Case Report / *Olgu Sunumu*
DOI: 10.5152/tftrd.2014.56254
Turk J Phys Med Rehab 2014;60:353-5
Türk Fiz Tıp Rehab Derg 2014;60:353-5

Hodgkin's Lymphoma: Case Report of Mimicking Sacroiliitis

Sakroiliiti Taklit Eden Hodgkin Lenfoma: Olgu Sunumu

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Abstract

Musculoskeletal syndromes may be associated with neoplastic diseases. We hereby report a case of sacroiliitis that was diagnosed as Hodgkin's lymphoma during the follow-up process. Though sacroiliitis was confirmed with magnetic resonance imaging (MRI), a diagnosis of malignancy associated with sacroiliitis may be delayed and cause a clinical challenge. We aimed to discuss the presentation of lymphoma with sacroiliac joint involvement and its probable mechanisms and review the literature.
Key Words: Sacroiliitis, Hodgkin's lymphoma, low back pain

Özet

Kas iskelet sistemi sendromları neoplastik hastalıklarla ilişkili olabilir. Bu yazıda, kliniğimizde sakroiliit tanısı alan ve takiplerde Hodgkin lenfoma tanısı koyduğumuz olguyu tanımladık. Sakroiliit tanısı manyetik rezonans görüntüleme ile de doğrulansa dahi sakroiliitle ilişkili olabilecek malignite tanısı gecikebilir ve bu durum klinik bir karışıklığa yol açabilir. Amacımız literatürü gözden geçirerek, malignite varlığında sakroiliak eklem tutulumun olası mekanizmasını tartışmaktır.
Anahtar Kelimeler: Sakroiliit, Hodgkin lenfoma, bel ağrısı

Introduction

Musculoskeletal syndromes, such as hypertrophic osteoarthropathy, carcinoma polyarthritis, dermatomyositis/polymyositis, and paraneoplastic vasculitis, fasciitis, panniculitis, erythema nodosum, Raynaud's syndrome, digital gangrene, erythromelalgia, and lupus-like syndromes, may be associated with neoplastic diseases (1). Although the association of malignancy and rheumatologic disorders is well described in the literature, only few cases have reported sacroiliitis as an initial presentation of lymphoproliferative disorder.

In this report, we describe a patient with sacroiliitis and diagnosed with Hodgkin's lymphoma 6 weeks after admission to our clinic.

Case Report

A 31-year-old man presented with right hip and buttock pain and left hip pain of 3 weeks in duration. The pain began acutely, initially involving the right side and involving the right buttock. He described marked morning stiffness lasting for 2 hours. In the course of this period, he was admitted to the emergency service of our hospital several times, and nonsteroidal anti-inflammatory and myorelaxant drugs were suggested, and he was referred to the physical medicine and rehabilitation outpatient clinic. He denied experiencing any symptoms of fever, weight loss, peripheral joint pain, skin lesions, urethritis, conjunctivitis, and venereal or gastrointestinal disease. The family history was negative for rheumatic diseases. He had no smoking

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Received/Geliş Tarihi: August/Ağustos 2013 Accepted/Kabul Tarihi: December/Aralık 2013

history. In the physical examination, right sacroiliac compression and Gaenslen tests were positive. Sacroiliac joint was exquisitely painful to direct pressure. The cervical, thoracic, and lumbar spine and peripheral joint examination was unremarkable.

In the laboratory examination, serum C-reactive protein (CRP) was 109.9 mg/L (normal range 0-5 mg/L), hemoglobin was 10.7 g/dL (normal range 12-15.5 g/dL), erythrocyte sedimentation rate (ESR) was 105 mm/h (normal range 0-20 mm/h), and white blood cell count was 10,600/mm^3 (normal range 3500-10,500/mm^3). The patient was HLA-B27-negative. A plain radiograph of the sacroiliac joint revealed minimal sclerotic changes in the iliac region of the right sacroiliac joint.

Magnetic resonance imaging of the sacroiliac joint showed bone marrow edema of the iliac bone and sacroiliac joint and gadolinium enhancement with hyperintense areas that may be associated with sacroiliitis (Figure 1).

The patient initially received sulfasalazine 1000 mg twice a day and indomethacin perorally 25 mg 3 times a day. At the first follow-up after 2 weeks, the patient's clinical response to therapy was excellent. There was no pain or morning stiffness. ESR was still 97 mm/h and CRP was 44.3 mg/L at this time. After 3 weeks, the patient experienced fatigue and detected multiple 1-2 cm non-tender cervical and axillar lymph nodes in the physical examination. The liver and spleen were not enlarged. Abdominal ultrasonography revealed multiple hypoechoic, regular-shaped solid lesions in the liver and spleen. An abdominal MRI confirmed hyperintense heterogeneity with contrast material in the liver and spleen that may be associated with lymphoproliferative disorder or granulomatous disease. Excisional biopsy of the cervical lymph node showed Hodgkin's lymphoma nodular sclerosis subtype. He was referred to the hematology clinic to receive a chemotherapy program. Positron emission tomography (PET), performed in the hematology clinic, revealed hypermetabolic areas in the right iliac bone, which may be associated with bone marrow involvement. Also, multiple hypermetabolic areas in the cervical, supraclavicular, mediastinal, and infradiaphragmatic lymph nodes and a hypermetabolic area in right fourth costa that may be associated with metastatic bone lesions were reported.

Discussion

Hereby, we reported a patient with low back pain and diagnosed as sacroiliitis, and the cervical and axillar lymph nodes directed with to perform advanced research with a suspicion of malignancy. We also aimed to emphasize the importance of a malignancy mimicking rheumatic conditions and reviewed the literature.

Although musculoskeletal disorders may be associated with neoplastic disease, an unusual presentation of a malignancy may be a cause of the delayed diagnosis of disease. Primary synovial neoplasm or metastasis to the bone or synovium may be the reason of musculoskeletal complaints. Periarticular metastases are not infrequent and may present as acute arthritis. These are neoplastic lesions of the bone without inclusion of the joint or deterioration of the joint because of metastases and non-neoplastic reactions in the adjacent joint and neoplastic invasion of the joint (2). In addition to the paraneoplastic phenomenon, the unusual presentation of malignancies mimicking rheumatic disorders has been a clinical challenge and may be the cause of the delayed diagnosis of the main disease.

In the literature, there are 4 cases of sacroiliitis associated with hematologic malignancy. One of them was acute lymphoblastic leukemia, in which a complete blood count revealed pancytopenia (3). Our patient's complete blood count was unremarkable, and the clinical response to medication was excellent. He had no complaint at the first follow-up, but he presented with constitutional symptoms, like fatigue and night sweats, and the physical examination determined lymphadenomegaly in the cervical chain at the next follow-up. Although there was an improvement in low back pain of our patient's, serum ESR and CRP levels were still high. These findings directed us to perform further diagnostic techniques. Two cases have been reported with sacroiliitis who were diagnosed with Hodgkin's disease 12 and 5 months after sacroiliitis, respectively. The interval between our patient's sacroiliac joint pain and the exact diagnosis of Hodgkin lymphoma was shorter than the previous report, because the constitutional symptoms and lymph nodes occurred rapidly (4). In a case report, sacroiliitis and enthesopathies developed due to diffuse, well-differentiated lymphocytic lymphoma (5).

However, the initial presentation of our patient was similar to sacroiliitis: ESR level was over 100 mm/h and remained high despite non-steroidal anti-inflammatory drug and sulfasalazine medication.

The etiology of sacroiliitis may be classified into inflammatory and non-inflammatory conditions. In particular, secondary carcinoma must take part in this list (2). The presentation of the patient was similar to spondyloarthritis, including ankylosing spondylitis, reactive arthritis, psoriatic arthritis, and enteropathic arthritis. We determined a high level of ESR, which was suspicious for inflammatory sacroiliitis. If ESR and CRP levels are higher than expected, the clinician must check out the initial diagnosis. When the expected clinical response is not achieved, with the persistence of symptoms, the diagnosis must be reconsidered. Also, constitutional symptoms may direct the clinician for an advanced examination with suspicion of malignancy.

The histopathologic diagnosis may be surprisingly different; however, an MRI indicating edema of the sacroiliac joint seems to meet the definition of sacroiliitis issued by the Assessment of the Spondyloarthritis International Society (ASAS). Bereau et al. (6) reported a case with sacroiliitis according to an MRI study that was diagnosed with Hodgkin's disease with radioguided biopsy of the iliac joint.

Rheumatic syndromes associated with paraneoplastic syndromes include rheumatoid arthritis, Sjogren's syndrome, systemic sclerosis, temporal arteritis, carcinoma polyarthritis, dermatomyositis, lupus erythematosus, and vasculitis. Sacroiliitis is not defined as a paraneoplastic syndrome. According to our

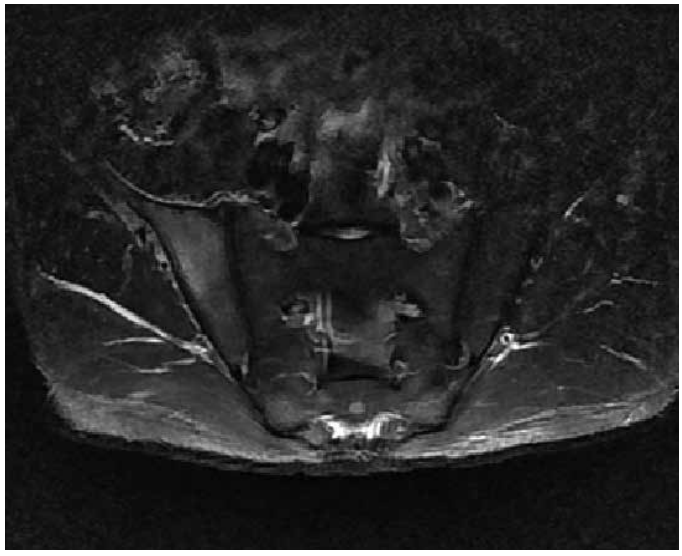

Figure 1. Bone marrow edema at the sacroiliac joint by magnetic resonance imaging

findings, we do not consider the sacroiliac pain of our patient as a paraneoplastic phenomenon but the direct involvement of the iliac bone by lymphoma mimicking unilateral sacroiliitis. Even though we could not confirm the sacroiliac joint involvement by Hodgkin lymphoma with the histopathological findings, the PET results confirmed our hypothesis. Also, our report is similar to previous reports: one of them was of sacroiliac joint involvement of metastatic adenocarcinoma, and the other one was prostatic carcinoma mimicking sacroiliitis (2,7). The self-criticism of this report is that the histopathological sample should be taken during the initial visits with a suspicion of malignancy even a well clinical response achieved.

Conclusion

In rheumatic syndromes, fever; weight loss; thromboembolic events; and neurological, endocrinological, gastrointestinal, or cutaneous disorders may be the signs of underlying neoplastic disease (8). In patients with rheumatic syndromes, ESR, complete blood cell count, and serum chemistries are almost always obtained during the routine evaluation. In respect to laboratory findings, elevated ESR and anemia may both present in malignancy and primary rheumatic syndromes. Both of these findings have limited value for the differential diagnosis between cancer-associated disorders and primary rheumatic diseases (9). More detailed investigations with a suspicion of malignancy are not recommended initially if there are no specific findings that are suggestive of malignancy. Therefore, the clinician must recognize the clues that may be the features of a malignancy associated with rheumatic syndromes.

In conclusion, sacroiliitis may be the consequence of a malignancy with sacroiliac joint involvement; however, morning stiffness and good response to non-steroidal anti-inflammatory drug treatment accompany the sacroiliac pain. The physician must be aware of a malignancy with elevated serum ESR levels over 100 mm/h, despite a clinical response achieved with medication. An unusual presentation of malignancies mimicking rheumatologic disorders can mask the exact diagnosis of the disease.

Informed Consent: Written informed consent was obtained from patient who participated in this case.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - Ş.A.Y.; Design - Ş.A.Y., A.M.E., A.B.S.; Supervision - C.Ç.; Funding - Ş.A.Y., A.M.E.; Materials - Ş.A.Y., A.M.E.; Data Collection and/or Processing - Ş.A.Y., A.M.E.; Analysis and/or Interpretation - A.B.S., Ş.A.Y., B.K.; Literature Review - Ş.A.Y.; Writer - Ş.A.Y.; Critical Review - C.Ç.; Other - C.Ç., A.B.S.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

Hasta Onamı: Yazılı hasta onamı bu olguya katılan hastadan alınmıştır.

Hakem değerlendirmesi: Dış bağımsız.

Yazar Katkıları: Fikir - Ş.A.Y.; Tasarım - Ş.A.Y., A.M.E., A.B.S.; Denetleme - C.Ç.; Kaynaklar - Ş.A.Y., A.M.E.; Malzemeler - Ş.A.Y., A.M.E.; Veri toplanması ve/veya işlemesi - Ş.A.Y., A.M.E.; Analiz ve/veya yorum - A.B.S., Ş.A.Y., B.K.; Literatür taraması - Ş.A.Y.; Yazıyı yazan - Ş.A.Y.; Eleştirel İnceleme - C.Ç.; Diğer - C.Ç., A.B.S.

Çıkar Çatışması: Yazarlar çıkar çatışması bildirmemişlerdir.

Finansal Destek: Yazarlar bu çalışma için finansal destek almadıklarını beyan etmişlerdir.

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