last 11 years in our institution have been documented. Despite the locally aggressive nature of ameloblastoma, patients commonly present late for treatment and majority of them had segmental jaw resection as a mode of treatment. The recurrence rate was fairly high especially following conservative treatment. Three gross surgical configurations of the specimens were described.

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