Unilateral Coxalgia Revealing Acute Leukemia: One Case Report and Review of Clinical Literature

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ABSTRACT

Introduction: The osteoarticular manifestations revealing acute leukemia in children are diverse and can be source of diagnostic delay, especially in the absence of other signs that point to a malignant origin.

Case Report: We report through this case of a 4-year-old boy without any medical history and who consulted in our department for isolated severe pain in the left hip bone causing sleep deprivation, occurred after a minimal trauma and causing lameness, all symptoms dated back to 15 days before admission. Clinical examination was normal. Radiological assessment was normal as well. Mild anemia with neutropenia was noted on the blood count with no inflammatory signs. The blood smear was normal. The bone marrow aspiration confirmed the diagnosis of acute B-cell lymphoblastic leukemia.

Discussion: Acute lymphoblastic leukemia (ALL) is the most common childhood malignancy. Osteoarticular manifestations of ALL in children are rare, they are revealing ALL in 14% of cases. The musculoskeletal symptoms can be so prominent or even the only presenting sign that sometimes the diagnosis of leukemia is delayed.

Conclusion: Knowledge of orthopedic manifestation of leukemia is important in the diagnosis, supportive treatment and follow up of patients in order to improve their survival.

Keywords
Coxalgia, Acute lymphoblastic leukemia (ALL)-B, Children.

Introduction
Leukemia is the most common cancer’s children, and may virtually affect any organ or system [1]. The disease is always spreading at the time of diagnosis, and the diagnosis is often obscure when the patient is first seen. It has a peak incidence between the ages of 2-6 years and is more common in males [1]. Bone pain is present in 5% of cases at diagnosis and can be revealing [2]. The presence of the complaints with normal peripheral blood counts, with no organ involvement or lymphadenopathy normally, diverts the attention from ruling out a bone marrow disease [3]. We emphasize through this case the importance to think to acute leukemia in some situations of bone pains.

Medical Report
Our patient is a 4-year-old male child with no particular medical history. He presented initially pain in the left hip fluctuating during the day causing insomnia, which occurred following a minimal trauma (fall from a height of less than 1 meter) evolving 15 days before the admission and causing limb weakness and lameness the child received nonsteroidal anti-inflammatory drugs (NSAIDs) without any improvement. Clinical exam in the department rounds found a child with no fever, his general condition was well-kept. His weight was 15 kg (-1DS) and height was 99 cm (-2 SD). He presented an intense pain at the pressure of left iliac crest without particular irradiation, the hip joint was free, without bruises or inflammatory signs, there were no signs of inequality in length of both lower limbs. The other joints were without notable abnormality.
The child did not have any known tumor, hemorrhagic syndrome or peripheral lymphadenopathy. Pelvis and hip X rays were normal. Biological assessment had noted a slight normochromic normocytic anemia at 10.7 g / dl, white blood cells at 2680 / mm³, neutropenia at 680 / mm³, a platelet count at 224000 / mm³, ESR at 27 mm at the 1st hour and CRP at 2 mg / l. The reticulocyte level was 54320 / mm³ and the blood smear was normal. After 2 control of CBC, which showed bicytopenia, bone marrow aspiration (BMA) was done and it found infiltration with blasts ant 66%. Cytology has shown lymphoblast and MPO negative. Immunophenotyping showed positivity of B markers and patient was classified B-ALL, Karyotype showed a hyperdiploid clone with 62 chromosomes and clonal abnormalities of number. Chemotherapy according to national Protocol MARALL-2006 was done. After two weeks; child have presented no pain and he was on CR after induction. Now, patient is at consolidation phase with a good improvement.

**Discussion**

Acute lymphoblastic leukemia (ALL) is the most common childhood malignancy, accounting for 25% of cancers in children [4]. Usually presents with fever, pallor, petechiae, ecchymosis, hepatosplenomegaly, lymphadenopathy [5]. Osteoarticular manifestations of ALL in children are rare. They are revealing ALL in 14% of cases [6]. The pain is usually diffuse, usually inflammatory and most often occurs in the axial skeleton, or in long bones (femur, tibia) related to specific bone damage or bone necrosis [2].

It can be nocturnal and disrupt sleep, even if it remains for the most part diurnal. It often fluctuates over time and has no triggering factor. This variability is a major difficulty in the diagnostic process because at the time of the tests pain may have regressed or disappeared between two examinations [7].

Patients having musculoskeletal symptoms as chief complaint sometimes have normal or slightly abnormal peripheral blood findings [8]. On the other hand, musculoskeletal symptoms can be so prominent or even the only presenting sign that sometimes the diagnosis of leukemia is delayed, and established only after subsequent detailed hematological workup [9].

Meehan PL et al, Kobayashi D et al, and Jonsson OG et al. cited the misdiagnosis of the patients with musculoskeletal features. They were not detected for weeks or months because the hematologic values were relatively normal [10]. The smear may be normal in patients with orthopedic manifestations, especially in leukemia of childhood. At initial presentation, many patients have non-specific signs and symptoms as well as lab diagnosis [10]. That was the case in our patient.

In children, acute leukemia at presentation can mimic several orthopedic conditions so that a variable delay of the correct diagnosis is often reported [11]. The diagnostic difficulty raised in our observation was acute pain in the hip bone without any other clinical or radiological signs that may point to a malignant origin. In the study of Maman E et al. high rate of orthopedic complaints among the children with B cell precursor (BCP) leukemia was found, 209 patients out of 576 (36.3%), compared to only 25 out of 176 children with T cell leukemia (14.2%) [9]. Findings reported in the literature indicate that these complaints correlate with: lower white cell counts, lower percentage of blast cells in the peripheral blood and lower incidence of organomegaly - that may delay the decision of bone marrow aspiration [3].

In terms of laboratory features, lower neutrophil and platelet levels were associated with ALL, and this was more important and significant for children who presented with fever and/or elevated inflammatory markers [4]. Treatment of ALL comprises is given in four important phases: remission induction, consolidation, reinduction (delayed intensification), and continuation (maintenance). Chemotherapy is administered based on stratified risk classification, as determined by clinical factors [12] and lasts for 2-2.5 years [13]. Contemporary childhood ALL studies have shown improved 5-year overall survival (OS) rates exceeding 90% [12,13]. In the end, Knowledge of orthopedic manifestation of leukemia is important in the diagnosis, supportive treatment and follow up of patients in order to improve their survival [14].

**Conclusion**

Osteoarticular manifestations of acute leukemia are infrequent and may be the only revealing sign of the disease at the beginning. The clinician must know how to think about the malignant etiology in front of any joint or bone pain regardless its location or its clinical presentation and increase the number of para-clinical investigations to avoid any diagnostic delay.

**References**