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Acute myocardial infarction caused by coronary artery aneurysm in Behçet's disease and review of the literature

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ABSTRACT

Behçet's disease is a chronic relapsing disease with multiple organ system involvement characterized clinically by oral and genital aphthae, cutaneous lesions, and ophthalmological, neurological, and/or gastrointestinal manifestations. It is an autoimmune vasculitis with rare incidence of vascular involvement, and especially of coronary arteries. The case present in this paper has coronary artery aneurysm in associated with Behçet's disease. Coronary angiography has revealed an aneurysm on the left anterior descending coronary artery of a 45-year old male patient who admitted our clinic with acute myocardial infarction. Successful coronary artery bypass grafting operation with left internal mammary artery to left anterior descending coronary artery (LAD) was performed after proximal and distal ligation of aneurysm.

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1. Introduction

Postmortem and antemortem studies have indicated that the incidence of coronary artery aneurysms (CAA) change between 1.5% and 5% (Syed and Lesch, 1997). However, it is rather difficult to determine the incidence of giant coronary artery aneurysms. Mainly coronary atherosclerosis, but also Kawasaki's disease, trauma, autoimmune diseases (polyarteritis nodosa, systemic lupus erythematosus, scleroderma) and coronary dissection can cause CAA (Ebina et al., 2009). Coronary artery involvement in Behçet's disease is rare. In this case report, we described a patient with coronary artery aneurysm associated with Behçet's disease.

2. Case

A 45-year old male patient was admitted to the cardiology clinics with typical retrosternal pain. Electrocardiography showed 0.5 mm ST elevation and pathological Q waves in the V1–6 derivations. His medical history included Behçet's disease and a surgically treated popliteal artery aneurysm four years ago. Physical examination revealed a blood pressure of 130/60 mmHg, heart rate of 76 beats per minute, partial loss of vision in the left eye. Laboratory investigations revealed that cardiac enzymes and troponin levels had increased. Acute myocardial infarction diagnosis was made. Primary percutaneous coronary intervention was not attempted because of the chest pain and ST segment elevation had receded. The next day, coronary angiography was performed by Judkins technique through the right femoral artery. Coronary aneurysm was seen located proximally in the left anterior descending coronary artery (LAD) with a serious narrowness at the distal end (diameter 16X48 mm) (Fig. 1). The left circumflex and the right coronary arteries were normal. Left ventriculography showed advanced hypokinesis on the anterior wall. After the procedure, revascularization

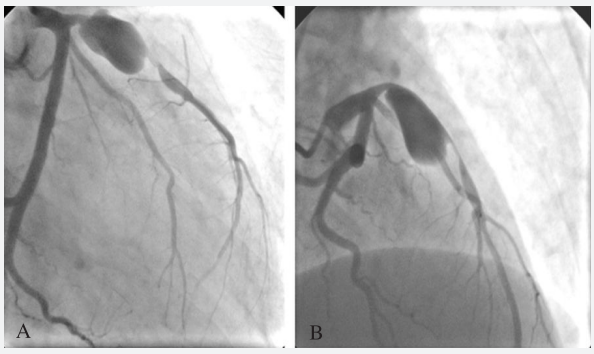

Fig. 1. A. Right anterior oblique and caudal angulation view of left coronay angiography **B.** Right anterior oblique and cranial angulation view of left coronay angiography

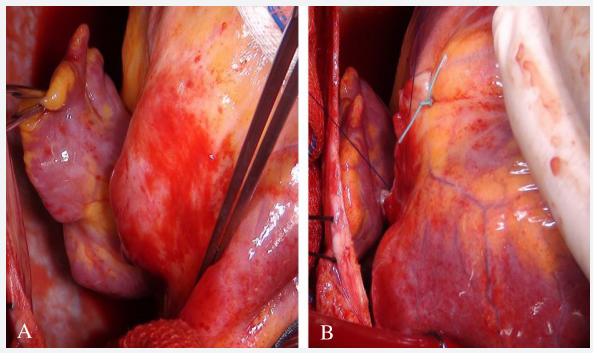

Fig. 2. Coronary aneurysm during surgery. **A.** Left anterior descending artery aneurysm is shown before ligation. **B**. After ligation

with graft coated stent was considered. However, the length of the aneurysm did not allow for this technique and surgical revascularization was performed. Successful coronary artery bypass grafting operation with left internal mammary artery to LAD artery was performed after proximal and distal ligation of aneurysm (Fig. 2). Surgery was performed via a middle sternotomy using standard cardiopulmonary bypass. The patient tolerated the procedure well and was discharged on postoperative day eight.

3. Discussion

The most frequently encountered cause of coronary aneurysms is atherosclerosis (Dogan et al., 2006). It is coincidentally met during coronary angiography performed on the asymptomatic patients or those presenting with chest pain. Investigation of the cases reported in the literature indicates that it is frequently localized on the right coronary artery (Ebina et al., 2009). Although conservative treatment is the preferred approach, surgical treatment is performed in symptomatic cases.

Although the arterial aneurysms are rare in Behçet's disease, their pathophysiology is well understood. Formation of immune deposits in the vaso vasorum results in inflammatory obliterative endarteritis causing fibrosis and loss of the elastic support and aneurysmal dilatations, and even the rupture of the arterial wall (Matsumoto et al., 1991). CAAs are seen in large arteries like the abdominal aorta, pulmonary artery and femoral artery. In the literature, coronary artery aneurysms associated with Behçet's disease have been reported in only few cases (Barçın et al., 2004; Ozeren et al., 2004; Geyik et al., 2005; Dogan et al., 2006; Cuisset et al., 2007; Porcu et al., 2008; Cevik et al., 2009; Kasapis et al., 2009; Cook et al., 2010; Doğan et al., 2011; Greenhouse et al, 2011; Okutucu et al., 2011; Spiliotopoulos et al., 2011; Yildiz et al., 2012). Investigations of these reports have indicated that unlike the atherosclerotic CAA, in Behçet's disease the CAAs are located more frequently on the

Table 1. Reported cases of coronary artery aneurysm in patients with Behçet's disease

No	Author	Max diameter (mm)	Sex	Age (years)	Coronary	Presentation	Additional vessel involvement	Treatment
1	Doğan A.	Unknown	M	32	LAD	STEMI (Anterior)		Surgery
2	Cuisset T.	Unknown	M	27	LAD	NSTMI		Medical
3	Porcu P.	25	M	29	LAD	Chest pain (UAP)		Surgery
4	Cevik ve ark.	Unknown	M	41	LAD, RCA	Chest pain (SAP)		Medical
5	Okutucu S.	Unknown	M	34	LAD	Dyspnea	Cerebral artery aneurysm	Medical
6	Özeren M.	50	M	33	LAD, RCA	Chest pain (SAP)	Femoral artery aneurysm	Surgery
7	Doğan SM.	Unknown	M	67	LMCA	Chest pain		Surgery
8	Geyik B.	Unknown	M	40	LAD	Chest pain		Medical
9	Barçın C.	Unknown	M	49	LMCA	Chest pain		Surgery
10	Cook AL.	43.7	F	16	LAD	Chest pain	Thrombotic occlusion of the superior vena cava	Surgery
11	Greenhouse DG.	67	F	19	LAD	Asymptomatic		Surgery
12	Kasapis C.	53	F	30	RCA	Chest pain	Multiple deep venous thromboses, pulmonary embolism, and cerebral transverse sinus thrombosis	Covered coronary stent
13	Spiliotopoulos K.	44	F	22	LAD	Chest pain, presyncope, palpitation, dyspnea		Surgery
14	Yıldız A.	20	F	37	RCA	Cardiac tamponade		Medical

M: Male; **F:** Female; **LAD:** Left anterior dessending artery; **RCA:** Right coronary artery; **STEMI:** ST segment elevation myocardial infarction; **NSTMI:** Non-ST segment elevation myocardial infarction; **UAP:** Unstable angina pektoris; **SAP:** Stable angina pektoris

LAD (Table 1). The most frequent symptom is chest pain. In the literature, apart from ours, only one other case report with ST elevated MI has been encountered (Doğan et al., 2011). However, one case with MI without ST elevation (Cuisset et al., 2007), one case with unstable angina (Porcu et al., 2008) and one case with stable angina pectoris (Cevik et al., 2009) have been diagnosed.

In our case we have encountered a history of surgical treatment of popliteal artery aneurysm four years previously. There have been very few earlier reports on coronary artery aneurysms together with aneurysms at other locations. One case has been reported with cerebral artery aneurysm, and one with femoral artery aneurysm (Ozeren et al., 2004; Okutucu et al., 2011).

In the literature, conservative approach is the most frequently used option for CAAs. Surgical treatment was recommended in cases with ischemia or high rupture risk. Only Kasapis et al. have successfully treated an aneurysm on the right coronary artery (RCA) with a 3.0x26 mm coated coronary stent. Although a similar approach was considered in our case the length of the aneurysm did not allow for this, therefore, surgical treatment had to be performed.

In conclusion, CCA's in association with Behçet's disease are very rare. Presence or the history of other aneurysms in these patients could be a warning criterion for coronary artery involvement. Although there have not been enough numbers of cases to recommend the mode of therapy, the conservative approach may be suitable in the asymptomatic cases.

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