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CASE REPORT

RS3PE WITH PROSTATE ADENOCARCINOMA

ABSTRACT

Relapsing seronegative symmetrical synovitis with pitting edema (RS3PE) is a clinical syndrome characterized by acute onset of symmetrical joint involvement with pitting edema of the hands and feet. The etiology and pathogenesis of RS3PE is still unknown, though environmental and infectious agents may play a role in its development. RS3PE might be associated with rheumatologic diseases; while coexistence of RS3PE with neoplasms is also reported. Prognosis of RS3PE is good, with high response to low dose corticosteroids. A 72 year old man, who also suffers from prostate adenocarcinoma, with RS3PE syndrome is reported. He had no symptoms of prostatism or systemic symptoms. Adenocarcinoma was detected during the investigation for a possible coexisting malignancy. He responded well to the steroid treatment. Aged patients diagnosed with RS3PE syndrome should be carefully investigated for coexistence with neoplasms and followed for a future malignant process. Low dose corticosteroids should be initiated in all patients, together with the appropriate treatment for the malignancy in patients with neoplastic RS3PE.

Key Words: Aged; Prostate; Synovitis.



OLGU SUNUMU

PROSTAT ADENOKARSİNOM TANISI ALAN BİR RS3PE OLGUSU

Öz

Te krarlayıcı seronegatif pitting ödemli simetrik sinovit sendromu (RS3PE), ani başlangıçlı, eller ve ayaklarda gode bırakan ödem ile karakterize bir klinik sendromdur. RS3PE sendromunun etyolojisi ve patogenezi hala bilinmemektedir ancak gelişiminde çevresel ve enfeksiyöz ajanlar rol alıyor olabilir. RS3PE romatolojik hastalıklarla ilişkili olabilir ayrıca, neoplazilerle birlikteliği de rapor edilmiştir. RS3PE sendromu düşük doz kortikosteroide iyi yanıt veren, prognozu iyi bir sendromdur. Bu makalede 72 yaşında prostat adenokarsinom tanısı alan RS3PE sendromlu bir hasta sunulmaktadır. Herhangi bir prostatizm ve sistemik semptomu olmayan hastanın malignensi ile birlikteliği ihtimali için yapılan ileri araştırmasında prostat adenokarsinom tanısı tespit edilmiştir. Hasta steroidde iyi yanıt vermiştir. RS3PE sendromu tanısı alan yaşlı hastalar malignensi birlikteliği ya da bu sendromu takiben çıkabilecek neoplazi açısından dikkatlice değerlendirilmelidir. Neoplastik RS3PE hastalarında düşük doz kortikosteroid tedavisi altta yatan neoplazinin tedavisi ile birlikte başlanmalıdır.

Anahtar Sözcükler: Prostat, Sinovit, Yaşlı.

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INTRODUCTION

Relapsing seronegative symmetrical synovitis with pitting edema (RS3PE) is a clinical syndrome characterized by acute onset symmetrical joint involvement with pitting edema of the hands and feet, first described by McCarty in 1985 (1). It is especially reported in men over 60 years of age. The etiology and pathogenesis of RS3PE are still unknown. Neither rheumatoid factor (RF) nor antinuclear antibodies (ANA) is positive (2). The prognosis is good; RS3PE generally responds to low dose corticosteroid treatment, and complete remission is reported within 18 months. RS3PE may coexist with or precede a malignant state. We report a 72-year-old man with RS3PE syndrome diagnosed with prostate adenocarcinoma during the investigation for a possible coexisting malignancy.

CASE

A 72-year-old male was admitted to the outpatient clinic with a 2-week history of bilateral painful swollen hands and feet. He reported morning stiffness lasting for half an hour. There was no history of trauma, fever, weight loss or any other metabolic or rheumatologic disease. He had smoked 30 cigarettes a day for 48 years between the ages 20 and 68.

On physical examination, there was pitting edema on the dorsum of the hands and feet bilaterally (Figure 1). Range of motion of wrists, ankles and metacarpophalangeal and interphalangeal joints of the hands and feet were limited and painful in all directions. Routine laboratory investigation was as normal except for mild microcytic anemia (hemoglobin:11.1 g/dL Mid Corpuscular Volume:76.4) and elevated

erythrocyte sedimentation rate (ESR) (83 mm/h) and C reactive protein (CRP) (6.66 mg/dl). RF and anti-cyclic citrullinated peptide (anti-CCP) were negative. Plain radiographs of the hands and feet showed soft tissue swelling but no evidence of articular erosions. A chest radiograph revealed no abnormality. Clinical impression was RS3PE and 5 mg prednisolone/day was started. We commenced further evaluation in order to rule out coexisting malignancy or rheumatologic disorder. ANA, HLAB27 and anti-ds-DNA were negative. Serum electrolyte levels, kidney, liver and thyroid function tests were within normal limits. Prostate specific antigen (PSA) was elevated: 72.25 ng/ml (0-4 ng/ml).

The patient was consulted to Urology Department. Prostate biopsy revealed prostatic adenocarcinoma with a Gleason score of 4. The patient refused the operation so radiotherapy was given for the treatment of prostatic adenocarcinoma. One month later his symptoms regressed completely. The edema had almost disappeared within ten days (Figure 2) and ESR was markedly decreased at the first month. At third month follow up he had no signs or symptoms of RS3PE. The patient continued to take prednisolone treatment for 6 months. At first year follow up there was no recurrence.

A typical patient with RS3PE fulfills the following criteria: (1) bilateral pitting edema of the hands, feet or both, (2) sudden onset of polyarthritis, (3) age over 50 years, and (4) seronegativity for RF. RS3PE was described by McCarty in 1985 for the first time in eight aged men and two aged women with symmetric polysynovitis with acute onset. He found no evidence of erosions on radiographs. Serologic tests such as RF, antiCCP, and ANA were negative (1). The HLA-B7 phenotype is reported to be positive in patients with



Figure 1— Before treatment.



Figure 2— After treatment.



RS3PE syndrome (1, 2, 5); however, HLA-B7 assay was not possible in our case. A proportion of RS3PE patients are associated with or develop features of rheumatic diseases like rheumatoid arthritis, spondyloarthropathy or connective tissue diseases (2).

In RS3PE syndrome, ESR is moderately high at the time of diagnosis but falls after treatment, which presumably indicates an inflammatory process. The etiology of RS3PE syndrome is still unclear; an infectious agent is presumed to be the triggering factor but none has been confirmed (5). RS3PE syndrome has also been described in neoplastic conditions. The first observation was RS3PE appearing 4 months before a non-Hodgkin's lymphoma (3). Since then solid tumors such as colon, gastric, breast, prostate and hematological malignancies (4-9) have been reported. Coexistence of RS3PE and malignancies gave rise to the thought that it might be a paraneoplastic syndrome. Review of the literature indicates that RS3PE syndrome may coexist with or precede a malignant state (10). Concerning the reports about RS3PE syndrome being associated with malignancies, some patients have constitutional symptoms with a deterioration in general health (3), whereas others have no difference from the classical clinical scene of RS3PE (5,11). Our patient had no symptoms other than the edema and arthritis of the hands and feet. Further, he had no symptoms of prostatism. Systemic symptoms in patients having RS3PE syndrome should alert the clinician about coexisting malignancies. Furthermore, patients diagnosed with RS3PE syndrome in whom any underlying pathology was not detected should be followed for a future malignant process (11).

Another issue with neoplastic RS3PE syndrome is the response to corticosteroids. Patients with idiopathic RS3PE show a good response to low dose corticosteroids (12). There are reports of a poor response of RS3PE patients with associated neoplasia (3,6). Good response to corticosteroids, similar to the idiopathic cases, is also reported in neoplastic RS3PE syndrome (5,11). The best management of paraneoplastic RS3PE syndrome is to treat the malignant process; most patients seem to respond after successful treatment of the underlying malignancy after surgical treatment of the tumour and/or chemotherapy or radiotherapy (4-6). Our patient responded to low dose steroid treatment in a short time; clinical and laboratory findings were normalized at the first month without any relapse.

CONCLUSION

Patients diagnosed with RS3PE syndrome should be carefully investigated for a possible neoplasm and followed for

a future malignant process. Systemic symptoms and inadequate response to glucocorticoids should alert the physician about a coexisting malignancy. Prostate adenocarcinoma should be kept in mind in aged men with RS3PE syndrome, even in the absence of symptoms of prostatism.

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