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Original Research Article

Factors Contributing to Poor Prognosis in Malignant Bone Tumours in the Paediatric Surgery Department of Donka National Hospital

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Abstract: ***Introduction*:** The objective was to identify certain factors contributing to the poor prognosis of malignant tumours of the limbs. ***Materials and Methods*:** This is a descriptive cross-sectional study with retrospective data collection spanning eight years, conducted in the paediatric surgery department of Donka National Hospital. The parameters studied were epidemiological, diagnostic, therapeutic and evolutionary. ***Results*:** We collected 14 patient files of patients admitted, hospitalised, treated and followed up for malignant bone tumours of the limbs, including 12 cases of osteosarcoma and 2 cases of Ewing's tumour. The frequency of bone tumours compared to other tumours was 4.75%, with a clear predominance of osteosarcoma (85.71%). The average age was 12.5 years (7 to 17 years), with a sex ratio of 1.8. The average time between onset and consultation was 5.6 months (1 to 24 months). The reasons for consultation were dominated by pain and swelling of the limb in all patients. The mode of detection was traumatic fracture in 6 cases (42.86%). The tumour site was the distal femur in 8 cases (57.14%). The left pelvic limb was affected in 8 cases (42.86%). CT scans were performed in 11 cases (78.57%). Biopsies were performed in all patients. Amputation was performed in 8 patients. The 1-year Survival rate was 2 patients (14.28%). ***Conclusion*:** Malignant bone tumours of the limbs are a cause for concern due to delays in diagnosis and treatment. Multidisciplinary consultation involving the authorities and partners could improve treatment.

Keywords: Malignant Tumours, Limbs, Children, Prognosis.

INTRODUCTION

Malignant bone tumours in children remain a relatively rare entity, accounting for only 5% of all paediatric malignant tumours. However, this figure should be weighted, because when considering all bone tumours in childhood, 15% of them are malignant. This means that any tumour in the musculoskeletal system must be examined and treated with this incidence in mind in order to avoid any errors or delays in diagnosis and treatment. For this reason, treatment guidelines have been established involving multiple stakeholders, with surgery being just one link in the chain [1]. The vast majority of malignant bone tumours are primary, mainly represented by osteosarcoma and Ewing's sarcoma (90%), with a slight predominance of osteosarcoma. Each year in France, approximately 150 children are treated for osteosarcoma or Ewing's sarcoma [2]. There are many ways in which these tumours are discovered and many reasons for consultation, which often lead to delays in diagnosis due to the lack of specific clinical signs and the rarity of this pathology. As a result, the diagnosis is rarely considered as a first-line option. For one team, the average time between the onset of symptoms and the final diagnosis varies from three months for osteosarcoma to seven months for Ewing's sarcoma, which obviously affects the prognosis, since metastases can develop during this period, reducing the cure rate accordingly [1].

However, in the African context, this delay in diagnosis is exacerbated by the practice of traditional healers who link the pathology to an evil spell. Patients most often present at an advanced stage of the disease, settling for palliative treatment. Added to this situation is the lack of adequate technical facilities and the lack of resources for parents to pay for additional tests and the cost of treatment.

The management of a bone tumour requires the definition of a possible diagnostic strategy based on an assessment of the lesion, which often presents with common clinical symptoms, but with certain indicative signs such as the patient's age or the location of the

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tumour. Imaging in all its forms can sometimes confirm, or at least approximate, the diagnosis. However, even the most modern imaging techniques cannot provide a histological diagnosis. Biological tests may reveal abnormalities that suggest a diagnosis, but cannot confirm it. This strategy must include an initial surgical procedure, which is essential for an accurate diagnosis of the lesion and determines the options for further surgery : tumour biopsy. Surgical biopsy is preferable because it carries a lower risk of diagnostic error. It must be performed by the treating surgeon in accordance with strict rules. The future of this treatment does not lie in modifying surgical techniques or developing prostheses, but in molecular biology [3].

Thus, the poor prognosis for malignant bone tumours in our context is linked to a lack of financial resources, with its corollaries of delayed diagnosis and treatment, weak technical facilities, and a lack of coordinated strategy and appropriate protocols.

The objective is to study bone tumours in order to identify the factors contributing to the poor prognosis of bone tumours of the limbs in our setting.

MATERIALS AND METHODS

This is a descriptive cross-sectional study with retrospective data collection over 8 years, covering 14 cases of malignant bone tumours. The parameters studied were epidemiological, clinical, therapeutic and evolutionary. We included all patients admitted, hospitalised and treated as a first or second line of treatment for bone tumours of the limbs who agreed to our protocol s for the treatment of osteosarcoma or Ewing's sarcoma, regardless of the stage of tumour progression.

The treatment protocol consisted of a complete clinical examination of the patient upon admission to the paediatric oncology unit, an initial additional assessment including standard X-rays of the affected limb and chest, a CT scan of the limb with thoracoabdominal incidence to look for tumour spread, and exploratory biological tests consisting of a complete blood count, blood urea, creatinine, transaminases, prothrombin time (PT), activated partial thromboplastin time (APTT), and blood typing. The follow-up tests included lactate dehydrogenase (LDH) and blood ionogram. After these various additional tests, which are paid for by the parents, a multidisciplinary team meeting (MDT) is held between the paediatric oncologist, paediatric surgeon, pathologist and radiologist, and sometimes with the Franco-African Paediatric Oncology Group (GFAOP) via Zoom conference. The decision to perform a biopsy is made after a pre-anaesthesia consultation has been approved. Our protocol did not allow neoadjuvant chemotherapy before the pathology results were available.

RESULTS

During the study period, we collected 14 cases of bone tumours ( ), including 12 cases of osteosarcoma and 2 cases of Ewing's tumour. The frequency of bone tumours compared to other tumours was 4.75%, with a clear predominance of osteosarcoma (85.71%) and 13.29% for Ewing's sarcoma.

Table I: Distribution by age group

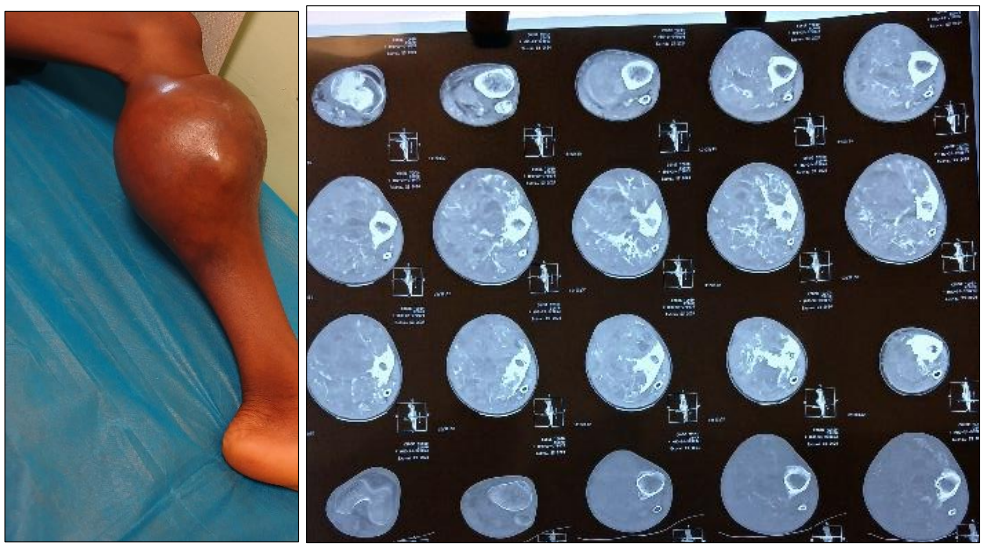
Age	Number of cases	Percentages
7 to 11 years	5	35.71
12 to 16	7	50
17	2	14.29
Total	**14**	**100**

Average age 12.5 years (7 to 17 years).

There were 8 boys and 6 girls, with a sex ratio of 1.8.

Nine patients were from the capital Conakry and five from the interior of the country. The average time to consultation was 5.6 months (1 to 24 months).

The main reasons for consultation were pain and swelling of the limb in all patients. Associated signs were weight loss in 8 patients (57.14%) and physical asthenia in 7 patients (50%). The circumstances of onset or mode of presentation were traumatic fracture in 6 cases (42.86%), traumatic wound in 4 cases (28.57%) and limping in 4 cases (28.57%). The tumour was located in the distal femur in 8 cases (57.14%) and in the proximal tibia in 6 cases (42.86%). The right pelvic limb was affected in 6 cases (42.86%), and the left pelvic limb in 8 cases (57.14%). Twelve out of 14 patients had undergone traditional treatment by bone setters prior to admission. All patients had undergone standard radiography showing either osteolysis, periosteal apposition, or soft tissue calcifications. A CT scan was performed in 11 (78.57%) patients who had pulmonary metastases on admission and 3 (21.43%) during hospitalisation. All patients had satellite lymph node, especially in the inguinal region. Biopsies were performed in all patients. The average time to perform the biopsy was 2 months (1 week to 1 month), the average time to receive the results was 6 weeks (range 3 weeks to 2 months), and the time to chemotherapy was more than 2 weeks after the biopsy. Amputation was performed in 8 patients in the presumed healthy area, and 3 patients underwent debridement surgery. The 1-year survival rate was 2 patients (14.28%). At a mean follow-up of 2 years, no patients are alive.



DISCUSSION

Bone tumours have become increasingly common in Guinea since the creation in 2017 of the paediatric oncology unit, which operates on a multidisciplinary model. Despite this structure, enormous difficulties remain in the management of these tumours. These include a lack of coordination of activities, inadequate technical facilities, the absence of a treatment protocol adapted to local realities, and frequent disruptions in the supply chain for anti-cancer drugs. The lack of accommodation for parents within the referral facility must also be taken into account.

Osteosarcoma and Ewing's sarcoma account for approximately 90% of all malignant bone tumours [4]. 80% occur in children and adolescents, with an average age of onset of 14 years [5]. In our series, we found an average age of 12.5 years. Male predominance is reported in most epidemiological studies, 1.7/1 [6]. According to Philip, cited by Ndour [7], age under 12 years is a factor for poor prognosis.

Osteosarcoma is the most common malignant tumour of the skeleton. It accounts for 15 to 35% of primary malignant bone tumours, depending on the series [5]. French paediatric tumour registries report a standardised rate for the global population of 3.1 osteosarcomas per million person-years. It is estimated that there are 150 new cases per year in France among those under 19 years of age [5].

The average time to consultation was higher in the series by N'Dour in Senegal [7], at 16 months (2 months to 4 years), which highlights the severity of this condition in Africa.

The preferred locations for osteosarcomas are the knee, shoulder and hip [7, 8]. In our series, the location was exclusively the distal femur and proximal tibia, even for the two cases of Ewing's sarcoma. Even in Ndour's series, the knee was the site of origin in 87.5% of cases, with the distal end of the femur being the most common site in 81.25% of cases.

While progress has been made in developed countries in terms of early treatment, this is not the case in sub-Saharan Africa. The first difficulty is the delay in diagnosis, which has a significant impact on treatment. There are several reasons for this delay:

In the African context, traditional beliefs mean that any swelling is mistakenly taken to be coagulated blood, and the first instinct is to go to a traditional healer to have this blood removed. Twelve of our 14 patients had undergone traditional treatment, as confirmed by the scarification marks found during physical examination. The danger of this traditional treatment must be emphasised. These scarifications involve making small cuts in the skin in order to draw out blood, and the risks of bleeding have been demonstrated, as it promotes tumour spread and the risk of decompensated anaemia, significantly altering the patient's general condition [9].

Another factor that explains this delay in diagnosis is the care provided at peripheral health facilities. When traditional treatment does not work, parents often turn to the nearest health facility. At this level, the swelling is often mistaken for simple inflammation or infection, and an X-ray is not always systematically requested or is not always available. The various factors mentioned above mean that most of our patients are seen with a large tumour mass and an often altered general condition, making hospital care more difficult. There are many ways in which the disease is discovered and many reasons for consultation, which often lead to delayed diagnosis due to the lack of specific clinical signs. As a result, the diagnosis is rarely considered as a first option. For one team, the average time between the onset of signs and the final diagnosis varies from three months for osteosarcoma to seven months for Ewing's sarcoma, which obviously affects the

prognosis, since metastases can develop during this period, reducing the cure rate [10].

The diagnosis of primary malignant bone tumours is based primarily on imaging and pathology guided by clinical findings. Imaging must establish the presence of a progressive lesion originating in the bone, define the diagnostic hypotheses and guide the biopsy. Standard X-rays, CT scans, MRI scans and bone scans are useful for optimal local staging. These investigations must be carried out very quickly before the biopsy and be coordinated from the outset by the paediatric oncologist [11]. Currently, attention is focused on digital radiography, which is increasingly used for the quality of the images it provides and the study of adjacent softs tissus [3]. MRI is the gold standard in the study of bone tumours. It is mandatory for any suspicious tumour. It is of great value in diagnosis and surgical strategy. It examine the tumour and gives its spatial location in relation to anatomical landmarks. It provides the best possible examination of the medullary canal and searches for skip metastases. It gives exact measurements of the areas to be resected in length and guides the biopsy. It must always be performed before the biopsy. It allows the reduction or extension of the tumour volume after adjuvant chemotherapy to be assessed. In our series, standard X-rays and CT scans were the most commonly used examinations prior to biopsy for diagnostic guidance and staging. The cost of CT scans was another significant factor in delays in treatment, as once a scan had been requested, some parents were unable to afford it, while those who managed to arrange one had to wait a week or two. CT scans are useful in local diagnosis because they examine the bone structure. They provide a three-dimensional reconstruction of the tumour, which is particularly useful in difficult locations, such as the pelvis. They are the best indication in cases of osteoid osteoma and guide resection. Above all, they are of general interest in staging [3].

Biopsy is a procedure of paramount importance ; it must be performed and successful on the first attempt, as the entire treatment programme depends on it. In our setting, it is one of the most significant factors delaying treatment due to the long turnaround time for results, which is six weeks on average and was equal to two months (three weeks and two months) in nine patients (64.28% of cases).

Once a bone lesion is detected, it must be treated in a relatively standardised manner. Except in special cases where the image is characteristic, the lesion is presumed to be malignant, and the chronology must be respected. The management of bone tumours can only be conceived in a concerted manner, within a multidisciplinary team experienced in this practice. The various links in the chain include all the specialities that will be involved in the diagnostic, therapeutic and rehabilitation stages, including the psychological support that is essential to cope with such an ordeal [1]. In our series, difficulties in accessing chemotherapy were another major factor in delaying the treatment of our patients. Apart from the parents' lack of resources, access to anti-cancer drugs is limited, dictated by constant shortages in pharmacies and insufficient stocks provided by the GFAOPO. On the other hand, it is important to note the lengthy process of delivering these drugs to the port or airport, with the authorities completely unaware of the urgency of the situation.

The 1-year survival rate was 2 patients (14.28%). After an average follow-up period of 2 years, none of the patients are alive. They died either as a result of metastases or tumour recurrence because, between the biopsy, the results of which take a long time to comeback, and the start of treatment, the tumour grows rapidly and metastases develop. We therefore advocate neoadjuvant chemotherapy immediately after the biopsy, which is already performed late, and even before the biopsy results are available. An illustrative case is that of a 17-year-old patient followed up for a granulating wound on the proximal third of the leg with no other clinical or radiological signs, with a delay of 3 weeks before admission. One week after the biopsy, the tumour flared up and radiological signs characteristic of osteosarcoma of the proximal tibia appeared. Given the delay in the biopsy, some tissue samples were sent to the Trousseau Hospital in Paris and the results were returned within a week, confirming a **diagnosis of telangiectatic osteosarcoma**. The sudden onset of dyspnoea led us to repeat the staging assessment, which had previously been unremarkable, and the result was unequivocal: balloon-like metastases and death within a week while he was being prepared for chemotherapy. All of this happened within two months and the results of the local histology were not yet available.

CONCLUSION

Malignant bone tumours in our context mainly consist of osteosarcoma and, in rare cases, Ewing's sarcoma, and their frequency is increasing due to the existence of a paediatric oncology referral unit that operates as part of a multidisciplinary team. However, treatment faces enormous structural difficulties due to the implementation of a policy of early diagnosis, supervision of traditional medicine, training of peripheral health workers and equipping of technical facilities. It is important to emphasise the need to adapt the protocol for the treatment of malignant bone tumours to the local environment, with the involvement of the authorities in creating a village for parents near the paediatric oncology unit and making medicines accessible and free of charge.

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