A patient with ankylosing spondylitis and hemoptysis: is there a hidden disease? 
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Background An overlap syndrome of Behçet’s disease and ankylosing spondylitis is a rare autoimmune disease of connective tissue in which a patient presents with symptoms of two diseases.

Case presentation We report a case of non-smoking 25 years old known to have ankylosing spondylitis presented by haemoptysis due to a cause not usually associated with his primary disease leading to the search for another cause.

Conclusion The coexistence of Behçet’s disease and ankylosing spondylitis may be encountered in clinical practice, although it is rare. If there is a resistance to the conservative treatments, TN-alpha blocking agents may be an alternative therapeutic option in these diseases.

Comment An overlap syndrome is an autoimmune disease of connective tissue in which a patient presents with symptoms of two or more diseases. Diagnosis depends on the diseases for which the patient shows symptoms and for which the patient has positive antibodies in laboratory serology. Although the prevalence of coexisting Behçet’s disease and ankylosing spondylitis is much debated, it has been reported in few studies.
Ankylosing spondylitis is a connective tissue disease of unknown pathogenesis [1]. Young male individuals aged 20–40 years are the most commonly affected; 90% of them are HLA-B27-positive, with male : female ratio of 10 : 1 [2]. The reported prevalence of pulmonary disease ranges from 0 to 30%, including apical interstitial lung disease with or without apical fibrobullous disease, pleural diseases (such as pleuritis, exudative pleural effusion, apical pleural thickening, and pleural calcifications), thoracic cage immobility, and hemothorax, which may result from apical cavitory disease or rarely capillaritis. Other clinical manifestations include peripheral arthritis, enthesitis (inflammation of tendons and ligaments), and extra-articular organ involvement [1].

Behçet’s disease is a multisystem disease characterized by a triple-symptom complex consisting of genital ulcer, aphthous stomatitis, and iritis, frequently accompanied by vasculitis [1]. Behçet’s disease was reported to have a strong association with HLA-B51 rather than with HLA-B27; however, the number of reported patients with coexisting Behçet’s disease and ankylosing spondylitis (overlap syndrome) has increased. The role of proinflammatory cytokines such as tumor necrosis factor–α (TNF–α) and genetic factors is important in the pathogenesis of both diseases [3].

Reports concerning the coexistence of these two diseases are found rarely in the studies. Dubost et al. [4] reported three patients with coexisting Behçet’s disease and ankylosing spondylitis among 11 patients with Behçet’s disease. We found increased erythrocyte sedimentation rate and C-reactive protein levels, suggesting that the patient was in the active phase in terms of both diseases. Similar to our patient, ankylosing spondylitis may coexist with Behçet’s disease [5,6]. Therefore, this coexistence should be kept in mind. TNF–α blocking agents are commonly used in the treatment regimens for ankylosing spondylitis [7], which was not given to the patient in our study because of unavailability.

Conclusion
The coexistence of Behçet’s disease and ankylosing spondylitis may be encountered in clinical practice, although it is rare, and this condition should be kept in mind. If there is a resistance to the conservative treatments, TNF–α blocking agents may be an alternative therapeutic option in patients with these diseases.

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Conflicts of interest
None declared.

References