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Malignant orofacial neoplasms in children and adolescents: A clinicopathologic review of cases in a Nigerian tertiary hospital

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Summary

Objective: The aim of the study was to determine the relative frequency of orofacial malignant neoplasm in children and adolescents.
Methods: A retrospective review of malignant orofacial tumours in children and adolescents ≤19 years from January 1992 to December 2003 from the records of the Department of Oral Pathology and Biology of the Lagos University Teaching Hospital, Nigeria was carried out. All the cases were analysed for age, gender, site distribution and histologic types.
Results: A total of 353 tumours and tumour-like lesions of the orofacial region were seen in patients ≤19 years during the period of the study. Of these, 47 (13.3%) were malignant tumours. This represented 3.3% (47 out of 1431) of all the tumours and tumour-like lesions seen during the period. The mean age (S.D.) of patients was 11.0 (±4.5) years (range, 2.5–19 years). Male-to-female ratio was 2.9:1. Burkitt's lymphoma (38.3%) was the most frequent malignant tumours. Lymphomas (53.2%) were the most common malignancy, followed by sarcomas (36.2%) and carcinomas (10.6%). Carcinomas exclusively affected patients in the 2nd decade of life and were predominantly glandular carcinomas. Osteosarcoma and rhabdomyosarcoma were the most common sarcomas. Burkitt's lymphoma (72%) occurring mostly in the first decade of life was the most common lymphoma.

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Conclusions: Malignant neoplasm constituted 13.3% of orofacial tumours and tumour-like lesions in children and adolescent in our centre. In agreement with previous reports from Africa, Burkitt's lymphoma is the most common malignant tumour and carcinoma is relative rare in this age group.

1. Introduction

Oral and pharyngeal malignant neoplasia are the sixth most common malignancy in the world [1]. They are reported to affect all age groups [1–5]. However, a considerable proportion of these tumours are reported to affect adult population, and only about 3–5% of head and neck tumours occur in children [6,7]. Oral carcinoma is considered the most common malignant tumour of the orofacial region constituting over 50% of these lesions [2,3]. Oral carcinoma is an age-related disease, and 98% of patients are reported to be over the age of 40 years [8,9]. In contrast, sarcomas of the orofacial region are reported to be less common than carcinomas [9,10]. They tend to affect a considerably younger age group than carcinomas [9,10]. Lymphomas are malignant lesions that can arise from any type of lymphocyte, but most frequently from B-cells [9]. They comprise Hodgkin's and the more common non-Hodgkin' lymphoma [9]. Burkitt's lymphoma is a common childhood tumour in tropical Africa [11]. It is predominantly extranodal. It is unusual in that its onset is in childhood and the jaw is the single most common initial site [9].

There are numerous reports of orofacial tumours in children from various parts of the world [5–7,12–15]. However, due to their rarity, reports focusing only on orofacial malignant tumours in children are scarce in the literature.

The aim of this study was to determine the relative frequency of orofacial malignant neoplasm in children and adolescents, and compare our findings with reports in the literature.

2. Material and methods

The histopathology records of the Department of Oral Pathology and Biology, Lagos University Teaching Hospital, Nigeria, were reviewed for all tumours and tumour-like lesions in patients aged 19 years or less from January 1992 to December 2003. All cases with histologic diagnosis of malignancy were selected and subjected to analysis of age, sex, site of occurrence and histologic type.

Data was analyzed using the software SPSS for Windows (version 12.0: SPSS, Chicago, IL). For analysis, simple frequency charts, descriptive statistics, and test of significance were used. A level of $P < 0.05$ was considered to be statistically significant.

3. Results

A total of 353 of tumours and tumour-like lesions were seen in patients ≤19 years during the period of the study. Of these, 47 (13.3%) were malignant tumours. This represented 3.3% (47 out of 1431) of all the tumours and tumour-like lesions seen in all age groups during the period. The mean age (S.D.) of patients was 11.0 (±4.5) years (range, 2.5–19 years). Majority (57.4%) of the patients were in the second decade of life at the time of diagnosis (Table 1). There were 35 boys and 12 girls with male-to-female ratio of 2.9:1. Burkitt's lymphoma (38.3%) was the most frequent malignant tumours. Lymphomas (53.2%) were the most common malignancy, followed by sarcomas (36.2%) and carcinomas (10.6%). No case of malignant odontogenic tumour was recorded. The most common site of occurrence was maxilla/maxillary antrum (38.3%), followed by mandible (29.8%) (Table 2).

Table 1 Age distribution of orofacial malignant neoplasm

Histologic types	Age (years)		Total (%)
	0–9	10–19	
Carcinomas			
Squamous cell carcinoma	0	1	1
Adenocarcinoma	0	2	2
Adenocystic carcinoma	0	1	1
Mucoepidermoid carcinoma	0	1	1
Sarcomas			
Osteosarcoma	0	4	4
Fibrosarcoma	0	3	3
Rhabdomyosarcoma	2	2	4
Ewing's sarcoma	1	2	3
Myxosarcoma	0	1	1
Malignant fibrous histiocytoma	0	2	2
Lymphomas			
Non-Hodgkin's lymphoma	17	7	24
Hodgkin's lymphoma	0	1	1
Total	20	27	47

Table 2 Site and gender distribution of patients

Histologic types	Gender[a]		Site distribution[b]									
	M	F	Man	Max	MM	Palate	Tongue	Lip	Cheek	Par	NS	Total
Carcinomas												
Squamous cell carcinoma	1	0	1	0	0	0	0	0	0	0	0	1
Adenocarcinoma	2	0	1	0	0	1	0	0	0	0	0	2
Adenocystic carcinoma	1	0	1	0	0	0	0	0	0	0	0	1
Mucoepidermoid carcinoma	1	0	0	0	0	0	1	0	0	0	0	1
Sarcomas												
Osteosarcoma	3	1	3	0	0	0	0	0	0	0	1	4
Fibrosarcoma	2	1	3	0	0	0	0	0	0	0	0	3
Rhabdomyosarcoma	2	2	0	3	0	0	0	1	0	0	0	4
Ewing's sarcoma	3	0	2	0	0	0	0	0	1	0	0	3
Myxosarcoma	0	1	0	0	0	0	0	0	0	0	1	1
Malignant fibrous histiocytoma	0	2	0	1	0	0	0	0	0	1	0	2
Lymphomas												
Hodgkin's lymphoma	1	0	1	0	0	0	0	0	0	0	0	1
Non-Hodgkin's lymphoma	19	5	2	14	7	1	0	0	0	0	0	24
Total	35	12	14	18	7	2	1	1	1	1	2	47

[a] M: male; F: female.
[b] Man: mandible/mandibular gingival; Max: maxilla/maxillary antrum; MM: mandible-maxilla; Par: parotid; NS: not specified.

3.1. Carcinomas

There were five patients with histologic diagnosis of carcinomas, constituting 10.6% of all the malignant tumours. All the cases exclusively occurred in the second decade of life (range, 11—18 years), and in boys. The most common histologic type was adenocarcinoma (Tables 1 and 2).

3.2. Sarcomas

There were 17 patients in this group, constituting 36.2% of the malignant tumours. Sarcomas predominantly occurred in the 10—19 years age-group (Table 1) and the male-to-female ratio was 1.4:1 (Table 2). Most cases (53%) were found in the mandible (Table 2). Rhabdomyosarcoma and osteosarcoma were the most frequent lesions in this group. Osteosarcomas were exclusively seen in the second decade while rhabdomyosarcoma occurred with equal ratio in the first and second decade of life (Table 1). Histologic variants of the rhabdomyosarcoma were embryonal (two cases) and alveolar (two cases).

3.3. Lymphomas

Lymphomas constituted 53.2% of the malignant tumours. Of these, 24 (96%) were non-Hodgkin's lymphomas. Only 1 (4%) case of Hodgkin's lymphoma was diagnosed in a 19-year-old boy. Lymphomas predominantly occurred in the 0—9 years age-group (Table 1); and 80% of the cases were found in boys.

Eighteen (75%) of the 24 cases of non-Hodgkin's lymphomas were Burkitt's lymphomas (BL). Mean age (S.D.) of patients with diagnosis of BL was 8.0 ± 2.6 years (range, 2.5—13 years). Males were eight times more affected than females (M:F ratio of 8:1), and majority (55.6%) of the cases involved the maxilla. Other sites of occurrence were mandible (11.1%) and bimaxillary (33.3%).

4. Discussion

The aetiology of most orofacial tumours remains obscure. Genetic predisposition has been suggested [1,6,16], while environmental factors such as viral infection, dietary deficiencies, trauma, and alcohol and tobacco intake have been implicated [5]. Epstein—Barr virus (EBV) has been implicated in BL and is consistently found in tumour cells of Burkitt's lymphoma which is endemic in East and Central Africa, where it is considered an aetiologic factor with malaria also considered a pathogenic co-factor [2,9,17].

Orofacial malignant tumours in children and adolescents accounted for 3.3% of all tumours and tumour-like lesions seen in all age groups. This figure is lower than 14.4% [5] and 8% [18] previously reported from Nigeria. Of all tumours seen in children and adolescents in our institution during the period of the study, 13.3% were malignant. This finding contrasts those of Arotiba [18] and Aregbesola et al. [5] who reported a prevalence of 40.2% and 51%, respectively. However, Bhaskar

[6] examined 293 cases of orofacial tumours among American children and found that only 9% were malignant.

Lymphomas, predominantly Burkitt's lymphomas were the most frequent malignant tumours in the present study, followed by sarcomas and carcinomas. This agrees with reports in the literature [2,5,18,19]. No case of malignant odontogenic tumours was recorded in the present series. Malignant odontogenic tumours are rare in adults, and very rare in children and adolescents [20,21]. They are reported to constitute less than 1% of all odontogenic tumours in America [22], although higher incidence are reported from Africa and Asia [21,23]. Arotiba [18] and Aregbesola et al. [5] also reported no case of malignant odontogenic tumour in children and adolescents.

In the present series, malignant tumours affected more males than females in the ratio of 3:1. Previous studies [2,5,18] on malignant tumours attest to this fact, albeit at lower male-to-female ratio. Aregbesola et al. [5] reported a male-to-female ratio of 2.5:1 in their study.

Burkitt's lymphoma accounted for 53% of the malignant tumours and 72% of the lymphomas. Males were eight times more affected than females, and maxilla was the predominant site of occurrence. This is consistent with previous reports from Africa [2,5,18,19]. Burkitt's lymphoma is a common childhood malignancy in a number of African countries [2,5,18,19,24,25]. The Epstein—Barr virus is consistently found in tumour cells of Burkitt's lymphoma which is endemic in East and Central Africa, where it is considered an aetiologic factor with malaria also considered a pathogenic co-factor [2,9,17]. It is predominantly extranodal. It is unusual in that its onset is in childhood and the jaw is the single most common initial site [9]. Aregbesola et al. [5] reported that Burkitt's lymphoma predominantly occurred in boys and in the maxilla and constituted 89% of tumours seen in children and adolescents in their study. The mean age (8.0 ± 2.6 years) of patients with Burkitt's lymphoma in the present study is similar to 8.8 ± 3.4 years reported earlier from Nigeria [26].

Rhabdomyosarcoma (RMS) and Osteosarcoma were the two predominant sarcomatous lesions in the present study. This is in agreement with the findings of Chidzonga [2] and Aregbesola et al. [5]. Osteosarcoma is a highly malignant tumour and is the most common primary malignant neoplasm of bone although rare in the maxillofacial region [10]. They are reported to affect a considerably younger age group than carcinomas [9,10]. In a recent report, about 30% of patients with Osteosarcoma were found below 18 years [10]. In contrast, RMS is an aggressive malignant skeletal neoplasm arising from embryonal mesenchyme [27]. It is reported to account for 4—8% of all malignancies in children less than 15 years of age [28]. This tumour is more common in Caucasians and most studies show a slight predominance in males [27]. The most common site in children is head and neck region, and oral cavity accounts for 10—20% of all head and neck RMS [27,29,30]. In the present study, RMS was seen in the first and second decade of life. RMS can occur in all age groups, but it is reported to be primarily seen in the first and second decades of life with a peak incidence between 2 and 6 years [31]. Congenital presentation of RMS has also been reported [31].

Carcinomas were the least common malignant tumours seen in the children and adolescents in the present series. This is a common finding in most reports [2—5,18]. Oral carcinoma is considered the most common malignant tumour of the orofacial region in adults [2,3]. It is an age-related disease, and 98% of patients are reported to be over the age of 40 years [9]. It is reported to be rare in children under 18 years and extremely rare in children under 10 years [2,3]. All the cases in the present series were seen in children between 11 and 18 years. However, previous authors [3,5] have reported few cases in children below 10 years.

5. Conclusions

Malignant neoplasm constituted 13.3% of orofacial tumours and tumour-like lesions in children and adolescent in our centre. In agreement with previous reports from Africa, Burkitt's lymphoma is the most common malignant tumour and carcinoma is relative rare in this age group.

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