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Large Conventional Osteosarcoma of the Proximal Humerus in a 13-Year-Old Child: Case Report

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Abstract

Introduction: Osteosarcoma is the most common primary malignant bone tumor in children. It is highly aggressive and has a poor prognosis. A late presentation modifies and makes difficult the management affecting the survival of children. We report the case of a large conventional osteosarcoma in a 13-year-old girl. **Case Presentation:** Adolescent girl admitted for painful swelling of the left shoulder with absolute functional impotence of the thoracic limb and severe anemia. The painful swelling was thought to have been caused by a minor trauma that had occurred six months previously. The patient's general condition was poor, and she presented with a large, shiny, painful mass over the shoulder and upper 2/3 of the left arm, measuring 28 cm long by 28 cm wide and 57 cm in circumference, and a large fistulous axillary adenopathy. CT scan showed a tumour lesion of the left humerus with liver and lung metastases, raising suspicion of osteogenic osteosarcoma. The tumor was classified according to TNM staging: T2N1M1(a + b). Management was modified when uncontrolled bleeding developed. It consisted of an extended amputation of the left thoracic limb. Pathological analysis showed a high-grade conventional osteosarcoma. Quality improvement was obtained for thirty days, followed by the onset of dyspnea. The evolution was towards death at forty days post-operatively. **Conclusion:** Osteosarcoma is a highly aggressive cancer. Delayed treatment leads to a fatal outcome. Early diagnosis is one of the challenges to be met in order to improve survival.

Keywords

Osteosarcoma, Child, Conventional, Case Report

1. Introduction

Osteosarcoma, the most common type of primary malignant bone tumor, is defined by the presence of malignant mesenchymal cells producing osteoid or immature bone. It represents 30% to 80% of primary skeletal sarcomas [1] [2] [3]. It preferentially affects young subjects with a peak in adolescence [1]-[9]. Osteosarcomas of the thoracic limb are rare, with an incidence of 0.18% in the United States [3], with a predominant location at the proximal end of the humerus. Conventional osteosarcoma is the most common type and accounts for 80% of all osteosarcoma cases [8], it is generally high grade and has a poor prognosis. The survival rate varies depending on the country, with a clear improvement obtained by more than 60% 5-year survival in developed countries with considerable progress in therapeutic protocols [1] [3] [8]. Late presentation modifies and makes management difficult, affecting the survival of children [2] [9]. We report the case of a large conventional osteosarcoma of the humerus in a 13-year-old girl with the aim of clarifying the difficulties of diagnostic and therapeutic management.

2. Observation

We report the case of a 13-year-old girl admitted for painful swelling of the left shoulder with absolute functional impotence of the left thoracic limb and severe anemia. The painful swelling of the left shoulder was caused by a minor trauma sustained during a jostle six months earlier, and was treated with tradi-therapy consisting of herbal massage and self-medication with analgesics (paracetamol 500 mg 3 to 4 times a day) and anti-inflammatories (dose not elucidated). The patient's symptoms worsened, with intense, insomniac tugging pain, recrudescing at night, waking her in the second half of the night and not relieved by rest, an alteration in general condition, the progressive onset of absolute functional impotence of the thoracic limb and the appearance of a zone of necrosis in the axillary fossa, leaking blood. The patient could only sleep in a sitting position, with her head resting on the lump. She was the fourth of four siblings, two of whom died of unexplained causes, and one of whom was in apparently good health. On admission, the patient presented with poor general condition, fever 38˚ Celsius: heart rate 112 beats per minute, respiratory rate 24 cycles per minute Height 1.50 metres Weight: 50 kilograms, body mass index at 22.22 kilograms/square meter, no dehydration or undernutrition folds, a voluminous glistening mass with areas of fluctuation and collateral venous circulation of the left shoulder extending to the upper 2/3 of the arm, painful, measuring 28 centimetres in length by 28 centimetres in width and 57 centimetres in circumference (**Figure**

1), with a large fistulated axillary adenopathy leaking fetid fluid mixed with blood and tumour necrosis (Figure 2). Active and passive mobilization was painful and impossible, with no sensory-motor deficit and perceptible distal pulses. Pulmonarily, the thorax was symmetrical with good thoracic ampliation, vocal vibrations and vesicular murmur were diminished in the right hemichamber, with tympany in the upper 1/3, with no sign of respiratory distress. An emergency blood count showed severe hypochromic microcytic anemia, with a hemoglobin level of 3.1 grams per deciliter and a predominantly granulocytic hyperleukocytosis of 13,860 elements per cubic millimeter. Standard radiography showed a grass-cut image (Figure 3). The CT scan showed a tumoral lesion of the left humerus affecting the epiphysis and diaphysis (periosteum, cortex, medulla) associated with "grass fire" osteolytic destruction of the humerus, infiltrating the surrounding soft tissues, with inhomogeneous hepatosplenomegaly, calcified nodular alveolar fillings scattered over the right and left lung parenchyma with a metastatic appearance, a lesion that raised suspicion of osteogenic osteosarcoma of the left humerus (Figure 4). The tumor was classified according to TNM staging: T2N1M1(a + b). Several transfusions were necessary due to episodes of anemia. The administration of step three analgesics and appropriate psychological treatment made it possible to achieve pain control. An essential multidisciplinary consultation meeting allowed the modification of the management indicating salvage surgery without neoadjuvant chemotherapy in the face of the appearance of bleeding that is difficult to control due to enlargement of the fistula (Figure 5). It consisted of tumor resection according to the requirements of oncological surgery for bone tumors. The approach was anterior on a macroscopically healthy area with careful dissection of the tissues, ligation of the axillary vessels, section of the clavicular bone with a Gigli saw, performing an enlarged single-piece amputation of the left thoracic limb taking 2/3 distal to the clavicle and scapula, the weight of the surgical specimen was 14 kilograms (Figure 6). Intraoperative hemodynamic instability throughout the operation due to massive hemorrhage upon removal of the pressure dressing requiring several transfusions of liquid blood products (red cell pellet and fresh frozen

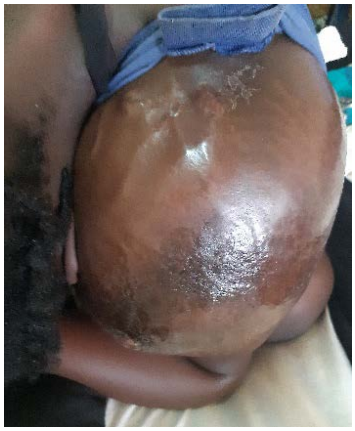

Figure 1. Large, shiny mass with areas of fluctuation and collateral venous circulation.

Figure 2. Large fistulized axillary lymphadenopathy.

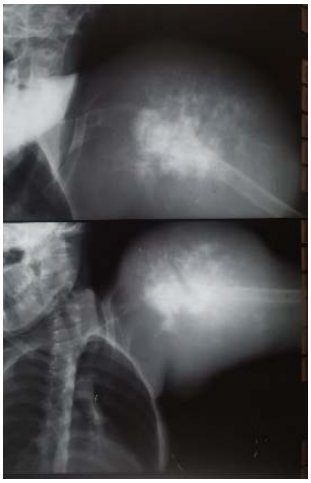

Figure 3. Radiological images of grass fires.

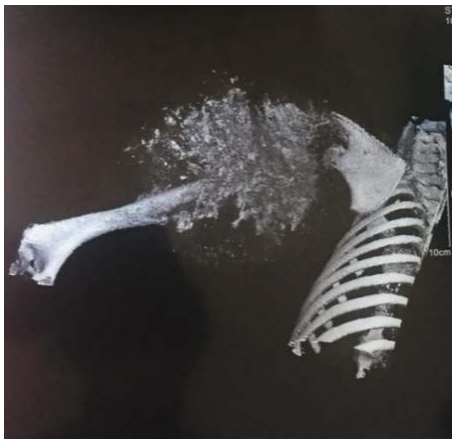

Figure 4. CT images: Tumor lesion of the left humerus with "grass fire" osteolytic destruction, infiltration of the surrounding soft tissues and the scapula.

plasma). Microscopic anatomopathological analysis using hematoxylin-eosin staining at 20 magnification showed a sarcomatous tumor proliferation with triple osteoblastic, chondroblastic and microplastic components, suggesting a high-grade conventional osteosarcoma (**Figure 7**). An improvement in quality was obtained

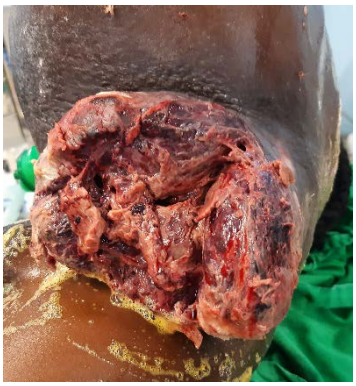

Figure 5. Enlargement of the fistula.

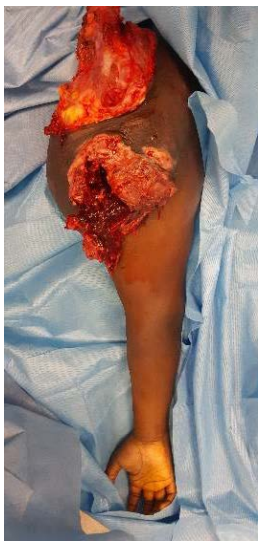

Figure 6. Operating room weighing 14 kilograms.

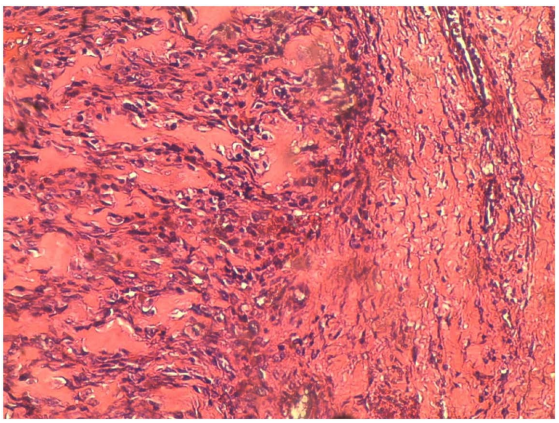

Figure 7. Sarcomatous proliferation with triple osteoblastic, chondroblastic and fibroblastic components (Magnification 20, Hematoxylin Eosin).

for thirty days, followed by the onset of a wet cough with polypnea. The physical examination revealed a syndrome of right pleural fluid effusion, confirmed by radiography (extensive pleurisy). An evacuation puncture performed thirty-six

days postoperatively revealed hematic fluid. The progression is towards a worsening of respiratory symptoms. Death occurred 40 days postoperatively and on the first day of multiagent chemotherapy (cysplatin, doxorubicin and high-dose chemotherapy).

3. Discussion

Osteosarcoma is the most common type of bone cancer in children. 80% of cases are located in the metaphysis of long bones, but can also occur in the diaphysis of long bones as well as in the axial skeleton [1] [2] [3] [8]. Its preferred site is the distal metaphysis of the femur [1]-[9]. Thoracic limb involvement is rare, and the proximal end of the humerus is the preferred site, accounting for 12% of osteosarcomas diagnosed in the US population [3]. Two-thirds of childhood cases occur between the ages of 10 and 14 [1] [2] [3] [5] [6] [7] [8] [9]. The incidence is similar in girls and boys up to the age of 14, with some authors noting a slight male predominance [1] [3]. Symptoms of osteosarcoma may include bone pain, swelling, redness and fracture [1]-[9], and in the form of tumours larger than 8 cm in 47.9% of cases in the US series [3]. At initial diagnosis, 15% - 20% of patients have overt lung metastases, while 40% develop metastases at a later stage [1] [2]. On radiographs, osteosarcoma can be osteolytic or osteoblastic, or both [8], grass-burning images are quite pathognomonic [2] [6]. On histology, proof of bone or osteoid production by tumor cells is a necessary condition for diagnosis [8]. Conventional osteosarcoma is the most common histological type and accounts for 80% of all osteosarcoma cases primarily affecting individuals in the first and second decades of life [8]. It can be subdivided into osteoblastic, chondroblastic and fibroblastic groups based on the predominant characteristics of the cells [8]. In the American meta-analysis [3], osteogenic and osteoblastic types were found in 66.8% of cases with high-grade tumors in 64.3% of cases. Osteosarcomas often have a high malignancy potential with a fulminant course. This means that any tumor point of call at the level of the musculoskeletal system must be examined and managed taking this aspect into account in order to avoid any diagnostic and therapeutic delay. In our 13-year-old adolescent, whose reason for consultation was the large tumor which developed fairly quickly in six months, a notion of minimal trauma was found, and fulminant clinical symptoms testify to the aggressive nature of the osteosarcoma. Considerable advances have been made in the early management of osteosarcomas in developed countries [1] [3] [5] [6] [7]. In developing countries, particularly in Africa, serious difficulties are still present, namely: frequent diagnostic delay, since the circumstances of discovery are atypical and often related to benign trauma; the difficulties of therapeutic management, both medical and surgical, in these growing young people [2] [9]. Therapeutic modalities, perfectly codified, use national protocols [1] [2] [5] [7] [8] [9]. They combine neoadjuvant chemotherapy, followed by tumor resection, then adjuvant chemotherapy which varies depending on the percentage of residual living cells on the resection specimen [1] [2] [3] [5] [6]

[8]. The chemotherapy protocol adopted almost by all is that of the North American Children's Oncology Group using cisplatin, doxorubicin and high-dose methotrexate [1] [3]. In our patient, faced with the large hemorrhagic tumor, a primary amputation was performed followed by adjuvant chemotherapy initiated more than thirty days after surgery. This treatment is linked to the diagnostic delay and the fact of an economically burdensome therapeutic protocol as reported by Ndour in Senegal [2]. In Western countries, there is a decline in mortality each year with a five-year survival rate greater than 60% [1] [3] [7] [8] compared to a rate less than 20% in Africa [2]. The morbidity and mortality found in our patient is linked to the association of different factors: the seriousness of the clinical symptoms, the delay in diagnosis and that of the results of the anatomopathological analysis, economic difficulties, the non-availability of pharmaceutical drugs despite a well-established national chemotherapy protocol.

4. Conclusion

Conventional high-grade osteosarcoma is a highly aggressive primary bone malignancy. Delayed diagnosis, inadequate technical resources and low living standards are the main causes of the difficulties encountered in treating this disease. A number of measures are needed to improve management: awareness-raising and information campaigns at two levels: the general public and medical and paramedical staff, with training programs; multi-disciplinary collaboration between the various players; improved technical facilities and availability of chemotherapy drugs.

Authors' Contributions

All the authors contributed to the conduct of this research work; they read and approved the final version of the manuscript.

Informed Consent of Parents

We certify that the parents of the child have been informed and have given their agreement for the publication of this case report.

Conflicts of Interest

The authors declare no conflict of interest.

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