Turk J Rheumatol 2011;26(3):271-272
doi: 10.5152/tjr.2011.045

Letter to the Editor

Coexistence of Behçet's Disease and Ankylosing Spondylitis

Behçet Hastalığı ve Ankilozan Spondilit Birlikteliği

Canan ÇELİK, Halil UÇAN, Aslı ÇALIŞKAN UÇKUN, Ebru ALEMDAROĞLU

2nd Department of Physical Medicine and Rehabilitation,
Ankara Physical Medicine and Rehabilitation Training and Research Hospital, Ankara, Turkey

Whether Behçet's disease (BD) is of the seronegative spondyloarthropathy (SSpA) group has been a subject of debate for many years.[1] Behçet's syndrome is now not considered to belong to the SSpA group[2] but is considered to be a systemic vasculitis.[3,4] There are many reasons why BD should not be classified as one of the SSpA group.[3] On the basis of some findings, including the lack of association with HLA B27,[2] the lack of familial association, and the association with HLA B51 rather than the B27 cross-reactive group, it is suggested that BD is not a seronegative spondyloarthropathy.[3] However, back pain is distinctly uncommon, and studies of sacroiliac joint involvement have not demonstrated an increased prevalence in Behçet's patients.[4-5] The coexistence of BD and ankylosing spondylitis (AS) has been rarely reported in the literature.[6] It was reported that AS was seen in 1.5% of Behçet's patients in Iran, which is 2.43 times greater than the general population, and this data showed that BD is one of the causes of secondary AS.[4]

A 34-year-old male patient came to our clinic complaining of bilateral knee pain and swelling. The culture of the aspirated synovial fluid was negative. He had an acute attack of anterior uveitis in his right eye six years ago. His mother has been treated for AS. He described recurrent oral aphthae, genital ulcers, and visual blurring. A pathergy test was positive. He had no history of inflammatory lower back pain, neck pain, morning stiffness, alternating buttocks pain, psoriasis, infection before the occurrence of arthritis, or inflammatory bowel disease. Upon physical examination, his spinal motions were limited but painless. The modified Schober's test was 2.5 cm and chest expansion was 2 cm. The sacroiliac joints and enthesis points were painless with compression. He had bilateral knee arthritis. The erythrocyte sedimentation rate (ESR) was 45 mm/h and the C-reactive protein level was 8.7 mg/dL (0-0.5). Other results of routine biochemicals were normal, and RF and anti-cyclic citrullinated protein antibodies (anti-CCP) were negative. The HLA phenotype was positive for both HLA B27 and HLA B51. In an X-ray examination, bilateral grade 4 sacroiliitis (SI) was determined. An anteroposterior lumbar spine X-ray examination revealed the sclerosis of facet joints and syndesmophytes. A lateral view of the lumbar spine revealed the squaring of vertebral bodies. Considering all these findings, the patient was diagnosed as having BD and AS.

Dilsen et al.[1] evaluated 334 Turkish patients with BD. Ten percent (n=33) of these patients were diagnosed as having AS, and in 34% (n=112) of the patients, SI was determined. Both the HLA B51 and B27 antigens were reported to be more frequent in patients with coexisting BD and AS. Yazıcı et al.[5] found no evidence of an increased frequency of SI in patients with BD and described only one patient with definitive AS among 114 patients with BD. In another study, Yazıcı et al.[7] pointed out that the inter-observer variation might be a major cause of discrepancies in interpreting pelvic

Received: May 12, 2010 ***Accepted:*** May 21, 2010

Correspondence: Canan Çelik, M.D. Ankara Fizik Tedavi ve Rehabilitasyon Eğitim ve Araştırma Hastanesi, 2. Fizik Tedavi ve Rehabilitasyon Kliniği, 06100 Sıhhiye, Ankara, Turkey. Tel: +90 312 - 310 32 30 e-mail: ccelik@hotmail.com

radiographs for SI. Chang et al.[8] evaluated 58 cases with BD, 56 cases with SSpA, and three cases with AS and BD. They conducted intergroup comparisons including the control group and found that none of the BD patients had HLA B27 while sacroiliitis or enthesitis had slightly increased in severity in those with BD. In the patients with BD, positive HLA B51 was significantly higher than in those with a disease of the SSpA group. The patterns of eye involvement in the BD group were different. One of three cases at the AS and BD clinic was referred to the hospital complaining of right knee pain and swelling. Similar to our case, the major complaint of this patient was peripheral arthritis. In this patient, HLA B27 and HLA B51 were determined to be positive. Similar results also were found in our patient.

Çimen et al.[9] presented a case with clinical features of AS that occurred after the diagnosis of BD. A 36-year-old male patient was diagnosed as having BD 16 years ago. He had acute knee arthritis. In his physical examination, the range of movements of his neck and lower back were severely limited, and the HLA B27 and HLA B51 antigens were positive. Upon radiologic examination, high grade findings of AS were found. Although AS has a functionally and radiologically progressive course, the patient had been asymptomatic for years. This was probably due to the suppression of clinical symptoms of AS by the usage of drugs for BD. There were some similarities between this case and our case. Both patients were male and similar in age. Our patient also had bilateral knee arthritis. Another common point between these two cases was that the HLA B27 and HLA B51 antigens were positive in both cases, but there was no usage of any drug that suppresses the symptoms of AS in our patient's history.

According to our case, we concluded that with the absence or slight presence of symptoms such as inflammatory lower back pain and morning stiffness, the presence of peripheral arthritis as the predominant sign along with both HLA B27 and HLA B51 positivity can be considered as the coexistence of AS and BD.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Dilsen N, Konice M, Aral O. Why Behçet's disease should be accepted as a seronegative arthritis. In: Lehner T, Barns CG, editors. Recent advances in Behçet's disease. London: Royal Society of Medicine Services; 1986. p. 281-4.
2. Yazici H, Yurdakul S, Hamuryudan V, Fresko I. Behçet's syndrome. In: Hochberg MC, Silman AJ, Smolen JS, Weinblatt ME, Weisman MH, editors. Rheumatology. Spain: Mosby; 2003. p. 1665-9.
3. Yazici H, Yurdakul S, Hamuryudan V. Behçet's syndrome. In: Klippel JH, Dieppe PA, editors. Rheumatology. London: Mosby; 1998. p. 7.26.1-7.26.6.
4. Davatchi F, Shahram F, Chams-Davatchi C, Shams H, Nadji A, Akhlaghi M, et al. Behcet's disease: from East to West. Clin Rheumatol 2010;29:823-33.
5. Yazici H, Tuzlaci M, Yurdakul S. A controlled survey of sacroiliitis in Behcet's disease. Ann Rheum Dis 1981;40:558-9.
6. Chang HK, Cho EH, Kim JU, Herr H. A case of coexisting Behçet's disease and ankylosing spondylitis. Korean J Intern Med 2000;15:93-5.
7. Yazici H, Turunç M, Ozdoğan H, Yurdakul S, Akinci A, Barnes CG. Observer variation in grading sacroiliac radiographs might be a cause of 'sacroiliitis' reported in certain disease states. Ann Rheum Dis 1987;46:139-45.
8. Chang HK, Lee DH, Jung SM, Choi SJ, Kim JU, Choi YJ, et al. The comparison between Behçet's disease and spondyloarthritides: does Behçet's disease belong to the spondyloarthropathy complex? J Korean Med Sci 2002;17:524-9.
9. Cimen OB, Sahin G, Biçer A, Erdogan C. Behçet's syndrome coexisting with clinically occult ankylosing spondylitis. South Med J 2002;95:1355-6.