Case Report

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# Giant Aneurysms of the LAD and CX Arteries associated with Behçet's Disease: A Very Rare Association

Yakup Alsancak[1], Zeynep Demir[1], Muhammed Fatih Kaleli[1], Ahmet Seyfeddin Gürbüz[1], Ahmet Soylu[1]
[1]Department of Cardiology, University Necmettin Erbakan, Konya, Türkiye

## Abstract

Behçet's disease (BD) is a chronic multisystem vasculitis involving arteries and veins of all sizes. Coronary artery involvement is rare but potentially life-threatening. Giant coronary artery aneurysms associated with BD are uncommon and may present with acute coronary syndrome in young patients without traditional cardiovascular risk factors. A 23-year-old man presented to the emergency department with retrosternal squeezing chest pain radiating to the neck and shoulder, worsening with inspiration and positional changes. He had a recent upper respiratory tract infection and a history of recurrent pericarditis. Electrocardiography was unremarkable, while transthoracic echocardiography demonstrated anterior and lateral wall hypokinesia, mildly reduced left ventricular ejection fraction (45%), and minimal pericardial effusion. Laboratory tests revealed elevated troponin and inflammatory markers. Coronary angiography, performed due to progressive decline in left ventricular function, showed a giant aneurysm with pseudoaneurysm formation in the proximal left anterior descending artery accompanied by severe distal stenosis, as well as a saccular aneurysm of the circumflex artery. Further evaluation revealed genital scars and a positive pathergy test, leading to the diagnosis of Behçet's disease. The patient was treated with immunosuppressive and optimal medical therapy, and surgical intervention was planned. Coronary artery aneurysm secondary to Behçet's disease should be considered in young patients presenting with chest pain and minimal cardiovascular risk factors. Early diagnosis and multidisciplinary management are crucial because coronary involvement may rapidly progress and significantly increase morbidity and mortality.

**Keywords:** Behçet's disease, chest pain, coronary artery aneurysm, giant aneurysm

## Introduction

Behçet's disease, or Behçet's syndrome, is a chronic inflammatory condition primarily affecting the body's blood vessels. Common symptoms include recurring painful oral and genital ulcers, inflammation of the eyes (uveitis), and skin lesions. It can also impact other areas, such as the gastrointestinal tract, joints, nervous system, and blood vessels, leading to complications (1).

Coronary artery disease (CAD) in BD requires awareness by the clinician, as it typically affects young subjects and may manifest as ischemic heart disease. Myocardial infarction due to coronary arteritis was reported in up to 17% of BS patients with cardiac involvement (2) and in 25% of cases may be the presenting feature of cardio-BD (3,4).

Cardiovascular manifestations have been observed in 7% to 46% of patients with BD, and mortality is reported in up to 20% of patients with significant vascular involvement. Reports of cardiovascular involvement include cases of endocarditis, myocarditis, pericarditis, acute myocardial infarction, aortic aneurysms, ventricular thrombosis, congestive cardiomyopathy, and valve dysfunction (5).

Vascular involvement is a common complication of BD. Venous involvement predominates over arterial involvement. Arterial lesions, such as arterial occlusions, stenosis, and aneurysmal formations, are uncommon complications in patients with BD. However, they closely correlate with patient mortality and morbidity. Coronary artery involvement in BD is rare and may manifest in the form of myocardial infarction or angina, and its frequency is yet to be defined. In this regard, a 2019 study by Zhou et al (6) found coronary aneurysms and pseudoaneurysms to be present in 7 of 69 patients, which amounts to 10.1% of the population studied; the mean age of the patients was 30.5 ± 11.3 (7).

## Case Report

A 23-year-old male patient presented with ongoing chest pain that was characterized a retrosternal squeezing sensation, radiating to the neck and shoulder for, worsening on

**Corresponding Author:** Zeynep Demir
**e-mail:** dr.zeyneppdemir@gmail.com
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inspiration and change of position one week after an upper respiratory tract infection. His physical examination revealed as normal and there was no risk factors for coronary artery disease except smoking. No pathological signs were detected with electrocardiogram (Figure-1) and the transthoracic echocardiography demonstrated an anterior and lateral walls hypokinesis of the left ventricle with a 45 % estimated ejection fraction and minimal pericardial effusion.

Laboratory tests showed no abnormality except elevated troponin and white blood count levels. So, colchicine 0,5 mg 2*1, high dose acetylsalicylate 500 mg*4, beta-blocker and angiotensin converting enzyme inhibitor therapy received for a pre-diagnosis of myopericarditis. Moreover, we learned that the patient was hospitalized for two times because of pericarditis episodes in a year from his past medical history. Although the patient was treated with colchicine and nonsteroidal anti-inflammatory in previous pericarditis attack, we detected that the patient did not regularly use after-discharge treatments. In addition, it was observed that the patient was evaluated for the etiology of recurrent pericarditis after the second episode and that the markers of connective tissue disease such as antinuclear antibodies (ANA) or rheumatoid factor were within normal limits. On the other hand, we noticed that the patient's left ventricula systolic function had a progressive reduction in one year. For that reason, we decided to perform coronary angiography to exclude the premature atherosclerotic coronary artery disease. Coronary angiogram revealed a large aneurysm and pseudoaneurysm of the proximal LAD associated with 90% stenosis at the distal level of the aneurysm and saccular circumflex artery aneurysm with a normal right coronary artery (Figure-2)

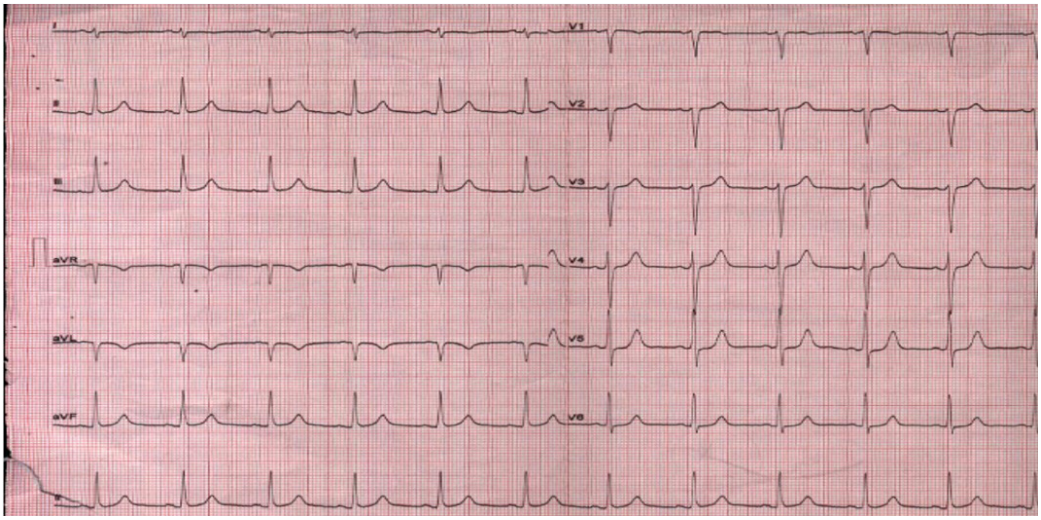


**Figure 1.** Electrocardiogram

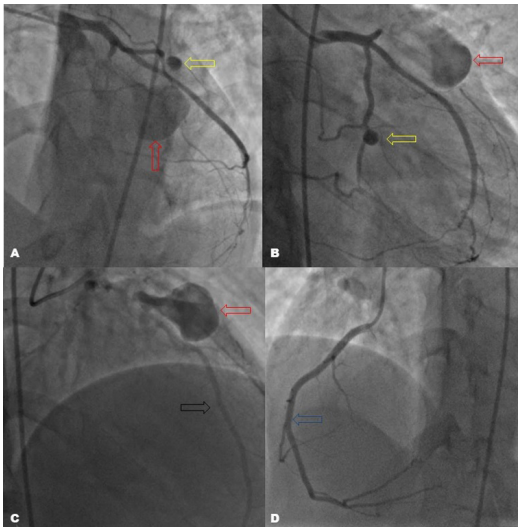


**Figure 2.** Coronary angiographic images demonstrating giant aneurysms in the left anterıor descending (LAD) and circumflex (CX) arteries.
(A) Proximal LAD aneurysm (arrow)
(B) Aneurysmal dilatation of the CX artery (arrow)
(C) Close-up view of the LAD aneurysm (arrow)
(D) Distal coronary segment without significant aneurysmal involvement

The patient was examined again for possible connective tissue diseases due to detection of coronary aneurysms. The physical examination at the department showed genital scars and pathergy test was considered as positive. The eye examination was normal in term of uveitis, but the ANA and ANCA level were detected high. At that time, the C-reactive and erythrocyte sedimentation rate levels were elevated to 2.77 mg/dl and 38 mm/h, respectively. The patient was diagnosed with Behçet's syndrome.

After the patient was evaluated together with the heart team and rheumatology department, conservative management with aspirin, metoprolol, and atorvastatin, angiotensin-converting enzyme inhibitor, high dose prednisolone,immunosuppressive therapy and colchicine was started. After the acute treatment period, surgical correction was planned and the patient was discharged.

## Discussion

BD, first described in 1937 by Hulusi Behcet, is an autoimmune vasculitic disease. Its distribution was noted in Mediterranean countries, Iran, China, and Japan. It is a multi-system disorder affecting eye, skin, and musculoskeletal, neurological and cardiovascular systems (8,9). The ICBD criteria is considered a highly sensitive and specific tool for diagnosis and classification of this disease. Ocular lesions, oral aphthosis and genital aphthosis are each assigned 2 points, while skin lesions, central nervous system involvement and vascular manifestations 1 point each. The pathergy test, when used, was assigned 1 point. A patient scoring ≥ 4 points is classified as having BD (10).

Vascular aneurysms are the other significant cardiovascular manifestation and complication of vascular Behçet's disease. Arteritis and inflammatory obliterative endarteritis and immune deposition of the vasa vasorum with fibrotic deterioration of the media are the major pathologic findings that causes to predispose the arterial wall to aneurysm formation (11).

Since the rates of coronary aneurysms in patients with Behcet's disease are low, there is no sufficient clinical information about the long-term prognosis of aneurysms. Coronary involvement is quite rare in BD and its prevalence is about 0.5% (12). It might be considered that coronary aneurysm with behçet's disease may have a higher rupture risk due to the nature of active vasculitis disease. So, a surgical approach to coronary aneurysm, such as coronary artery bypass grafting or resection for aneurysms and percutaneous interventions are known as a modality of treatment. In general, experience based approached suggests that adjuvant therapy choice with corticosteroids and immunosuppressive agents, colchium, and possibly with anticoagulants or antiaggregants, may be effective in reducing postoperative recurrences or graft occlusions or other surgical complications.

But, the management of coronary aneurysms may be very complicated due to its nature and effects on cardiac function. Coronary artery aneurysm in Behcet's disease may present as angina, myocardial infarction or sudden cardiac death due to rupture, embolization or thrombosis. Coronary aneurysm associated with severe stenosis can make this situation more complicated, especially in patients with non-received medical therapy. A previously published study has investigated the cases for behçet disease and myocardial infarction with coronary aneurysm. They showed that the patients were generally young, usually male, without vascular risk factors, except smoking (13). Coronary aneursym, Behçet's disease and accompanied acute myocardial infarction may be one of the most difficult puzzle that encounter of an interventional cardiologist, because of appropriate therapeutic approach is currently not clear. Different therapeutic options were reported by some authors such as high dose of corticosteroids, thrombolytic agents or primary percutaneous coronary interventions. Successful percutaneous stenting has been described for a giant right coronary aneurysm with a covered coronary stent, but there are case reports that has been associated with a high risk of subacute thrombosis and pseudoaneurysm formation at the site of stent placement (12, 14). And also a hybrid approach of distal CABG and proximal aneurysm coil embolization for a huge left anterior descending coronary artery aneursym with stenosis after acute myocardial infarction has also been reported (15). In addition, endovascular coil embolization of an aneurysm related with Behçet's disease has been reported due to failure of surgical suture (16). Moreover, anticoagulant and antiplatelet therapy have been offered as an option in these patients. In a published report, patient with Behçet's disease have been hospitalised due to acute coronary syndrom and recieved an intravenous infusion of glycoprotein IIb/IIIa receptor inhibitor after demonstrating right coronary aneursym with thrombosis. But, a rapidly grown up left anterior descending coronary artery aneursym and right coronary aneursym have been shown after three weeks (17). This process may explain with an active inflammation during acute coronary syndrome on vascular endothelial surface. We think that progression of aneurysm is not associated with antithrombotic therapy, but it may a cause of occurring a pseudoaneurysm. Another problem that needs to be answered is intensive corticosteroid therapy after a myocardial infarction for new diagnosis of a Behçet's disease. Treatment should be individualized according to the patient's clinical condition and myocardial performance.

Aneurysms have been shown to progress even in patients receiving adequate treatment, when most of the cases in the literature are evaluated together. It may also be thought that there is no relationship between the duration time of diagnosis and development of aneurysms. It can be seen that some of aneurysm have been diagnosed after the long illness period and some of them have appeared with the disease

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diagnosis at younger ages. Cardiologists should keep in minds the diagnosis of Behçet's disease in young patients after myocardial infarction. Furthermore, the entire vascular system must be evaluated for involvement, when a coronary aneurysm is detected in patients with Behçet's disease. Additional vascular system involvement has been observed in the majority of cases in the literature. Magnetic resonance angiography of our patient was in normal range. Another important issue is the management of patients with a large aneurysm. Although surgical or percutaneous treatment has been effective in early period, recurrent aneurysms can be annoying problem at minor traumatic segments.

## Conclusion

In conclusion, the clinical condition of the patient, the aneurysms character and the specific treatment for Behçet's disease should be evaluated together to decided appropriate approach to aneurysm. We think that percutaneous or surgical invasive intervention is most reasonable strategy after a short period of immunosuppressive treatment for Behçet's disease except emergency cases.

**Conflict of interest:** None

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