

RESEARCH ARTICLE

An 8-year Pattern of Orofacial Sarcoma from the National Referral Hospital in United Republic of Tanzania

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ABSTRACT

Aim: This study aimed at evaluating the pattern of head and neck sarcomas among patients treated at Muhimbili National Hospital, Tanzania.

Background: Sarcomas of the head and neck are very rare, representing only 1% of all primary tumors arising within the head and neck region and accounting for 4 to 10% of all sarcomas, with more than 50 distinct existing histologic subtypes.

Materials and methods: It was a retrospective study whereby histological results of the lesions arising from oral and maxillofacial region between 2008 and 2016 were analyzed. Patient's demographic data, histopathological diagnosis, and the type of sarcoma were recorded. The lesions were broadly grouped as soft and hard tissue sarcomas (HTSs). Data analysis was done using Statistical Package for the Social Sciences version 19 computer program.

Results: Sarcomas accounted for 7% of all lesions occurring in orofacial region. Male to female ratio was 1:1.4. The age ranged from 3 to 81 years, mean age being 33 ± 16 years. The most affected age groups were of 30 to 39 followed by 20 to 29 years. Approximately half of the patients were aged below 30 years and three quarters below 40 years. Kaposi's sarcoma (KS) and rhabdomyosarcoma were the most common soft tissue sarcoma (STS), while osteosarcoma and chondrosarcoma were the commonest HTS.

Conclusion: The analysis demonstrated that the head and neck sarcomas are a very rare group of neoplasm, with approximately 20 cases per year. The STSs are more common than the HTSs, while KS is the most common, followed by osteosarcoma and rhabdomyosarcoma. Generally, patients less than 40 years of age are the mostly affected, with a slightly higher female predominance.

Clinical significance: By contributing to the identification of the incidence of sarcomas at a tertiary hospital in Tanzania, this study promotes scientific understanding of pattern of occurrence and underscores the necessity of early detection of sarcomas, since the affected are young individuals.

Keywords: Head and neck, Kaposi's sarcoma, Osteosarcoma, Sarcomas.

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INTRODUCTION

Sarcomas are malignant neoplasms derived from cells of mesenchymal component with diverse tissue of origin including bone, cartilage, fibrous, vascular, fatty, muscular, and neural tissue.¹ Sarcoma of the head and neck is very rare, representing only 1% of all primary tumors arising within the head and neck region and accounting for 4 to 10% of all sarcomas.^{2,3}

They represent multiple malignancies rather than a single cancer,⁴ with more than 50 distinct histologic subtypes existing, many occurring at any age, and are not restricted to a specific location of the body.⁵ Despite several histological subtypes, sarcomas may be grouped together because of resemblances in prognostic factors, clinical presentation, embryonic derivation, and overall outcome.⁶

Sarcomas in head and neck region can be broadly divided into soft tissue sarcoma (STS) and hard tissue sarcoma (HTS). The STS is classified according to the adult tissue that they resemble; similarly, HTS is usually classified according to the type of matrix production: Osteoid-producing sarcomas are classified as osteosarcomas and chondroid-producing sarcomas are classified as chondrosarcomas.⁷

The most common sarcoma is debatable, depending on age, site, and possibly racial factors. Osteosarcoma is reported to be the commonest oral and maxillofacial sarcoma in adults,⁸ while in children rhabdomyosarcoma is predominant.⁹ Several studies reported higher incidence of sarcomas in younger age groups, especially below age of 40 years.¹⁰⁻¹²

Regardless of lower incidence rates of sarcomas, considering that they occur more frequently in young adults and adolescents compared with other cancers, the years of life lost can often be substantial.⁵ The epidemiological characteristics of disease vary in geographical areas, and

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numerous studies have demonstrated geographical variation affects the incidence of different types of sarcomas, age and gender prevalence, making conduction of such studies undeniable.¹³

The aim of this study was to evaluate the pattern of head and neck sarcomas among patients treated at Muhimbili National Hospital (MNH), Tanzania.

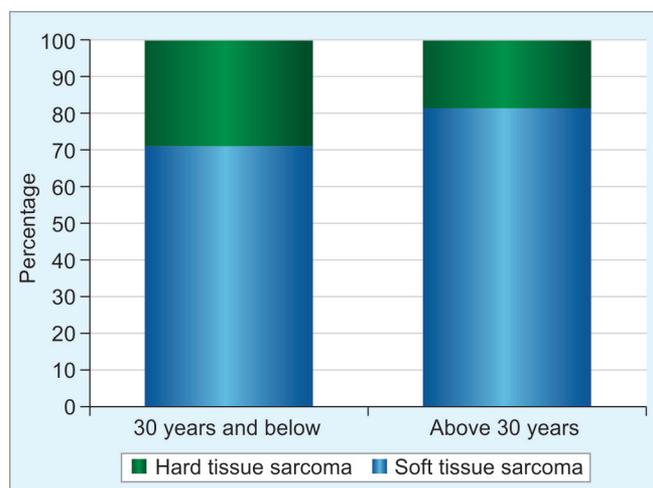
MATERIALS AND METHODS

This was a retrospective descriptive study whereby histological results of patients who had lesions affecting the oral and maxillofacial region from January 2008 to September 2016 were retrieved from the archives of the Oral and Maxillofacial Surgery firm at MNH in Dar-es-Salaam, Tanzania. The age, sex, file number, and histopathological number of the patient were recorded. Diagnosis was recorded as reported in the biopsy result slip of the patient. The histological result slip which had sarcomas as final diagnoses were obtained. Reports of fine needle aspiration cytology were excluded. Those reports which had no final diagnosis were excluded as well. In case where a single patient had more than one result, as one for presurgery incisional biopsy and another of postsurgical excision of the lesion, the postoperative results were included. The data for histological diagnosis and demographic data were entered into a dataset and data analysis was done by Statistical Package for Social sciences computer program version 19. The statistical tests that were used in analysis included chi-square test, t-test, and Fisher's exact test, where by $p \leq 0.05$ was considered statistically significant.

RESULTS

A total of 2,205 biopsy results slip of patients with maxillofacial tumors were retrieved from the archives of Oral and Maxillofacial Surgery firm in MNH, for a period of 8 years (2008–2016). Sarcomas accounted for 7% ($n = 154$) of all lesions occurring in orofacial region.

Out of 154 biopsy results of sarcomas obtained, females composed 57.8% ($n = 89$), with male to female ratio of 1:1.4; however, the difference observed was not statistically significant ($p = 0.389$). The age distribution of the patient was between 3 and 81 years. The overall mean age for occurrence of sarcomas was found to be 33 ± 16 years. Most of the patients were in the age group 30 to 39 years (27.9%) followed by age group 20 to 29 years (24%) and 10 to 19 years (16.2%). Patients who were aged 30 years and below comprised 47.4% ($n = 73$), while 74% ($n = 114$) of the patients were aged 40 years and below. The overall mean age for female was 33.1 ± 15.9 years, while for male was 32.9 ± 16.6 years; however, the observed difference in mean age was statistically insignificant ($p = 0.996$).



Graph 1: Distribution of main categories of sarcomas between the young and older patients

The STSs were the most common type when compared with their hard tissue type counterparts, each accounting for 76.6 and 23.4% respectively. The ratio of STSs to HTSs was 3.3:1. The difference in distribution of STS and HTS between the young patients (30 years and below) and older patients (above 30 years) was not statistically significant ($p = 0.133$) (Graph 1).

Age group 30 to 39 years was the most predominant among the STSs, with overall mean age of occurrence of 33.4 ± 15.8 years; in females, the mean age was 32.5 ± 14.3 years and for males 34.6 ± 17.8 years; however, there was no statistical difference between the mean ages of female and male ($p = 0.488$). For HTSs age group 20 to 29 years was the most predominant, with overall mean age of occurrence of 31.8 ± 17.4 years; in females, the mean age was 34.9 ± 21 years and for males 27.9 ± 11.1 years; however, there was no statistical difference between the mean ages of females and males ($p = 0.24$).

Of the 14 different types of sarcomas encountered in this study, Kaposi's sarcoma (KS) was the most prevalent lesion (52.6%), followed by osteosarcoma (16.9%) and rhabdomyosarcoma (9.7%) (Table 1). The KS was more common in females than males, with male to female ratio of 1:1.7. It was found to be predominant in the age group 30 to 39 years, with mean age of 35.5 ± 11.5 years (Table 2). The mean ages for males and females were 38.1 ± 15.5 and 33.6 ± 11.5 years respectively, though the difference in mean of ages was not statistically significant ($p = 0.11$).

Osteosarcoma showed no sex predilection, with male to female ratio of 1:1. It was found to be predominant in the age group 20 to 29 years, with mean age of 30.2 ± 16.2 years (Table 2). The mean ages for male and female were 27.6 ± 12.1 and 32.9 ± 19.6 years respectively, though the difference in mean ages was not statistically significant ($p = 0.414$).

Table 1: Types and sex distribution of orofacial sarcomas

Histological types of sarcoma	Gender of the patient		Total
	Male	Female	
Kaposi's sarcoma	30	51	81
Osteosarcoma	13	13	26
Rhabdomyosarcoma	6	9	15
Chondrosarcoma	3	6	9
Fibrosarcoma	4	3	7
Malignant fibrous histiocytoma	2	2	4
Hemangiopericytoma	0	3	3
Liposarcoma	2	0	2
Malignant schwannoma	1	1	2
Angiosarcoma	1	0	1
Ewing sarcoma	0	1	1
Fibromyxoid sarcoma	1	0	1
Leiomyosarcoma	1	0	1
Neurofibrosarcoma	1	0	1
Total	65	89	154

Rhabdomyosarcoma was more common in females than males, with male to female ratio of 1:1.5. It was found to be predominant in the age group of 10 to 19 years with mean age of 24.33 ± 21.1 years (Table 2). The mean ages for male and female were 16.7 ± 18 and 29.4 ± 22.4 years respectively, though the difference in mean of ages was not statistically significant ($p = 0.265$).

The sarcomas which were encountered in moderate frequency in this study ($n = 3-9$) included chondrosarcoma, fibrosarcomas, malignant fibrous histiocytoma, and hemangiopericytoma. Sarcomas with least frequency of encounter ($n = 1$ or 2) included liposarcoma, malignant schwannoma, angiosarcoma, Ewing's sarcoma, fibromyxoid sarcoma, leiomyosarcoma, and neurofibrosarcoma (Table 1).

DISCUSSION

The findings of this study showed that the most common sarcomas of head and neck region are KS, osteosarcoma, rhabdomyosarcoma, chondrosarcoma, and fibrosarcomas. The occurrence of these sarcomas was predominantly in younger age groups. Sarcomas of the head and neck are very rare, representing only 1% of all primary tumors arising within the head and neck region.² Epidemiological evaluations in different countries are limited and they present different results about prevalence of various sarcomas.¹³ Taking this into account, this study was conducted to assess the pattern of head and neck sarcomas among patients treated at MNH, Tanzania, so as to appreciate their trends in comparison to trends in other parts of the world.

In this study, we found that sarcomas accounted for 7% of all lesions occurring in maxillofacial region, almost similar prevalence of 6% was reported by Alishahi et al¹³ in a study from Iran. Budhy et al¹⁴ reported a prevalence of 4% of maxillofacial lesions from Indonesia, while Rafindadi and Ayuba⁸ in a study from Nigeria reported the prevalence of 4.9%. In view of these results, it is apparent that the sarcomas are rare lesions.

The findings of this study found higher prevalence of female patients (57.8%), with female to male ratio of 1.4:1. Higher prevalence of sarcomas in females was also reported by de Bree et al¹⁵ in a study from Netherlands and Guevara-Canales et al¹² in a Peruvian study. Contrary to our findings, higher occurrence of sarcomas among males has been reported in studies from Japan,¹ Zimbabwe,¹⁰ Brazil,¹⁶ and Portugal.¹⁷

It was ascertained in this study that the overall mean age for occurrence of sarcomas was 33 ± 16 years, with 74% ($n = 114$) of the patients being aged 40 years and

Table 2: Age group distribution of patients with orofacial sarcoma

Histological diagnosis	Age group							Mean age (years)
	0-9	10-19	20-29	30-39	40-49	50-59	60+	
Kaposi's sarcoma	2	5	16	33	18	4	3	35.3
Osteosarcoma	2	3	11	6	2	-	2	30.3
Rhabdomyosarcoma	2	8	1	-	-	2	2	24.3
Chondrosarcoma	-	1	2	3	1	-	2	37.7
Fibrosarcoma	-	1	3	-	1	-	2	40.7
Malignant fibrous histiocytoma	-	1	1	-	1	1	-	32.6
Hemangiopericytoma	-	2	-	1	-	-	-	19.0
Liposarcoma	-	1	-	-	1	-	-	33.5
Malignant schwannoma	-	1	1	-	-	-	-	20.0
Angiosarcoma	-	-	1	-	-	-	-	21.0
Ewing sarcoma	-	1	-	-	-	-	-	19.0
Fibromyxoid sarcoma	-	-	1	-	-	-	-	26.0
Leiomyosarcoma	-	-	-	-	-	-	1	81.0
Neurofibrosarcoma	-	1	-	-	-	-	-	11.0
Total	6	25	37	43	24	7	12	

below. Almost similar findings were reported in a study from Kenya by Kamau et al¹⁸ whose findings were: Age range of 3 to 90 years, common age group 20 to 29 years, 70.29% of cases occurred in patients less than 40 years of age, and the mean age of 31.7 years. Another study from Africa, by Adebayo et al¹⁹ in a Nigerian population, reported a mean age of 31 ± 15 years. Contrary to the findings from African studies, a study from Portugal by Barosa et al¹⁷ reported a higher mean age of 45.9 ± 25 years; similarly, Yamaguchi et al¹ reported mean age of 42 years in a study from Japan, and in an Iranian study, the mean age of 41 years was reported by Alishahi et al.¹³

In Africa, KS has been described as one of the most common malignant tumors, being prevalent in countries with higher human immunodeficiency virus/acquired immunodeficiency syndrome prevalence.¹⁸ In a Kenyan study done by Chindia et al,²⁰ KS was the most prevalent sarcoma, while Kamau et al¹⁸ reported it to be the second most common sarcoma, while in another study from Kenya, the mean age of occurrence was 36.6 years and with no sex predilection. The mean age of 34.7 years with male predominance was reported by Lager et al.²¹ Male KS predominance has also been reported in other studies.^{22,23} Despite the findings of this study concurring with mean age of occurrence of KS to other African studies, there was slight female predilection in our study, with male to female ratio (M:F) of 1:1.7. The gender differences in KS are of interest considering previous suggestions of lower female risk, possibly attributed to hormonal factors including human chorionic gonadotropin in pregnant women.²⁴ The high frequency of patients aged 20 to 49 years in our study is representative of the sexually active population and noticeably suggests the association of KS development with sexual behavior.

Osteosarcoma is the most common bone sarcoma affecting all ages; the clear peak incidence is the period of puberty and adolescence followed by another peak in the seventh decade.²⁵ Osteosarcoma was the most frequently diagnosed HTS, and this is consistent with data from other parts of Africa.^{10,18,20} Our study found no sex preference, with M:F of 1:1 in contrary to the findings of Guevara-Canales et al¹² and Kamau et al¹⁸ that reported female predilection. On the contrary, Alishahi et al,¹³ Adebayo et al,¹⁹ and Chindia et al²⁰ reported male predominance. The difference between male and female reported in these studies is, however, marginal; the mean age of occurrence of osteosarcoma in this series was concurrent with most reports from different literature;^{12,13,18,26} however, it did not coincide with the mean age reported by Adebayo et al.¹⁹

Rhabdomyosarcoma, a malignancy of primitive muscle, predominates in pediatric patients in the first decade of life, making it the most common maxillofacial

sarcoma in childhood but can be seen at any age.^{18,27} In this study, rhabdomyosarcoma was the third most common sarcoma accounting for 9.7%. Kamau et al¹⁸ reported that rhabdomyosarcoma was the fourth most common sarcoma accounting for 9.9% of all tumors. Adebayo et al,¹⁹ on the contrary, reported rhabdomyosarcoma to be the third most common, accounting for 10% of all sarcomas. In contrast to the above studies and our study, Chidzonga and Mahomva¹⁰ reported prevalence of rhabdomyosarcoma to be 23.8% in a study from Zimbabwe, and Pacheco et al¹⁶ reported prevalence of 25%. An Iranian study¹³ reported rhabdomyosarcoma to be the fourth common sarcoma, accounting for 7.61%. We found female predominance over male counterparts; similar results were also found in the literature,^{12,13} but this was in contrast to other studies that reported higher prevalence in males.^{1,12,19,20} As is evident, many literature reports early age of onset of rhabdomyosarcoma and a strong male predominance. However, this male predominance was more pronounced in embryonal subtype of rhabdomyosarcoma, while there were no sex differences in alveolar subtype incidence.²⁸

Chondrosarcoma is a slow-growing but malignant tumor with a relatively high local recurrence rate, representing 10 to 20% of all malignant bone tumors, and of these, 1 to 12% originate in the head and neck region.²⁹ Chondrosarcoma was found to be present in 5.8% of all sarcomas in this series, with higher incidence in females compared with males (2:1), with mean age of 37.7 years. Most of the studies report almost similar mean age for occurrence of chondrosarcoma in the head and neck region.^{12,13,19,29,30} We report higher incidence of chondrosarcoma in female patients, similar to reports from Peru.¹² Two studies from Asia, however, reported no sex preference,^{13,29} while a Nigerian study and Brazilian study reported higher prevalence in males.^{19,30}

Fibrosarcoma is a malignant tumor whose occurrence in the head and neck is rare, accounting for less than 5% of fibrosarcomas of all sites.²⁰ In this series, the incidence of fibrosarcomas was 4.5%, with a mean age of occurrence being 40.7 years, having slightest higher prevalence among male patients. Some studies from different parts of the world show similar male predominance,^{12,20} others report of equal predilection,^{13,19} while Kamau et al¹⁸ found a higher prevalence in females. As far as the incidence of occurrence of head and neck fibrosarcomas is of concern, higher incidence than our findings has been reported in Kenya^{18,20}; also, another almost similar incidence to our findings has been reported in Peru¹² and Iran.¹³ Most of the studies have reported lower mean age of occurrence compared with our findings.^{12,13,18,20}

Other sarcomas that were found in this study included malignant fibrous histiocytoma, hemangiopericytoma, liposarcoma, malignant schwannoma, angiosarcoma,

Ewing's sarcoma, fibromyxoid sarcoma, leiomyosarcoma, and neurofibrosarcoma. However, in view of the small number of lesions identified in the current study, no specific observations could be made regarding these lesions.

Findings of this study might need some caution in interpretation for some inherent limitations. The study being of retrospective design, enrolled biopsy results are found in the archives of the Department of Oral and Maxillofacial Surgery, and not all the biopsy results were believed to be found, since some of the few results might have been misplaced. In addition to that, incomplete patients' detail in the files made information of the patients difficult to attain for evaluating the location and size of tumor, signs and symptoms, treatment rendered, and prognosis of the disease.

The findings may be compared with other studies reported in the region, but the interpretation must be done with caution because of the disparities in genetic composition among the population, diagnostic capacity, and difference in management capability existing among institutions, especially in the developing nations. Therefore, coordinated multicenter study on these lesions is very crucial in order to understand the extent of the problem in the region.

It is a challenge for surgical pathologists to diagnose sarcomas into different types and subtypes basing on histology due to several facts including wide array of sarcomas with morphologic heterogeneity in combination with rarity of these lesions in common practice, hence, being susceptible to be misdiagnosed.³¹ Considering this fact, it is high time for surgical pathologists to use multiple diagnostic techniques to supplement the traditional histopathological analysis, by use of immunohistochemistry and where feasible molecular studies, as this will give better incidences of different sarcomas, hence, more accurate prevalence of different types of sarcomas.

CONCLUSION

Based on the results obtained, the head and neck sarcomas in Tanzania are a very rare group of neoplasm, with approximately 20 cases per year. The STSs are more common than the HTSs, while KS is the most common, followed by osteosarcoma and rhabdomyosarcoma. Generally, patients less than 40 years of age are mostly affected, with a slightly higher female predominance. Difference in genetic background and overall management capabilities may be the reason for taking caution when comparing results from different countries.

CLINICAL SIGNIFICANCE

By contributing to the identification of the incidence of sarcomas at a tertiary hospital in Tanzania, this

study promotes scientific understanding of pattern of occurrence and underscores the necessity of making the professionals as well as general public aware of the importance of early detection of sarcomas, since the affected are young individuals, who are the workforce of any society.

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