



# Infliximab treatment in co-existing behcet's disease and ankylosing spondylitis case presentation

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## Abstract

Only a few cases of coexisting Behcet's disease and Ankylosing Spondylitis have been reported in the literature. In this article, we aimed to present a male patient with co-existing Behcet's disease and ankylosing spondylitis. However, the effectiveness of infliximab therapy in treatment co-existing Behcet's Disease and Ankylosing Spondylitis which was reported in this paper makes it interesting.

**Keywords:** Ankylosing spondylitis, behcet's disease, coexistence, infliximab, therapy

## Introduction

Behcet's disease (BD) is characterized by oral and genital aphthous and involvement of various organs such as eyes, skin, joints, central nervous system and blood vessels. Vasculitis is the main anatomic lesion [1,2]. Sacroiliitis is rarely seen in BD [3]. Ankylosing Spondylitis (AS) is the prototype of the seronegative spondyloarthropathy (SPA) group which involves the sacroiliac joint and the spine and results in spinal disability [1,2]. The claim whether BD is a sub-group of SPA is controversial [1].

In the recent years, co-existing BD and AS cases are more frequently identified [4,5] in the literature and attention on the co-existence of these two inflammatory rheumatic diseases for which auto-immune mechanisms are held responsible in the etiology is becoming more pronounced. In this article, we present the clinical features of the case involving a 29 years-old male diagnosed with co-existing BD and AS and the results of Tumor Necrosis Factor-alpha (TNF- $\alpha$ ) blocker (infliximab) treatment.

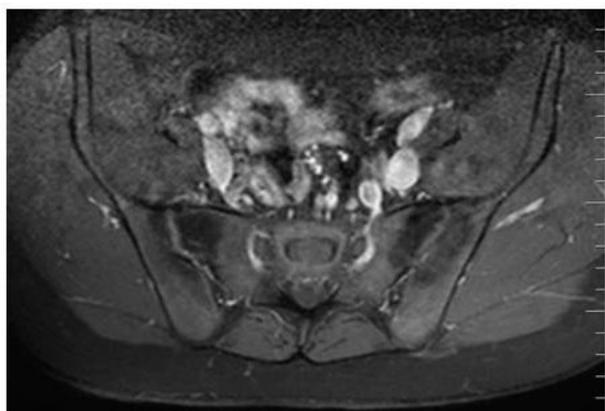
## Case presentation

Twenty nine years-old male patient applied to the Physical Medicine and Rehabilitation Clinic with a complaint of back pain and morning stiffness lasting since 5 years and aggravating since 6 months. Diagnosed with AS about 13 months ago, the patient was describing back pain with inflammatory characteristics which aggravates with rest and relieves with exercise despite his ongoing sulfasalazine 2000mg/day and indomethacin 75 mg/day treatment. Sacroiliitis was detected on his magnetic resonance imaginings (Figure 1).

Back movements were painful in the physical examination.

Tenderness detected at enthesi points. Sacroiliac joints were sensitive to compression. Gaenslen's test was bilateral positive. Finger to ground distance was 16 cm. Schober's test was 2.5 cm and modified Schober's test was 3.6 cm. Neck, back and pelvis examination was normal. Recovering oral aphthosis and painful genital aphthosis was detected in the examination (Figure 2). The patient was diagnosed as Behçet's Disease according to the International Criteria for Behçet's Disease (ICBD) [5]. Neurological examination was normal. Erythrocyte sedimentation rate was 64mm/hour, C-reactive protein (CRP) was 16mg/dl (0-6), and rheumatoid factor was negative in laboratory examinations. Other routine blood examinations were normal. HLA-B27 and HLAB51 were positive. No rheumatologic medical history in patient's family.

Patient's Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) value was scored as 6.6 and Bath Ankylosing Spondylitis Functional Index (BASFI) value as 6.4. Infliximab treatment was planned for the patient with uncontrolled disease activity with previous Nonsteroidal antiinflammatory drug (NSAID)'s and sulfasalazine use. Cardiologic and thoracic consultations of the patient were performed. Patient was taken isoniazid prophylaxis upon twice null Purified Protein Derivative (PPD) scores. Patient was administered a weekly 400 mg (5mg/kg) intravenous infliximab infusion on Week 0, Week 2, Week 6 and every 8 weeks subsequently. Sulfasalazine treatment was discontinued. Patient's BASDAI value was determined as 3.2 and BASFI value as 3.1 at the fifth cure of the treatment. Finger to ground distance was 13 cm. Schober's test was 3.2 cm and modified Schober's test was 3.6 cm. Neck, back and pelvis examination was normal. Recovering Also, laboratory



**Figure 1.** Magnetic resonance imaging of bilateral sacroiliac joint inflammation.



**Figure 2.** Aphthosis of glans penis was detected in the examination.

examinations were found to be normal.

## Discussion

BD is an inflammatory disease characterized with oral and genital aphthous, uveitis, and skin lesions [4]. Arthritis is usually mono- or oligo-articular in BD. Joint manifestations are 51%, arthralgia 35%, arthritides 23% and AS 2% [5]. Knee, ankle, and wrist involvement is very frequent, while distal interphalangeal joint, vertebra and sacroiliac involvement is very rare [7]. AS is a chronic inflammatory disease in which vertebra and sacroiliac joint is involved [6,8]. Etiology of BD and AS is not completely clarified. However, genetic predisposition, infection and environmental factors are the most emphasized subjects in both clinical pictures.

There are disputes on whether BD being a sub-group of SPA as sacroiliitis is rarely seen in BD [2]. There are studies

on co-existence of AS and sacroiliitis in patients with BD. Taarit et al., found that 6% of 309 BD patients with joint involvement had sacroiliitis and 2 patients had AS [3]. Oliveri et al., observed 6 sacroiliitis cases out of 20 patients with Behcet's disease and 1 sacroiliitis case out of 20 controls [6]. Dubost et al., [9] reported three cases of BD and AS coexistence among 11 patients with BD. Borman et al., detected HLA-B27 positivity and HLA-B51 negativity in a 29 years-old female patient with co-existing AS and BD [7]. Similarly, Etaouil et al., found HLA-B27 positivity and HLA-B51 negativity in 2 cases with combined AS and BD [10]. Yazıcı et al., assessed 184 BD patients in Turkish population and found AS only in 1 case [11]. Yazıcı et al., indicated that the sacroiliac joint antero-posterior assessment differ greatly from person to person, therefore suggested the prevalence of sacroiliitis in BD to be unclear [12]. Olivieri et al., recommended Computerized Tomography in order to reduce the differences arising from errors in the radiologic assessment of the sacroiliac joint. A relationship between HLA phenotypes of Behcet's disease and AS cases was detected in the literature [13]. There is HLA-B27 positivity in approximately 90% of SPA patients and HLA-B5 positivity in 60 to 80% of BD patients [8,11]. Incidence of HLA-B27 is more frequent in patients with co-existing BD and SPA [8,13]. Kallel et al., found that AS risk was higher in BD patients with negative HLA-B5 [2].

Ocular involvement in patients with BD and patients with AS also differ. Anterior part of the eye is affected in HLA-B27-related AS uveitis and it is benign. On the other hand, both anterior and posterior uveitis develop in BD and it may lead to vision loss in 25% of the patients [2,8].

Most commonly used anti-TNF agent in Behcet's disease is infliximab. Infliximab is used for the treatment of ocular involvement in particular, central nervous system (CNS) involvement, gastrointestinal system (GIS) involvement, muco-cutaneous findings, vascular involvement and arthritis. Intestinal BD is characterized by intestinal aphthous and gastrointestinal symptoms. The medical treatment of intestinal BD includes corticosteroids and immunosuppressants. There have been several reports of tumor necrosis factor- $\alpha$  blockers being successful in treatment of refractory intestinal BD. Chung et al., [14] reported on a patient who was diagnosed with intestinal Behcet's disease despite treatment with the fully humanized tumor necrosis factor  $\alpha$  blocker (adalimumab) for underlying ankylosing spondylitis. This patient achieved clinical remission through the addition of a steroid and azathioprine to the adalimumab regimen. Early and positive responses were received in a study where infliximab treatment was evaluated retrospectively; etanercept treatment was started because of relative decrease in response, no infliximab treatment was considered necessary in one case and other 6 cases stayed in remission with their every 8 week infliximab treatment [3].

## Conclusion

The coexistence of these two diseases is a rare entity but

the number of case reports is increasing. The conditions will become more intelligible if many other studies are done. This paper reports the satisfied therapy of infliximab treatment in co-existing BD and AS.

### Competing interests

The authors declare that they have no competing interests.

### Authors' contributions

Authors' contributions	BO	AYK	SCD	LT	SK	ES
Research concept and design	✓	✓	✓	✓	✓	✓
Collection and/or assembly of data	✓	✓	✓	✓	✓	✓
Data analysis and interpretation	✓	✓	✓	✓	✓	✓
Writing the article	✓	✓	✓	✓	✓	✓
Critical revision of the article	✓	✓	✓	✓	✓	✓
Final approval of article	✓	✓	✓	✓	✓	✓

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