Coexistence of Behçet's Disease and Ankylosing Spondylitis

Sami Küçükşen[1], Sinan Bağçacı[1], A. Yavuz Karahan[2], Muhammed Şahin[1], Hatice Uğurlu[1]

ABSTRACT

Behçet's disease (BD) is a disease which has effects on different systems. Genital ulcer, aphthous stomatitis and iritis are characterized by triple symptom complex of BD. BD is considered to be a systemic vasculitis. BD was previously accepted in spondyloarthropathy (SSpA) group, but there are many reasons for not classifying BD as one of SSpA group. Ankylosing spondylitis (AS) is a prototype of seronegative spondyloarthropathy, and mainly axial skeleton is affected. In this paper, we aimed to present a-33-year old female patient with coexistence of BD and AS. In addition, the coexistence was aimed to be discussed.

Key words: *Behçet's disease, ankylosing spondylitis, coexistence*

Behçet Hastalığı ve Ankilozan Spondilit Birlikteliği

ÖZET

Behçet hastalığı (BH) farklı sistemler üzerine etkileri olan bir hastalıktır. Genital ülser, aftöz stomatit ve iritis, üçlü semptom kompleksi ile karakterizedir. BH sistemik bir vaskülit olarak kabul edilir. BH daha önceleri spondiloartropati (SSPA) grubu içerisinde kabul edilirdi, ancak BH' yi SSPA grubu içerisinde sınıflandırmamak için pek çok neden vardır. Ankilozan spondilit (AS) özellikle aksiyal iskelet tutulumu ile seyreden seronegatif spondilartropati grubunun bir prototipidir. Bu yazıda BH' na eşlik eden AS tablosuyla izlenen 33 yaşındaki bir kadın hastayı sunmayı ve bu iki hastalığın birlikteliğini tartışmayı amaçladık.

Anahtar kelimeler: *Behçet hastalığı, ankilozan spondilit, birliktelik*

INTRODUCTION

Behçet's disease (BD) is a disease which has effects on different systems. BD is characterized with oral or orogenital ulcers and various systemic (eye, skin, joint, central nervous system, and blood vessels) symptoms. The basic anatomical lesion is vasculitis (1,2). Ankylosing spondylitis (AS), a prototype of seronegative spondiloartropathy (SSpA) group, is a chronic inflammatory disease of the axial skeleton primarily involving the sacroiliac joint and vertebra. The coexistence of BD and AS has been rarely reported. Whether BD is one of SSpA group and whether BD progresses with sacroiliitis development have been subjects of debate (2). In this paper, we aimed to present a-33-year old female patient with coexistence of BD and AS. In addition, the coexistence was aimed to be discussed.

CASE

A-33-year old woman was admitted to our clinic with low back pain lasting for three months. She was suffering from oral ulcers frequently (4 times a month). BD had been diagnosed seven years ago. She had a history of erythema nodosum. A pathergy test was performed, and its finding was positive. Morning stiffness continu-

[1]Department of Physical Medicine and Rehabilitation of the Medical Faculty of Konya University, [2]Department of Physical Medicine and Rehabilitation of the State Hospital of Karaman

Correspondence: *Ali Yavuz Karahan*
Yunusemre mh Nurani Sk No:1 Meram/Konya, Türkiye
E-mail: *ayk222@hotmail.com*

Received: 13.02.2012, Accepted: 28.04.2012

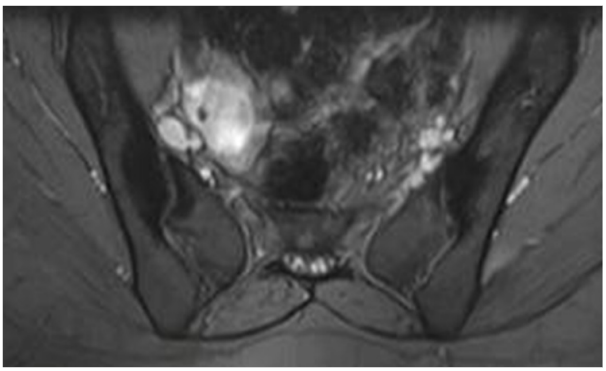

***Figure 1**. Sacroiliac MRI showing bilaterally sacroiliitis*

ing over 30 minutes was accompanied by back pain. On physical examination, spinal extension was limited and painful, and other spinal motions were open and painless. Tenderness on bilateral sacroiliac joints was present with compression. Schober test was:14,5 cm, and modified Schober test was 20cm. The erythrocyte sedimentation rate (ESR) was 25 mm/h, and the C-reactive protein level was 4 mg/dl (0-8). Other biochemical routine test results were normal. RF was negative. HLA-B27 and HLA-B51 were both positive. Sacroiliac Magnetic resonance imaging (MRI) showed bilaterally sacroiliitis (Figure 1).

Ankylosing spondylitis was diagnosed under the new Assessments in Spondyloarthritis International Society (ASAS) classification criteria (1), and the case met the Internal Study Group's (ISG) diagnostic criteria (2) for BD. The patient was administered colchicine 1 mg / day and started on sulfosalazine 2 g/day and indomethacin 150 mg/day. Two months later, patient's complaints significantly faded away.

DISCUSSION

There is a discussion on whether Behçet's disease is in the seronegative spondyloarthropathy group. BD is accepted as a vasculitic syndrome. AS is considered to be in SspA group. Dilsen et al. (3) carried out a study including 334 Turkish patients with BD. Among this study population, 10 % was reported to be AS, and 34% of the patients had sacroiliitis. HLA B51 and B27 were more frequently found to be positive in patients with coexisting BD and AS than normal population. Yazıcı et al. (4) reported only a single case defined AS among 114 patients with BD. This study showed no relationship between AS and BD. In our case, both HLA B51 and B27 were positive. In another study, Yazıcı et al. (5) mentioned that the inter-observer variation may be the major cause for discrepancies in the evaluation of pelvic radiography for sacroiliitis.

In 2004, ASAS developed a new diagnostic criteria for SSpA to be used in the early stage of disease (6). Before the development of the criteria, New York criteria mainly depending on radiographic sacroiliitis was being commonly used for the diagnosis. Radiographic evaluation for sacroiliitis often reflects no changes in early period (7). While utilizing the criteria, the observer could fail to detect the inflammation on radiographies at early stages. Radiography shows structural changes occuring due to inflammation in sacroiliac joints rather than the signs of inflammation. Inflammation in sacroiliac joints can be detected using MRI earlier than radiographies (8-11). In our case, we diagnosed sacroiliitis via sacroiliac MRI. AS was diagnosed under ASAS classification criteria about 3 months earlier than the beginning of symptoms. In other words, the diagnosis of AS was at an early stage in our case. The coexistence of AS and BD is a rare entity. On the other hand, the number of studies reporting this coexistence is increasing (12). Some medications like NSAIDs may be effective in the treatment of BD, and this may mask the symptoms of AS.

As the number of studies related to the coexistence between AS and BD increases, the relationship between the conditions will become more understandable. Some medications like NSAIDs may be used in BD, and this may mask symptoms of AS. Therefore, the symptoms of AS in patients with BD should be investigated meticulously to reveal the coexistence.

REFERENCES

1. *Zeidler H, Amor B. The Assessment in Spondyloarthritis International Society (ASAS) classification criteria for peripheral spondyloarthritis and for spondyloarthritis in general: the spondyloarthritis concept in progress. Ann Rheum Dis 2011;70:1-3.*
2. *O'Neill TW, Rigby AS, Silman AJ, Barnes C. Validation of the International Study Group criteria for Behçet's disease. Br J Rheumatol 1994;33:115-7.*
3. *Dilsen N, Koniçe M, Aral O. Why Behçet's disease should be accepted as a seronegative arthritis. In: Lehner T, Barnes CG eds. Recent advances in Behçet's disease. Royal Society of Medicine Services, London 1986: 281-4.*

4. Yazici H, Tuzlaci M, Yurdakul S. A controlled survey of sacroiliitis in Behçet's disease. Ann Rheum Dis 1981;40:558-9.

5. 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.

6. Rudwaleit M, van der Heijde D, Landewé R et al. The development of Assessment of Spondyloarthritis international Society classification criteria for axial spondyloarthritis (part II): validation and final selection. Ann Rheum Dis 2009;68:777-83.

7. van der Linden S, Valkenburg HA, Cats A. Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York criteria. Arthritis Rheum 1984;27:361-8.

8. Braun J, Bollow M, Eggens U, Konig H, Distler A, Sieper J. Use of dynamic magnetic resonance imaging with fast imaging in the detection of early and advanced sacroiliitis in spondylarthropathy patients. Arthritis Rheum 1994;37:1039-45.

9. Grigoryan M, Roemer FW, Mohr A, Genant HK. Imaging in spondyloarthropathies. Curr Rheumatol Rep 2004;6:102-9.

10. Oostveen J, Prevo R, den Boer J, van de Laar M. Early detection of sacroiliitis on magnetic resonance imaging and subsequent development of sacroiliitis on plain radiography. A prospective, longitudinal study. J Rheumatol 1999;26:1953-8.

11. Bennett AN, McGonagle D, O'Connor P et al. Severity of baseline magnetic resonance imaging-evident sacroiliitis and HLA-B27 status in early inflammatory back pain predict radiographically evident ankylosing spondylitis at eight years. Arthritis Rheum 2008;58:3413-8.

12. Yoo WH. Coexisting Behçet's disease and ankylosing spondylitis presented with deep venous thrombosis: a case report and review of the literature. Rheumatol Int 2010;30:1439-9.