A Takayasu arteritis case presented with pulmonary involvement in a male patient with diabetic foot ulcers: Case report

Sadriye Pulak¹, İnan Anaforoğlu¹, Şakir Özgür Keşkek²*, Ekrem Algün³, Esin Ertuğrul⁴

¹Associate Professor, Department of Internal Medicine, Kanuni Training and Research Hospital, Trabzon, Turkey
²Associate Professor, Department of Internal Medicine, Numune Training and Research Hospital, Adana, Turkey
³Professor, Department of Internal Medicine, Rize University School of Medicine, Rize, Turkey
⁴Division of Rheumatology, Kanuni Training and Research Hospital, Trabzon, Turkey

Abstract

Takayasu’s arteritis is a rare form of chronic systemic vasculitis. It predominantly affects young adults, women in particular. We report an unusual case of Takayasu’s arteritis with pulmonary involvement in a 47-year-old male patient with diabetic foot ulcers. Patient was admitted to the hospital with high blood sugar, unhealed foot ulcers, weakness and weight loss. He developed sudden dyspnea during the treatment. The diagnosis of Takayasu’s arteritis was made using physical examination and computed tomography angiography.

Keywords: Takayasu’s arteritis, Pulmonary involvement, Diabetes, Diabetic foot ulcers.

INTRODUCTION

Takayasu’s arteritis (TA) is a chronic, progressive vasculitis of unknown etiology that primarily affects the aorta and its major branches. The first case of TA was described in 1908 by Takayasu, a Japanese ophthalmologist [1]. TA is a very rare systemic inflammatory disease with an estimated incidence of only about three cases per million of the U.S. population [2]. While 80 to 90 % of all cases are diagnosed in women, the most common age of onset is between 20 and 30 years. Most patients present with symptoms related to vascular insufficiency of the upper extremities, usually with fatigue or weakness in the arms (claudication). Physical examination findings include weak or absent pulse and a difference of >10 mmHg in systolic blood pressure between the arms. The non-specific early clinical features may lead to delay or misdiagnosis of TA [3].

We present an unusual case of TA with pulmonary involvement in a 47-year-old man with a history of type 2 diabetes mellitus and diabetic foot ulcers.

CASE REPORT

A 47-year-old man was admitted to the emergency department with complaints of pain on left shoulder, ulcers on the bottom of the feet and high blood sugar. He has had type 2 diabetes mellitus (DM), hypertension (HT), peripheral vascular disease, and dyslipidemia for 10 years, had unhealed ulcerous wounds on the bottoms of his feet for a year, and further developed anemia during the past 6 months. He also had loss of appetite and weakness, and lost approximately 5 kg in the last few months. Physical examination showed that posterior tibial, dorsalis pedis, popliteal and femoral arterial pulses were present in both legs. Respiratory and cardiovascular system examinations were normal. There were infected wounds of 2-3 cm in diameter on the bottom of both feet. Laboratory findings included a hemoglobin of 10.3 g/dL, WBC 12.7 10⁹/L, MCV 74 fL, glucose 259 mg/dL, HbA1c 11.4%, erythrocyte sedimentation rate 103 mm/h, C-reactive protein 26.7 mg/dL, creatinine 1.6 mg/dL, ferritin 110 ng/mL, iron 12 mg/dL, and iron binding capacity 104 mg/dL. X-rays of both feet were normal. Complete urine analysis tested positive only for leukocytes. Intensive insulin therapy was administered to control high blood sugar. Intravenous ampicillin-sulbactam (3*1 gr) and ciprofloxacin (2*200 mg) were administered for foot ulcers and urinary tract infection. Blood tests showed anemia of chronic disease.
Twenty-four hours after hospitalization, the patient developed sudden dyspnea. He was transferred to the intensive care unit due to hypoxia in arterial blood. Electrocardiogram showed sinus tachycardia. Serum D-dimer level was measured as 500 ng/mL, leading to suspicion of pulmonary embolism. Contrast-enhanced computed tomographic (CT) scan of the chest was not performed due to high creatinine level. Low-molecular-weight heparin was administered. Blood pressure was 100/50 mmHg in the left arm and 150/90 mmHg in the right arm. There was no palpable radial pulse in the left arm. These findings led us to consider the possibility of TA as the differential diagnosis. Unenhanced CT of the thorax revealed ground-glass opacities in both lungs, more prominent in the right lower lung. Following the decrease in creatinine levels, CT angiography was performed for pre-diagnosis of TA, and showed total occlusion of the left subclavian artery. There was 60-70% occlusion of the right common carotid artery (Figure 1), and an aneurysmatic dilatation enlarging the neck (1 cm) at the right side of the main pulmonary artery. Ground-glass opacity was observed in both lungs (Figure 2). Prevascular, para-aortic, para-tracheal, precarinal, and bilateral hilar lymph nodes (<1cm) were found. CT angiography performed for lower extremity and other branches of the aorta showed no pathology, except for an aneurysmatic dilatation at the distal left femoral artery (Figure 3). Based on physical examination and laboratory findings, a diagnosis of Takayasu’s arteritis with pulmonary involvement was made, and the treatment was started. The general condition of the patient was good during the treatment, and his blood sugar was under control. Routine follow-up visits were planned, and the patient was discharged.

**DISCUSSION**

TA is a progressive, chronic, systemic vasculitis of unknown etiology. It occurs mainly in young adults, women in particular. Diagnosis of TA in older male patients, such as our patient, is rare. Its etiopathogenesis is thought to be caused by cellular immunity. Anti-endothelial cell antibodies (AECA) are increased in patients with TA, and the AECA titers have been shown to correlate with disease activity. In a study conducted in Turkey, interleukin (IL) 12, IL-2 and IL-6 gen polymorphism were suggested to cause TA sensitivity.

Most cases exhibit nonspecific symptoms in the early periods of the disease, including fever, night sweats, fatigue, weight loss, myalgia, arthralgia and mild anemia. These non-specific early symptoms may cause a delay in diagnosis. However, although rarely, TA can also be diagnosed without systemic symptoms, using incidental signs including hypertension, arterial blood pressure and pulse differences between the arms. Our patient had long-term nonspecific symptoms such as weakness, fatigue, and weight loss.

The abnormalities in test results are generally non-specific in TA. Laboratory findings on admission to hospital may include increased C-reactive protein (CRP) and gamma globulin, decreased hemoglobin, and leukocytosis. Erythrocyte sedimentation rate (ESR) is generally high, especially in the exacerbation periods. Our patient also had low hemoglobin levels and an increased ESR.

Coronary artery disease and pulmonary arterial involvement are common in TA. Ulcers and gangrene may develop later due to extremity vascular occlusion. Our patient’s unhealing diabetic foot ulcers, which had been followed for the past year, might be associated with ischemia and inflammation caused by lower extremity vascular occlusions.

The classification criteria developed by the American College of Rheumatology in 1990 helps to distinguish TA from other vasculitis cases. According to the classification criteria, the diagnosis should include three or more of the listed criteria. The angiography, on the other hand, has been the golden standard investigation for the diagnosis of TA. In the present case, CT angiogram showed total occlusion of the left subclavian artery.

Prognosis of TA is dependent on the localization, distribution and complications of vasculitis. Hypertension, retinopathy, severe aortic insufficiency and aneurysm are the indicators of poor prognosis. The treatment of TA includes antiaggregants, corticosteroids, and cytotoxic drugs such as methotrexate. Activity of the disease can be evaluated by ESR, systemic complaints, and new and progressive vascular findings. Although 20% of all cases are asymptomatic, 80% of the cases may have active disease and need treatment during the diagnosis. Abnormal chest imaging findings are observed in about 45-80% of all cases. Additionally, angiographic abnormalities are found in 30-74% of the cases present. However, predominant pulmonary symptoms
and pulmonary involvement are rare \cite{14,15}. Our patient developed sudden dyspnea during the follow-up period. CT thorax showed ground-glass opacities. These findings might indicate symptoms of pulmonary involvement in TA.

**CONCLUSION**

In this case report, TA was predominantly presented with pulmonary symptoms. The case we presented was unusual because the patient was male, he was not young, and he further had diabetes and unhealed foot ulcers. This case shows that TA may also occur in a rare clinical picture. The diagnosis of TA further underscores the importance of physical examination.

To our knowledge, this is the first report of a TA with pulmonary involvement in a patient with diabetes mellitus and diabetic foot ulcers. In conclusion, TA may be accompanied by diabetes mellitus and diabetic foot ulcers. Foot ulcers that have been present for a long period may be related to chronic inflammation associated with rheumatic diseases. Therefore, we should also consider vasculitis in patients presenting with unhealed, long-lasting diabetic foot ulcers. Lastly, TA may also present with sudden dyspnea caused by pulmonary involvement.

**REFERENCES**