



A Prodrome of Acute Lymphoblastic Leukemia Mimicking Sacroiliitis

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ABSTRACT

Rheumatic syndromes can be seen in the course of neoplasia either accompanying, antedating or postdating the diagnosis of cancer. Herein, a 40-year-old female patient with acute lymphoblastic leukemia (L3) preceded by a presentation mimicking sacroiliitis is reported.

Key words: *Sacroiliitis, hematological malignancy, leukemia*

Sakroiliit Benzeri Prodromla Başvuran Bir Akut Lenfoblastik Lösemi Olgusu

Malignite tanısı konmadan önce, malignite seyri sırasında ya da tanısının ertesinde romatolojik bulgular ortaya çıkabilir. Burada sakroiliit benzeri bir klinik tablo ile başvurup akut lenfoblastik lösemi (L3) tanısı konan 40 yaşında bir kadın hasta sunulmaktadır.

Anahtar kelimeler: *Sakroiliit, hematolojik malignite, lösemi*

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INTRODUCTION

Malignancy associated with rheumatological syndromes has been described in different settings. Herein, a 40-year-old female patient with acute lymphoblastic leukemia (L3) preceded by a presentation mimicking sacroiliitis is reported.

CASE

On May 2003, a 41-year-old woman had been admitted to a hospital because of a new onset low backache and left leg pain. The laboratory examination had revealed a high erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), anemia and thrombocytosis. The report of the plain radiograph of the sacroiliac joints had stated narrowing of both sacroiliac joint spaces and sclerosis on both iliac bones. This clinical presentation along with the laboratory and radiographic findings had been found suggestive of spondyloarthropathy and she was then put on corticosteroids, salazopyrine, colchicine and indomethazin. After a treatment of four months, expected clinical and laboratory response had not been achieved. Bone scintigraphy had showed increased iliac, acetabular and femur head uptake and a hypoactive area in the left sacroiliac region. Magnetic resonance imaging (MRI) of sacroiliac joints had been reported to display a nonhomogeneous signal change of L5 vertebra, sacral vertebrae and iliac bones. Since a clinical deterioration had been observed, she was referred to our hospital in November 2004 for further evaluation.

On admission, her physical examination revealed pain at both sacroiliac joints that was more prominent on the left and restricted range of movement at the left hip joint. When questioned, the low backache and the pain on her left leg that she had been suffering for over a year had progressed gradually; it was continuous during rest and activity, intense in strength and not responding to non-steroid antiinflammatory drugs (NSAID) and corticosteroids. The laboratory examination revealed an ESR of 80 mm/hour, CRP of 40 mg/L. Her peripheral blood count and peripheral blood smear were normal. Plain pelvis radiography showed significant sclerosis of both iliac bones. Former magnetic resonance tomography of sacroiliac joints was reevaluated. Bone marrow reconversion and red marrow dominance was significant at T1-weighted images (Figure 1A). There was abnormal signal intensity at right sacrum and L5 vertebra at T2-

weighted images; multiple infiltrating lesions were observed at iliac crests (Figure 1B). Bone marrow trephine biopsy from the left iliac crest showed that immunohistochemically, the cells expressed TdT. Bone marrow aspirate smear showed that 75% of the cells were blasts. The immunophenotyping of the marrow was consistent with acute lymphoblastic leukemia (L3) and the patient was transferred to hematology unit for further treatment. After the chemotherapy, the patient had a complete relief of pain.

DISCUSSION

Rheumatic syndromes accompanying, antedating or postdating the diagnosis of cancer seem to appear through different mechanisms. They can occur in the context of paraneoplastic syndromes, by the local inflammatory reaction as a result of invasion of musculoskeletal structures by tumor cells and by hyperuricemia as a result of massive cell turnover either spontaneously or in response to treatment. Paraneoplastic syndromes are induced by the tumor via mediators which are normal cell products excreted in excess because of increased tumor mass, the products of highly undifferentiated cells or the result of nonapoptotic death of tumor cells. These mediators may damage vascular endothelial cells, synovial epithelia and mesenchymal tissues. Paraneoplastic rheumatic disorders may precede the diagnosis of cancer by utmost 2 years (1).

Among rheumatic manifestations of hematologic malignancies, bone pain seems to be the most common symptom. Arthritis is not frequent but has been described. Leukemia associated arthritis has been reported in both acute and chronic forms of the disease. It is much more common in children compared to adults and may result from leukemic infiltration of the synovium and subperiosteal tissue (2,3,4,5,6). It is striking that in most of the reported cases, rheumatic symptoms preceded the diagnosis of leukemia (7). In a study where rheumatic manifestations preceding adult acute leukemia were studied during a 10-year period in 139 patients, 5.8 % of the patients were found to present with rheumatic manifestations (2). The incidence was reported as 4% and 16.5% in previous studies (8,6). In the study group with 139 patients, the average duration of rheumatological prodrome was 3 months (2). Asymmetric involvement of large joints and back pain were the most common symptoms followed by symmetric polyarthritis mimick-



Figure 1. Magnetic resonance tomography of sacroiliac joints. A) T1-weighted image: Significant bone marrow reconversion; dominance of red marrow. B) T2-weighted image: Abnormal signal intensity at right sacrum and L5 vertebra; multiple lesions at iliac crests.

ing early rheumatoid arthritis. Distinctive characteristics that would remind of a paraneoplastic process were severe pain disproportionate to physical examination, poor response to conventional antirheumatic treatment as NSAID and corticosteroids and abnormal radiologic findings like lytic bone lesions or early osteopenia.

The case described here was a 40-year-old woman diagnosed as biphenotypic leukemia with a presentation mimicking spondyloarthropathy. The disproportion between the severity of her pain and the physical findings and her unresponsiveness to conventional antirheumatic treatment was striking. It is also worth to emphasize the importance of a careful interpretation of sacroiliac MRI which is an important imaging technique in the diagnosis and follow-up of spondyloarthropathy. Our literature search shows that the mean duration of the rheumatological prodrome predating acute leukemias is three months with the asymmetric involvement of large joints and back pain being the most common symptoms. No case is described that is diagnosed as acute leukemia after a one-year period of rheumatological complaints.

In conclusion, rheumatic syndromes can be seen in the course of neoplasia either predating the diagnosis, accompanying it or postdating. When a patient presents with a rheumatological syndrome, existence of atypical features should alarm the physician to search for an underlying malignancy for a timely diagnosis.

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