

A clinical study of Sarcoma of jaw and orofacial region

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Abstract:

Background: Sarcoma of jaw and orofacial soft tissue is rare, constituting between 4-8% of all malignancies in the region. A few case reports of individual tumors are available while reviews of significant series is lacking. An observational descriptive study (March' 2007 to Feb' 2011) was performed at Oral and Maxillofacial Surgery department, Dhaka Dental College and Hospital. This study presents 20 cases of sarcoma collected over 4 years at a tertiary oral care centre in Dhaka, Bangladesh. **Objectives:** To find out the distribution & pattern of Sarcoma among all oral malignancy and to find out the age, sex, site, clinical presentation and the histological types of these tumors **Method:** Histopathological types of the sarcomas were analyzed to indicate the numbers that occurred and also the pattern of occurrence according to age, gender, site and clinical presentation. **Result:** There were 138 maxillofacial malignancies of which 20 (14%) were sarcomas. Seven histopathologic types were found of which osteosarcoma (30%), fibrosarcoma (20%), Ewing's sarcoma (20%), malignant fibrous histiocytoma (15%) were predominant. The male to female ratio was 1.86:1. Patients with sarcoma were between 3.5 years and 70 years (mean age 34.3±20.3 years) with most patients (35%) in 35 to 45 years of life. Case presented with symptoms such as swelling (100%), pain (70%) and tissue ulceration (30%). Surgery was performed for 70% of cases treated while chemotherapy was used for 50%. **Conclusion:** In Dhaka Dental College Hospital, sarcomas account for 14% of all maxillofacial malignancies with the osteosarcoma as the predominant type. Most affected were people in the fourth decade of life. Surgery was the main modality used for treatment while some patients had no treatment due to self discharge and late presentation.

Keywords: Oral Malignancy, Sarcoma, Osteosarcoma

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Introduction:

Sarcomas are malignant neoplasm derived from cells of mesenchymal origin. The originating tissue is diverse and includes bone, cartilage, muscular, fibrous, vascular, fatty and neural tissue. In the oral and maxillofacial region, sarcomas are uncommon. Compared to carcinomas, sarcomas are rare. Incidence of sarcoma in jaw and orofacial soft tissue area varied depending upon different population reported by researchers. Sadat et al² reviewed 139 cases of oral malignancies from Bangladeshi population among which sarcomas were 12.9%, Budhy et

al¹ found 4% sarcoma while squamous cell carcinoma made up 70% from East Java, Indonesia. Adebayo et al³ examined 406 maxillofacial malignancies from Kaduna, Nigeria of which 80 (20%) were sarcoma. Sarcomas may appear at any age, the earliest reported being Gallagher et al³⁰ in a 16 months old baby, Adebayo et al³ reported in one 24 months old baby, while Hoffman et al²⁸ reported one in patient 84 years old. It tends to affect considerably younger group than that of carcinomas. Male are slightly more affected than female by jaw and orofacial soft tissue sarcoma. According to Yamaguchi et al⁵ mean age was 42 years; male to female ratio was 3:1. The median age was 46 years and male to female ratio was 2:1 in 36 head and neck soft tissue sarcoma reviewed by Rabindra.¹² 31.3 ±14.1 years were mean age and male to female ratio were 2.3:1, reported by Pandey.²⁶ Clinical presentation of orofacial sarcoma depends upon the histological types and location of the tumor. Adebayo et al³ reported that orofacial sarcoma presented with symptoms such as swelling (100%), pain (54%), and tissue ulceration (26%). Pandey et al²⁷ reported that symptoms of their patients were progressive swelling

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or mass which was present in all cases, painful in three (13.6%), painless in the rest, facial nerve palsy was present in one case and bleeding was present in another. The median tumor size was 5.5cm (range 0.6-13cm) according to Robindra et al.¹² Any type of sarcoma can affect the oral tissue. Sarcoma in the maxillofacial area has wide variety of histological types. Weber et al²¹ reported that the most common soft tissue sarcoma occurring in the head and neck area is rhabdomyosarcoma (RMS), followed by malignant fibrous histiocytoma (MFH), fibrosarcoma and neurofibrosarcoma. Osteosarcoma is regarded as the tumor most frequently manifested in the bones and the lower jaw⁶, chondrosarcoma one half as frequent as the osteosarcoma but twice as common as Ewing's tumor.²⁹ Adebayo et al³ found maxillofacial sarcoma as osteosarcoma (28%), chondrosarcoma (17%), rhabdomyosarcoma (12%) and fibrosarcoma (12%). Hard tissues are more affected (72%) than soft tissue (11%) by sarcoma in maxillofacial region.³ The primary sites of sarcoma according to Yamaguchi⁵ included the maxilla, maxillary sinus, mandible, buccal mucosa, temporomandibular fossa and submandibular region. Sarcomas grow rapidly, are invasive destroy surrounding tissues and usually spread by the blood stream. Their occurrence result in considerable morbidity and mortality. Head and neck sarcomas have high mortality rate with a high risk of recurrence. According to Penel et al²⁵ 2 years overall survival was 71% and the 5-year overall survival was 52.3%. Rabindra et al¹² reported overall survival was 49% at 5 year. In Bangladesh, few reports of maxillofacial sarcoma have been published but detailed studies are lacking. So this present study was designed to evaluate the clinicopathologic characteristics of sarcoma in maxillofacial region which would shed some more light on this lesion.

Materials & Methods:

The study was carried out in the department of Oral and Maxillofacial Surgery, Dhaka Dental College Hospital from March 2007 to February 2011. Patients admitted with maxillofacial sarcoma irrespective of age and sex were selected for the study. Sample size of the study was 20, of them 13 were male and 7 cases were female. Histopathological types of the sarcomas were analyzed to indicate the numbers that occurred; and also the pattern of occurrence according to age, gender, site and clinical presentation. A standardized structured data collection sheet was used to collect necessary information of the subject group. Data sheet included all of the variables

regarding to the study. Data were screened and cleaned for any discrepancy. After cleaning data were entered into template of SPSS@16 software. Descriptive statistics were generated to see the distribution of baseline characteristics of the patient.

Results:

Table-I
Distribution of orofacial sarcoma in maxillofacial Region.

Type	Number	Percentage (%)
Squamous Cell Carcinoma	110	79.71
Maxillofacial Sarcoma	20	14.49
Other Malignancies	8	5.79
Total	138	100

There were 138 cases of malignant neoplasm of the oral and maxillofacial region within the study period of which 20 (14%) were sarcoma as compared to 110 (80%) cases of squamous cell carcinoma.

Table-II
Distribution of the respondents by Age

Age in group	Frequency	Percent
Less than 15 years	4	20.0
15 to 30 years	5	25.0
30 to 45 years	7	35.0
45 to 60 years	1	5.0
More than 60 years	3	15.0
Total	20	100

Mean 34.3±20.3, Min-3.50 and Max-70.00

Majority of the respondents (35%) were in the age between 30 to 45 years, 5(25%) were aged between 15 to 30 years, 4(20%) were less than 15 years of age 3 (15%) were more than 60 years of age and only 1(5%) were found at the age between 45 to 60 years. Mean age was 34.3±20.3, min-3.5 years and max-70 years.

Table-III
Distribution of the respondents by sex

Histopathological diagnosis	Male	Female	Total
Osteosarcoma	4(20%)	2(10%)	6(30%)
Malignant-fibrous histiocytoma	2(10%)	1(5%)	3(15%)
Fibrosarcoma	3(15%)	1(5%)	4(20%)
Chondrosarcoma	1(5%)	-	1(5%)
Ewing's sarcoma	2(10%)	2(10%)	4(20%)
Rhabdomyosarcoma	-	1(5%)	1(5%)
Others	1(5%)	-	1(5%)
Total	13(65%)	7(35%)	20(100%)

Among the 20 respondents 13(65%) were male and rest 35% were female.

*Others- Ameloblastic fibrosarcoma

By the site, most 14(70%) of the lesion were found at mandible, 2(10%) at maxilla and 1(5%) each at the site of buccal mucosa, oral cavity, sinus and TM joint.

The above table shows that 12(60%) diameter of the lesion were < 25 sq.cm, 6(30%) were size between 25 - 50 sq.c.m and only 10% were found > 50 sq.c.m in diameter. Mean diameter was 28.4±27.0 and min-5 sq. cm and max- 120 sq. cm.

Among the 20 respondents majority 6 (30%) were diagnosed as Osteosarcoma, 3(15.0%) were found

Malignant fibrous histocytoma, 4(20%) were diagnosed as fibrosarcoma, 1(5%) was chondrosarcoma, 4(20%) were Ewing's sarcoma, 1(5%) rhabdomyosarcoma and 1(5%) was other and all 20(100.0%) had a complaint of swelling, 14(70.0%) were presented with pain, 1(5.0%) with nasal bleeding, 6(30.0%) with ulceration, 2(10.0%) had paresthesia, 3(15.0%) had toothache, 3(15.0%) were presented with loose tooth, 4(20.0%) were limitation of mouth opening and 1(5%) had other clinical feature.

Table-IV*Distribution of the Lesion in Maxillofacial region*

Histopathological diagnosis	Mandible	Maxilla	Buccal mucosa	Oral cavity	Sinus	TM joint
Osteosarcoma	4	2	-	-	-	-
Malignant fibrous histocytoma	3	-	-	-	-	-
Fibrosarcoma	3	-	1	-	-	-
Chondrosarcoma	-	-	-	-	-	1
Ewing's sarcoma	3	-	-	-	1	-
Rhabdomyosarcoma	-	-	-	1	-	-
Ameloblastic fibrosarcoma	1	-	-	-	-	-
Total (20)	14(70%)	2(10%)	1(5%)	1(5%)	1(5%)	1(5%)

Table-V*Distribution of the respondents by Diameter of the lesion*

Diameter of the lesion (Sq.cm)	Frequency	Percent
< 25 sq.c.m	12	60.0
25 - 50 sq.c.m	6	30.0
> 50 sq.c.m	2	10.0
Total	20	100.0

Mean 28.4±27.0, Mini-5 sq. cm and Max-120 sq. cm

Table-VI*Distribution of the lesion by Histopathological findings and Clinical features*

Clinical feature	Osteosarcoma	Malignant fibrous histocytoma	Fibro sarcoma	Chondro sarcoma	Ewing's sarcoma	Rhabdomyo sarcoma	Others	Total
Swelling	6(100%)	3(100%)	4(100%)	1(100%)	4(100%)	1(100%)	1(100%)	20
Pain	5(63.3%)	3(100%)	2(50.0%)	1(100%)	3(75.0%)	0(.0%)	0(.0%)	14
Ulceration	1(6.7%)	1(33.3%)	2(50.0%)	1(100%)	0(.0%)	1(100%)	1(100%)	06
Paresthesia	1(6.7%)	0(.0%)	1(25.0%)	0(.0%)	0(.0%)	0(.0%)	0(.0%)	02
Nasal Bleeding	0(.0%)	0(.0%)	0(.0%)	0(.0%)	1(25.0%)	0(.0%)	0(.0%)	01
Toothache	0(.0%)	1(33.3%)	2(50.0%)	0(.0%)	0(.0%)	0(.0%)	0(.0%)	03
Loose tooth	0(.0%)	1(33.3%)	0(.0%)	0(.0%)	1(25.0%)	0(.0%)	1(100%)	03
Limitation of mouth opening	1(6.7%)	0(.0%)	1(25.0%)	1(100%)	0(.0%)	1(100%)	0(.0%)	04
Others	0(.0%)	0(.0%)	1(25.0%)	0(.0%)	0(.0%)	0(.0%)	0(.0%)	01
Total	6 (30%)	3(15%)	4(20%)	1(5.0%)	4(20%)	1(5.0%)	1(5.0%)	20(100%)

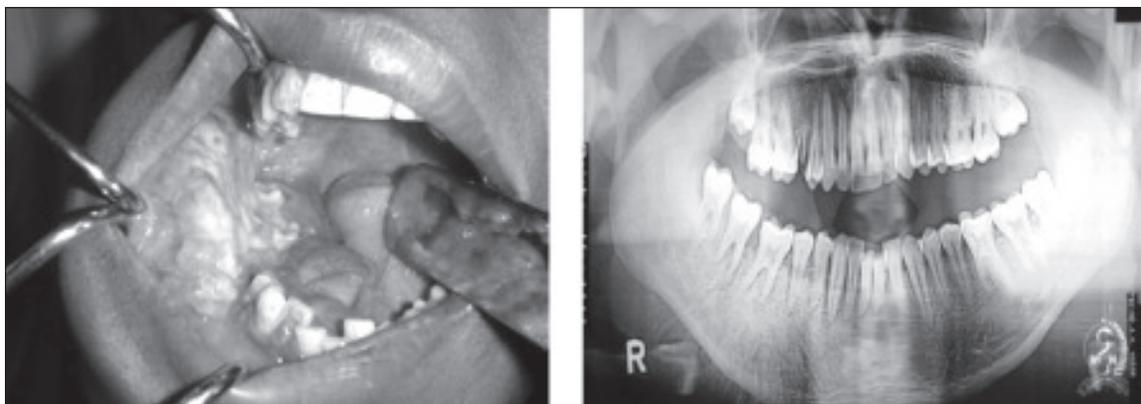


Fig: Clinical & Radiological image of a case of Osteosarcoma.

Discussion:

The incidence of maxillofacial sarcoma is unknown. Budhy et al¹ examined 994 histopathological specimens of maxillofacial malignancies from East Java, Indonesia. They found 42 (4%) sarcomas in their sample. Adebayo et al³ examined 406 maxillofacial malignancies from Kaduna, Nigeria of which 80(20%) were sarcoma. Sadat et al² reviewed 139 cases of oral malignancies from Bangladeshi population among which sarcoma were 12.9%. In this study out of 138 maxillofacial malignancies seen at our centre within the last 4 years, 20 (14%) were sarcomas. However we believe that a yearly sarcoma incidence of 5 cases in the estimated population served by our center shows rarity of this lesion in the population.

The most common sarcoma is controversial. It depends on age group, site and possibly racial factors. According to Soule et al¹⁴ rhabdomyosarcoma is the commonest oral and maxillofacial sarcoma of childhood while in adults; osteogenic sarcoma (osteosarcoma) is predominant.⁷ It is the belief of Miller and Dalager¹⁶ that rhabdomyosarcomas are commoner among Caucasians than Negroes speculating a genetic factor in the Caucasoid stock.

In Ibadan, Nigeria, osteosarcoma accounts for 37% of sarcomas over a 15 years period from the report of Daramola et al.¹⁵ Yamaguchi et al⁵ reviewed 32 cases sarcomas involving the oral and maxillofacial region of them 9 (28%) cases of osteosarcoma. As with other reports, the most common sarcoma in our series was osteosarcoma (30%) (Table-VI). This is similar to the 30% incidence from Pretoria, South Africa.¹⁷ Among 6 cases of osteosarcoma found in our series, there were fewer females than males, ratio was 1:2. This is slightly higher than male to female ratio of 5:3 found from 8 cases.⁸

Unlike in the rest of the human body where osteosarcoma occurs mostly in the 2nd decade, that of the maxillofacial

region occurs in older persons (mean age 38years) from the study of Harrison and Lund.²² Six patients (30%) in our series with osteosarcoma were between 18-60 years old showing its bias for occurrence in adults (Table-II).

Sarcomas can originate in any part of the body but certain types show a predilection for some parts of the maxillofacial region. Considering specific neoplasm's, osteosarcoma occurs more in the maxilla 8(58%) than the mandible 6 (42%) from the work of Mardinger et al.²³ Though soft tissue involvement by the tumor can occur it is rare. Table 4 showed that among 6 cases of osteosarcoma more occurred in the mandible (66%) than maxilla (34%).

Sarcomas could be detected during routine examination for other conditions or due to non-specific symptoms. Since cure of orofacial malignancies is enhanced by early detection and initiation of adequate treatment, the dental surgeon or medical practitioner has a vital role to play in early detection particularly at the asymptomatic stage through opportunistic screening.¹⁸ The presenting features of sarcomas are non-specific and depend on tumor location, size, rate of growth, duration and the level of cancer awareness of the individual.¹⁸ Cases of osteosarcoma reported by Doval et al⁸, presented as swellings of the jaws while a few had pain, tooth loosening, derangement of teeth and ulceration. In our 6 patients main features encountered were swellings (100%), pain (83%), ulceration (16.7%), paresthesia (16.7%) and limitation of mouth opening (16.7%) (Table-VI).

Chondrosarcoma is less common than osteosarcoma from reports, Rafindadi and Ayuba⁷, had 12% incidence of chondrosarcoma. This is found higher (5%) than our study (Table-VI). The lesion occurs more in males than females, ratio 2:1,^{31, 32} but in our study only one case was found out of 20 maxillofacial sarcomas (Table-VI). In the mouth and jaws, the lesion afflicts younger persons than in other

parts of the body.²² In this study, one chondrosarcoma male patient was 65 years old. Primary chondrosarcoma affects the maxilla more than the mandible (Adekeye et al and Arlen et al). The site of the lesion in our patient was mandible and presented with painful swelling and limitation of mouth opening. Larger series are needed to ascertain the actual site predisposition for chondrosarcoma among our population.

Among three forms of sarcoma reported by Rafindadi and Ayuba⁷, fibrosarcoma accounted for 38%. This is much higher than our result. There was sexual bias in 4 fibrosarcomas of the maxillofacial region found in our series which contrasts with the slight female predominance by a ratio of 1.3:1 in the Dutch report.⁹ Fibrosarcomas occur in the soft tissues of the maxillofacial region followed by the maxillary sinus, other paranasal sinuses and the nasopharynx.¹⁹ Slootweg and Müller⁹ considered the lesion in the jaws of Dutch population and found more in mandible (n=5) than maxilla (n=2). Our 4 cases were distributed between the mandible 75% and buccal mucosa 25%. Harrison and Lund²² noted that difficulties occur in distinguishing maxillary lesions on the basis of origin from soft tissues such as periosteum or intraosseous. The large sizes of tumors seen in this series emphasize this problem. Slootweg and Müller⁹ found that out of seven cases of fibrosarcoma, painful swelling occurred in 3 persons with tooth loosening, pathological fracture, trismus and paraesthesia of the lower lip in one case each. Out of four cases in our study all are presented with swelling with or without pain, ulceration, toothache, paresthesia and limitation of mouth opening.

Tumor rarity makes incidence of malignant fibrous histiocytoma among other oral and maxillofacial sarcomas difficult to find. It accounted for 15% of sarcomas in this study. There are slightly more males than females with malignant fibrous histiocytoma in the maxillofacial region.²⁰ We had three patients among them two were male and one were female. Harrison and Lund²² stated that the lesion occurs mostly in the 6th decade. Cases of malignant fibrous histiocytoma were between 13 and 54 years old (mean 34).²⁷ In our series, one patient was 9 years, one was 45 years and another was 70 years old (mean 41) demonstrating its predilection for older people unlike rhabdomyosarcoma. Malignant fibrous histiocytoma was found more in the hard tissues such as bone than in soft tissues.²⁶ Out of three lesions they reported, two were in the jaws while one occurred in the scalp. Our three cases were in the mandible (n=3) (Table-IV). In this series, the lesion presented with swelling, pain, ulceration, and

derangement of teeth. These are not dissimilar to features of malignant fibrous histiocytoma in another report.

Ewing's sarcoma is a highly malignant tumor which develops from medullary tissue of bone. It accounts for 4 to 5 percent of all primary bone tumors.²⁹ Ewing's sarcoma is the second most common malignant bone tumor of childhood and adolescent, yet it is a rare tumor. Less than 3% of all Ewing's sarcoma originates in the maxillofacial region, usually involving the mandible, 90% occur in the first three decades of life and males are more often affected than females. Clinical symptoms such as swelling, pain and sensory disturbances are rather unspecific and sometimes be misleading.¹⁰ We had 4 cases of Ewing's sarcoma patients of them 2 cases were male and 2 cases were female. Three cases were involved in the mandible and rest was in the sinus. Patients age range was 3.5 years to 45 years (mean age was 25 years), we had two patients those age were above 40 years which is similar to Proamate et al¹³ study. Our Ewing's sarcoma patient's main clinical presentation was swelling and pain with ulceration, nasal bleeding and loose tooth.

Rhabdomyosarcomas can occur at any age but the lesion is commonest in the first decade of life making it the commonest maxillofacial sarcoma of childhood.¹⁴ Five cases were recorded in India by Pandey et al²⁷, whose mean age was 16 years (range 4-33years) with 80% in the 1st and 2nd decades. We had one case of rhabdomyosarcoma which involved in the oral cavity and patient age was 12 years, clinical presentation was swelling, ulceration and limitation of mouth opening.

Muller Susan et al²⁴ reported five cases of ameloblastic fibrosarcoma, the malignant counterpart of the ameloblastic fibroma, is a rare odontogenic tumor characterized by benign epithelium and malignant fibrous stroma. The mean age of the patients was 14.6 to 22 years. Park Hae Ryoum¹¹ reported a highly malignant ameloblastic fibrosarcoma located in the right retromolar region. The patient was 17-years old male and his complaint was painful mass in this region. Clinically, the patient had an exophytic strawberry-appearing mass in the right retromolar area with loosen the lower right second molar. We had a male case of ameloblastic fibrosarcoma that was involved in the right retromolar area, presented with painless swelling with loose tooth; size was about 3×3cm.

Especially in developing countries such as Bangladesh, poverty, ignorance about medical problems and poorly developed medical infrastructure contribute to morbidity and mortality from malignant conditions such as sarcomas and carcinomas. While patient management is improved

with adequate diagnostic and treatment facilities, health care must be accessible for the population to benefit. To improve the patient survival and freedom from recurrence, there is need for increased cancer awareness and funding for the health sector in Bangladesh. Also, regional cancer treatment centers are necessary to cope with the prevalence of malignancies in our environment.

Conclusion:

In Dhaka Dental College Hospital, sarcomas account for 14% of all maxillofacial malignancies with the osteosarcoma as the predominant type. Most affected people were in the fourth decade of life. Surgery was the main modality used for treatment while some patients had no treatment due to self discharge and late presentation. The need for improved medical awareness and upgrading of infrastructure was stressed.

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