# An Audit of Childhood Orofacial Tumours Seen in a Tertiary Hospital in South-South Nigeria

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# ABSTRACT

**Background:** Several orofacial tumours affect children, and these tumours pose a major health challenge due to the associated morbidity and mortality, mostly with malignant tumours.

**Objective**: To determine the clinico-pathologic pattern of childhood orofacial tumours seen in a tertiary health institution in South-south Nigeria.

**Method**: A cross-sectional retrospective study from the histopathology archives of the Department of Oral and Maxillofacial Pathology and Medicine, University of Benin Teaching Hospital (UBTH), Benin City, over 15 years (2008–2022). All the tumours of the orofacial region histopathologically diagnosed in patients 16 years and below were assessed and reviewed.

Results: A total of 105 cases were included. There were 61 males (58.1%) and 44 females (41.9), giving a male-to-female ratio of 1.4:1. The age range was 2 to 16 years with a mean age of 9.55+3.973 years. The peak age of the tumours was observed in the 13 to 16 years age groups (n=31, 29.5%). The mandible (n=43, 41.0%) was the site most commonly affected, followed by the maxilla (n=18, 17.1%). The lymphomas were the most common tumours observed (n=34, 32.4%), consisting mostly of Burkitt's lymphoma (n=22, 21.1%). The lymphomas were mostly seen among the 5 to 8 years age group (n=19, 55.9%), with 73.5% of them occurring more in males (n=25) and affecting the mandible (n=13, 38.2%) mostly. Conclusion: This study shows that orofacial tumours in children are diverse, and the most prevalent were Lymphomas, especially the Burkitt's type. A good understanding of these tumours in children by clinicians would assist with the timely identification of cases and actions instituted to adequately manage them.

**Keywords:** Childhood, Orofacial, tumours, histopathological diagnosis, Benin City

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# INTRODUCTION

Orofacial tumours affect any age, including children.<sup>1</sup> These tumours pose a major health problem due to the increasing morbidity and mortality associated with the tumours, especially the malignant tumours.<sup>1,2</sup> Orofacial tumours are heterogeneous groups of pathologic disorders with various histologic types and clinical behaviour.<sup>2,3,4</sup> They affect hard and soft tissues of the oral and maxillofacial region.<sup>5,6</sup>

In Sub-Saharan Africa, these tumours and their management constitute a major health challenge resulting from the late presentation of patients at advanced stages of the tumours and the low socioeconomic status.<sup>2,5</sup> These tumours result in the destruction of facial bone with compromise to the airway and digestive system and adjacent structures when neglected.<sup>4</sup> Various reports within and outside Nigeria have discussed the frequency, clinical presentation, histopathological characteristics and management of orofacial tumours and tumour-like lesions in children.<sup>1,3,7,8,9</sup> In our environment, an earlier study by Omoregie and Akpata,7 reported a prevalence of 13.5% of orofacial tumours in children. This present study is aimed at auditing the clinicopathologic pattern of childhood orofacial tumours seen in a tertiary health institution in South-south Nigeria.

#### **METHODS**

A cross-sectional retrospective record review from the histopathology archives of the Department of Oral and Maxillofacial Pathology and Medicine of the University of Benin Teaching Hospital (UBTH), Benin City, Nigeria, over 15 years (2008-2022). All the tumours of the orofacial region histopathologically diagnosed in patients 16 years and below were assessed and reviewed. Cases that had complete clinical and histopathological data were included, while those without were excluded. Also, reactive lesions were excluded. The tumours were grouped using four-year intervals among the age groups: 0-4 years, 5-8 years, 9-12 years and 13-16 years. Data such as age, gender, site and histopathologic diagnosis were entered into IBM SPSS 21 and analysed. Data was summarised with frequencies, mean and standard deviation.

# RESULTS

A total of 1,525 histopathologically diagnosed orofacial lesions were seen within the study period,

of which 118 were orofacial tumours in children, constituting 7.7% of all the lesions overall. Of the 118 cases of tumours in children, 105 cases had complete data and were included in this review. From the 105 cases included in this study, there were 61 males and 44 females, giving a male-to-female ratio of 1.4:1. The age range was 2 to 16 years, with a mean age of  $9.55\pm3.97$  years. The peak age of the tumours was observed in the 13 to 16 years age group 31(29.5%), with the highest frequency in patients that were 14 years old 13(12.4%). The mandible 43(41.0%) was the site most commonly affected, followed by the maxilla 18(17.1%) [Table 1]. Seven tumours affected more than one site (Multiple site lesions), constituting 6.7% of the cases.

There were 57 benign tumours (54.3%) and 48 malignant lesions (45.7%) [Table 2]. The Lymphomas 34(32.4%) were the most common tumours observed, consisting mostly of Burkitt's lymphoma 22(21.1%) [Figure 1 and 2]. The second most common tumour was ameloblastoma 26(24.8%). The lymphomas were seen mostly among the 5 to 8 years age group 19(55.9%), with 73.5% occurring in males (n=25) and affecting mostly the mandible 13(38.2%).

#### Table 1 Site distribution of the orofacial tumours

Site	Frequency n (%)
Cheek	3 (2.9)
Face	9 (8.6)
Gingiva	8 (7.6)
Lip	3 (2.9)
Mandible	43 (41.0)
Multiple sites	7 (6.7)
Maxilla	18 (17.1)
Oropharynx	1(1.0)
Palate	5 (4.8)
Parotid region	4 (3.8)
Submandibular	4 (3.8)
Total	105 (100.0)

Table 2. Frequency of the orofacial tumours

Histopathologic diagnosis	Frequency n (%)		
Malignant Salivary Gland tumours (Adenocarcinomas)	3 (2.9)		
Adenoma (Pleomorphic Salivary Adenoma)	4 (3.8)		
Ameloblastoma	26 (24.8)		
Adenomatoid Odontogenic tumour	5 (4.8)		
Calcifying Cystic Odontogenic tumour	1 (1.0)		
Central Giant Cell Granuloma	5 (4.8)		
Chondrosarcoma	2 (1.9)		
Haemangioma	4 (3.8)		
Langerhans Cell Histiocytosis	1(1.0)		
Lipoma	2 (1.9)		
Lymphoma	34 (32.4)		
Lymphangioma	5 (4.8)		
Malignant Fibrous Histiocytoma	3 (2.9)		
Neurofibroma	1(1.0)		
Peripheral Ossifying Fibroma	2 (1.9)		
Rhabdomyosarcoma	1(1.0)		
Round Blue Cell tumour	1(1.0)		
Squamous Cell Carcinoma	4 (3.8)		
Wegener's Granulomatosis	1 (1.0)		
Total	105 (100)		



Fig. 1. Burkitt's lymphoma is characterised by progressive orofacial swelling in a male child involving the upper and lower jaws



Fig. 2 Burkitt's lymphoma. Histopathologic photomicrograph showing malignant lymphocytes (black arrow) interspersed by pale staining macrophages (blue arrow), giving a starry sky appearance (H & E x100).

Table 3. Relationship between the histopathologically diagnosed orofacial childhood tumours among the age groups

		Total			
Histopathologic diagnosis	0-4	5 – 8	9–12	13 - 16	
Malignant Salivary Gland	2(66.7%)	-	1(33.3%)	-	3
tumours (Adenocarcinomas)					
Adenoma (Pleomorphic	-	-	2(50.0%)	2(50.0%)	4
Salivary Adenoma)					
Ameloblastoma	-	3(11.5%)	10(38.5%)	13(50.5%)	26

Adenomatoid Odontogenic	-	-	1(20.0%)	4(80.0%)	5	
tumour						
Calcifying Cystic Odontogenic	-	-	-	1(100%)	1	
tumour						
Central Giant Cell Granuloma	-	3(60.0%)	-	2(40.0%)	5	
						org
Chondrosarcoma	-	-	-	2(100%)	2	d.6
Haemangioma	1(25.5%)	2(50.0%)	-	1(25.5%)	4	hap
Langerhans Cell Histiocytosis	-	-	1(100%)	-	1	ę.
Lipoma	1(50.0%)	-	-	1(50.0%)	2	þdį
Lymphoma	7(20.6%)	19(55.9%)	7(20.6%)	1(2.9%)	34	×.
Lymphangioma	2(40.0%)	-	3(60.0%)	-	5	
Malignant Fibrous	-	1(33.3%)	1(33.3%)	1(33.3%)	3	
Histiocytoma						
Neurofibroma	1 (100%)	-	-	-	1	
Peripheral Ossifying Fibroma	-	-	1(50.0%)	1(50.0%)	2	
, , , ,						
Rhabdomyosarcoma	-	1(100%)	-	-	1	
Round Blue Cell tumour	-	1(100%)	-	-	1	
Squamous Cell Carcinoma	-	-	3(75.0%)	1(25.0%)	4	
Wegener's Granulomatosis	-	-	-	1(100%)	1	

# Table 4. Relationship of the histopathological diagnosis of the orofacial tumours with gender

Histopathologic diagnosis	Gender	
	Male n, %	Female n, %
Malignant Salivary Gland tumours (Adenocarcinomas)	-	3 (100)
Adenoma (Pleomorphic Salivary Adenoma)	1(25.0)	3 (75.0)
Ameloblastoma	20 (76.9)	6 (23.1)
Adenomatoid Odontogenic tumour	-	5 (100)
Calcifying Cystic Odontogenic tumour	-	1 (100)
Central Giant Cell Granuloma	3 (60)	2 (40.0)
Chondrosarcoma	1(50.0)	1 (50.0)
Haemangioma	3 (75.0)	1 (25.0)
Langerhans Cell Histiocytosis	1 (100)	-
Lipoma	-	2 (100)
Lymphoma	25 (73.5)	9 (26.5)
Lymphangioma	3 (60.0)	2 (40.0)
Malignant Fibrous Histiocytoma	-	3 (100)
Neurofibroma	1 (100)	-
Peripheral Ossifying Fibroma	-	2 (100)
Rhabdomyosarcoma	1 (100)	-
Round Blue Cell tumour	-	1 (100)
Squamous Cell Carcinoma	2 (50.0)	2 (50.0)
Wegener's Granulomatosis	-	1 (100)

Histopathologic diagnosis	Anatomical sites n(%)										
j i i	Cheek	Face	Gingi	Lip	Mand	Maxi	Orop	Palate	Paroti	Subma	Multipl
	n (%)	n (%)	n (%)	n (%	n (%)	n (%)	har n (%)	n (%)	d n (%)	n n (%)	e sites n (%)
MSGT(Adenocarci nomas)	1(33.3	-	-	-	1(33.3	-		-	-	1(33.	-napc
Adenoma (PSA)	1(25.5	1(25.5	-	-	-	-		1(25.5	1(25.5	-	- d
Ameloblastoma	-	1(3.8)	2(7.7	-	19(73.1	1(3.8	-	1(3.8	1(3.8	1(3.8	-   <u>2</u>
AOT	-	-	-	-	1(20.0	4(80.0	-	-	-	-	-
Calcifying Cystic Odontogenic tumour	-	1(100	-	-	-	-	-	-	-	-	-
Central Giant Cell Granuloma	-	-	-	-	4(80)	(20	-	-	-	-	-
Chondrosarcoma	-	-	-	1(50	-	-	-	-	1(50	-	-
Haemangioma	-	1(25	-	-	2(50	-	-	1(25	-	-	-
Langerhans Cell Histiocytosis	-	-	-	-	-	1(100	-	-	-	-	-
Lipoma	-	-	-	-	-	1(50	-	-	-	1(50	-
Lymphoma	1(2.9	5(147	1(2.9	-	13(38.2	-		-	1(20	1(20	-
Lymphangioma	-	-	-	2(40	1(20	-	-	-	1(20	1(20	-
Malignant Fibrous Histiocytoma	-	-	-	-	-	2(66.7	-	1(33.3	-	-	-
Neurofibroma	-	-	-	-	-	-	-	-	-	-	1(100
Peripheral Ossifying Fibroma	-	-	2(100	-	-	-	-	-	-	-	-
Rhabdomyosarco- ma	-	-	1(100								
Round Blue Cell tumour	-	-	-	-	1(100	-	-	-	-	-	-
Squamous Cell Carcinoma	-	-	2(50	-	1(25	1(25	-	-	-	-	-
Wegener's Granulomatosis	-	-	-	-	-	-	1(100	-	-	-	-

Table 5. Relationship of the histopathologically diagnosed orofacial childhood tumours with site

MGST – Malignant Salivary Gland Tumour, PAS – Pleomorphic Salivary Adenoma, AOT – Adenomatoid Odontogenic Tumour, Gin – Gingiva, Man – Mandible, Max – Maxilla, Pal – Palate, Par – Parotid, Subman – Submandibular

## DISCUSSION

An understanding of the clinico-pathologic pattern of orofacial tumours in children is vital for early diagnosis, treatment planning and overall management of cases when they present. This audit is to help raise awareness among Dental surgeons, especially the Paedodontist and other clinicians that these tumours are seen in children in our environment. When the children present with orofacial lesions, the differential diagnosis should be expanded. More so, their caregivers should be motivated to ensure that patients carry out all the necessary investigations early enough to enable early diagnosis and early treatment.

The prevalence of Orofacial tumours in children in this present study (7.73%) among all the lesions

histopathologically diagnosed among all ages within the study period in our environment is low. This finding is similar to the 7.15% seen among the total number of biopsies in the study by Ulmansky<sup>10</sup> in Israel but lower than the earlier report of 13.5% in our environment by Omoregie and Akpata,<sup>7</sup> the 20.8% reported by Orikpete et al.<sup>8</sup> in their 11-year study of all the biopsied lesions, the 19.3% reported by Arotiba,<sup>9</sup> and the 33.9% reported by Taiwo et al.<sup>4</sup> This lower prevalence in our study compared to Taiwo et al.<sup>4</sup> and Arotiba<sup>9</sup> may be due to their reporting of the prevalence of the childhood orofacial tumours among tumours only whereas our study looked at the prevalence of the childhood orofacial tumours across all the lesions (tumours and non-tumours) within the study period. Several studies have reported male preponderance.<sup>1,4,11</sup>

The male-to-female (M: F) ratio in this study is similar to Aregbashola et al.,<sup>11</sup> who reported M: F ratio of 1.4:1 and Taiwo et al.,<sup>4</sup> who observed a M: F ratio of 1.5:1 but it in contrast to Braimah et al.,<sup>5</sup> who reported a slight female preponderance at a M: F ratio of 1:1.08. The mean age in this present study is similar to the earlier report of  $9\pm4.3$  years reported in our environment<sup>7</sup> but slightly younger than the mean age of 10.4 ± 4.1 years reported by Orikpete et al.<sup>8</sup> Similar to previous studies<sup>7,8</sup> the mandible is the most frequent site in our review whereas Akhiwu et al.,<sup>12</sup> observed the maxilla as the most common site in their study.

Lymphomas were the most common tumours observed, and Burkitt Lymphoma was the most common type of lymphoma in this study. This agrees with previous studies.<sup>7,11,12</sup> This is not unexpected because Burkitt's lymphoma is endemic in sub-Saharan Africa affecting African children.<sup>13,14</sup> The lymphomas in this study were prevalent in the 5 to 8 years age groups, affecting males mostly and involving the mandible especially, followed by the maxilla. About 17.6% of the Lymphoma involved multiple sites. This is because Burkitt lymphoma is a rapidly progressing malignant lesion with aggressive behaviour seen mostly in African children.<sup>13</sup> In contrast to our findings, Orikpete<sup>8</sup> reported pyogenic granuloma, a reactive lesion, as the most prevalent, while Unicystic Ameloblastoma was the most common odontogenic tumour in their report. Pleomorphic adenoma was the most common

tumour overall in the study by Jaafari-Ashkavandi and Ashraf. $^{3}$ 

Ameloblastoma was the second most common tumour encountered in this present study. This is in contrast to the Adenomatoid Odontogenic tumour seen as the second most common tumour after Burkitt lymphoma in the study by Akhiwu et al.<sup>12</sup> However, in the studies of childhood jaw tumours by some authors; Ameloblastoma was only reported as the most common tumour among the odontogenic tumour.<sup>9,3</sup> The variations that exist in the patterns of histopathologically diagnosed orofacial the childhood tumours in our environment in comparison with other studies may be due to the clinical, demographic and geographic variations. This study did not undertake inferential analysis since these tumours are heterogeneous. Therefore further assessment will help categorise the tumour into groups to determine the extent of association of these tumours with the gender, age groups and the site of occurrence.

#### CONCLUSION

This audit showed that the orofacial tumours seen in children in our centre are diverse, with a relatively lower prevalence compared with previous studies. Overall, the tumours were more frequent in males, mostly from five years and above and the mandible was the most common site involved. The most prevalent among the tumours was the Lymphomas and the Burkitt's type was the most common lymphoma seen. A good understanding of these diverse tumours in children in our environment by clinicians would assist with the timely identification of cases and actions instituted to adequately manage them.

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