

Left Circumflex Pseudoaneurysm and Left Anterior Descending Aneurysm in Coronary Artery Due to Behcet's Disease

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To the Editor: A 33-year-old Chinese male, with a 5-year history of recurrent oral and genital ulcers, was admitted to our hospital for chest tightness, fever, and tiredness from 2 weeks. The patient had been diagnosed with Behcet's disease (BD) 3 years before admission to our hospital, suffered from deep venous thrombosis related to BD, and was being treated with prednisolone. Physical examination revealed multiple varisized lesions occurred in the skin of periorbital and lower extremities. Electrocardiogram and chest X-ray were negative, and laboratory data, including creatine kinase-MB and cardiac troponin, were within normal limits. The patient persisted with shortness of breath and thoracic oppression; therefore, a conventional contrast-enhanced coronary computed tomography angiography (CTA) was performed and showed a mass lesion of mixed density (46 mm × 40 mm × 65 mm) arising from the left circumflex coronary artery and compressing the left principal bronchus and left auricle [Figure 1]. In addition, CTA demonstrated an aneurysm measuring approximately 7 mm in diameter in the left anterior descending coronary artery. Based on CTA finding and preventing fatal complications due to this rapidly expanding pseudoaneurysm, urgent surgery was deemed necessary. No pericarditis or pericardial effusion was found preoperatively. The pseudoaneurysm sac was opened, and the thrombus was evacuated before restored blood flow with a bypass had been rapidly elected for controlling the pseudoaneurysm and revascularization. Moreover, basic treatment with prednisolone and cyclophosphamide was also adopted postoperatively. The patient was discharged 8 days later. At his 6-month follow-up, he was asymptomatic and had recovered full activity with no dyspnea or angina. However, we regret that the patient refused to receive coronary CTA review because of his good feeling and high expense of CTA examination.

BD is a rare multisystemic chronic inflammatory disease of unknown etiology, first described in 1937.^[1] It is suggested that there may be a genetic predisposition (human leukocyte antigen [HLA], HLA-B51) and that certain microorganisms may play a role. This disease presents in young adults from the Mediterranean region, the Middle East, and the Far East, with an equal prevalence

in both genders, but more severely affecting males. In 2010, in an effort to improve clinical sensitivity in diagnosing BD, revised diagnostic criteria were proposed by the international criteria for BD.^[2] These criteria included oral aphthosis, genital ulcers, ocular manifestations, skin lesions, vascular manifestations of different sizes in various organs, central nervous system involvement, and positive pathergy test. Getting four or more points indicates BD diagnosis (oral aphthosis two points, genital ulcers two points, ocular manifestations two points, and the remaining each one point). In our case, clinical characteristics and symptoms of this patient with BD were coincided with previous reported literatures.

Arterial involvement is a late complication with the prevalence from 3% to 5% in the population of BD, and it occurs a median of 4 years after the initial diagnosis of BD. The arterial affection includes aneurysms and occlusion, mostly not only involving the abdominal aorta and its branches but also the thoracic aorta; in fact, BD is the most common cause of pulmonary artery aneurysm. However, coronary arteries are seldom involved, and extracranial carotid pseudoaneurysms are rarer than those affecting the aorta, pulmonary, and femoral arteries. Although coronary artery aneurysms are usually encountered in patients with BD, as far as we know, there have been few reports of existence of both aneurysm and pseudoaneurysm formations in the coronary artery simultaneously. The development of aneurysm and pseudoaneurysm at arteries in BD can be explained by the pathologic process in the wall of the blood vessels. Immune complex deposition in small vessels leads to complement fixation and polymorphonuclear leukocyte activation. The neutrophil activation and perivascular infiltration in the vessel wall lead to degeneration and occlusion of the vasa

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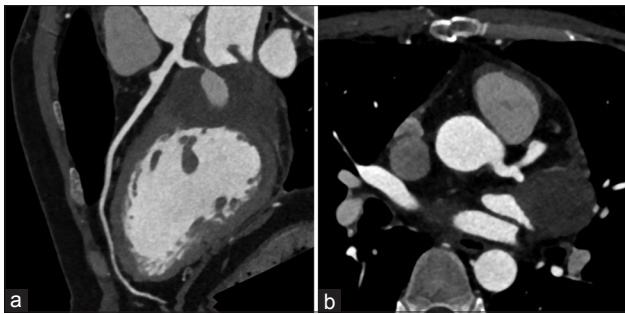


Figure 1: A 46 mm × 40 mm × 65 mm pseudoaneurysm with surrounding thrombus arising from the left circumflex coronary artery and an aneurysm measured 7 mm in diameter located on the left circumflex coronary artery were detected on CTA. (a) 3D spiral CTA section. (b) Axial section. CTA: Computed tomography angiography; 3D: Three-dimensional.

vasorum. Therefore, the main difference between true aneurysm and pseudoaneurysm is the lack of an adventitia in pseudoaneurysm, where the whole wall is lacerated.^[3,4]

Arterial involvement is the leading cause of death for patients with BD, with 30% of patients with aneurysms dying within 2 years and 2-year mortality rate after the onset of hemoptysis being 50%. Therefore, it is very important to diagnose the vascular involvement because of its relevance to the patient's prognosis and its high mortality. As an excellent noninvasive technique of diagnosis, CTA can not only reveal aneurysmal dilatation of the arteries or their branches but also display the thrombus in the lumen of aneurysm; therefore, CTA is widely used for diagnosing arterial involvement in patients with BD.^[4]

Systemic corticosteroids are required for disease exacerbation, and sometimes, adding some other form of immunosuppressive treatment is necessary when serious end-organ involvement occurs. Steroid therapy may sometimes be effective for treating unruptured

aneurysms, as these may show some regression. However, the rupture, a pseudoaneurysm, or aneurysm can be fatal, and the operation is recommended. The methods adopted by surgeons include open surgery, such as restoring blood flow with a bypass, as this case, or repairing with a patch and endovascular therapy, such as administering stents, vein-covered stent, or coils.^[5]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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