CASE REPORT AND LITERATURE REVIEW

Rheumatology Quarterly

DOI: 10.4274/qrheumatol.galenos.2024.51523
Rheumatology Quarterly 2024;2(2):95-7

ANKYLOSING SPONDYLITIS COEXISTING WITH TAKAYASU'S ARTERITIS IN A FEMALE PATIENT: A RARE ENTITY

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Abstract

Ankylosing spondylitis (AS) is a chronic inflammatory disease that mainly affects the sacroiliac joints, spine, and enthesis. Typical symptoms include stiffness and gradual functional restriction of the axial skeleton. Takayasu's arteritis (TA) is a condition characterized by chronic inflammation of blood vessels, usually affecting major vessels like the aorta and its branches. It leads to constriction, obstruction, and the development of aneurysms in the systemic and pulmonary arteries. Both AS and TA are rare inflammatory conditions, and their occurrence together in one person is even rarer. Autoimmune mechanisms are likely to have a substantial impact on the onset of TA, similar to AS. Hence, therapy involving steroids and biological medications like infliximab and tocilizumab is expected to be advantageous. In this case report, we present a 24-year-old female diagnosed with TA and AS.

Keywords: Ankylosing spondylitis, Takayasu's arteritis, female, anti-TNF

INTRODUCTION

Ankylosing spondylitis (AS) is a persistent inflammatory condition that primarily affects the sacroiliac joints, spine, and enthesis. Stiffness and gradual functional restriction of the axial skeleton are typical symptoms of this condition, which may also be associated with sleep disturbances. Under the age of 40 years, males are the most likely to be affected by the disease (1-3). AS has a strong genetic component closely linked to the human leukocyte antigen B27 (4). Takayasu's arteritis (TA) is an uncommon chronic inflammation of blood vessels that typically impacts large vessels such as the aorta and its branches. It results in narrowing, blockage, and the formation of aneurysms in the systemic and pulmonary arteries (5). Clinical symptoms consist of pain resulting from inflammation in the arterial wall and, in a later phase, ischemic symptoms due to the narrowing of the arteries. Computed tomography (CT), magnetic resonance (MR), and ultrasound (US) imaging can reveal arterial thickening and stenosis. Classical angiography reveals narrowing or complete blockage of the affected arteries (6). Here we present the case of a 24-year-old female patient diagnosed with AS and TA.

CASE REPORT

Upon initial presentation, the patient reported experiencing pain in the waist, hips, knees, and heels and morning stiffness lasting for 1 hour. Pain was alleviated by movement. The pain was waking her up from sleep. She had oral canker sores more than three times a year. On physical examination, a murmur was detected over the left carotid artery, and pain upon palpation was detected in the knees, ankles, heel, and sacroiliac region. During the patient's most recent hospitalization, her joint pain persisted. The patient with AS was treated with adalimumab for 5 years.

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Received: 13.04.2024 **Accepted:** 23.05.2024

She discontinued the treatment 3 months 5 months ago and then resumed it. She was diagnosed with cardiac arrhythmia and hypertension. The patient was prescribed amlodipine, calcium dobesilate monohydrate, and doxazosin and was admitted to us for treatment planning. Brain MR imaging angiography revealed that the distal segment of the left vertebral artery was narrower than that of the right, and the right vertebral artery showed slight tortuosity. Both the distal segment of the vertebral arteries and the basilar artery were open. Carotid color Doppler US imaging detected stenosis in the carotid artery. The patient was suspected to have active vasculitis. The carotid walls were found to have stenosis and irregularities. Inflammation of the wall of the superior mesenteric artery was detected. A stenosis of approximately 60% was found in the common carotid artery. Carotid color Doppler US showed that the wall thickening was caused by vasculitis, leading to the development of jet flow due to stenosis, resulting in an audible murmur. Because of CT angiography interpretation, the diameter of the ascending aorta was 35 mm and was found to be at the upper limit of normal for age. The diameter of the aortic arch is 22 mm, and that of the descending aorta is 19 mm. It is in normal filling calibration. In the proximal localization of both renal arteries, secondary to vasculitis, there were vascular wall irregularities in a segment of 1.5 cm on the right and 2 cm on the left, 95% stenosis in the right renal artery, and 80% stenosis in the left renal artery. Inflammatory changes secondary to vasculitis. In the superior mesenteric artery, vascular wall irregularities and inflammatory changes cause 50% stenosis the entire trace. Distally, the superior mesenteric artery and its branches were recanalized with collaterals and were in normal filling calibration. The subclavian arteries, celiac artery, inferior mesenteric artery and its branches, iliac arteries, and proximal parts of the femoral arteries are in normal filling calibration (Figure 1).

The patient with a preliminary diagnosis of TA was scheduled to receive anti-tumor necrosis factor. The patient, who was recently diagnosed with TA, experienced an episode while receiving adalimumab. As a result, adalimumab was withdrawn and replaced with infliximab. In addition, we prescribed methotrexate, folic acid injection, and 16 mg of prednisolone.

DISCUSSION

Both AS and TA are uncommon inflammatory conditions, and their coexistence in one individual is even more infrequent. Research has shown that the presence of TA and AS may be coincidental or simultaneous because of certain underlying factors (1,2). AS typically affects the spine, hip, and enthesis, leading to a limited range of motion. TA is an inflammatory disease primarily affecting the aorta and its branches, leading to limb claudication pain (1,2,7). CT, MR, and US imaging can detect indications of inflammation, such as arterial thickness and stenosis. Classical angiography visualizes the narrowing or complete blockage of the arteries in question (8). A study by Rivière et al. (7) examined 14 patients with both TA and spondyloarthritis (SpA), including AS. Of these patients, 11 individuals with AS were of Caucasian ethnicity, and 10 of them were women. In addition, SpA occurred before TA in 13 individuals, with an average age at onset of 43.5 years, which was higher than the typical age of TA onset. These findings validate that AS is very prevalent in patients who have both SpA and TA simultaneously, and unlike the overall population with AS, the occurrence of TA is elevated. Thus, it is quite probable that the occurrence of these conditions in patients is not accidental but rather a result of an underlying component (7,9).

All of the individuals described in the literature are thought to have large vessel vasculitis characterized by aortic and collateral lesions. Our patient had bilateral sacroiliitis findings that were radiologically indistinguishable from AS. According to the case series documented by Mielnik et al. (2), if we exclude patients with unknown imaging results, peripheral arthritis was observed in 50% of the patients (2,8).

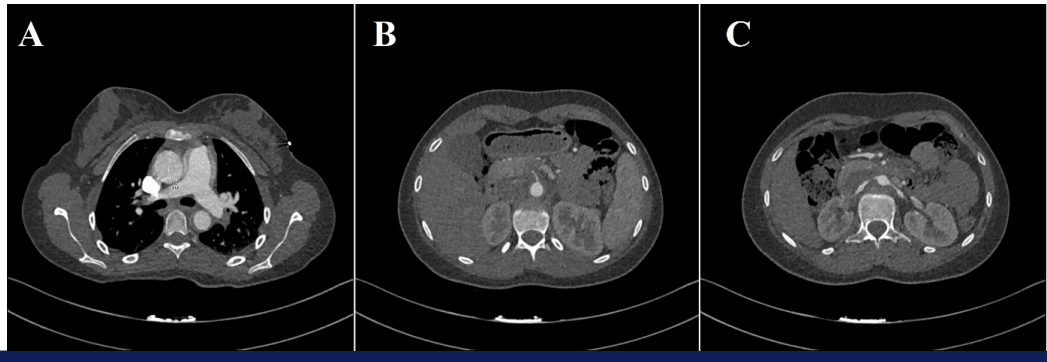

Figure 1. CT angiography images. A) Ascending aorta, B) superior mesenteric artery, C) right and left renal arteries
CT: Computed tomography

TA, like AS, is probably influenced by autoimmune processes. As a result, treatment with steroids and biological agents like tocilizumab is expected to be beneficial (10). Patients who develop TA during AS experience a sudden rise in C-reactive protein (CRP) levels, as observed in this case (1). We measured the peak CRP value in this study at 62.54 and reduced it to 8.03 over a period of 2 weeks.

CONCLUSION

Both diseases share characteristic findings, such as cytokine abnormalities. Although uncommon, AS typically occurs before TA in patients with both diseases, and TA usually develops at an older age.

Our patient was diagnosed with AS 6 years ago, and we diagnosed TA with our recent examinations. While it is possible that treatment for AS may prevent the development of TA, the simultaneous presence of both conditions is intriguing, necessitating further investigation into their relationship.

Ethics

Informed Consent: Written and verbal consent was obtained from the patient.

Authorship Contributions

Concept: M.A.D, B.T., Design: M.A.D, Data Collection or Processing: M.A.D, B.T., Analysis or Interpretation: M.A.D, B.T., Literature Search: M.A.D, Writing: M.A.D.

Conflict of Interest: The authors have no conflicts of interest to declare.

Financial Disclosure: The authors declared that this study received no financial support.

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