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Letter to the Editor

Anti-TNF-Alpha Therapy for Concomitant Behçet's Disease and Ankylosing Spondylitis

Behçet Hastalığı ile Ankilozan Spondilit Birlikteliğinde Anti-TNF-Alfa Tedavisi

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We have read the manuscript titled "Coexistence of Behçet's Disease (BD) and Ankylosing Spondylitis" authored by Çelik et al.[1] and would like to share our opinions and experience with regard to the subject matter and content of this paper.

A 31-year-old male patient presented at our outpatient clinic with an increase in lumbar and back pain, bilateral pain of the knees with swelling of the left knee, and rashes on both the upper and lower extremities. At the age of 21, while the patient was performing compulsory military service, he complained of lumbar and rare hip pains as well as morning stiffness that lasted for about 30 minutes, and ankylosing spondylitis (AS) was diagnosed. At 24 years of age, the patient experienced swelling of both knees along with difficulty walking and was treated with sulfasalazine and non-steroidal anti-inflammatory drugs (NSAIDs). This treatment regimen was stopped when the patient reached 28 years of age, at which point etanercept was started to alleviate the increased lumbar and back pain and the development of arthritis in both knees. He continued receiving etanercept for two years and, as his condition improved, the patient decided to end the treatment on his own. He has been taking NSAIDs as required for the past seven months.

The patient's lumbar and back pain has increased significantly in the last four months. It has been especially severe in the morning, and the patient has experienced morning stiffness lasting for at least two hours. His complaints have increased with rest and decreased with exercise and physical activity. Although he has complained of fatigue, the patient has not had a fever, night pain, or weight loss. While he has not had any symptoms in the eyes, chest, or heels, he has had bilateral knee pain for the last month, with swelling of the left knee.

During the physical examination of the patient, lumbar lordosis was loss with the increase in dorsal kyphosis. The Schober's test measurement was 2 cm, chest expansion was 2.5 cm, and lumbar spinal motions were limited in all directions. He had no cervical pain or complaints, and his range of joint motion was normal. The sacral compression, Gaenslen's, Mennell's and Patrick's tests were positive. The straight leg extension test and neurological examinations were completely normal. The patient had bilateral effusion of the knees, which was significant on the left side. Also, both knees were warm and painful with compression or movement.

In the laboratory tests, the erythrocyte sedimentation rate (ESR) was found to be 66 mm/h, the C-reactive protein (CRP) was 14 mg/L (normal 0.1-0.5), and the rheumatoid factor (RF) was negative. The patient was human leukocyte antigen (HLA)-B27 positive. Anteroposterior pelvis radiographs showed grade III bilateral sacroiliitis,

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and lateral lumbar radiographs revealed a decrease in lordosis, squaring of the vertebral bodies, and syndesmophyte formation.

A dermatology consultation was requested for the rashes, and the dermatologist reported several oral lesions associated with aphthous stomatitis which were noted to have appeared repeatedly (4 or 5 times per month) over a four-year period. In addition, erythema nodosum and papulopustular lesions were detected. A typical genital ulcer history, which had appeared only once 15 years previously, was also obtained from the patient. The skin lesions had been particularly active in the past six months. Moreover, the patient had a previous diagnosis of uveitis, and a pathergy test was positive. In light of these results and based on the criteria set by the International Study Group for Behçet's Disease (BD), the patient was diagnosed as having BD.[2] The lesions of the patient improved after treatment with colchicine (0.5 mg 3x1) and methylprednisolone (16 mg/day). In addition, the patient was diagnosed as having uveitis from an ocular examination which followed complaints of stinging, pain, and blurriness in the left eye, and appropriate treatment was initiated.

The Bath Ankylosing Spondylitis Activity Index (BASDAI) is the most widely used instrument for assessing disease activity in patients with AS.[3] It is easy to use, reliable, valid, comprehensive, and sensitive to change. It has been translated from the original English into several other languages, including Turkish.[4] The Assessments in Ankylosing Spondylitis (ASAS) International working group consensus statement proposed using the BASDAI to evaluate active disease in AS patients before the beginning of anti-TNF-alpha (α) therapy.[5] The patient was not being followed up in our hospital; therefore, we had no records of the BASDAI scores either before the start of therapy, or after he gave up the therapy on his own seven months ago. However, seven months before admission to our polyclinic, he had no complaints and stopped the anti-TNF-α therapy, which suggests that the therapy provided an obvious benefit.

Although it is the subject of a long-running debate, BD is not currently included in the seronegative spondyloarthropathy group. The combination of AS and BD in a single patient has been reported in only a few case presentations in the literature.[6] Therapy for chronic diseases is difficult, and patient satisfaction is for the most part poor. For a patient who has more than one chronic disease, therapy is complicated in terms of drug interactions and side effects. The use of anti-TNF-α agents is preferred for patients who are refractory to treatment with NSAIDs and sulfasalazine for AS.[5] In addition, anti-TNF-α agents have been shown to be effective with regard to the mucocutaneous manifestations of BD and inflammatory eye disease in selected patients.[7-9] This data suggests that a patient with both AS and BD association can be treated by anti-TNF-α therapy. In this respect, Yıldız et al.[10] reported positive results with adalimumab therapy in a patient with both AS and BD. In the present case, when the patient stopped etanercept treatment, there were increases in the frequencies of oral aphthae and erythema nodosum related to BD together with the emergence of uveitis. This suggests that the anti-TNF-α therapy suppressed the mucocutaneous manifestations of BD and inflammatory eye disease. Moreover, this case indicates that anti-TNF-α therapy results in delayed diagnosis of comorbidities due to the suppression of symptoms by the immunosuppressive therapy.

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