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Research Article

Incidence of Malignant Bone Tumors and Short-Term Functional Outcome of Amputation

Hassan Mohammed Hassan Elbahri[1], Osman Mohamed Mohamed Ahmed[2], Mohammed Mubarak Mohammed Ahmed[3], Dafaalla Abdelmoneim Dafaalla Salih[4] and Hozifa Mohammed Ali Abd-Elmaged[5]*

[1]Associated Professor of Orthopedic Oncology, Department of Orthopedic International University of Africa - Khartoum, Sudan

[2]MD Orthopedics and Trauma, Sudan

[3]MD Orthopedics and Trauma, Ibrahim Malik Teaching Hospital, Sudan

[4]Medical Student at Al-Zaiem Al-Azhari University, Khartoum, Sudan

[5]Assistant Professor of Orthopedic, Department of Orthopedic Al-Zaiem Al-Azhari University- Khartoum, Sudan

***Corresponding Author:** Hassan Mohammed Hassan Elbahri, Associated Professor of Orthopedic Oncology, Department of Orthopedic International University of Africa - Khartoum, Sudane.

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Abstract

Background: A malignant bone tumour is a neoplastic growth of tissue in bone which can be benign or malignant. Amputation is the removal part or all of a limb. When used to treat cancer, amputation removes the limb part with the tumour, some healthy tissue above it, and everything below it. The prognosis depends on the type of tumour. The outcome is expected to be good for people with benign tumours, although some types of benign tumours may eventually become malignant.

Methods: Observational descriptive cross-sectional hospital-based study was conducted in Ibrahim Malik Teaching Hospital in Khartoum State in the period from January 2018 to September 2018. Data was collected using a datasheet. Data entered and analyzed using SPSS version 25.0.

Results: This study covered 50 study participants, most of them (96%) were less than 40 years of age. Our study found that only (16%) of the study participants were fully aware of their disease while (40%) were not aware at all. Furthermore, there was a delay between the presentation and starting the treatment among most of them (90%) while (82%) had tried other traditional or non-formal methods of treatment. Concerning the type of tumour, most of the study participants (84%) were diagnosed with osteosarcoma, while a small proportion of them was diagnosed with Chondrosarcoma (6%), Ewing sarcoma (6%), and Admantinoma among only (4%). The study found that the total number of patients with primary bone sarcoma were 65 patient from January to June 2018(hospital records) so the incidence of amputation at that time was 76%. Regarding the management applied, all study participants were amputated, of which (56%) had chemotherapy and (12%) had radiotherapy. The analysis found that there is a significant association between the level of education and the delayed presentation (pvalue < 0/001).

Conclusion: A high awareness of malignant bone tumours are essential to avoid diagnostic delays. Some might even be dangerous.

Keywords: Malignant Bone; Amputation; Chondrosarcoma; Ewing sarcom; Admantinoma

Introduction

A bone tumour is a neoplastic growth of tissue in bone, Which can be malignant or benign abnormal growth. The average five-year survival after being diagnosed with bone and joint cancer is 67%. The bone tumour can be classified as Primary tumours which originate in bone or from bone-derived cells and tissues, and secondary tumours which originate in other sites and spread (metastasize) to the skeleton(Carcinomas of the prostate, breasts, lungs, thyroid, and kidneys are the carcinomas that most commonly metastasize to bone). Secondary malignant Bone tumours are estimated to be 50 to 100 times as common as primary bone cancers.

Usually, bone tumours have no specific symptoms. A person may go weeks, months, and sometimes years before seeking help; the pain increases with the growth of the tumour. Additional symptoms may include fatigue, fever, weight loss, anaemia, and/or unexplained bone fractures. Many patients will not experience any symptoms, except for a painless mass. Some bone tumours may weaken the structure of the bone, causing pathologic fractures.

Bone tumours comprise 0.7% of all cancer diagnoses worldwide. For all ages, they have an age-standardized rate of 8 per 1,000,000 persons per year in males and 6 per 1,000,000 persons per year in females. Primary bone cancer is rarely diagnosed in children before the age of five or adults over the age of sixty. Incidence of osteosarcoma increases with age until a peak in late childhood or adolescence around the time of puberty. A second less pronounced peak occurs in older adults aged more than 65 years. Ewing sarcoma has an incidence peak in the late teenage years, whilst chondrosarcoma peaks in older adults (aged > 65 years).

In Sudan, the first National Population-based Cancer Registry (NCR) stated that bone tumours are considered one of the uncommon types of cancers. But generally, the bone cancer incidence data was behind portraying a cancer picture similar to that of the developed world.

Primary benign and malignant bone tumours are grouped in 15 different categories, including cartilage, fibrogenic, fibrohistiocytic, estrogenic, notochordal, hematopoietic, giant cell, lipogenic smooth muscle, vascular, and neural tumours, Ewing sarcoma/primitive neuroectodermal tumour, miscellaneous tumours and lesions, and joint lesions.

Management of bone tumours is highly dependent on the type of tumour which includes

- **Chemotherapy and radiotherapy:** Effective in some tumours (such as Ewing's sarcoma) but less so in others (such as chondrosarcoma).
- **Medication:** Such as non-hormonal bisphosphonates, Metatron, and Generic Strontium Chloride Sr-89 Injection UPS.

Surgical management.

Amputation

Amputation is the removal of all or part of an extremity or limb. This surgical procedure may be used to remove malignant bone tumours from the arm or leg. Amputation for bone cancer is usually reserved for cases in which the limb would otherwise be left without good function. When amputation is used to treat cancer, the surgery removes the limb with the tumour as well as healthy tissue above it. The orthopaedic surgeon will use MRI scans to examine the tissue to help decide how much of the limb needs to be removed Muscles and skin will be formed around the remaining bone so that an artificial limb can be used.

Amputation indicates that Surgical treatment of primary bone malignancies requires extensive resection, massive tumours invading vessels and nerves, resection of which would leave the limb nonfunctional, and if conservative resection is impossible.

Types of Amputation are Lower Limb Amputation (Syme, below knee, above knee, Hip disarticulation, and Hemipelvectomy which is the most radical type), Upper Limb Amputation (below elbow, upper elbow, Shoulder disarticulation, Forequarter).

The prognosis depends on the type of tumour. The outcome is expected to be good for people with noncancerous benign tumours, although some types of benign tumours may eventually become cancerous malignant. With malignant bone tumours that have not spread, most patients achieve a cure, but the cure rate depends on the type of cancer, location, size, and other factors. Throughout this context, this study aims to study the amputation following bone tumours in terms of factors influencing the possibility of amputation, indications, prevalence and the outcome according to MSTS scale among patients attending the department of orthopaedic and oncology, Ibrahim Malik Teaching Hospital, Khartoum state, Sudan, from January to September 2018.

Methodology

Study design

Observational, descriptive, cross-sectional hospital-based study.

Study area

Orthopaedic oncology unit at Ibrahim Malik Teaching Hospital which is a public hospital that shoulders all cases of musculoskeletal tumours, soft and bone, benign, malignant and trauma.

Study duration

The study was conducted within the period from January to September 2018.

Study population

This study covered all patients who attended the study area within the study period

and who fulfil the following criteria.

Inclusion criteria

Patients underwent amputation following malignant bone tumours.

Exclusion criteria

- Benign tumours
- Other modalities of treatment such as Limb salvage surgery
- Amputation following complicated limb salvage procedures such as infection.

Sample size and technique

The total coverage method was applied. All cases that fulfill the study population criteria and recruited within the study area and period.

Data collection tools and methods

The data will be collected through a datasheet. It will cover all patients, influencing factors, indication, rate and the overall short outcome for all study participants recruited under the study.

Study variables

The study variable can be classified as shown in the following table

Main Categories	Categories	Variable
Independent	General background	Age
		Sex
		Residence
		Occupation
		Socioeconomic status
	Influencing factors	Awareness of the patients
		Delay after symptoms
		Tumour type
	Indications	Large tumour invading neurovascular structure
		Affecting the neurovascular bundle
		Nonfunctioning limb
		Unresponsive for chemotherapy
		Other indication
Dependent	Management	Radiotherapy
		Chemotherapy
		Others
	Prevalence of amputation	Amputation
		Type
	Outcome Musculo skeletal	Tumour Society
		(MSTS) Rating Scale

Table 1

Outcome Measurement

Musculoskeletal Tumor Society (MSTS) Rating Scale

The MSTS Rating Scale is a used functional instrument which was developed in 1983 and later modified by the MSTS in 1993. It is composed of six items, including pain, function, emotional acceptance, use of any external support, walking ability, and gait alteration. Each item was rated on a scale of 0 to 5. The total score ranges from 0 to 30, with higher scores indicating better function. The TESS questionnaire is a self-administered questionnaire evalu-

ating functional difficulties. The TESS questionnaire of the lower extremity was comprised of 29 questions rated on a 5-point scale, including "impossible to do," "extremely difficult," "moderately difficult," "a little bit difficult," and "not at all difficult".

Plan of analysis

Data entry, analysis and presentation

- Data entered, cleaned, and analyzed using SPSS version 25.0
- Descriptive statistics in terms of frequency tables with percentages and graphs.
- Means and standard deviations are presented with relevant graphical representation for quantitative data.
- Bi-variable analysis to determine the associations between the outcome variables and the other relevant influencing factors with the Chi-square test (for categorical variables) and t-test (quantitative variables) statistical tests.
- The relation between quantitative variables is assessed by Pearson correlation coefficients.
- Relative risk is calculated to assess the relationship between influencing factors with the outcome (re-bleeding) with the selected level of significant
- A p-value of 0.05 or less is considered statistically significant.
- Data was represented after analysis in form of uni-variable tables, cross-tabulation (bi-variable tables), figures and narrative illustration.

Ethical considerations

- Written ethical clearance and approval for conducting this research obtained from Sudan Medical Specialization Board ethical Committee.
- Written Informed consent was obtained from all participants.
- Written permission was obtained from the Administrative Authority of Ibrahim Malik teaching hospital.
- Study data/information used for the research purposes only. The privacy issues intentionally considered.

Results

This study covered 50 participants; most of them (96%) were less than 40 years of age, with a male: female ratio of 0.9:1. The study found that more than a third of the study participants were from Khartoum state (34%). Nearly two-thirds of them were from low socioeconomic class (66%) and less than half of them had not been involved in any type of formal education as detailed in table 1. The study found that the rate of amputation at that time was 76% (the total number of patients with bone sarcomas was 65 patients in that period from January to June 2018).

Our study found that only (16%) of the study participants were fully aware of the disease while (40%) were not aware at all. Furthermore, there was a delay between the presentation and starting the treatment among most of them (90%) while (82%) had tried other traditional or non-formal methods of treatment.

Concerning the type of tumour, most of the study participants (84%) were diagnosed with osteosarcoma, while a small proportion of them was diagnosed with Chondrosarcoma (6%), Ewing sarcoma (6%) and Admantinoma among only (4%).

The study found that among patients diagnosed with Osteosarcoma, (60%) of them were affected in the distal femur, (14%) in the proximal tibia, (and 4%) in the hand. Moreover, (6%) of the study participants who had been diagnosed with Chondrosarcoma were affected in their proximal femurs, Admantinoma in their tibia, while Ewing sarcoma affected them in proximal femur (2%), and proximal tibia (2%).

The study found that all patients diagnosed with Chondrosarcoma, Ewing sarcoma and Admantinoma were affected on their left side, while (44%) of patients 24 who were diagnosed with Osteosarcoma had been affected on their right side as shown in table 2.

Regarding the management applied, all study participants were amputated, (56%) had chemotherapy and (12%) had radiotherapy. More than three-quarters of them had been amputated above the knee (76%), hip dislocation (10%), Forequarter Amputation (6%), Below Elbow Amputation (4%) and Below Knee Amputation among only (4%). The most common indications for amputation were Large tumor (90%), affecting the neurovascular bundle (62%), Non-functioning limb (56%), unresponsive for chemotherapy (8%) and Fungating (10%).

The musculoskeletal Tumor Rating Scale (MSTS) was calculated for all patients enrolled in this study, the mean score was 10.6 ± 4.6

and nearly half of the patients in the study (48%) had MSTS less or equal to 10.

In this study, the relationship was studied between the Musculoskeletal Tumor Rating Scale (MSTS) With some other patients' characteristics. The study found that the patient with Admantinoma obtained the highest MSTS score (13), followed by Osteosarcoma (11.1), while Chondrosarcoma had the lowest score (6.3). Moreover, according to the amputation level, the lowest MSTS was obtained with Hip Disarticulation (7.2), followed by Below Knee Amputation [9], and the highest MSTS was by Fore Quarter Amputation (21)followed by Below Elbow Amputation (13).

The study assessed the average Musculoskeletal Tumor Rating Scale (MSTS) with a management line applied. The study found that the lowest MSTS was for the patients who had amputation alone, followed by the patients who 25 performed amputation with chemotherapy (11.2) than, the patients who had amputation with radiotherapy (25).

Discussion

This study aimed to study the amputation following bone tumours in terms of factors influencing the possibility of amputation, indications, prevalence and the overall outcome among patients attending the department of orthopaedic and oncology, Ibrahim Malik Teaching Hospital, Khartoum state, Sudan, from January to June 2018 and covered 50 study participants.

Our study found that most of the study participants (96%) were less than 40 years of age. According to Rhonda S. Robert., *et al.* data, the median age at diagnosis for cancer of the bones and joints was 40 years of age. Approximately 29.0% were diagnosed under the age of 20; 15.4% between 20 and 34 10.5% between 35 and 44, 13.0% between 45 and 54; 11.4% between 55 and 64; 8.3% between 65 and 74, 9.1% between 75 and 84; and 3.5% over 85 years of age. (42). In a Nigerian study by A Ajibade., *et al.* They found that The patients were mostly males and below the age of 40.

Our study found that the male: female ratio of 0.9:1. Another study by Janneke C., *et al.* Was in agreement that in general, there is no significant gender predilection, although some tumours (e.g., Paget's sarcoma, chordoma) show a higher prevalence in males. Furthermore, there was a delay between the presentation and

Demographical background		Frequency	Per cent
Age - years	0 - 10	17	34.0
	11 - 20	22	44.0
	21 - 30	6	12.0
	30 - 40	3	6.0
	> 40	2	4.0
Gender	Male	23	46.0
	Female	27	54.0
State of residence	Khartoum	17	34.0
	Kordufan	9	18.0
	Darfur	8	16.0
	Northern	6	12.0
	White Nile	3	6.0
	Gazira	3	6.0
	River Nile	2	4.0
	South Sudan	2	4.0
Socioeconomic class	Low	33	66.0
	Middle	17	34.0
	High	0	0.0
Education	Illiterate	24	48.0
	Primary	18	36.0
	Secondary	3	6.0
	University or above	5	10.0

Table 2

starting the treatment among most of them (90%) while (82%) had tried other traditional or non-formal methods of treatment. Another study by Erstad DJ., *et al.* (47) claimed that delay before recognition of the bone tumour on radiographs; results of diagnostic imaging; inaccurate diagnoses; type of intervention based on these inaccurate diagnoses; and outcome of survival. There were forty high-grade sarcomas and twenty-eight low-grade sarcomas. (47) another study by Goedhart LM., *et al,* concluded that prolonged delay in diagnosis does not result in lower survival.

Metastatic disease has a pronounced effect on survival. Aggressive tumour behaviour results in shorter delays. Minimizing the patient's delays could be achieved by adopting a lower threshold for obtaining plain radiographs at the prehospital stage. on the other

hand, Erstad DJ., *et al.* Concluded that for patients who ultimately need amputation, timing (early vs. delayed) does not affect survival.

Concerning the type of tumour, our study found that most of the study participants (84%) were diagnosed with osteosarcoma, while a small proportion of them was diagnosed with Chondrosarcoma (6%), Ewing sarcoma (6%) and Admantinoma among only (4%). The study found that among patients diagnosed with Osteosarcoma, (60%) of them were affected in the distal femur, ((14%) in the proximal tibia, (and 4%) in the hand. Moreover, (6%) of the study participants who had been diagnosed with Chondrosarcoma were affected in their proximal femurs, Admantinoma in their tibia, while Ewing sarcoma affected them in proximal femur (2%), and proximal tibia (2%). The study found that all patients diagnosed with Chondrosarcoma, Ewing sarcoma and Admantinoma were affected on their left side, while (44%) of patients diagnosed with Osteosarcoma had been affected on their right side. Another American study by Colin M., *et al.* Was in agreement that the most common diagnoses were sarcoma (55%) and squamous cell carcinoma (25%).

The study found that the rate of amputation at that time was 76% (the total number of patients with bone sarcomas was 65pateint in that period from January to June 2018). Regarding the management applied, all study participants were amputated, (56%) had chemotherapy and (12%) had radiotherapy. Studies (34) explained that the benefits of surgical resection and reconstruction after neoadjuvant chemotherapy include complete removal of all tumour cells, including potentially 40 drug-resistant cells, thus minimizing local recurrence while also providing a stable construct for ambulation or upper extremity function (34).

In our study, the musculoskeletal Tumor Rating Scale (MSTS) was calculated for all patients enrolled in this study, the mean score was 10.6 ± 4.6 and nearly half of the patients under the study (48%) had MSTS less or equal to 10. In another study by Janneke C. etal, they found that the mean MSTS score was 21.5 ± 7.1.then, they concluded that depending on the location and the progressive status of the sarcoma, amputation surgery may still be necessary, despite the cost of compromised body image. Furthermore, they stated that questionnaires and functional performance tests showed no significant difference in functional outcomes between 2 years and 7 years after surgery ($P < 0.05$).

The study had some limitations. The relatively limited number of study participants (50 study participants from one state only) may affect negatively the probability of finding more significant relevant findings among patients with amputation bone tumors in Sudanese hospitals.

Another limitation is follow-up. Some outcomes - such as a long-term outcome or the presence of long-term complications - may need to be followed over time for a longer period. So, a long-term prospective cohort follow-up design may be useful for a more detailed description of the confirmatory practices regarding this important risky group [1-22].

Conclusion

A high index of suspicion and awareness of clinical features of malignant osseous foot tumours are both essential to avoid diagnostic delays.

Patients should be counselled regarding the alternative treatments that are used instead of a doctor's medical treatment. They should know that many of these methods have not been proven to work. Some might even be dangerous.

Provision of rehabilitative and therapeutics services and prosthetic limbs for low-income patients through state health.

Outlying protocol for preparation of the patients pre and post-surgical amputation and provision of centers to support the amputated patients psychologically and financially.

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