Original Article

Osteosarcoma of the Jaws Seen in a Nigerian Tertiary Health Institution

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Abstract

Introduction: Jaw morbidity and mortality among African descend are rarely caused by bone tumors of malignant origin. Osteosarcoma being the most frequent primary malignant bone tumor account for 5.3%–9.5% of malignancies in some part of Africa, Nigeria, included. Its late occurrence and higher survival rate helps to differentiate it from that of a long bone and other locations. **Patients and Methods:** This is a retrospective study of patients seen at the Oral and Maxillofacial Clinic between January 2003 and December 2015. **Results:** Osteosarcoma constituted about 3% of all tumor seen within the study period with a high male dominance (60%). The mean age was 31.3 years, and the mandible was 74.3% more affected than the maxilla. **Conclusion:** Osteosarcoma of the jaws remains a challenge to the oral and maxillofacial surgeon in this part of the world.

Keywords: Jaw, malignancy, maxillofacial, oral, osteosarcoma, surgery

Introduction

Osteosarcoma is the most frequent primary malignant bone tumor; accounting for 5.3% of all malignancies in Kenya,[1] 6% in Southwestern Nigeria,[2] and 9.5% in Northwestern Nigeria.[3] Its late occurrence and higher survival rate helps differentiate it from that of a long bone and other locations.[4] The lesion is aggressive and characterized by the formation of osteoid tissue or immature bone.[5,6] It affects men more than women and peak second, third, and fourth decades.[5]

Osteosarcoma is generally classified into primary and secondary types based on the etiology; although numerous histopathologic forms have been described. The primary type formerly occurs in younger patients, while the secondary types are usually associated with patients more mature in age.[4] The etiology is unknown, but the primary type may be due to environmental factors or genetic disorder. The secondary type occurs in patients with preexisting Paget's diseases, fibrous dysplasia, and history of irradiation or trauma.[4,5]

The last series of osteosarcoma was published from our center in 1987.[7] This report aims at the trend and pattern of this lesion over 12 years' period and highlights the challenges of management in our tertiary care facility.

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Patients and Methods

A retrospective study of patients seen at the Oral and Maxillofacial Clinic of Ahmadu Bello University Teaching Hospital, Zaria, Nigeria, between January 2003 and December 2015 was conducted. Patients' record examined included case files, operation records, and histopathologic results. Cases of osteogenic sarcomas were selected from all cases of sarcomas which were selected from other malignancies and analyzed for age, sex, tumor site, clinical features, radiographic reports histopathologic types, treatment, and follow-up reports. Tumor staging and grading were not done routinely within the study period.

Results

A total of 98 (8.4%) cases of sarcoma out of 1170 tumor of the orofacial region were seen within the period and osteosarcoma constituted 35 (3%) cases. There were more

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males (*n* = 21, 60%) than females (*n* = 14, 40%). The age range was between 7 and 67 years with a mean of 31.3 years. The majority of patients were in the third decade of life followed by the second then fourth decade. The rest is as described on the table. The ratio of mandible to maxilla was 2.9:1 (mandible 74.3% and maxilla 25.7%). The time from the first symptom to presentation was between 3 and 18 months. Clinical features were swelling (*n* = 35, 100%), pain (*n* = 21, 60%), ulceration (*n* = 12, 34.3%), and previous tumor (*n* = 1, 2.8%). No predisposing factors such as irradiation, chromic oxide, implant treatment, Paget's disease of the bone, or trauma. One patient, however, developed the osteosarcoma from a long-standing fibrous dysplasia.

It was recorded that spread in the neighboring tissues through the cheek to the skin; from the mandible through the origin of the tongue, to the lateral pharyngeal space and tonsillar fossa as shown in Figure 1a, or from the condylar and coronoid processes into the zygoma as shown in Figure 2, orbital floor and base of skull. Maxillary lesions invaded the zygoma, temporal bones, and the pterygoid fossa. Figure 1a shows a mandibular lesion with extension to the pterygoid fossa. And 1b shows extension to the left with the temporal region spared.

Radiographic description was not documented in about 77.1% of patients. The rest (8, 22.9%) were radiolucent (*n* = 3, 37.5%), radiopaque (*n* = 4, 50%), or mixed type (*n* = 1, 12.5%). The predominant histological subtype was osteoblastic osteosarcoma (*n* = 28, 80%), followed by chondroblastic osteosarcoma (*n* = 5, 14%), fibroblastic osteosarcoma (*n* = 1, 3%), and mixed type (*n* = 1, 3%).

The treatment rendered were surgery (*n* = 19, 54.3%) and surgery combined with chemoradiation (*n* = 15, 42.9%), chemoradiation only (*n* = 15, 42.9%) while the rest declined treatment (*n* = 5, 14.3%).

Patients treated using surgery alone had mandibulectomies (*n* = 14, 4%) which were either resection with disarticulation (*n* = 9), hemimandibulectomy (*n* = 1), subtotal mandibulectomy (*n* = 3), and total mandibulectomy (*n* = 1). More females (*n* = 11, 57.9%) were operated than males (*n* = 8, 42.1%). The maxillary (*n* = 5, 26%) procedures were four total maxillectomies (*n* = 4) and one extended maxillectomy. Because of spread to the lateral pharyngeal wall, the intubation was generally difficult. Two (10.5%) patients had tracheostomy while one (5.25%) was intubated through fiberoptic laryngoscopy.

The follow-up was from 1 to 60 months and within this period of follow up, 14.3% of cases (5) died. The rest were lost to follow-up except for two that were followed up to 52 and 60 months, respectively. There was a recurrence rate of 47.4% within the first 9 months. Furthermore, patients (*n* = 2) who presented earlier had a better prognosis than their counterpart who presented late.

Discussion

Hard-tissue sarcomas are rare as compared to soft-tissue sarcoma, constituting 3% of total biopsy within the period of the study while soft-tissue sarcoma was 5.5%.[8] In Nigeria, the incidence of sarcoma is not known.[7] Hard-tissue sarcomas are rare in the jaw bone unlike in the long bones.[5]

According to Chaudhary and Chaudhary,[4] males seem to be more commonly affected, but Laskar *et al*.[9] stated that there was equal sex distribution. In our study, there were more males than females in the ratio of 1.5:1. The male predominance observed in this study confirms the literature from other parts of the world[4,5,9,10] including report from Southwest Nigeria.[11] This was however contrary to other reports including our center[7,10] where females were more than males including Kamau *et al*.[1] from Kenya and Chidzonga and Mahomva[12] from Zimbabwe. The difference in sexual predilection from reports indicates that it was improbable that osteosarcoma had any real sexual predilection.

The majority of the patients were in the second (22.8%) and third decades (31.4%) of life [Table 1] and a mean age of 31.3 years with a range between 7 and 67 years. This supports earlier reports of mean age and age ranged of 31.6 years and 17–70 years, respectively in the literature.[1,7,12,13] The mandible was affected more than the maxilla in majority of the literatures from authors in Nigeria and abroad also described the mandible

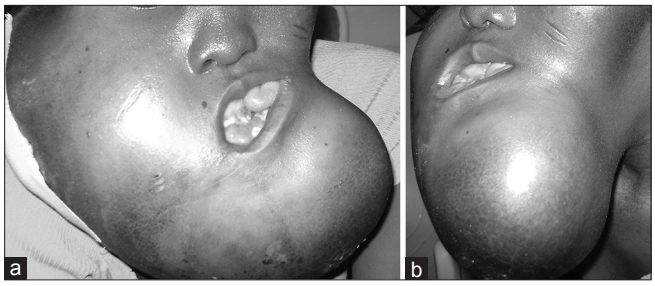

Figure 1: (a and b) a female patient with right recurrent osteosarcoma extending to lateral pharyngeal site and the opposite site mandible. Mouth opening shows the tongue pushed to the side

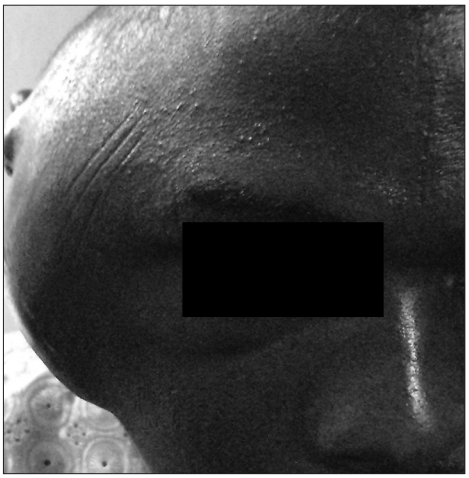

Figure 2: A male patient with osteosarcoma extending from the condylar region to the temporal

Table 1: Age, sex, and site distribution of the 35 cases of osteosarcoma seen in the study

Age (years)	Gender		Site			
	Male	Female	Mandible	Maxilla	Extension to other region of the head and neck	Total
0-10	1	0	0	1		1
11-20	5	4	7	2	Retromolar, lateral pharyngeal wall	9
21-30	7	4	8	3	Zygomatic region, pterygoid fossa, base of skull, tonsillar fossa, tongue root	11
31-40	4	3	5	2	Cheek, condylar region, pterygoid fossa, zygomatic region	7
41-50	1	2	3	0		3
51-60	3	0	2	1		3
61-70	0	1	0	1		1
Total	21	14	26	9		35

as the most implicated site,[7,9,10,13-15] but some reported equal distribution.[4,5] These are contrary to reports by Daramola *et al*.[11] from Nigeria and Kamau *et al*.[1] from Kenya with 57% and 54% of this lesion affecting the maxilla. Some authors reported more maxillary lesions in females,[5] but this was contrary to our findings [Table 1] that show 71% of female had mandibular lesions. The risk factors in osteosarcoma of the jaws are identical with those of long bones. About 5.5%–6.3% of osteosarcomas develop from radiotherapy.[5,16] Trauma also has been viewed as a cause of sarcoma, but none of the patients in this series volunteered such a history. There is, however, a patient (female) who presented with a long-standing fibrous dysplasia of the right maxillary region which transformed to conventional osteogenic sarcoma and this was previously reported our series on fibro-osseous lesions.[17]

Laskar *et al*.[9] and Kammerer *et al*.[5] in their series had no patients with either history of previous bone disease, irradiation nor trauma. Neither was it reported by Adekeye *et al*.[7] and Adebayo *et al*.[13] from the same center. Other possible etiological factors are dental implant placement[18] and exposure to environmental factors such as ionizing radiation and chromic oxide, a radioactive scanning agent.[4]

Osteosarcoma patients have nonspecific features, dependent on tumor location, size, rate of growth, duration, and the level of cancer awareness of the individual.[13] Some patients relate the occurrence of tumor to the previous dental treatment, most commonly, dental extractions. The reason for this is most likely to be rapid growth of tumor seen immediately after tooth extraction, a phenomenon often shown by bone tumors.[4]

Swelling was the most common clinical presentation in 100% of the cases, pain in 60%, and ulceration 34.3%. These are in agreement with previous findings that swelling rather than pain is the most common symptom in osteosarcoma of the jaw.[5,7,9,11,13,16,19] Duration of the symptom was between 3 and 18 months which is within the reported Nigerian standard of 0.5–18 months as given by Adebayo *et al*.[13] Patients who presented early had a better prognosis than those who presented late.

Conventional radiographs are of limited worth in evaluating head- and neck-osteosarcomas because of the superimposed bony structures.[20] However, they can be very useful adjuncts to computed tomography (CT) for maxillary and mandibular tumors when there are extensive metallic tooth fillings or permanent dentures.[20] Majority of our patients could not afford CT, they therefore took plain radiographs. The findings were basically those of bony destruction.

In our study, osteoblastic subtype was the most common form seen (80%) followed by the chondroblastic (14%). This is similar to other previous findings[1,5,6,7,10] but contradict workers who found the chondroblastic and fibroblastic as more common.[9,19,21,22] As prognosis does not seem to be influenced by the histologic subtype,[23] their significances are as yet undetermined. Clark *et al*.[24] had reported that chondroblastic subtype has a better prognosis, but this has not been confirmed by other workers.

Reports of distant metastases are less frequent, but Chaudhary and Chaudhary[4] and Garrington *et al.*, found that distant metastases occurs in approximately 50% of the patients with jaw osteosarcoma. Kammerer *et al*.[5] reported 5% metastasis to the lungs. They also reported high-grade tumors in 87%–90% of osteosarcoma of the jaws compared to previous literature that reported a lesser grade of differentiation than osteosarcoma of long bones. In our study, staging of the lesions was not recorded in the patient records reviewed, meaning that treatment offered were not often standardized. This reflects the inadequacy of retrospective clinical data, but more seriously, could imply that the quality of care given to these patients may not have been adequate and based on the current evidence. The challenge of poor data recording in Nigerian health institutions warrants serious attention in view of its ethical- and medico-legal importance.

Treatment options have been surgery, chemoradiotherapy, employed in accordance with the age of the patient, staging and location of the tumor.[5] The surgery with a wide margin and follow-up has been recommended.[7] The anatomical relationship in the craniofacial area often make it difficult to achieve tumor-free surgical resection margins with a poor prognosis for such patients, especially, that we do not do frozen section in our center. The mandibular procedures included disarticulation due to the posterior location of the lesions. This is in conformity with the findings of Wang *et al*.[10] and Kedar *et al*.[6] treatment options for disease conditions, especially,

malignant lesions in Nigeria are also dependent on the stage of presentation and whether the patient can afford treatment. The absence of universal health insurance coverage reduced social spending for health, widespread poverty and ignorance means that most patients initially consult traditional healers and present to orthodox practitioners when their conditions are advanced, without adequate resources to pay for the treatment. Therefore, there is a need for governments in developing countries like Nigeria to enforce and fund the universal health coverage, especially for life-threatening conditions such as malignancies. Health professionals also need to provide the health education while collaborative efforts have to be made to reduce poverty in the population to improve the health status.

Osteosarcomas of the jaws have a high tendency toward locoregional relapse.[5] They have some distinct features such as older age at presentation, longer median survival, rare locoregional lymphatic node metastases, and local recurrences which are difficult to control, typically leading to reduced quality of life and mortality.[4,5]

The challenges of patients' faces during return for long-term follow-up visits after treatment has been reported by several workers in our environment. While in the 1st month after discharge, 100% of the patients returned for follow-up, only one patient reported for follow-up to 60 months. Five (26.3%) patients died by the 5th month at which only about 40% of the patients still turn up and at the 9th month, 9 (47.3%) patients have had recurrence. Laskar *et al*.[9] recorded 42% death and 12% recurrence.

Inability to follow-up patients did not permit us to assess the prognosis. Furthermore, the cost of treatment may have had an influence on the options available as radiotherapy and chemotherapy remain very expensive modalities, unaffordable to most patients seen in this study center, majority being peasant farmers. These patients patronize the traditional healers initially, only reporting to the hospital after they have exhausted the limited resource.

Conclusion

Osteosarcoma of the jaw remains a challenge to the oral and maxillofacial surgeon in this part of the world, due to the advanced stage at which they present and treatment even though radical, is not curative. In a few cases, the surgery has been curative as they presented quite early.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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