Original Article

Ameloblastoma of the jaws in children: an evaluation of cases seen in a tertiary hospital in South-Eastern Nigeria

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SUMMARY

Background: Ameloblastoma is one of the most common benign odontogenic tumours in Nigeria. It is considered uncommon in children.

Materials and methods: This is a retrospective study of pediatric patients with histopathological diagnosis of ameloblastoma seen over seven years at the Oral and Maxillofacial Surgery Department of the University of Nigeria Teaching Hospital, Enugu, Nigeria. Relevant information was retrieved from patients' records and data obtained were analysed using SPSS version 23, the chi-square test was used to compare qualitative variables, a p-value of <0.05 was considered as significant.

Results: One hundred and thirty-six cases of ameloblastoma in all age groups were seen within the period. Thirty of the cases met the requirement. The mean age of the patients was 14.4 STD 2.03 (range from 10-17) years. Fourteen (46.7%) patients were male while 16 (53.3%) were female giving a ratio of 1:1.1. The duration of the lesion ranged from 3 months to 72 months (mean 15.07 months). Histologically, the follicular type (n=20, 66.7%) constitute the majority, while the clinical types were solid-multicystic (n=18, 60%) and unicystic (n=12, 40%). Enucleation was the treatment of choice in most (n=18, (60%)) of the patients.

Conclusion: Ameloblastoma is relatively uncommon in children, especially those less than ten years of age. The solid-multicystic variety was the predominant type in the children studied. Most patients presented long after the onset of the tumour and enucleation with mechanical curettage produced satisfactory results in these patients.

Keywords: Ameloblastoma, odontogenic tumour, children, resection, enucleation.
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INTRODUCTION

Ameloblastoma is a benign odontogenic tumour that exhibits a variably aggressive biological behaviour as shown by its infiltrative growth pattern and marked tendency for recurrence and malignant transformation.[1] It is the most common odontogenic tumour and affects the jaws exclusively.[2] Previous Nigerian study by Odukoya reported that odontogenic tumours (OT) constituted 19% of all orofacial tumours and tumour-like lesions. Ameloblastoma was the commonest OT, (58.5%) and showed a predilection for males and the posterior mandible.[3] It is commonly found around the 3rd and 4th decades of life and rarely in children. However, it occurs in almost all age groups.[4] About 10%–15% of ameloblastoma cases occur in the paediatric population.[1] Small and Waldron[5] described

Ameloblastoma as a slow-growing lesion that often starts developing around early childhood and young age without manifesting until adulthood.

It ranks top among the odontogenic tumours in Africa and Asia and the second most common in America.[6,7,8,9] Ameloblastoma arises from tooth forming apparatus including remnants of the dental lamina, enamel organ, epithelial lining of an odontogenic cyst, Hertwig's root sheath and basal cells of the oral epithelium.[10] Well known downstream signal pathways like sonic hedgehog, nitrogen activated protein kinase and WNT/β-catenin pathway associated with deregulation of multiple genes are strongly linked to molecular and cytogenetic factors that promote the oncogenic transformation of odontogenic epithelium and rest cells to ameloblastoma.[1]

Ameloblastoma presents with jaw swelling typically producing buccolingual expansion, dental anarchy and tooth mobility. Clinicosurgically, ameloblastomas are classified into 4 groups namely Conventional (solid) also known as solid-multicystic, Unicystic, Peripheral, and Malignant. The most aggressive clinicopathologic association in the benign types is seen in the conventional type, which is associated with the highest recurrence rate of up to 90 % with conservative operations such as enucleation and curettage.[11,12,13] Radiographically, it could present as Uni or Multilocular radioluscency.

These presentations could affect the choice of treatment which ranges from simple enucleation and curettage to resection of the affected jaw bone.[14] Treatment in children could be a challenge to the surgeon as they have to balance issues of recurrence with problems associated with radical surgical option in this age group. Several studies on this topic have been reported globally but no study has been done in South Eastern Nigeria, thus the purpose of this study is to determine the prevalence of this tumour in South Eastern Nigeria particularly among children to add to the literature on the subject.

METHODS

This is a retrospective study of pediatric patients with histopathological diagnosis of ameloblastoma seen over a period of 7 years from July 2009 to June 2016 at the Oral and Maxillofacial Surgery Department of our hospital in Enugu, Nigeria. The record of all the patients seen within the study period was obtained from the departmental register and case files of the patients retrieved from the Record Department of the hospital. Relevant information such as biodata, site, duration of the tumour, clinical and histopathologic type, radiologic appearance, treatment options and outcome were extracted from the case files, histological slides were re-evaluated to authenticate the previous diagnosis.

Data obtained were analysed using IBM SPSS Statistics for Windows Version 23 Armonk, NY, the chi-square test was used to compare qualitative variables, a p-value of <0.05 was considered as significant. Included in the study were those whose data were complete. Patients who were more than 18 years of age at the time of presentation were excluded from the study.

Ethical Consideration

Due to the retrospective nature of this study, it was granted an exemption in writing by the University of Nigeria Teaching Hospital Health Research and Ethics Committee (NHREC/05/01/2008B-FWA0000248-IRB00002323).

RESULTS

One hundred and thirty six cases of ameloblastoma in all age groups were studied between July 2009 and June 2016. Thirty of these cases were children below the age of 18 years which constitute a prevalence of (22.1%).
The age range of the patients was 10-17 years with a mean age ± SD of 14.4 ± 2.0 years. There were 14 males and 16 females; giving a male: female ratio of 1:1.1. The duration of the swelling ranged from 3 months to 72 months, mean ± SD of 15.1 ± 16.3 months, the mean duration in females (18.9 months) was almost twice that of the males (10.7 months), however, this was not statistically significant (p=.157).

Histological types were follicular 20 (66.7%), plexiform 9 (30%) and acanthomatous 1 (3.3%) while the clinical types were solid 18 (60%) and cystic 12 (40%). Seventeen (56.7%) patients had multilocular radiolucent lesion and 13 (43.3%) had unilocular radiolucency. In 28 (93.3%) cases, the tumour occurred in the mandible while 2 (6.7%) occurred in the maxilla.

Fifteen (83.3%) of the solid type were of follicular and 3 (16.7%) were plexiform while 5 (41.7%) of the cystic type were follicular, 6 (50%) plexiform and 1 (8.3%) was acanthomatous. This shows a slight statistical significant relationship (p=0.05) between histological and clinical types of ameloblastoma. The two cases affecting the maxilla were of follicular type, while clinically, each was of cystic and solid types respectively.

The site distribution of the tumour on the mandible shows anterior 13 (46.4%), body 11 (39.3%) and posterior 4 (14.3%). Tumour extending to both sides of the lower jaw (bilateral) and those on the right side, accounted for 10 (35.7%) each while the left side contributes 8 (28.6 %). Eighteen (60%) patients were treated by enucleation while 12 (40%) patients had jaw resection.

Table 1 Distribution of the histological, clinical and radiographic types among various age groups and sex

	Age (%)		Sex (%)	
Hjstologic type	**10-13**	**14-17**	**Male**	**Female**
Follicular	6(60.0)	14(70.0)	11(78.6)	9(56.3)
Plexiform	3(30.0)	6(30.0)	2(14.3)	7(43.8)
Acanthomatous	1(10.0)	-	1(7.1)	
Total	10	20	14	16
P value		0.350		0.145
Clinical type				
Solid-multicystic	3(30.0)	15(75.0)	8(57.1)	10(62.5)
Cystic	7(70.0)	5(25.0)	6(42.9)	6(37.5)
Total	10	20	14	16
P value		0.018		0.765
Radiologic type				
Multicystic	3(30.0)	14(70.0)	6(42.9)	11(68.8)
Unicystic	7(70.0)	6(30.0)	8(57.1)	5(31.3)
P-value		0.037		0.153

Table 1 shows that follicular type was the commonest histologic type seen amongst the patients. There is a statistically significant relationship between the age of the patients and the clinical and radiological types.

Table 2 Distribution of treatment type against clinical types of Ameloblastoma

		Clinical type			
Treatment Type		Solid-multicystic	Cystic	Total	
	Enucleation with mechanical curettage	8 (44.4%)	10 (55.6%)	18 (100%)	p=0.03
	Resection	10 (83.3%)	2 (16.7%)	12 (100%)	

Table 2 shows that there is statistical significance relationship (p=0.03) between the clinical types of ameloblastoma and treatment options used in this study.

Table 3 Distribution of Treatment type, Complications and Recurrence within 2 years of follow up

Age groups	**Treatment Type**		**Complications**			**Recurrence (in 2 years)**
	Enucleation with mechanical curettage	Resection	Nil	Root Resorption	Lip Paraesthesia	
10 -13	7 (23.3%)	3 (10%)	10 (33.3%)	-	-	1(3.3%)
14 – 17	11(36.7%)	9 (30%)	14 (46.7%)	5(16.7)	1 (3.3%)	-

Table 3 shows that enucleation was the commonest treatment option among the younger age group while resection was the commonest among the older age group.

DISCUSSION

Ameloblastoma is a clinically important odontogenic tumour; its relevance cannot be overemphasised due to its high health burden, high prevalence rate, aggressiveness, high potential for recurrence, and occasionally malignant transformation.[1,7,15,16] Management of this tumour is challenging especially in resource-limited environment like Nigeria.

In Nigeria and other developing countries, the typical presentation in most of the patients is obvious jaw swelling of buccolingual expansion pattern and dental anarchy. Occasionally, few cases are diagnosed accidentally from radiographic investigations in asymptomatic patients.

Ameloblastoma is known to be uncommon in children and young adults and studies show that its prevalence varies due to factors ranging from race and location where the study was done, the age limit chosen for the paediatric population and duration of study.[2,16] The prevalence of ameloblastoma seen in this study is similar to previously reported multicenter study in Nigeria by Arotiba et al[6]. Butt et al[17] reported a prevalence of 21.3% which also appears similar to that of this study. Our finding is significantly higher than some other African studies by Olaitan et al[18], Olasoji et al[19] and Chidzonga et al[20] where a prevalence of 14.6%, 16.8% and 17.9% respectively were reported. Prevalence as low as 6.8% has been reported among children in some western studies.[21,22]

Meanwhile, Ord et al[2] and Al-khateeb and Ababneh[23] both reported prevalence rates much higher than the African studies. This finding signifies variations possibly due to race or location and may also suggest that these cases could be under-reported in our environment. Our finding is similar to the reported average of 14.7 years by Arotiba et al[6], Olasoji et al[20] and Belardo et al[24] in their respective studies. None of the patients in this study was under the age of 10 years which supports the findings that the tumour is rare within the first decade of life.[2,19,21,24,25]

Although a higher mean age (16.0 years) was reported in a Jordanian study, a much lower mean age (10.8years) was reported in Argentina.[22,24] The sex distribution seen in this study agrees with that reported by Al-khateeb and Ababneh [24]and Khan et al [26]who noticed a higher female ratio. However, it differs from other studies in Nigeria and other parts of the world.[6,19,20,23,26,27] Very few studies considered the duration of the tumour at presentation. The mean duration of the tumour at presentation in this study was significantly lower than that reported by Olasoji et al[20] in their study of Northern Nigeria population although their peak duration was 60 months as against 72 months in our study. It was also noticed that the mean duration in female was almost twice that of the male although this is not statistically significant. The reason for this is not clear but it can be attributed to sociocultural discrimination among the female gender as most families tend to cater for their male children more than the female.

There is scanty information on the histologic distribution of this tumour among this group of patients. The commonest histologic type seen in this study is the follicular type which disagrees with the finding of Olasoji et al [20] who reported the plexiform type as the commonest histologic type. This study also showed a significant relationship between histologic types and clinical types as most of the follicular types were solid-multicystic while the cystic types were mostly plexiform ameloblastoma.

Clinical types of ameloblastoma include solid-multicystic, unicystic and peripheral types. In this study, there was significantly more solid type than the unicystic type. Arotiba et al[6], Huang et al[23] and Wenlu et al[28] all reported higher ratio of multicystic types than the unicystic types. On the contrary, Ord et al[2], Abdulai[29] reported higher number of the unicystic type in their various studies, hence, our finding appeared to differ with the notion that most ameloblastoma in children arises from odontogenic cysts.[2,15]

The solid type seen more in the older paediatric age group conformed with the other studies that suggested that this type of tumour is the commonest form found in the African child and resembles the adult variety[2],even though it could also be due to the late presentation of these patients. The common radiographic classification of ameloblastoma in children is unilocular and multilocular.[29] The essence of radiographic assessment in the management of this tumour is that it aids in determining the extent of the lesion thereby assisting the surgeon to achieve a tumour free margin during surgical treatment. It is also very useful in making diagnosis of the tumour.[14] Majority of the patients had multilocular radiolucent lesion in our study (Table 1).

This finding is comparable to most African studies where the researchers reported multilocular radiolucency as the commonest radiographic presentation.[6,19,20,21] On the contrary, Huang et al[23], Khan et al[26], and Wenlu et al[29] reported unilocular type to be more common. The mandible is the commonest site of presentation of the tumour in this study. This agrees with other studies that reported the preponderance of ameloblastoma in the mandible.[2,6,19,29,30] Of the 28 cases in the mandible, anterior tumours with bilateral extension were the commonest pattern seen. This agrees with that of Olasoji et al[20] who reported that the anterior mandible accounts for a majority of the site of the tumour followed by the body. Ord et al[2] and Arotiba et al[6] both reported the angle and posterior ramus region to be the predominant sites in their various review of the literature. The reasons for the high frequency in the anterior region is not clear however, a number of researchers suggested that this could be due to poor oral hygiene and irritation from calculus deposited commonly at this region.[20,21] We also notice that the right mandible was more affected than the left which coincides with similar study reported by Agbaje et al.[30]

Management of ameloblastoma in children poses a lot of challenges because the facial bones and other structures in this age group are growing. As a result, radical surgery has the potential to disrupt this growth process leading to severe facial deformities and unacceptable aesthetics.[6,21] Due to the high recurrence rate of ameloblastoma, resection of the jaw with healthy margin of about 1.5cm is considered the best treatment option; however, due to the challenges associated with radical surgery more conservation approaches have been explored.[14]

The choice of treatment in these patients is determined by the patient age, extent and location of the lesion, histological type and growth rate.[14] There is a strong statistically significant relationship ($p = 0.03$) between the clinical type and treatment option of this lesion as more than half of the solid types were treated by resection while 83.3% of the cystic types were treated by careful and thorough enucleation with mechanical curettage using vulcanized burs (Table 2). The reason for this finding may be attributed to the fact that there are limited facilities for adequate reconstruction of the affected jaw with flaps after resection in these children.[20,29,31]

These findings in this study, therefore agree with the growing support for conservative surgical treatment for ameloblastoma in the paediatric population and young adults.[20] Zhang et al[28], Wenlu et al[29], Huang et al[23] and Abdulai et al[30] reported that (78.4%), (76.7%), (73.3%) and (83.3%) patients respectively were treated conservatively. It has also been suggested that enucleation could be a way of 'buying time' for the jaws to fully develop before a more definitive treatment is carried out.[30]

Though a number of the cases presented long after the onset of the tumour, the outcome of the conservative surgical option adopted in this study was encouraging as only 1 patient had recurrence within 2 years of follow up which was resected again (Table 3).

CONCLUSION

Ameloblastoma is relatively uncommon in children and it is rarely seen before the age of 10 years. The anterior mandible was the commonest site while the solid-multicystic variety was the predominant type. Most patients in this study presented long after the onset of the tumour and enucleation appears to be a satisfactory treatment of option for these set of patients.

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