# Adult-onset Still's Disease Detected in Ankylosing Spondylitis Patient using Adalimumab

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Ankylosing spondylitis (AS) is a chronic inflammatory rheumatic disease where symptoms, such as fever and weight loss, are not expected, and may be part of a condition associated with these symptoms.[1-3] Adult-onset Still's disease (AOSD) is a rare systemic condition and is diagnosed by exclusion of other diseases. It includes high fever, rash, arthritis and various systemic symptoms. The combination of AS and AOSD is rare; and several cases have been reported in the literature.[2] Here, a case of AOSD that developed in a patient receiving adalimumab for a diagnosis of AS is reported.

A 62-year male, who had been diagnosed with AS in 2005 despite having symptoms since 1985, and who had been using adalimumab, 40 mg, every 15 days for 9 years, presented with sore throat, fever, chills, shivering, joint pains and weight loss for 4 days. Laboratory tests showed severe inflammation: C-reactive protein (CRP) was 192 mg/L; erythrocyte sedimentation rate (ESR) was 104 mm/1st h; white blood cell (WBC) count was $11.65\times10^3$/uL; hemoglobin, 13.20 g/dL; platelets, $401\times10^3$/uL; neutrophils, $9.03\times10^3$/uL; procalcitonin, 0.057 ng/ml, while liver function tests were normal.

Otorhinolaryngology and infectious disease consultants were consulted, who diagnosed it as pharyngitis, and ampicillin-sulbactam was administered. Acute phase reactants and fever persisted despite antibiotherapy. The search for an infectious disease was negative; a series of blood cultures at the time of febril peaks, throat and urine cultures, viral serology (HIV, hepatitis B and C, syphilis, EBV), anti-nuclear antibody (ANA), rheumatoid factor (RF), chest X-ray and echocardiography for endocarditis were all negative. Cervico-thoraco-abdominal computed tomography was performed to look for malignancy; there was no pathology except hepatomegaly.

After antibiotic treatment, non-pruritic rashes started and the skin lesions of the patient were evaluated as drug reaction by the dermatology specialist and antibiotherapy was discontinued. On 10$^{th}$ day of hospitalisation, laboratory tests were requested due to non-regressive acute phase reactants, persistent fever and existing skin rash. Ferritin level was 1397 ng/mL, methylprednisolone 100 mg, intravenously was initiated when the patient's clinical features complied with the diagnostic criteria for AOSD.[2] On corticosteroid treatment, fever subsided; sore throat, arthralgia and fatigue complaints disappeared, and ESR, CRP, and ferritin levels decreased. After observing response to methylprednisolone treatment, methotrexate 15 mg/week and folic acid were added. The patient has been in remission for 18 months.

Increased steroid dose and methotrexate supplementation are not always sufficient in the treatment of refractory AOSD. Tumor necrosis factor-alpha (TNF-α) blockers have been reported to be the most widely used biological drugs in the treatment of refractory AOSD.[4] However, their effects have been reported to be low when compared to other biological drugs (IL-1 antagonists, IL-6 inhibitors) in several publications.[5]

In conclusion, a patient with AS and receiving TNF inhibitor therapy, may develop super-added AOSD; and this should be kept in mind when a patient with AS develops fever, sore throat and various systemic symptoms. Methotrexate treatment was successful in the presented case. In AOSD occurring in a patient while using TNF-α inhibitor treatment, addition of methotrexate to the treatment may be effective.

**CONFLICT OF INTEREST:**
The authors declared no conflict of interest.

**AUTHORS' CONTRIBUTION:**
TID: Designed the work.
DYK: Drafted the work.
MO: Gave final approval.

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