Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Radiology

Primary Bone Lymphoma of the Knee: A Rare Entity

Youssef Bouktib[1*], K. Abidine[1], B. Boutakioute[1], A. El Hajjami[1], M. Ouali Idrissi[1], N. Cherif Idrissi El Guennouni[1]

[1]Radiology Department, Mother and Child Hospital Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Cadi Ayyad Marrakech University, Morocco

DOI: 10.36347/sjmcr.2023.v11i04.074 | **Received:** 17.03.2023 | **Accepted:** 25.04.2023 | **Published:** 28.04.2023

***Corresponding author:** Youssef Bouktib
Radiology Department, Mother and Child Hospital Mohammed VI University Hospital, Faculty of Medicine and Pharmacy, Cadi Ayyad Marrakech University, Morocco

Abstract **Case Report**

***Introduction*:** The World Health Organization (WHO) defined primary bone lymphoma as a single bone lesion with or without regional lymph node involvement or multiple bone lesions without distant visceral or lymph node involvement. It is a rare entity; indeed, primary bone lymphomas represent 1-3% of non-Hodgkin's lymphomas, 5% of extra-nodal non-Hodgkin's lymphomas and 3% of primary bone tumors [2, 3]. ***Objective*:** The objective of our work is to clarify the imaging of a rare entity of primary bone lymphoma in a 6-year-old boy, who had been complaining for 1 month of pain in his right lower limb without functional impotence. ***Case Report*:** A 6 year old boy, who had been complaining for 1 month of pain in his right lower limb without functional impotence or other associated signs. The clinical examination revealed a child in good general condition, apyretic, with a limp when walking, limitation of all the articular amplitudes of the left he knee, a swelling of the homolateral thigh with discreet quadricipital amyotrophy. Examination of the lymph nodes did not identify any adenopathies. Standard radiographs of the left knee showed multiple lesions involving the lower extremities of the femur and the upper extremities of the tibia, with confluent intraspongiosa and areas of osteocondensation associated with localized cortical fracture without periosteal reaction. The biological workup revealed no abnormalities, including an LDH in the normal range. An MRI of the right thigh was requested for a better characterization of the lesion and to establish a locoregional extension assessment. It showed a large epiphyseal-metaphyseal lesion process of the lower end of the femur and epiphyseal-metaphyseal of the upper end of the left tibia in T1 hyposignal, locally infiltrating T2 heterogeneous hyper signal with vascular enclosure and popliteal ADPs. The CT scan was requested as part of the extension work-up and did not reveal any secondary lesion. The patient underwent a bone biopsy, the immunohistochemical result of which was in favor of a diffuse large cell B lymphoma. After a PCR discussion, the child is a candidate for 6 courses of CHOP chemotherapy and subsequent radiotherapy. ***Conclusion*:** LOP is a rare malignant bone tumor. Multifocal forms may be confused with secondary bone localizations. An early diagnosis is necessary for a better therapeutic management.

Keywords: lymphoma, primary bone tumors, adenopathies, chemotherapy**.**

INTRODUCTION

In 2002, the World Health Organization (WHO) defined primary bone lymphoma as a single bone lesion with or without regional lymph node involvement or multiple bone lesions without distant visceral or lymph node involvement [1].

It is a rare entity; indeed, primary bone lymphomas represent 1-3% of non-Hodgkin's lymphomas, 5% of extra-nodal non-Hodgkin's lymphomas and 3% of primary bone tumors [2, 3].

CASE REPORT

A.I a 6 year old boy, who had been complaining for 1 month of pain in his right lower limb without functional impotence or other associated signs. The clinical examination revealed a child in good general condition, apyretic, with a limp when walking, limitation of all the articular amplitudes of the left he knee, a swelling of the homolateral thigh with discreet quadricipital amyotrophy. Examination of the lymph nodes did not identify any adenopathies.

Standard radiographs of the left knee showed multiple lesions involving the lower extremities of the femur and the upper extremities of the tibia, with confluent intraspongiosa and areas of osteocondensation associated with localized cortical fracture without periosteal reaction (Figure 1).

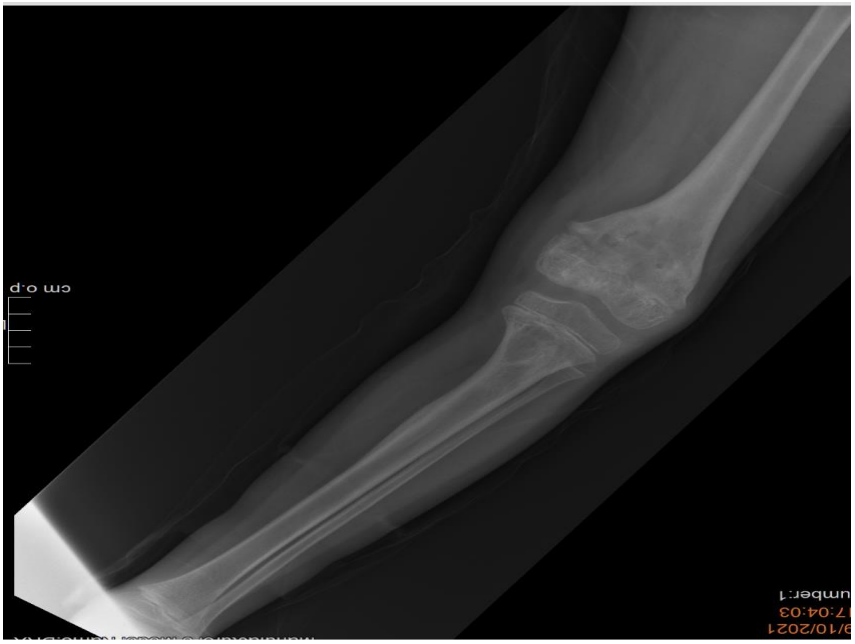
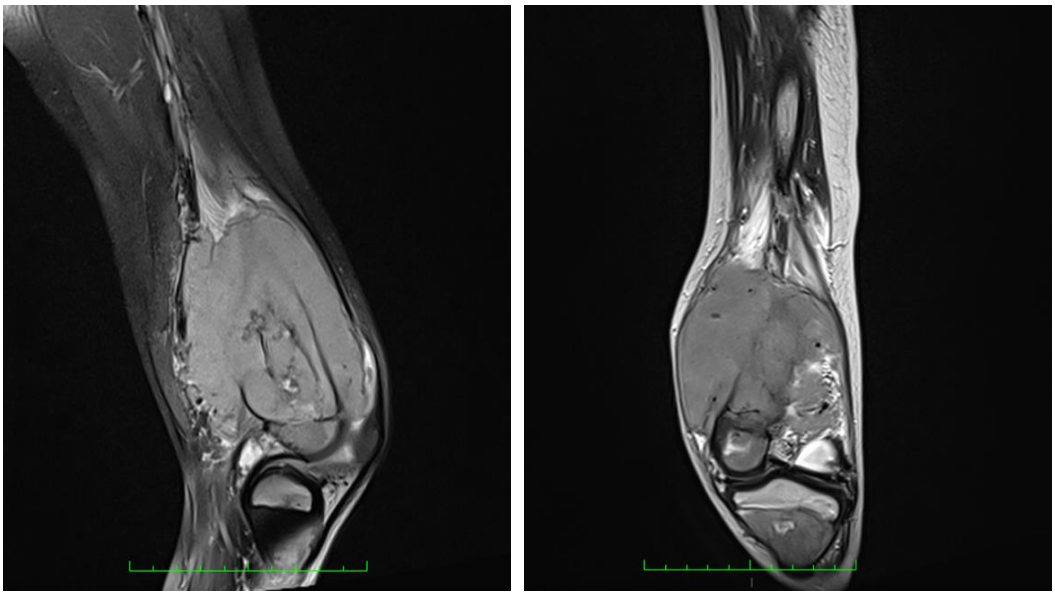


Figure 1: Multiple intraspongy epiphyseal-metaphyseal lacunae of the lower extremities of the left femur and upper tibia with blurred contours, confluent in poorly defined patches associated with patches of osteocondensation, densification of the surrounding soft tissues is noted

The biological workup revealed no abnormalities, including an LDH in the normal range.

An MRI of the right thigh was requested for a better characterization of the lesion and to establish a locoregional extension assessment. It showed a large epiphyseal-metaphyseal lesion process of the lower end of the femur and epiphyseal-metaphyseal of the upper end of the left tibia in T1 hyposignal, locally infiltrating T2 heterogeneous hyper signal with vascular enclosure and popliteal ADPs (figure 2, 3).



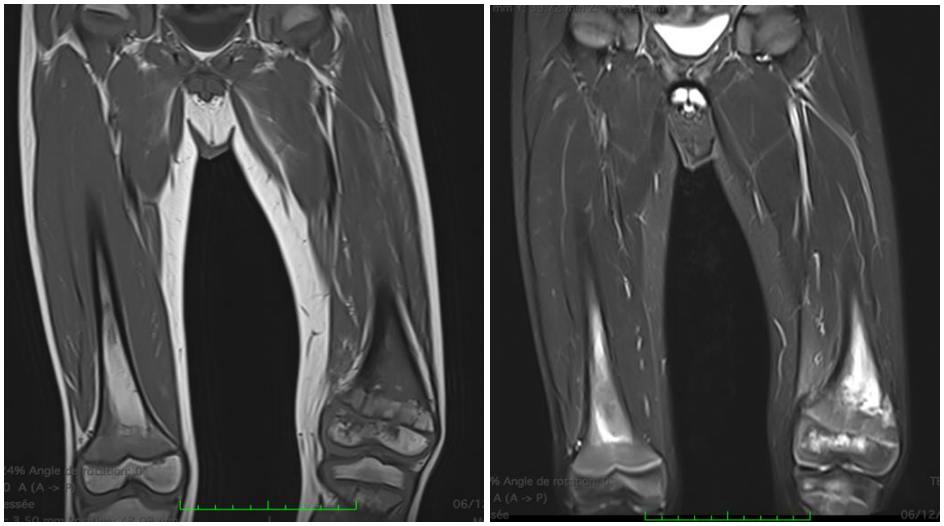
Figures 2, 3: Epiphyseal-metaphyseal lesion process of the lower extremity of the femur and epiphyseal-metaphyseal process of the upper extremity of the left tibia in T1 hyposignal, locally infiltrating T2 heterogeneous hyper signal with vascular envelopment and popliteal ADPs

The CT scan was requested as part of the extension work-up and did not reveal any secondary lesion.

The patient underwent a bone biopsy, the immunohistochemical result of which was in favor of a diffuse large cell B lymphoma.

After a PCR discussion, the child is a candidate for 6 courses of CHOP chemotherapy and subsequent radiotherapy.

A follow-up MRI was requested to evaluate the patient's therapeutic response, which showed a good therapeutic response with persistent femoral and tibial medullary infiltration associated with discrete infiltration of the surrounding soft tissues (Figure 4, 5).



Figures 4, 5: Good therapeutic response with persistent medullary and tibial infiltration associated with discrete infiltration of the surrounding soft tissues

DISCUSSION

LOP is a rare anatomical-clinical entity. It was described in 1928 by Oberling as bone reticulosarcoma [1], then as Parker and Jackson lymphoma [2] in 1939. Its frequency is estimated to be less than 1% of all lymphomas and 3 to 4% of primary malignant bone tumors [3]. It affects men more frequently than women, with a sex ratio of 1.3 to 2. It occurs at any age, with a peak in frequency around the age of 45 [4].

The topography of the lesions involves the long bones, in particular the femur, but also the axial skeleton with a decreasing frequency in the spine, the iliac bone, the sternum and the ribs. The involvement is often metaphyseal diaphyseal, more rarely epiphyseal [5] as in our patient's case. Multifocal LOP is mainly localized around the knee (21% of cases) and the majority of patients have involvement of both the knee and the skull [6].

The clinical signs are dominated by chronic insidious pain, sometimes associated with swelling related to the extension to the surrounding soft tissues as in our patient. Sometimes a pathological fracture or signs of spinal cord compression in spinal locations may reveal the diagnosis of LOP [3, 8].

The average time from the first sign to diagnosis is 7 months.

Biologically, blood calcium and alkaline phosphatase levels are usually normal, but there is an elevation of LDH, the main marker of the tumor mass, associated with an inflammatory syndrome [7].

The radiological appearance is often that of a lytic, metaphyseal, elongated lesion with blurred permeative or worm-like boundaries [9]. The condensing appearance of LOP is much rarer and may point to Hodgkin's lymphoma [10].

A periosteal reaction is inconstant and found in about 60% of cases [6]. It is usually spiculated or lamellar, indicating an aggressive lesion. The cortical bone remains preserved for a long time. Bone sequestration is visualized in 11% of cases and corresponds to an OPL [8, 9].

The CT scan allows a more precise analysis of the lesions and their extension to the soft tissues and bone marrow. However, MRI remains the most effective imaging technique: PBL appears in T1 hyposignal and in variable T2 hypersignal enhanced to a greater or lesser extent after injection of Gadolinium [6, 10]. Diagnosis is based on bone biopsy with immunohistochemical study that specifies the degree of aggressiveness and the tumor phenotype: B or T (the B phenotype being more frequent than the T phenotype) [11, 12].

The lesion assessment should rule out other lymph node or visceral locations; isolated OLP is classified as Ann Arbor stage IE, whereas multifocal involvement is considered stage II [3].

The treatment of OLP is not consensual; the best results are obtained by the combination of radiation and chemotherapy, with a remission rate of 88%. The use of chemotherapy alone is a new therapeutic approach in LOP [5, 8, 13]

Poor prognostic elements are large cell histological type, multifocal bone involvement, and soft tissue extension [3].

In conclusion, LOP is a rare malignant bone tumor. Multifocal forms may be confused with secondary bone localizations. Early diagnosis is necessary for better therapeutic management.

CONCLUSION

LOP is a rare malignant bone tumor. Multifocal forms may be confused with secondary bone localizations. An early diagnosis is necessary for a better therapeutic management.

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