Ameloblastoma in Children and Adolescents: A Seven-year Study in Enugu, Nigeria

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ABSTRACT

Objective: To study the prevalence and clinicopathologic features of ameloblastoma in children and adolescent patients at a tertiary health centre in Enugu Southeast Nigeria.

Methods: This is a seven-year (2012 to 2018) retrospective observational study of children and adolescent patients below 19 years of age. Their biodata and clinico-pathologic information were extracted from the biopsy forms, histopathology reports and case files archived in the Records department. Further clinicopathologic information obtained included: gender, site of tumour, age at presentation, duration, type of tumour fluid aspirate (straw, dark-brown, serosanguinous, purulent, negative aspirate), presence of ulceration, complaint of pain, histologic type of ameloblastoma diagnosis, and the age-at-onset of tumour (the time of patients or guardians' awareness of occurrence of the tumour before presentation).

Results: Thirty-seven (34.4%) out of 93 cases of ameloblastoma were observed in children and adolescents. The mean age was 13.59±2.87 years, with age range of 6 years to 18 years. Male patients were 15 (40.5%) in number, and female patients were 22 (59.5%) with a male to female ratio of 0.7:1. Children in the study (1-10 years) constituted 13.5% (5) while adolescents (11-18 years) made up 86.5% (32). One (2.7%) recurrence was recorded. Pain was an attribute in 59.5% of the patients and was reported early in the lesion. Tumour fluid aspiration was positive in 83.8% of patients. Straw fluid was the commonest tumour aspirate in 61.3% of patients. Straw-coloured aspirates predominated in children while all the dark-brown aspirates were observed in adolescents.

Conclusion: Ameloblastoma is a common odontogenic tumour in children and adolescents in Enugu. Low recurrence rate, posterior mandibular location, pain and straw-coloured tumour fluids are typical clinical observations.

Key words: Adolescent, Ameloblastoma, Children, Tumour fluid aspirate

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Introduction

Ameloblastoma is a benign epithelial odontogenic tumour characterized by its cystic nature, invasive capability and high recurrence potential.[1] It is the commonest benign odontogenic tumour in Nigeria with a prevalence as high as 71.2%.[2] It is also reported as the most frequent odontogenic tumour in children and adolescents in Ibadan Western Nigeria.[3] On the other hand, some studies reported that ameloblastoma is extremely rare [4], or that it is not seen in the first ten years of life.[5] This could be because age-at-presentation was used as the foundation for identifying and classifying children with ameloblastoma. This strategy could rule out those who were diagnosed with ameloblastoma as children or teenagers but did not present until they were no longer in these age categories. The categorization of young patients as children or adolescents based on age-at-presentation could alter the prevalence values, and affect the aetiopathogenic understanding, clinical features and management considerations. The age-at-onset could be a more representative method of evaluating the prevalence of ameloblastoma in children and adolescents.

Many studies on ameloblastoma have focused more on management of ameloblastoma compared to the clinicopathologic features.[6] Clinicopathologic features such as tumour aspirates, presence or absence of pain,[7] and ulceration have not been properly investigated. Their role could be important in the management and prognostic evaluation of the disease.

There is paucity of reports on ameloblastoma in children and adolescents from southeast of Nigeria. Ameloblastoma in this group of patients presents a special challenge in the management because of the need to offer a conservative treatment that would take into consideration the continuing development and growth of the affected jaws.[7] In addition, to the best of the author's knowledge, no one has investigated ameloblastoma in children and adolescents using the age-at-onset method. Thus, this study focuses on the prevalence of ameloblastoma in children and adolescents in Enugu, South-Eastern Nigeria, using age-at-onset to categorize children and adolescents. It also investigates the rarely reported types of tumour aspirates of ameloblastoma in these age groups, as well as ulceration and pain complaint.

Methodology: This is a retrospective observational study of ameloblastoma in children and adolescent patients at a tertiary health institution in Enugu Eastern Nigeria, from 2012 to 2018. The clinical and histopathology records of all patients diagnosed of ameloblastoma were identified and retrieved from the Departmental archives. The lesions were confirmed as ameloblastoma by histopathological study based on histochemical staining using hematoxylin and eosin. Further clinicopathologic information of each patient was obtained including gender, site of tumour, age at presentation, duration, type of tumour fluid aspirate (straw, dark-brown, serosanguinous, purulent, negative aspirate), presence of ulceration, complaint of pain, histologic type of ameloblastoma diagnosis, and the age-at-onset of tumour. The age-at-onset is the patients' age when the child, adolescent or guardian became aware of occurrence of initial swelling or other associated symptoms before presentation to the clinic. This age-at-onset is obtained by subtracting the duration stated by the patient from the age at clinic presentation.

The data was analyzed with Statistical Product and Service Solutions (SPSS) version 20.0 (SPSS Inc, Chicago, IL). The test of significance was set at $P<0.05$. Categorical variables were analyzed as frequencies and percentages. The frequency tables of categorical variables were determined, while the associations of categorical variables were carried out using chi square tests. Quantitative variables were summarized as means and standard deviation.

All ethical issues were adhered to in accordance with the Declarations of Helsinki's recommendations. Patient's privacy and confidentiality were not compromised.

Result:

A total of 93 cases of ameloblastoma were identified. Thirty-seven children and adolescents (34.4%) were among those diagnosed with ameloblastoma, (Table I). Their age ranged from 6 to 18 years old, with a mean of 13.6 ±2.9 years. Male patients accounted for 15 (40.5%) and female patients accounted for 22 (59.5%), as well as male to female ratio of 0.7:1. In the study, children aged 1 to 10 years made up 13.5% (5) of the patients, while adolescents aged 11 to 18 years accounted for 86.5% (32).

Table II shows the ameloblastoma location based on age groups. The mandible was the most common location for tumors, accounting for 97.3% (36) of all cases. Ameloblastoma in adolescents were mostly seen in the posterior mandible (20) while female subjects had more posteriorly situated lesions. Conventional ameloblastoma was more common 30

(81.1%), while unicystic ameloblastoma occurred in 7 (18.9%) cases as shown in table III.

Table IV shows that pain could be attributed to tumour in 22 (59.5%) subjects. However, female gender accounted for 59.1% (13) of the pain associated with tumour. Pain was reported during the first year of the lesion in 48.6% (18).

Tumour fluid aspiration to exclude cystic and vascular lesions was negative in 6 (16.2%) cases but positive in 31(83.8%) of patients. Majority of the negative aspirations 83.3% (5), occurred in adolescent female patients (Table I). Straw fluid was the commonest aspirate 61.3 % (19), while serosanguinous, and purulent aspirates were infrequent as shown in Table V. Table I showed that most of the dark brown aspirates 62.5% (5) and straw aspirates 47.4% (9) were observed as early as the first year of tumour appearance. Children were observed to yield only straw-coloured aspirates (100%) (Table I). All the observed dark-brown aspirates 25.8% (8) were from tumours in adolescents. Table VI shows that five cases 13.5% (5) presented with ulceration, and mostly among the female gender 80.0% (4).

There were no significant differences in the results, in the associations at 95% confidence intervals. The p values for Tables II, III, IV, & VI were 0.581, 0.196, 0.956 and 0.314 respectively.

Table I: Clinicopathologic Features of ameloblastoma in children and adolescents (n=37)

S/N	Sex	Age-at-onset (years)	Duration (months)	Age-at-Presentation (years)	Tumour fluid Aspirates	Ulceration	Pain	Recurrence	Histologic type
1	M	18	48	22	Dark-brown	Nil	Yes	Nil	Solid
2	F	18	6	18	Nil	Yes	Yes	Nil	Solid
3	F	18	7	18	Dark-brown	Nil	Yes	Nil	Solid
4	F	17	10	18	Straw	Nil	Yes	Nil	Solid
5	M	17	12	18	Nil	Nil	Nil	Nil	Unicystic
6	F	17	5	17	Straw	Nil	Yes	Nil	Solid
7	M	17	5	17	Dark-brown	Nil	Yes	Nil	Solid
8	F	16	24	18	Straw	Nil	Nil	Nil	Solid
9	F	16	24	18	Straw	Nil	Yes	Nil	Solid
10	M	16	4	16	Straw	Nil	Nil	Nil	Solid
11	F	16	6	16	Straw	Nil	Nil	Nil	Solid
12	F	15	168	29	Dark-brown	Nil	Yes	Nil	Solid
13	M	15	84	22	Purulent	Nil	Yes	Nil	Solid
14	M	15	60	20	Straw	Nil	Yes	Nil	Solid
15	M	15	5	15	Dark-brown	Nil	Nil	Nil	Solid
16	F	14	96	22	Dark-brown	Nil	Yes	Nil	Solid
17	F	14	72	20	Straw	Nil	Yes	Nil	Solid
18	F	14	36	17	Nil	Nil	Nil	Nil	Solid
19	F	13	36	17	Purulent	Nil	Yes	Nil	Solid
20	F	13	36	16	Nil	Nil	Yes	Nil	Solid
21	F	13	1	14	Serosanguinous	Nil	Nil	Nil	Solid
22	F	13	4	13	Straw	Nil	Yes	Nil	Solid
23	M	13	6	13	Straw	Nil	Yes	Nil	Solid
24	M	13	6	13	Straw	Nil	Yes	Nil	Unicystic
25	M	12	120	22	Straw	Yes	Yes	Nil	Solid
26	F	12	120	22	Straw	Yes	Nil	Nil	Solid
27	M	12	24	14	Straw	Nil	Nil	Nil	Unicystic
28	F	12	9	13	Dark-brown	Nil	Nil	Nil	Unicystic
29	F	12	6	12	Nil	Nil	Nil	Nil	Solid
30	M	11	84	18	Purulent	Nil	Nil	Nil	Solid
31	M	11	36	14	Straw	Nil	Yes	Nil	Solid
32	F	11	9	12	Dark-brown	Nil	Nil	Nil	Unicystic
33	M	10	36	13	Straw	Nil	Nil	Nil	Solid

34	M	10	6	11	Straw	Nil	Yes	Nil	Unicystic
35	F	9	156	22	Straw	Yes	Yes	Yes	Solid
36	F	9	72	15	Nil	Yes	Nil	Nil	Solid
37	F	6	4	6	Straw	Nil	Yes	Nil	Unicystic

F: Female; M: Male.

Table II: Ameloblastoma location based on age groups

Site	Children (0-10yrs)	Adolescent (11-18yrs)	Total (%)
Mandible Anterior	3	11	**14** (37.8%)
Mandible Posterior	2	20	**22** (59.5%)
Maxilla Posterior	0	1	**1** (2.7%)
Total	5	32	**37 (100%)**

p value = 0.581

Table III. Histologic types of ameloblastoma in children and adolescent

	Child (0-10 years)	Adolescent (11-18 years)	Total n (%)
Solid/multicystic	3	27	30 (81.1)
Unicystic	2	5	7 (18.9)
Total	**5**	**32**	**37**

p value = 0.196

Table IV. Association of pain attributed to tumour based on gender

	Yes	NO
Male	9 (40.91%)	6
Female	13 (59.1%)	9
Total	22	15

p value = 0.956

Table V: Types and prevalence of aspirates of ameloblastoma in children and adolescents (n=31)

Type of Tumour Fluid Aspirate	Frequency (n)	% (n=31)
Straw	19	61.3
Serosanguinous	1	3.2
Dark-brown	8	25.8
Purulent	3	9.7
Total	**31**	**100.0**

Table VI. Presence of ulceration

	Presence of Ulceration		
	Yes	No	Total
Male	1	14	15
Female	4	18	22
Total	5	32	37

p value = 0.314

DISCUSSION

Many earlier Nigerian studies[1,2,8,4,9] may have reported the prevalence of ameloblastoma in children and adolescents based on age-at-presentation in place of the estimated age-at-onset. Since the duration of tumours often run into years before clinical presentation, ameloblastoma which commenced in children and adolescents were not considered when the age at presentation crossed the adolescent age. Age based studies in children and adolescents would need to use a methodology that assigns several children and adolescents with ages that reflect their true age-at-onset of the swelling. The age-at-onset of ameloblastoma employed in this study is more representative in determining the estimated period of the lesion manifestation and the age distribution pattern in children and adolescents.

The observed minimum age of occurrence at six years in this study was low and comparable to the four years reported in Lagos Nigeria by Ajayi et al.[10] A lower age of occurrence of plexiform unicystic ameloblastoma in a 3-year-old child was reported by Chaudhary et al.[11] while the least known age of occurrence was reported by Soumithran et al. [12] of mural unicystic ameloblastoma in a one-year-old child. Okechi et al.[4] did not observe ameloblastoma in children less than 10 years of age in their study probably because it relied on the age-at presentation instead of age-at-onset. The findings of this study (13.5%), as well as those of Arotiba et al.[1] (2.5%), Okoh et al.[2] (9.3%), Iyogun et al.[9] (5.6%), and Ajayi et al.[10] (8.5%), refute the concept that ameloblastoma is uncommon before the age of ten years. In a review of 233 cases of ameloblastoma in children and adolescents within the age range 4 years to 20 years, Zing et al. [13] observed a mean age of 14.5 years which is comparable to the mean age of 13.6 years obtained in this study. Similarly, Bansal et al. [14] studied 39 childhood/adolescent (15.2%) cases of ameloblastoma and reported a mean age of 13.6 years.

The ameloblastoma prevalence of 34.4% is within the upper value of the range of 14% to 35% reported in other Nigerian studies of children and adolescents.[1,2,8,4,10] An Indian study by Bansal et al.[13] reported 15.2% prevalence for patients below 18 years of age. It specifically observed that 17.9% were children less than 10 years while 82.1% were aged 11-18 years.

The prevalence of 18.9% for unicystic ameloblastoma obtained in the present study from Enugu, is comparable to the value for unicystic ameloblastoma reported by Ajayi et al.[10] and Arotiba et al.[1] in Lagos but differs from the 39.1% reported by Lawal et al.[3] in Ibadan. The reason for these varied results may be due to number of cases studied, duration of the study and the methodology.

The high female frequency in the present study, which is not statistically significant, contrasts with previous Nigerian studies that reported male predominance of occurrence.[1,3,10,14]

The mandible was the most common location for tumours in 97.3% of all cases and similar to that observed in a review of 233 cases of ameloblastoma in children and adolescents.[13] The posterior mandible which was the most frequent site of involvement in this study is similar to observations from other studies.[4,14] However, Arotiba et al.[1] reported anterior involvement (57.3%) as the most common mandibular site in a southwest Nigerian study. The site of ameloblastoma could have a prognostic relevance because it makes the tumour easily accessible for excision and without interfering with jaw development.

Ameloblastoma could be considered a painful tumour in children due to a high frequency of complaint of pain (59.5%) observed in children and adolescents in this series. Lower rates of pain occurrence (29%) were reported by Cadavid et al.[15] although in an adult population. Some cases of ameloblastoma have also been observed to be asymptomatic.[16] However, in this study, more children and adolescent patients presented within the first year of tumour appearance probably because parents responded quicker when their wards have pain or swelling.

The ameloblastoma fluid aspirates obtained during clinical examination of children and adolescents showed predominance of straw-coloured aspirates similar to those obtained in a cystic odontogenic lesion like dentigerous cysts.[17] This could suggest lowered aggressive potential in ameloblastomas with straw aspirates. This opinion would need evaluation by further study to demonstrate any statistical association of any ameloblastoma aspirate with aggressive biological activity. The low rate of recurrence (2.7%, 1/37) observed in this series was probably due to the short period covered by this study, since ameloblastoma has been known to recur after decades.[18]

CONCLUSION

Ameloblastoma is a common odontogenic tumor among children and adolescent in Enugu,

southeastern Nigeria. Some of the clinical features include a posterior mandibular position, pain, straw-colored tumor aspirates, and a low recurrence rate. In categorizing children and adolescents with ameloblastoma, the age-at-onset is more representative.

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