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Case Report

Early Relapse of Unusual Takayasu Arteritis with Infective Endocarditis

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Abstract

A 27-years-old female patient with lower limb rest pain and upper limb claudication. Blood culture revealed Streptococcus sanguinis. Diagnosis of Takayasu Arteritis (TA) with infective endocarditis was established based on her investigation result, patient started pulse corticosteroid then bridged to methotrexate. A few days later after response, a sudden early relapse occurred. We started Tocilizumab and she improved. Our case has an unusual presentation of TA with infective endocarditis. Tocilizumab is a promising treatment for TA especially in such cases with early relapse, but we have to emphasize its high efficacy in the long term.

Keywords: *Takayasu Arteritis; Infective Endocarditis; Vasculitis, Iliac Aneurysm; Tocilizumab; Alveolar Hemorrhage*

Introduction

Takayasu arteritis (TA) is a rare disease that was first reported in 1908 by a Japanese ophthalmologist, Mikito Takayasu [1]. It is a chronic granulomatous inflammatory disease affecting the large and medium sized vessels, mainly the aorta and its major branches. The incidence of TA in Japan among adults is estimated to be 2/1,000,000. However, TA is not limited to any ethnic group and is spread all over the world but most likely to be seen during the second or third decades of Asian female life, and is possibly a life-threatening condition [2,3]. Diagnosis of TA is based on 2022 American College of Rheumatology (ACR)/EULAR classification criteria, which requires that the age of the patient must be ≤ 60 years at the time of diagnosis and the presence of imaging evidence for vasculitis in addition to getting a score of ≥ 5 on a scoring system that consists of 10 clinical and imaging items [4]. The most effective treating agents are glucocorticoids (GCs) and immunosuppressive agents such as methotrexate (MTX), azathioprine (AZA), mycophenolic acid (MMF), and leflunomide (LEF). The biologic drugs including anti-TNF (rituximab) and anti-IL-6 (tocilizumab) seem to be promising especially in relapsed or refractory cases [5,6]. Hence, this report describes an unusual presentation of TA associated with infective endocarditis and the management course. The patient provided consent for the publication of the article and photographs in this case.

Case Presentation

A 27-years-old female patient complaining of left lower limb rest pain for 3 days associated right upper limb claudication, she had episode of right facial weakness with dysarthria for 1 hour, there is history of weight loss 20 kg for last 6 months, no history of pregnancy or abortion, no family history, no contact with tuberculosis patients or history of drug addiction. Upon examination there was systolic blood pressure difference in her arms more than 20 mmHg, unpalpable left common femoral artery with monophasic distal pulses, Intact sensation and motor-power and no tissue loss. There were unpalpable brachial and radial arteries with monophasic pulse. The erythrocyte sedimentation rate (ESR) was 53 mm/hour, and the C-reactive protein (CRP) was 171 mg/L. The blood culture showed Streptococcus sanguinis. The patient did Computerized Tomography Angiogram (CTA) of the abdomen and bilateral lower limbs (Figure 1) which showed left common iliac artery occlusion with abnormal wall enhancement with aneurysmal dilatation (3 × 1.7 cm). The diagnosis of Takayasu arteritis was made according to the 2022 ACR/EULAR classification criteria [4], our patient score was 8 namely, female sex, reduce upper extremity pulse, lower limb claudication, positive difference in bilateral upper limb blood pressure, and imaging findings. The patient started on methylprednisolone 1gm IV OD for 3 days then shifted to oral prednisolone 60 mg PO OD and 15 mg methotrexate SC once weekly. Antibiotics started Ceftriaxone 2 gm IV OD and Vancomycin 1 gm IV BID for infective endocarditis (IE) which was diagnosed by Echocardiography and confirmed by Trans-Esophageal Echogram (TEE) reported rheumatic mitral valve with severe stenosis with large vegetation (2 × 2.5 cm) and normal ejection fraction of 55%. Heparin infusion started and then bridged to warfarin. Magnetic Resonance Imaging (MRI) aortogram (Figure 2). Showed: Right mid subclavian artery is severely attenuated and total occlusion of the left common iliac artery with aneurysmal dilatation. Multidisciplinary team meeting conducted in the presence of vascular surgeon, cardiac surgeon Rheumatologist and infectious diseases team decision was made that she needs medical treatment in the form of antibiotics, in addition to TA treatment, then to be operated for open heart and mitral valve replacement after completing the treatment course. During admission she was diagnosed with alveolar hemorrhage by High resolution computerized tomography and treated for that. The patient had good clinical and laboratory responses to all given treatments and all symptoms were relieved. On day 20, sudden relapse and activity of the disease occurred. The patient started complaining of diffuse abdominal pain, back pain, ESR elevated to 52 and CRP to 219. CTA of the abdomen (Figure 3) showed new occluded aorta distal to the inferior mesenteric artery that extend to bilateral common iliac artery surronded by soft tissue thicking and fat stranding with left common iliac artery aneurysmal dilatation. We started the patient on Tocilizumab 162 mg SC once weekly with methylprednisolone 1 gm IV for 3 days. The patient improved and her symptoms were relieved entirely and her inflammatory markers returned to normal.

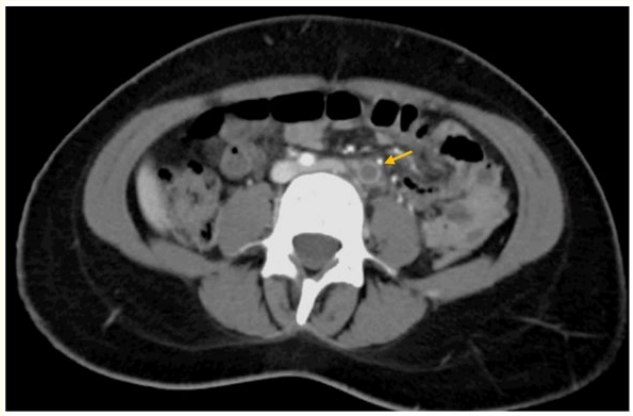

Figure 1: *Computerized tomography abdominal and lower limb angiogram showed: Left common iliac artery occlusion with abnormal enhancement and surrounding by soft tissue thickening with aneurysmal dilatation. "Marked with yellow arrow".*

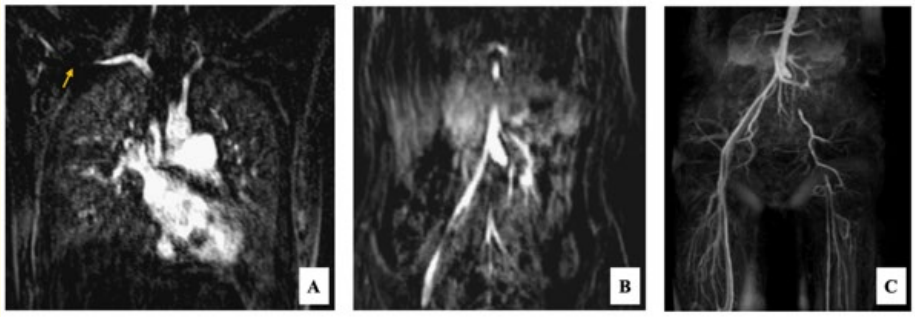

Figure 2: *Magnetic resonance aortogram. (A) Right attenuated mid subclavian artery. "Yellow arrow". (B) Left common iliac artery occlusion with aneurysmal dilatation. (C) Reconstructed image showing Left common iliac occlusion with collaterals.*

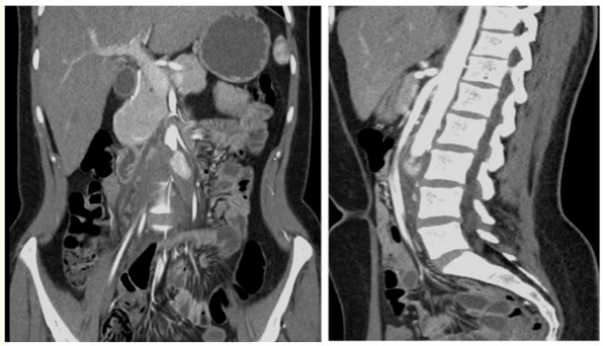

Figure 3: *Computerized tomography abdominal angiogram showed: Occluded aorta distal to the inferior mesenteric artery that extend to bilateral common iliac artery surrounded by soft tissue thickening and fat stranding with left common iliac artery aneurysmal dilatation.*

Discussion

Our patient was presented with right upper limb claudication and left lower limb rest pain and transient attack of neurological manifestation. The diagnosis of TA was made based on the ACR/EULAR criteria [4]. Then she had a sudden early relapse, so we started the Tocilizumab, after 5 days the patient improved clinically and her inflammatory markers returned to normal. Tocilizumab (TCZ) is anti-IL-6 therapy that has also been used successfully to induce remission and decrease the need for steroid therapy. The EULAR 2018 recommendation that considering adjunctive treatment with conventional immunosuppressive drugs at diagnosis of TA, anti-TNF or anti-IL-6 can be used as second line agents in case of relapsing disease and because of good- quality data on long- term treatment of TA are lacking, the duration of treatment must be decided on an individual basis [7]. The well-known cardiac manifestations of TA are aortic regurgitation, pulmonary hypertension, ischemic heart disease due to coronary artery involvement and left ventricular hypertrophy due to hypertension [8]. Most of the available large case series describe these cardiac manifestations in a significant proportion of patients with valvular heart disease occurring in more than 20% patients out of which aortic valve involvement contributing to the majority [9]. Cardiac failure is also known to occur in about 10–30% of the patients depending on the case series, but the symptoms are commonly described together with other vascular and systemic features [10]. There are several case reports where patients have presented with heart failure in Takayasu arteritis. A recent case report described a 15-year-old girl who presented with acute heart failure due to

myocarditis, however she had preceding arm claudication, systemic and cutaneous manifestations in the preceding one year [11]. Another case described a 27-year-old female who presented with features of heart failure was found to have severe pulmonary hypertension and dilated cardiomyopathy together with pulse and blood pressure difference between two arms [12]. Regarding our case patient had an unusual association with IE which is one of the well-known mimics for small vessels vasculitis however the vessels involvement in our case were medium to large vessels which is not known to be involved in case of IE and there are no reported cases neither data suggesting that involvement.

Conclusion

The unique presentation of our Takayasu patient is an accidental discover of unusual association of infective endocarditis and then the early relapse that had occurred in our case, nevertheless after Tocilizumab administration patient symptoms improved and relived, so we found that Tocilizumab is promising treatment for TA especially in early relapse cases, but we have to emphasize its high efficacy in the long term.

Declaration of Interests

The authors have no competing interests.

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