

ODONTOGENIC TUMOURS IN CHILDREN AND ADOLESCENTS: A REVIEW OF FORTY-EIGHT CASES

A.O. Lawal¹, A.O. Adisa¹ and B.O. Popoola²

1. Department of Oral Pathology, College of Medicine, University of Ibadan, Nigeria
2. Department of Child Oral Health, College of Medicine, University of Ibadan, Nigeria

Correspondence:

Dr. A.O. Lawal

Department of Oral Pathology,
College of Medicine,
University of Ibadan, Ibadan,
Nigeria.

E-mail: lawaloluwatoyin@gmail.com

Tel: +2348055133964

Fax: +23422411768

SUMMARY

Objectives: Odontogenic tumours comprise a large heterogeneous group of lesions originating from odontogenic epithelium and/or ectomesenchyme and its vestiges. The aim of this study was to analyze odontogenic tumours in children and adolescents seen at a tertiary institution in South Western Nigeria and compare with results from previous studies.

Material and Methods: Archival records of the Department of Oral Pathology, University College Hospital Ibadan were reviewed. All histologically diagnosed odontogenic tumours in patients 19 years and below spanning a period of 21 years (1990-2011) were retrieved. Data regarding age, gender, and tumor topography were analyzed using SPSS for Window (version 18.0; SPSS Inc. Chicago, IL)

Results: One hundred and forty seven jaw swellings were seen in children and adolescents aged 19 or less during the study period, out of which 48 (32.7%) were odontogenic tumours. More cases were seen in males than females with a male: female ratio of 7:5. The mandible was the commonest site of occurrence with mandible: maxilla ratio of 11:4. Ameloblastoma was the commonest odontogenic tumours with 14 (29.1%) solid ameloblastoma and 9 (18.8%) cystic ameloblastoma cases followed by fibromyxoma with 8 (16.7%) cases. calcifying epithelial odontogenic tumour, calcifying cystic odontogenic tumour and odontogenic fibroma were occasionally seen.

Conclusion: This study showed that ameloblastoma was the most common odontogenic tumour in children and no case of odontoma was seen. Odontogenic tumours in children and adolescents may not be as rare as previously reported by some authors and inclusion of keratocystic odontogenic tumour in this study slightly affected the relative incidence of odontogenic tumors in children and adolescents.

Keywords: Adolescents, Children, Odontogenic tumours

INTRODUCTION

Odontogenic tumours comprise a large heterogeneous group of lesions originating from odontogenic epithelium and or ectomesenchyme and its vestiges.¹ Odontogenic tumours include entities of hamartomatous nature (for example, odontoma), benign neoplasms, some of which are aggressive (for example, ameloblastoma and myxoma) and malignant neoplasms capable of metastasis.^{2, 3} Odontogenic tumours accounted for between 1% and 28% of oral lesions from various studies depending on the criteria and categories that were used for comparison.⁴ Odontogenic keratocysts were recently included in the WHO classification of odontogenic tumours as

keratocystic odontogenic tumours based on its clinical behaviour, genetic and molecular features.⁵

Previous reports examined odontogenic tumours in children as part of oral tumors^{6,7,8} or studied individual tumors such as ameloblastomas⁹ and adenomatoid odontogenic tumour (AOT)¹⁰, but few have examined Odontogenic tumours generally in children. Reports by Ajayi *et al*¹ and Adebayo *et al*² did not include keratocystic odontogenic tumour (KOT) because as at the time of carrying out their studies, KOT was still regarded as a cystic lesion. The aim of this study was to analyze Odontogenic tumour cases in children and

adolescents seen at a tertiary institution in South-Western Nigeria and compare with results from previous studies.

MATERIAL AND METHODS

Archival records of the Department of Oral Pathology, University College Hospital Ibadan, Nigeria were reviewed. All histologically diagnosed odontogenic tumours in patients 19 years and below spanning a period of 21 years (1990 - 2011) were retrieved. Data regarding age, gender, and tumor topography were analyzed using SPSS for Window (version 18.0; SPSS Inc. Chicago, IL)

RESULTS

One hundred and forty-seven neoplasms were seen in children and adolescents aged 19 or less during the study period, out of which 48 (32.7%) were odontogenic tumours. The peak age of occurrence was 19 years with age range between 3 to 19 years and mean age of 15.3 (± 3.3) years. Odontogenic tumours in children occurred more in males than females with a male: female ratio of 7:5. The mandible

was the commonest site of occurrence with mandible: maxilla ratio of 11:4 and just one case was located in the soft tissue. Ameloblastoma was the commonest odontogenic tumour in children with 14 (29.1%) solid ameloblastoma and 9 (18.8%) cystic ameloblastoma cases, followed by eight (16.7%) cases of fibromyxoma. Four (8.3%) cases each of ameloblastic fibroma (AF), KOT and AOT were diagnosed in this age group. (Table 1)

Ameloblastoma

Ameloblastoma was the most common odontogenic tumour in this series. A total of 23 ameloblastoma cases were seen, consisting of 14 solid and 9 cystic ameloblastomas. The solid ameloblastoma had a mean age of 15.4 (± 3.4) years (range 8-18 years), a male to female ratio of 11:3 and an exclusive mandibular occurrence. The cystic ameloblastoma on the other hand, had a mean age of 16.6 (± 2.1) years (range 13-19 years), it affected more females than males (male to female ratio 3:6) and had a predilection for the mandible (Mandible to maxilla ratio = 7:2).

Table 1: Demography of odontogenic tumors in children and adolescents

Types of odontogenic tumour	N (%)	Age		Site			Gender	
		Mean age	Age range	Mandible	maxilla	Soft tissue	Male	female
Solid								
Ameloblastoma	14(29.1)	15.36	8-18	14	0	0	11	3
Cystic ameloblastoma	9 (18.8)	16.56	13-19	7	2	0	3	6
Fibromyxoma	8 (16.7)	14.75	3-19	4	4	0	6	2
Ameloblastic fibroma	4(8.3)	16.00	14-19	4	0	0	4	0
Keratocystic odontogenic tumor	4 (8.3)	12.75	10-16	3	1	0	1	3
Adenomatoid odontogenic tumor	4 (8.3)	15.00	12-18	0	4	0	2	2
Calcifying odontogenic cyst	2(4.2)	15.00	13-17	2	0	0	0	2
Calcifying epithelial odontogenic tumor	2(4.2)	16.50	16-17	1	1	0	1	1
Odontogenic fibroma	1(2.1)	13.00	13	0	0	1	0	1

Fibromyxoma

Fibromyxoma was the second most common lesion with a mean age of occurrence of 14.8 (± 5.5) years (range 3-19 years). There was a male preponderance with male to female ratio of 6:2 but an equal site distribution of 4:4.

Ameloblastic fibroma

The mean age of occurrence for AF was 16.0 (± 2.5) years (range 14-19 years). The lesion was seen only in males and occurred exclusively in the mandible.

AOT

The age range for AOT was 12-18 years with mean age of 15.0 (± 2.5) years. The lesion had no sex preponderance in this series (male to female ratio 2:2). All four cases were seen in the maxilla.

KOT

The four cases of KOT seen had a mean age of 12.75 (± 2.50) years (range 10-16 years). KOT was seen more in females with a male to female ratio of 1:3 and had a mandibular predilection (mandible: maxilla ratio 3:1).

CEOT/CCOC

Two cases of CEOT were seen with a mean age of 16.5 (± 0.7) (range 16-17). The lesion had no sex or site predilection. (Table 1) CCOC occurred exclusively in females and was only seen in the mandible. The mean age was 15 (± 2.83) years (range 13-17 years).

OF

The only case of odontogenic fibroma was seen in a 13-year-old female and it was also the only case of extraosseous tumour in this study.

DISCUSSION

Previous reports found odontogenic tumours to be rare in children and adolescents.¹² Comparing results regarding the relative prevalence of odontogenic tumours in children with previous studies may be difficult because different studies used dissimilar ages to categorize odontogenic tumours in children and these groups were varied over a wide spectrum of oro-facial lesions.^{12,13,14} Odontogenic tumours in children in this series represented 32.7% of the total number of jaw neoplasms in children and adolescents. Previous publications of odontogenic tumours in children by Adebayo *et al*⁶ in Kaduna, Arotiba⁷ in Lagos, and Ulmansky¹⁴ in Israel reported 25.9%, 18.4% and 9.5% respectively. Our finding was however similar to results in studies that included both children and adult population with Adebayo¹⁵ in Kaduna and Arotiba *et al*⁶ in Ibadan reporting a relative occurrence of odontogenic tumours of 32% and 30% respectively. The relatively higher occurrence of

odontogenic tumours in children in this study may be due to the addition of KOT which accounted for 8.3% of cases. Odontogenic tumours in children in this series, had predilection for the mandible (68.8%) which was in agreement with most other studies^{6,11}, though, a study in Jordan⁸ found more (64%) cases in the maxilla. Most studies were in agreement with our finding of male predilection^{6,11,12} except for Ulmansky¹⁴ who reported a female preponderance.

Ameloblastoma was the most common odontogenic tumour in children and adolescents with a total of 48% of cases. Previous studies report ameloblastoma to be the most common odontogenic tumour both in general population and in children; Ajayi *et al*¹¹ in Lagos reported that ameloblastoma constituted 48.9% of odontogenic tumours in children while Adebayo *et al*² in Kaduna reported a slightly higher figure of 54% of odontogenic tumours in their series. Other authors found odontomas to be the commonest odontogenic tumours in children and adolescents with Guerrisi⁴ in Argentina reporting 50.9% of odontogenic tumours as odontomas while ameloblastoma accounted for just 18.3% of odontogenic tumours in their series. Although unicystic ameloblastoma is said to be more commonly seen in the second and third decades of life¹⁷, the solid ameloblastoma (29.2%) was more common than the unicystic type (18.8%) but this was higher than the 11% reported by Ajayi *et al*.¹¹

Fibromyxoma with 16.7% of cases was the second most common odontogenic tumour in this series. Adebayo *et al*⁶ also found fibromyxoma to be the second commonest odontogenic tumour in children with 19% of cases in their series but Ajayi *et al*¹¹ and Guerrisi *et al*⁴ reported lower figures of 8.7% and 8.5% respectively. Fibromyxoma was however reported to be the third most common odontogenic tumour in the series by Ajayi *et al* and was the commonest odontogenic tumour as reported by Ulmansky *et al*.¹⁴

The male predominance in this series is at variance with most previous studies which showed a female predilection^{11,12,14} but the equal mandible/maxilla distribution in this study, was corroborated by other studies.^{11,12,14}

Ameloblastic fibroma accounted for 8.3% of odontogenic tumours in this series. Other studies have reported between 1% and 8% of odontogenic tumours to be AF^{11,12}. The male and mandibular predilection is in agreement with most other reports except for Ajayi *et al*¹¹ who found a female predominance.

We found 4 (8.3%) cases of KOT in this study; previous publications did not include KOT in their reports^{11,12,15,16} either because they used older WHO classifications of oral tumors that considered KOT as a cystic lesion, or as Guerrisi *et al*⁴ claimed, for easy comparison with other studies. The finding that KOT affected more female than males is at variance with previous studies.¹⁸ AOT was the second and third most common odontogenic tumors in children in previous Nigerian studies from Lagos¹¹ and Kaduna⁶ constituting 19.6% and 9% in their respective studies. This was slightly different in this study with AOT accounting for 8.6% of cases. AOT was exclusively seen in the maxilla in this series; which was in keeping with several other studies.^{6,10,11,14}

Only one case of OF was diagnosed in children in this series. Adebayo *et al*⁶ did not report any case of OF in their study while Arotiba in Ibadan¹⁶ reported only two in their series. In contrast, Arotiba in Lagos⁷ and Guerrisi in Argentina⁴ reported four and five cases respectively.

Odontomas and malignant odontogenic tumours were not seen in this study which was in conformity with previous studies from Nigeria which either found few or no cases^{6,11,16} but at variance with some other studies that showed odontomas to be the commonest odontogenic tumours in their series¹⁴. Odontomas may be under reported, as it is usually asymptomatic and frequently diagnosed by routine panoramic radiographic examination that is not yet a routine in most clinical settings in Nigeria.

This study reviewed odontogenic tumours in children and adolescents using the WHO 2005 classification with results conforming relatively to the demography of the different tumor types in previous publications. The inclusion of KOT in this series appears to have affected the relative occurrence odontogenic tumours compared to previous studies but the demography of odontogenic tumours were essentially similar to those of previous studies. We conclude that odontogenic tumours in children and adolescents may not be as rare as previously reported by some authors, though, the higher age limit considered and the inclusion of KOT in this series may have accounted for the relatively higher percentage of odontogenic tumours in children in this study.

REFERENCES

1. **Ladeinde AP**, Ajayi OF, Ogunlewe MO *et al*. A review of 319 cases in a Nigerian teaching hospital. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005; 99: 191-195.

2. **Philipsen HP**, Reichart PA. Revision of the 1992 edition of the WHO histological typing of odontogenic tumors. A suggestion. *J Oral Pathol Med* 2002; 31: 253-258.
3. **Mosqueda Taylor A**, Meneses García A, Ruíz Godoy Rivera LM *et al*. Malignant odontogenic tumors. A retrospective and collaborative study of seven cases. *Med Oral* 2003; 8: 110-121.
4. **Guerrisi M**, Piloni MJ, Keszler A. Odontogenic tumors in children and adolescents. A 15-year retrospective study in Argentina. *Med Oral Patol Oral Cir Bucal* 2007; 12: E180-185.
5. **Philipsen HP**. Keratocystic odontogenic tumor. En: Barnes L, Eve-son JW, Reichart, Sidransky D, eds. WHO classification of tumors. Pathology and genetics of head and neck tumors. Lyon: IARC Press; 2005; p. 306-307
6. **Adebayo E T**, Ajike S O, Adekeye E O. Tumors and tumor-like lesions of the oral and perioral structures of Nigerian children. *Int. J. Oral Maxillofac. Surg.* 2001; 30: 205–208.
7. **Arotiba GT**. A study of orofacial tumors in Nigerian children. *J Oral Maxillofac Surg* 1996; 54:34-38.
8. **Al-Khateeb T**, AL Hadi Hamasha AA, Almasri NM. Oral and maxillofacial tumors in North Jordanian children and adolescents: a retrospective analysis over 10 years, *Int. J. Oral Maxillofac. Surg.* 2003; 32: 78–83
9. **Arotiba GT**, Ladeinde AL, Arotiba JT *et al*. Ameloblastoma in Nigerian children and adolescents: a review of 79 cases. *J Oral Maxillofac Surg* 2005; 63:747-751.
10. **Arotiba GT**, Arotiba JT, Olaitan AA, Ajayi OF. The adenomatoid odontogenic tumor: an analysis of 57 cases in a black African population. *J Oral Maxillofac Surg.* 1997; 55: 146-148.
11. **Ajayi OF**, Ladeinde AL, Adeyemo WL, Ogunlewe MO. Odontogenic tumors in Nigerian children and adolescents- a retrospective study of 92 cases. *World J Surg Oncol.* 2004 Nov 27; 2:39.
12. **Adebayo ET**, Ajike SO, Adekeye EO. Odontogenic tumors in children and adolescents: a study of 78 Nigerian cases. *J Craniomaxillofac Surg.* 2002; 30: 267-272.
13. **Asamoia EO**, Ayanlere AO, Olaitan AA, Adekeye EO: Paediatric tumors of the jaws in Northern Nigeria. *J Cranio Maxillofac Surg.* 1990; 18: 130–135.
14. **Ulmansky M**, Lustmann J, Balkin N: Tumors and tumor-like lesions of the oral cavity and related structures in Israeli children. *Int J Oral Maxillofac Surg.* 1999; 28: 291–294.

15. **Adebayo ET**, Ajike SO, Adekeye EO. A review of 318 odontogenic tumors in Kaduna, Nigeria. *J Oral Maxillofac Surg* 2005; 63: 811-819.
16. **Arotiba JT**, Ogunbiyi JO. Obiechina, AE. Odontogenic tumors: a 15-year review from Ibadan, Nigeria *Br J Oral Maxillofac Surg*. 1997; 35: 363-367.
17. **Rosenstein T**, Pogrel MA, Smith RA, Regezi JA. Cystic ameloblastoma-behavior and treatment of 21 cases. *J Oral Maxillofac Surg*. 2001; 59: 1311-1316.
18. **El-Hajj G**, Anneroth G. Odontogenic keratocysts - a retrospective clinical and histologic study. *Int J Oral Maxillofac Surg*. 1996; 25: 124-129.