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RESEARCH ARTICLE

A rare presentation of takayasu arteritis- swelling and allodynia of both ankles and feet

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ABSTRACT

Takayasu is a challenging large vessel vasculitis to diagnose and treat. Takayasu mostly occurred in young women, and its most common symptoms are pulselessness in limbs, hypertension, and claudication. In our case, we have an unusual presentation. A young woman with a history of IBD came to our clinic with pulselessness, coldness, edema and allodynia in lower limbs. In the investigations, an imaging diagnosis of Takayasu vasculitis was confirmed.

KEYWORDS:
Rare, swelling, presentation .

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INTRODUCTION

The patient was a white woman with 34 years of age who was referred to our clinic with pain, swelling, and paresthesia of both ankles and feet. She had arthralgia from five months ago that had been worsened, especially in the left foot over the past month. In her past medical history, she had ulcerative colitis that was in the remission. In family history, her mother was a known case of ankylosing spondylitis.

In the physical examinations, both ankles and feet were swollen and cold, and the range of motion of ankles was decreased. The swelling in metatarsophalangeal joints (MTP) 2nd and 3rd of the right hand and MTP 4th of the left hand were observed. Her gastroenterologist with suspicious of articular manifestations of UC prescribed 15mg prednisolone daily and adalimumab weekly. During the investigations, the gastroenterologist did not find anything to confirm the relapse of ulcerative colitis. The patient had no diarrhea, fever or rectal bleeding, and her calprotectin was normal. It seemed that her signs didn't have correlation to her ulcerative colitis. The gastroenterologist referred her to rheumatology clinic. In the examination allodynia in both limbs were observed. The paresthesia and ruptured blister, and some cyanosis on her fifth and third toes were also observed. The pulse of the right radial, dorsalis pedis, and posterior tibial were not detected on any sides. The left upper limb and both lower limbs were pulseless, bruit in right and left subclavian artery and the right arm was heard. Also, the pressure of blood could not be detected in the left arm.

Laboratory data

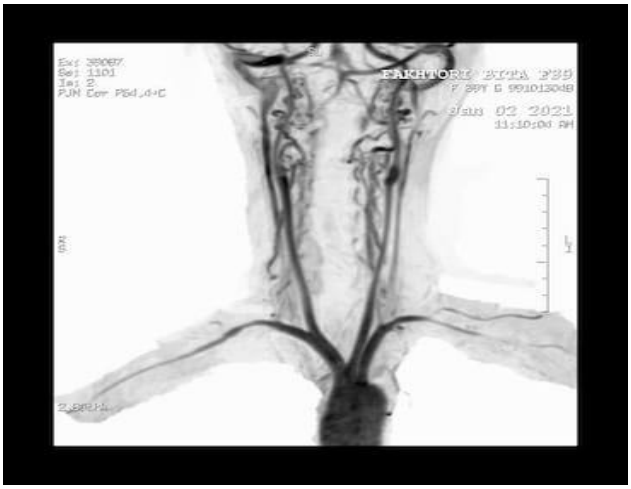
WBC:12400/cumm. (normal range: 4400-11000), Hemoglobulin: 11 (normal range: 12.3-15.3) plt283000(150000-450000)Bun:18 (8-25) Creatinin:1mg/dl (0.6-1.2) ESR:110 mm.CRP:3(normal range:up to 6)

AST, ALT, ALP, ANA, Antipoospholipid AB.RF, AntiCCP, CANCA. PANCA , C3, C4.CH50 and fecal calprotectin were negative. HLA Typing for HLA_B52 was negative.

Color Doppler Sonography of both lower limbs Arteries and Veins showed severe to near occlusion stenosis within both lower limbs arteries that begins from common femoral in the right side, and common iliac in the left side.

CT Angiography of the lower limb 3D Reconstruction showed multiple cut-off and run-off of both lower limbs as follows; in the distal aspect of external and origin of common femoral in the left side, the proximal and middle third of SFA in the left side, and the middle third of SFA in the right side. Popliteal trifurcation, ATA and PTA were open. The distal portion of the abdominal aorta and the major branches, and the left and right common iliac are open with normal diameter.

Fig. 1:



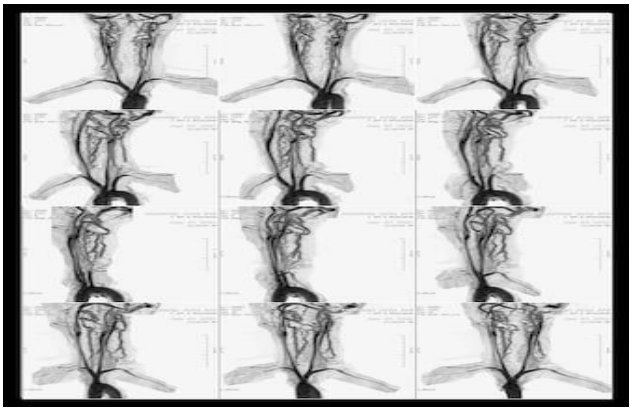


Fig.2:

MR angiography of the neck revealed a long segment narrowing in the left carotid artery extending up to the carotid bifurcation, highly suggestive of Takayasu disease. The MR Angiography of the thoracic aorta showed a long segment narrowing with some irregularities seen in both subclavian arteries extending toward the axillary on the left side. The MR angiography of the abdominal aorta was normal.

Above these findings, by diagnosing Takayasu disease, CellCept 3 gram and prednisolone 50 mg daily begun, but her signs did not disappear. Therefore, infliximab 3mg/kg begun and CellCept discontinued. One month later, the claudication and pulses were improved. Five months later in a follow-up, her radial pulse and posterior tibialis and dorsalis pedis were not detected on any side, and the left subclavian bruit was heard and in follow up lab test, ESR reduced to 45 mm.

DISCUSSION

Takayasu is a large vessel vasculitis, which is mostly observed in younger women. The incidence of TA is highest in the fourth decade [1]. Inflammations in the endothelium may lead to luminal occlusion, stenoses, thromboses, and aneurysmal dilatation [2]. Despite the fact that TA may manifest as different symptoms, most of the cases have signs of vascular insufficiency (from stenosis, occlusion, or aneurysm), systemic inflammations, or both. The signs that are majorly observed include pain in the legs or arm (35%), decreased or absent pulse (25%), carotidynia (20%), high blood pressure (20%), carotid bruit (20%) dizziness (20%), and unsymmetrical blood pressure of the arms (15%). Strokes, aortic regurgitations, and visual abnormalities may be observed at the initial phases in less than ten percent of cases [3]. In the year 1990, from the more typical features of Takayasu's arteritis, the American College of Rheumatology (ACR) introduced particular characteristics of this disease. However, angiography is still the gold standard method for diagnosing these patients [4].

Table 1: 1990 ACR criteria for classifying Takayasu arteritis [5].

Criterion	Definition
The start of this disorder in less than 40 years of age	Developing signs or manifestations associated with Takayasu arteritis at less than 40 years of age
Claudication of extremities	Developing exacerbated fatigue and pain in muscles of some extremities during activities, especially the upper extremities
Reduced pulses of brachial artery	Reduced pulses in one or both brachial arteries
Higher than ten millimeters of mercury difference between the blood pressures	Higher than 10 mm Hg difference between the systolic blood pressures of the two arms
Bruit on the subclavian arteries or aorta	Bruits that could be detected with auscultation on one or both subclavian arteries or abdominal aorta
Abnormalities in the arteriogram	Arteriographic narrowings or occlusions throughout the aorta, the major branches, and major arteries in the proximal upper or lower extremities, which are not induced by arteriosclerosis, fibromuscular dysplasia, and equivalent reasons; these changes are regularly focal or segmental.

* At least 3 of the mentioned criteria are required for diagnosing Takayasu's arteritis.

Six characteristics were identified for the diagnosis of the disorder using the traditional format, which are related to age, claudication, arterial pulses, blood pressure, bruit on the arteries or the aorta, and narrowings or occlusions of the aorta and its major primary branches. Detection of three or more criteria demonstrates 90.5% sensitivity and 97.8% specificity [6].

The sign and symptoms of Takayasu are variable. The constitutional signs of the disorder such as losing weight, mild

fevers, arthralgias, limb claudication, hypertension, skin lesions, gastrointestinal symptoms, and neurologic symptoms are some of the Takayasu features.

In our case regard to the history of ulcerative colitis, we must rule out that the articular presentation of the patient is not related to UC. Ulcerative colitis has some extraintestinal manifestation such as musculoskeletal involvement. The peripheral spondyloarthropathies in ulcerative colitis has two subtypes. Pauciarticular (Type 1) peripheral arthritis consists of self-limited episodes, which may last about 10 weeks or fewer, and it is associated with intestinal-IBD activities. Polyarticular (Type 2) peripheral arthritis has different manifestations including symmetrical arthritis that involves small joints, and is not associated with IBD activities [7]. In our case, the physical examination and laboratory data were not compatible to any of the musculoskeletal manifestation of ulcerative colitis. In our patient, further the swelling of the ankles and foot the allodynia, coldness and pulselessness were obvious and there was not any gastrointestinal sign and symptom, and these articular manifestations did not respond to prednisolone. According to the signs and high titer ESR and the criteria in Table1, the diagnosis of Takayasu was established and confirmed with MRI angiography.

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