Case Report

Recurrent first-trimester abortion in a young female: Rare presentation of Takayasu arteritis

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Abstract

Takayasu arteritis (TA) is a chronic, progressive, autoimmune, idiopathic, and large-vessel vasculitis that usually affects young adults, especially females. TA primarily affects the aorta and its major branches, the coronary arteries, and the pulmonary arteries. Recurrent pregnancy loss is usually defined as three or more consecutive losses occurring at <20 weeks' gestation of a clinically recognized pregnancy. Common causes of recurrent fetal loss include anatomic, chromosomal, hormonal, infectious, or antiphospholipid antibody syndrome. However, to the best of our knowledge, TA causing recurrent fetal loss has not been described in the literature. We present such a rare case of a patient who presented with hemoptysis as her presenting complaint and also had a recurrent first-trimester abortion.

Keywords: Fetal loss, hemoptysis, iliac arteries, Takayasu arteritis

Introduction

Takayasu's arteritis (TA) is a chronic inflammatory disease that mainly affects the large arteries such as, the aorta and its main branches. However, to a lesser extent, the common iliac arteries can be involved as well. Nonspecific arteritis was described first by Savory in 1856.[1] Takayasu,[2] in 1908, noted the ocular changes of the disease. Over the years, arteritis has gained interest of both clinicians and pathologists. Initially, it was thought that TA involved only aorta and its branches. However, later on, it was found that any medium and large artery may be involved in TA.

Case Report

A 25-year-old female presented to our hospital with the chief complaints of streaky hemoptysis for the past 5 days. There was no history of breathlessness, cough, fever, chest pain, weight loss, night sweats, transient visual obscurations, syncopal attacks, postprandial abdominal pain, lower limb ulcers, Raynaud's phenomenon, alopecia, or photosensitivity. However, the patient gave a history of bilateral lower limb claudication with decreasing claudication distance for the past 6 months. There was history of 3 consecutive first trimester abortions also. There was no family history of hypertension or renal disease.

On admission, she was afebrile, had tachycardia with a pulse rate of 110 beats per minute. His right and left brachial blood pressures were 176/98 and 180/94 mmHg, respectively. Her lower limb blood pressure was not recordable. There was no bruit over any of the vessels. There were feeble bilateral femoral pulses. Popliteal, dorsalis pedis, and posterior interosseus pulses were absent. No bruit was detected over the abdomen or elsewhere. The skin examination was normal. Her fundus examination revealed changes of Grade 2 hypertensive retinopathy. Neurological examination was normal.

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Laboratory investigations showed following results: Complete blood count, liver, renal function tests, random blood sugar, and urinalysis were normal. The Westergen erythrocyte sedimentation rate (ESR) was 126. The C-reactive protein was 8.8 mg/dl. Her serum was negative for rheumatoid factor, antinuclear factors, anti-phospholipid antibodies, and antineutrophil cytoplasmic antibodies. Hypercoagulable workup did not reveal factor v Leiden. Protein C, Protein S, and antithrombin three levels were normal. No prothrombin gene mutation was detected. The complement factors C3 and C4 were normal. The hepatitis B antigen, HIV, and venereal disease research laboratory was negative. The tuberculin skin test was negative. Thyroid function test and coagulation profile were normal. Chest radiography, renal Doppler, and echocardiography were normal ultrasonography abdomen, and pelvis was also normal. Computed tomography (CT) angiography of the great vessels revealed diffuse narrowing of descending thoracic and abdominal aorta (abdominal >thoracic) [Figure 1] with tortuous, irregular margins and diffuse wall thickening, especially in the suprarenal abdominal aorta. Calcific plaques were seen in mid-thoracic aorta. There was a diffuse narrowing of thoracic and abdominal aorta [Figure 2] with narrowing extending to bilateral iliac arteries. Right main pulmonary artery and ascending aorta were prominent. Aortic arch branches were unaffected [Figure 3].

A possibility of TA was kept and patient was started on prednisolone with an initial dose of 1 mg/kg/day and mycophenolate mofetil 1 g twice daily. The dose of prednisolone was maintained for 6 weeks and then a slow taper of the prednisolone was started. The patient was also given amlodipine for the control of hypertension. Currently, patient is on prednisolone 10 mg daily and mycophenolate mofetil and is doing fine. Blood pressure is 130/80 mm hg. The lower limb pulses have returned and are well-palpable now after 1 year of follow-up ESR is 20.

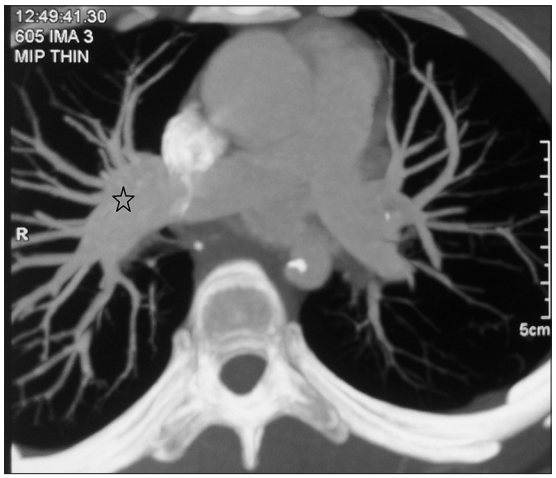

Figure 1: Axial maximum intensity projection image showing enlarged right main pulmonary artery (starred) with irregular margins (aneurysmal dilatation)

Discussion

Incidence of TA is 1.2–2.6/million per year in the western population, but the incidence is higher in Southeast Asia, Central and South America, and Africa.[3]

Females are affected in 80–90% of patients with TA, mostly in the second and third decades of life. The pathologic process in TA involves medium and large caliber arteries; thus, it is not surprising that the pulmonary arteries may be involved. TA is characterized by granulomatous inflammation of the arterial wall with marked intimal proliferation and fibrosis of the media and adventitia. The lesions are segmental with a patchy distribution.[4]

A study by Lupi *et al.*,[5] evaluated the radiological features of pulmonary artery involvement in TA. He found two out of six patients with pulmonary artery involvement to have pulmonary artery dilatation. Pulmonary artery hypertension may occur in 25% of cases with pulmonary artery involvement.[6,7]

Our patient had history of recurrent abortions. Hypercoagulable workup was negative. However, clinical examination pointed toward a disease process affecting the large vessels. As the inflammatory markers were positive a possibility of large vessel vasculitis was kept. CT angiography revealed involvement of thoracic, abdominal aorta, and common iliac arteries as well. As the uterine artery is a branch of the anterior division of internal

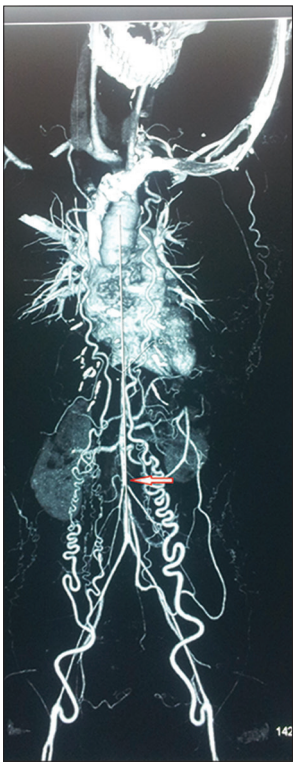

Figure 2: Coronal maximum intensity projection image showing diffuse narrowing of abdominal aorta (black arrow)

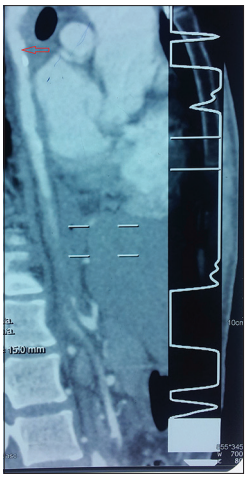

Figure 3: Sagittal maximum intensity projection image showing tortuous irregular wall thickening and diffuse narrowing of descending thoracic and abdominal aorta (abd > thoracic), especially in suprarenal abdominal aorta (straight arrows). Note is made of focal wall calcification in proximal descending thoracic aorta (curved arrow)

iliac artery. To our mind, narrowing of the common iliac artery lead to decreased blood flow in the uterine artery accounting for recurrent fetal losses.

Causes of recurrent fetal losses include genetic factors, endocrine causes, autoimmune disorders such as antiphospholipid antibody syndrome and uterine causes. TA has never been reported as an etiologic risk factor for recurrent fetal losses. Pistorius *et al.* reported a case of an isolated aortoilliac TA where a 30-year-old woman had acute limb ischemia complicated by second trimester abortion. Subsequently, the woman was found to have aortoiliac Takayasu disease.[8]

Common iliac involvement in TA is not a common event. Arnaud *et al.* in their series of 82 patients reported the involvement of right-sided iliac arteries in 18% and left-sided iliac arteries in 12% of patients.[9]

Our case is unique in the sense that TA presenting as recurrent abortions has not been reported in literature. Involvement of the descending thoracic and abdominal aorta is also not very common. Study from Jain *et al.*[10] showed the similar pattern of involvement in 3.8% of Indian patients. As the steroid resistant nature of the disease is being increasingly recognized and discussed. Such a rapid response was not anticipated and the further behavior of the disease in this patient will decide whether any further immunosupression is warranted or not.

Whether the recurrent abortions were purely due to the occlusive involvement of the iliac vessels leading to uterine hypoperfusion or a part of complications of the systemic vasculitis left untreated during pregnancy is an issue of debate.

Hence, we conclude that the list of differentials while investigating a case of recurrent abortions needs to be expanded. Yes, TA/large vessel vasculitis should be added and looked for, if no other etiology is found and there are indirect clinical markers for it as described in our case.

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Conflicts of interest

There are no conflicts of interest.

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