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# Descriptive Epidemiology of the Clinico-Pathologic Profile of Orofacial Sarcomas: A Retrospective Study of 52 Cases Seen in a Nigerian Tertiary Health Institution

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## Abstract

**Background:** In our environment, the clinico-pathologic profile of orofacial sarcomas has not been fully assessed.

**Objective:** To determine the clinico-pathologic profile of orofacial sarcomas seen in a Nigerian tertiary health institution in the south-south geopolitical zone.

**Methods:** This was a retrospective cross-sectional study from the archives of histopathologic records of the department of Oral Pathology and Medicine, University of Benin Teaching hospital within a 25 year period (1990 to 2014). Cases histopathologically diagnosed as sarcomas of the orofacial region were reviewed and analyzed based on their age, gender, site and histopathological pattern.

**Results:** There were 285 (18.7%) cases diagnosed as malignant lesions which consist of 52 (18.2%) sarcomas. The age range was 3 to 82 years with mean age of 30.2 ± 20.3. There was equal gender distribution. The highest occurrence was in the second decade (n=14, 26.9%). The mandible was the most common site. The most common sarcoma was Malignant fibrous histiocytoma (n=21, 40.4%) while the least common was Fibrosarcoma.

**Conclusion:** This study shows that orofacial sarcomas are relatively rare in our environment occurring mostly in the younger age with no gender predilection. Malignant fibrous histiocytoma was the predominant histopathological diagnosis.

**Keywords:** Clinico-pathologic; Orofacial sarcoma; Histiocytoma

## Introduction

The oral and maxillofacial region encompassing the jaw bones, mandible, maxilla, zygoma, sinonasal bones and related tissues can be the site of a multitude of neoplastic conditions [1]. Sarcomas are malignant neoplasm of mesenchymal origin that can affect any site in this region [2]. Sarcomas, though rare compared with carcinomas, are of concern to people globally because they can cause facial deformity, invade vital structures and due to their often-grave prognosis can result in morbidity and mortality in affected persons [2,3]. Sarcomas account for nearly 21% of all pediatric solid malignant tumours and less than 1% of all adult solid malignant tumours. More than 50 distinct histological sarcoma sub-types have been recognized [4,5]. Sarcomas include the soft tissue sarcomas and the malignant bone tumours [5,6]. The soft tissue sarcomas make up approximately 5-15% of all adult sarcomas. They are rare in the head and neck region, constituting less than 1% of all head and neck neoplasm [7-9]. Soft tissue sarcomas can range from relatively slowing lesions, with little destructive growth, to becoming locally aggressive with regionally destructive effect and having potential for metastases to distant sites [9,10]. Sarcomas of the jaw bones are infrequent, accounting for about 1% of all the malignant tumors that occur in the orofacial region [11,12]. Bone sarcomas in the oral and maxillofacial region are highly aggressive and as such require accurate diagnosis and effectively treatment [12,13]. Yamaguchi et al. [14] stated that adequate excision with safety surgical margin as the initial therapy is important for survival although the value of radiation therapy and/or chemotherapy is uncertain. They further reported from their study that the 5-year survival rate of primary sarcoma cases was 61%. Nagler et al. [15] in their review of a follow-up of 25 treated sarcoma cases, reported that the overall 2- and 5-year survival rates were 72% and 60% respectively. They observed that survival was significantly correlated with the histological type of sarcoma and that younger patients had better survival rates. Sarcomas like other malignancies present a health burden for the patient and management challenge to the surgeons. Although studies on orofacial malignancies have been reported, [16,17] not much has been done to determine the clinicopathologic profile of the Orofacial sarcomas in our environment. The aim of this study was to determine the prevalence and clinico-pathological patterns of orofacial sarcomas seen in a Nigerian population over a 24 year period.

## Materials and Methods

This was a retrospective cross-sectional study from the archives of the histopathologic records of patients diagnosed as sarcomas of the orofacial region in the documents of department of Oral Pathology and Medicine, University of Benin Teaching hospital, Benin City, Nigeria within a 25 year period (1990 to 2014). The cases were reviewed and analyzed based on their prevalence, age, gender, site and histopathological types of the lesions. Data were analyzed using the Statistical Package for Social Sciences (Chicago, SPSS; 2005 version 17.0) with confidence level set at 95% and $p < 0.05$ was regarded as significant.

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## Results

From the 1,523 biopsies with histological diagnosis recorded within the 25 year period, a total of 285 malignancies affecting the orofacial region were recorded consisting of sarcomas (n=52, 18.2%), carcinomas (n=160, 56.1%) and lymphomas (n=73, 25.6%). There was an equal gender distribution of the sarcomas giving a male (n= 26, 50.0%) to female (n= 26, 50.0%) ratio of 1:1. The age range of the sarcomas was between 3 to 82 years with a mean age of 30.2 + 20.3. The highest occurrence was in the second decade (n=14, 26.9%) (Table 1). The mandible (n=17, 32.7%) was the commonest site followed by the maxilla (n= 10, 19.2%) (Table 2). The predominance of the mandible in this study was statistically significant (p=0.043). Table 3 shows the frequency of occurrence of the orofacial sarcomas. The most common sarcomas were Malignant fibrous histiocytoma (n=21, 40.4%) affecting 9 males (34.6%) and 12 females (46.2%) giving a male to female ratio of 0.75:1. The highest prevalence of Malignant fibrous histiocytoma was in the second (n=5, 23.8%) and fourth decades (n=5, 23.8%) with mean age of 31+20.8 years. The mandible (n=8, 38.1%) was the most common site of the Malignant fibrous histiocytoma in this study. Figure 1 shows the photomicrograph of a malignant fibrous histiocytoma. Eight cases (15.4%) were diagnosed as Rhabdomyosarcoma which was the second most frequent sarcoma in this study. The patients were mostly in the first (n=3, 37.5%) and second (n=4, 50%) decades with a mean age of 13.5 ± 11.6 years, affecting 6 males (23.1%) and 2 females (7.7%) giving a ratio of 3:1. The cheek (n =3, 37.5%) was the most affected site. The third most frequent sarcoma in this study was Osteosarcoma of the jaws (n=6, 11.5%) found in the second to the seventh decades of life with 1 case (16.7%) in each decade. The mean age of the Osteosarcoma was 39.7 ± 40.9 years with equal sex predilection giving a male (n =3, 11.5%) to female (n =3, 11.5%) ratio of 1:1. The mandible was the most frequently involved site in this study accounting for 5(83.3%) of the Osteosarcoma. There were 5 cases (9.6%) of Chondrosarcoma identified occurring mostly in the second decade (n=3, 60%) with a mean age of 30.4 ± 18.2 years. There were 3 females (11.5%) and 2 males (7.7%) giving a female to male ratio of 1.5:1. The maxilla (n=2, 40%), palate (n=2, 40%) and the mandible were the affected sites. The rare sarcomas in this study includes Fibrosarcoma (n=1, 1.9%), Myxosarcoma (n=1, 1.9%) and Neurogenic sarcoma (n=1, 1.9%).

| Decade | Gender | | Total n (%) |
|---|---|---|---|
| | F (n) | M (n) | |
| 0 - 9 | 2 | 4 | 6 (11.5) |
| 10 - 19 | 10 | 4 | 14 (26.6) |
| 20 - 29 | 7 | 4 | 11 (21.2) |
| 30 - 39 | 4 | 3 | 7 (13.5) |
| 40 - 49 | 0 | 3 | 3 (5.8) |
| 50 - 59 | 1 | 2 | 3 (5.8) |
| 60 - 69 | 2 | 4 | 6 (11.5) |
| 70 - 79 | 0 | 1 | 1 (1.9) |
| 80 - 89 | 0 | 1 | 1 (1.9) |
| Total | 26 | 26 | 52 (100) |

**Table 1:** Age and gender distribution of the orofacial sarcomas.

| Site | n (%) |
|---|---|
| Cheek | 6 (11.5) |
| Orbit | 1 (1.9) |
| Floor of the Mouth | 1 (1.9) |
| Gingiva | 7 (13.5) |
| Mandible | 17 (32.5) |
| Maxilla | 10 (19.2) |
| Nasal cavity | 1 (1.9) |
| Palate | 4 (7.7) |
| Parotid | 2 (3.8) |
| Submandibular | 2 (3.8) |
| Zygoma | 1 (1.9) |

**Table 2:** Site distribution of the orofacial sarcomas.

| Histopathological diagnosis | n (%) |
|---|---|
| Carcinosarcoma | 1 (1.9) |
| Chondrosarcoma | 5 (9.6) |
| Fibrosarcoma | 1 (1.9) |
| Hemangioendothelioma | 3 (5.8) |
| Malignant Fibrous Histiocytoma | 21(40.4) |
| Primitive Neuroectodermal Tumour | 1 (1.9) |
| Myxosarcoma | 1 (1.9) |
| Neuroblastoma | 4 (7.7) |
| Neurogenic sarcoma | 1 (1.9) |
| Osteosarcoma | 6 (11.5) |
| Rhabdomyosarcoma | 8 (15.4) |
| Total | 52 (100) |

**Table 3:** Histopathological diagnosis of the orofacial sarcomas.

## Discussion

Several studies have reported different prevalence of the Orofacial sarcomas in their regions in Nigeria and other parts of Africa [2,3,17-20]. Sarcomas accounted for 18.2% of malignant lesions seen within the study period. This is similar to the 18% reported in Lagos, South-west Nigeria [17], and 18.9% reported in Kenya [2]. This differs slightly from the 16.7% in Jos [18] and the 20% reported in Kaduna [3] both in Northern Nigeria. In contrast, a lower prevalence of 7% was observed in Libya [19] and 8.9% was found in Ibadan [20]. This suggests variable prevalence in different regions in Nigeria and among African population. The overall equal gender distribution in this study is comparable to previous reports [3,20,21]. The overall mean age of 30 ± 20.3 years observed in this study is comparable to the 31 years reported by Adebayo et al. and the 29 years reported by Aldriane [21], but slightly higher than the 27.1 years observed by Adisa et al. [20] The

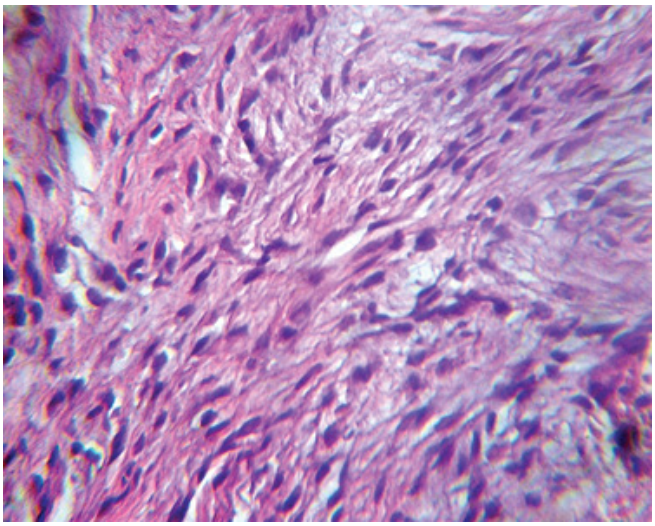

**Figure 1:** Malignant fibrous histiocytoma: Malignant fibroblast in storiform pattern and histiocytes in a fibro-cellular connective tissue stroma (H&E 400x).

preponderance of the lesions in the jaws- mandible and the maxilla in this present study, is similar to the findings previously reported that the maxillofacial bones were the most common location of orofacial sarcoma [2,20]. However a significant predilection for the mandible was observed in this study in contrasts to the report by Arotiba et al. [16] that found the maxilla as the commonest site for the orofacial sarcomas.

This study showed that malignant fibrous histiocytoma (40.4%) was the most common sarcoma, while Rhabdomyosarcoma (15.4%) and Osteosarcoma (11.5%) were the second and third most common sarcomas respectively. This is in contrast with findings of several authors [2,3,16,21]. A study on Malignant orofacial tumours in Lagos found Rhabdomyosarcoma as the most common among the sarcomas followed by Chondrosarcoma and Osteosarcoma [16]. Also, a Ugandan study reported that Kaposi's sarcoma accounted for 82.8% of the cases, followed by Rhabdomyosarcoma (6.9%) while Osteosarcoma and Chondrosarcoma each accounted for 2.5% of the cases [21]. Whereas, Kamau et al. in a Kenya population observed that Osteosarcoma and Kaposi's sarcoma were the two most commonly occurring sarcomas with both lesions accounting for 59% of all the sarcomas in their series [2]. The high incidence of HIV/AIDS in most of the east and central and southern Africa may have contributed to the high proportion of Kaposi sarcoma in their studies.

Although the occurrence of Malignant fibrous histiocytoma (MFH) in the orofacial region is reported to be rare [22] it has been observed severally in the soft tissues of the face and neck, the oral cavity, salivary glands and in the mandible and maxilla [9,23]. Previous studies by Guevera-Canal et al. [24] (in South America) and Amnirul et al. [25] (in Asia) reported a low prevalence of malignant fibrous histiocytoma of 10.3% and 14.3% respectively. Furthermore, Adebayo et al. [3] in Kaduna, Nigeria, reported 6% prevalence of malignant fibrous histiocytoma. These findings were relatively low compared to the high prevalence of 40.4% of MFH observed in our study. The diagnosis of MFH in this study was done using tissue histomorphology which was very adequate. Some studies have recommended the exclusion of some variants of MFH, reclassification of MFH and the use of immunohistochemistry in categorizing this lesion. This may address the apparent high prevalence of MFH in our environment. Also, this study observed a slight female preponderance of MFH and the mandible was the mostly involved site. This is in contrast to the observation by Effiom et al. [26] that studied 5 cases of malignant fibrous histiocytoma and reported a male predilection (60.0%) and the maxilla was the only site involved in all 5 cases. This difference from our findings may probably be due to the smaller sample size in their study.

Rhabdomyosarcoma is the second most common orofacial sarcoma seen in this study. Adebayo et al. [3] reported in there review that Rhabdomyosarcoma is the most common soft tissue sarcoma in the oral and maxillofacial region. Pappo [27] and Andrade et al. [28] stated that Rhabdomyosarcoma, a malignant tumor of skeletal muscle origin, accounts for about 5-10% of childhood cancers and more than 50% of pediatric soft tissue sarcomas. In this study, Rhabdomyosarcoma accounts for 15.4% of all the orofacial sarcomas. This finding is higher than the 13% reported by Adebayo et al. [3]. This study also showed that it occurred in younger patients within the first and second decades of life. The mean age of occurrence was 13.5 ± 11.6 years which is slightly less than the mean age of 14.3 years reported by Andrade et al. [28] in Brazil. There was a male predominance in 75% of the cases. In comparison, Andrade et al. [28] also reported a male predominance although with a lesser prevalence of 58.6% of the cases. The cheek was the most affected site and this agrees with previous reports [3,29].

About 6% to 10% of Osteosarcoma occurs in the craniofacial region [30,31]. Approximately 7% of all arise in the jaw bones [20,32-34]. Study by Adebayo et al. [3] reported Osteosarcoma (28%) as the most common orofacial sarcoma followed by Chondrosarcoma (17%) with Rhabdomyosarcoma and Fibrosarcoma accounting for 12% of cases each. Also, a study in Tokyo by Satoshi et al. [14] reported Osteosarcoma as the most common accounting for 28.1% of cases followed by malignant fibrous histiocytoma (21.9%), whereas in this study relatively lower prevalence of Osteosarcoma (11.5%) was observed. The mean age of onset of Osteosarcoma of the jaws is in the third to fourth decade of life [31,35]. A higher mean age of 39.7 ± 40.9 years for Osteosarcoma was found in this study compared to the mean age of 27.2 years reported by Ogunlewe et al. [13] in their analysis of 17 cases of Osteosarcoma of the jaw bones. The equal gender distribution of Osteosarcoma in this study agrees with previous studies [31,36]. However, some studies have reported slight male preponderance [13,34,37]. The predilection of Osteosarcoma for the mandible in our study is consistent with reports from the previous studies [3,38-40].

Chondrosarcoma is a rare primary malignant neoplasm of the head, neck, oral and maxillofacial regions [41]. After Osteosarcoma, it is the second most common primary bone malignancy [41,42]. Chowdhury et al. [43] and Kundu et al. [41] stated that the involvement of Chondrosarcoma in the craniofacial region is very rare accounting for less than 10% of all Chondrosarcoma. In the maxillofacial region, it accounts for 1-3% of all the Chondrosarcoma of the entire body [44,45]. In this study, Chondrosarcoma is the fourth most common sarcoma accounting for 9.6% of the orofacial sarcomas. Adriane [21] also found Chondrosarcoma as the fourth most common in the cases studied but with a lower prevalence of 2.5%. Chondrosarcoma was also the second most common sarcoma in the findings by Adebayo et al. [3] and Arotiba et al. [16] with higher prevalence of 13% and 18.9% respectively compared to our study. Previous reports assert that Chondrosarcoma occurs mostly between the 3rd and 6th decades of life [41,43]. Our study observed a mean age of 30 ± 18.2 years with the lesion occurring mostly in the second decade (60%). This is similar to the mean age of 30 years observed by Adriane [21] but differs slightly from the 32 years reported by Adebayo et al. [3] who observed the occurrence of this lesion in the 3rd to 6th decade of life. Also, our study showed a slight female preponderance which is in contrast with the findings from previous studies which reported that males are more commonly affected [3,21,41]. Eighty percent of the cases seen in this study involved the upper jaw while the mandible was involved in 20% of cases. This finding agrees with various literatures which stated that Chondrosarcoma in the orofacial region has a predilection primarily for the maxilla due to the presence of pre-existing nasal cartilage and that the occurrence of Chondrosarcoma in the mandible is rare [44-47].

Other orofacial sarcomas in this study such as Myxosarcoma and Neurogenic sarcoma were of low prevalence as reported in several literatures [17,48-50].

Surgery is reported to be the mainstay of treatment of the sarcomas. The operability of these tumours in terms of anatomic considerations, tumor size, stage at diagnosis and attainment of a surgical clear margin are critical factors for survival [14,51,52]. Similarly, other authors have reported variables such as gender, age, tumor location, tumour size, histopathological diagnosis and the type of treatment as prognostic factors [12,53,54]. However, treatment of these tumors should be provided by a multidisciplinary team to achieve optimal outcomes and immediate reconstruction should be an integral part of the surgical treatment [52].

## Conclusion

The findings from this study show that orofacial sarcomas are relatively rare in our environment occurring mostly in the younger adults with no gender predilection. Malignant fibrous histiocytoma was the predominant histological type followed by Rhabdomyosarcoma and Osteosarcoma in this study. Surgical treatment remains a very useful approach in management of cases. This study serves as baseline on the prevalence and clinicopathologic patterns of orofacial sarcomas in the South-south zone of Nigeria.

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