

# Left Anterior Descending and Right Coronary Artery-Left Ventricular Multiple Micro-Fistulas Presenting as Angina Pectoris: Case Report

## Sol Ön İnen ve Sağ Koroner Arter ile Sol Ventrikül Arasında Multipl Mikrofistüller Olan Hastanın Anjina Pectoris ile Prezantasyonu

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Geliş Tarihi/Received: 21.05.2010  
Kabul Tarihi/Accepted: 20.12.2010

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**ABSTRACT** A rare type of congenital coronary fistula to the left ventricle, known as ‘coronary artery-left ventricular multiple micro-fistulas’ was defined within last thirty years. Morphologic features and treatment options of these extremely rare fistulas differ significantly with respect to classical solitary coronary fistulas. A 63 year-old woman admitted to our cardiology clinic with a complaint of typical angina pectoris. After coronary angiography, left anterior descending and right coronary-left ventricular multiple micro-fistulas were diagnosed. She became asymptomatic after medical treatment and she did not experience any cardiac event in the past sixteen months. Coronary artery-left ventricular multiple micro-fistulas can be easily overlooked, so all physicians performing coronary angiography should be careful about these fistulas.

**Key Words:** Angina pectoris; microvascular angina; arterio-arterial fistula

**ÖZET** Son otuz yılda ‘Koroner arter-sol ventrikül multipl mikrofistüller’ olarak adlandırılan nadir bir konjenital koroner arter fistül tipi tanımlandı. Oldukça nadir rastlanan bu fistüllerin morfolojik özellikleri ve tedavi biçimi, soliter koroner arter fistüllerine göre belirgin farklılıklar göstermektedir. Altmış üç yaşında kadın hasta, kardiyoloji bölümümüze tipik anjina pectoris yakınması ile başvurdu. Koroner anjiyografi sonrası sol ön inen ve sağ koroner arterler ile sol ventrikül arasında çok sayıda mikrofistül tespit edildi. Medikal tedaviye cevap veren ve asemptomatik olan hasta 16 aylık takibinde herhangi bir kardiyak olay yaşamadı. ‘Koroner arter-sol ventrikül multipl mikrofistüller’ kolaylıkla gözden kaçabileceğinden, koroner anjiyografi yapan tüm kardiyologların bu konuda dikkatli olmaları gerekmektedir.

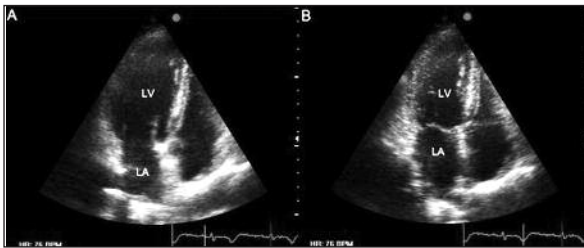
**Anahtar Kelimeler:** Angina pectoris; mikrovasküler anjina; arteriyo-arteriyel fistül

Türkiye Klinikleri J Cardiovasc Sci 2012;24(3):302-5

Congenital solitary coronary fistulas are seen rarely and most of them drain into the right heart chambers, great cardiac veins, coronary sinus or pulmonary arteries.<sup>1,2</sup> However, another special and even rarer type of congenital fistula between coronaries and left ventricle, known as ‘coronary artery-left ventricular multiple micro-fistulas (CA-LVMMFs)’ which was defined within last thirty years.<sup>3-5</sup> Morphologic features and treatment options of these extremely rare fistulas differ significantly from classical solitary coronary fistulas. We present a patient with typical angina pectoris caused by bilateral CA-LVMMFs and successful management of angina pectoris with medical treatment.

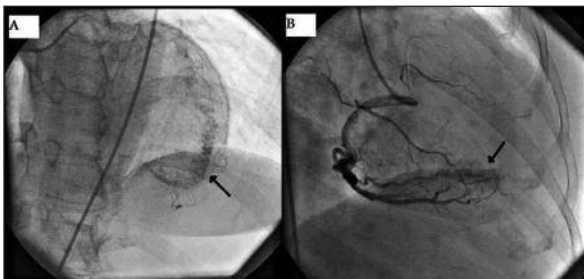
## CASE REPORT

Sixty-three year old woman with a past medical history significant for hypertension and mild diabetes mellitus admitted to our cardiology department with a complaint of typical angina pectoris. Physical examination was normal. Blood pressure was about 130/80 mm Hg and ECG was normal sinus rhythm with a rate of 74 bpm. Transthoracic echocardiography (TTE) revealed grade 1 diastolic dysfunction without left or right ventricular hypertrophy (left ventricular wall thickness, 9 mm), aortic dilatation or any other pathology that may cause cardiac chest pain (Figure 1). Coronary angiography showed non-obstructive atherosclerotic lesions, but revealed multiple microvascular fistulas draining into the left ventricle arising from left anterior descending and right coronary arteries (Figure 2). In venous phase, coronary sinus was clearly detected. Ventriculography was normal (Figure 3).

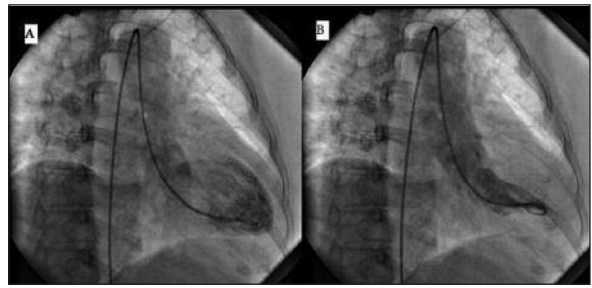


**FIGURE 1:** Diastolic (A) and systolic (B) transthoracic echocardiographic frames from apical four chamber view showing normal chamber sizes and ventricular wall thicknesses.

LA: left atrium, LV: left ventricle.



**FIGURE 2:** A) Left anterior oblique projection of left coronary angiogram showing opacification of left ventricular cavity (arrow) by micro-fistulas arising from distal parts of left anterior descending artery and B) Right anterior oblique projection of right coronary angiogram showing opacification of the left ventricular cavity (arrow) by micro-fistulas arising from distal parts of right coronary artery.



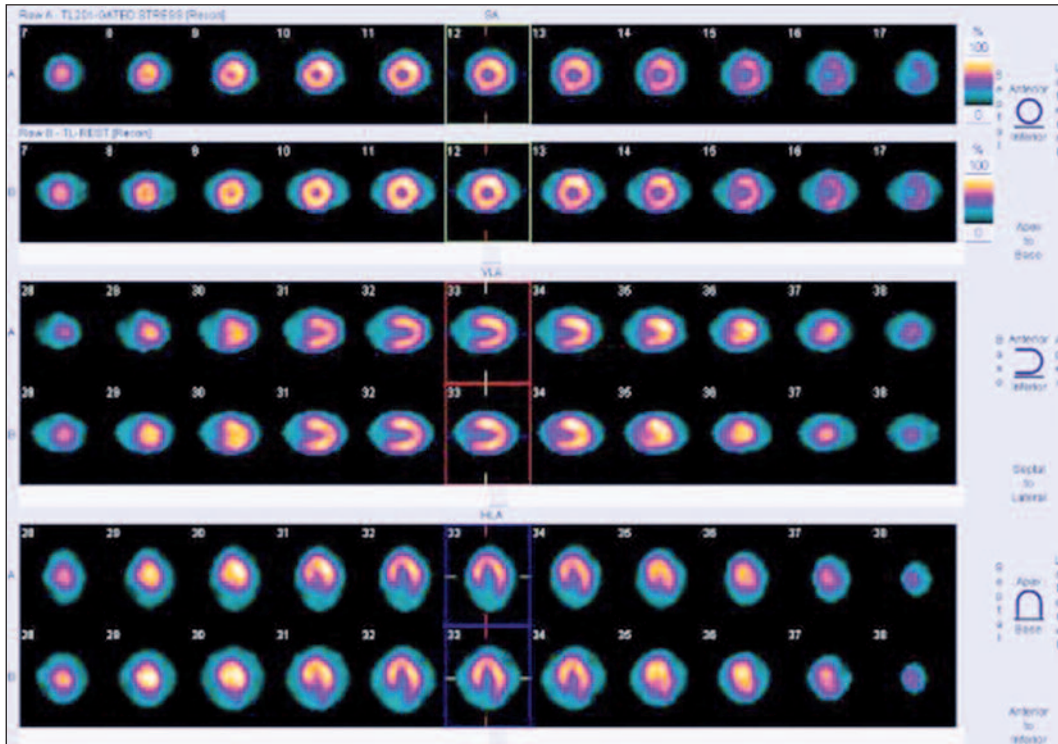
**FIGURE 3:** Left ventriculography in diastolic A and systolic B phases showing normal systolic function without additional pathology.

After exercise stress testing with thallium-201, no perfusion abnormalities or wall motion defects were detected on gated single photon emission computed tomography study. All the left ventricle walls showed homogeneous distribution of the radiotracer in short axis, horizontal long axis and vertical long axis (Figure 4). Acetylsalicylic acid and metoprolol were prescribed in addition to her antihypertensive and oral antidiabetic medications. Angina disappeared shortly after and she had not experienced cardiac events in the past 16 months after index event.

## DISCUSSION

In our patient, CA-LVMMFs were deliberated to be responsible pathology for stable angina pectoris. However, typical angina pectoris can also be seen in some other conditions besides coronary artery disease. Uncontrolled systemic hypertension, severe pulmonary hypertension, pulmonary embolism, right ventricular hypertrophy-ischemia, ascending aorta dilatation-dissection, left ventricular hypertrophy and aortic stenosis are some well known examples. None of the aforementioned conditions were detected on physical examination, TTE or during left heart catheterization. On the other hand, Syndrome X, which has similar clinical and laboratory features, is a diagnosis of exclusion and it is defined only in the setting of angiographically normal coronary arteries.

Microvascular fistulas between coronary arteries and left ventricle have been increasingly encountered since first report was published in 1982.<sup>3-5</sup> Although true incidence is not known, in a study from Said and van der Werf,<sup>4</sup> 20 cases were



**FIGURE 4:** ECG-gated single photon emission computed tomography (SPECT) images in short axis, horizontal long axis and vertical long axis showing normal perfusion during stress **A** and redistribution **B**.

(See for colored form <http://cardiovascular.turkiyeklinikleri.com/>)

diagnosed after a retrospective analysis of 30.829 coronary angiographies that are performed between 1996 and 2003. In agreement with our case, a predilection for female gender was present; and mean age of presentation was 67.3 years in this study. In contrast, left ventricle is not a common termination side for congenital coronary solitary fistulas<sup>1-2</sup> and all age groups can be affected without gender predilection.<sup>4</sup>

Angina pectoris is the most common symptom of CA-LVMMFs, but patients may also be presented with myocardial infarction or congestive heart failure.<sup>4</sup> Mechanism of ischemia seems to be 'coronary steal' even in the absence of atherosclerosis.<sup>6-8</sup> Severity may differ from patient to patient and absence of coronary sinus silhouette in venous phase of coronary angiography may give an idea about the extent of fistula.<sup>5</sup>

It is interesting that both resting ECG and stress tests did not reveal any objective findings of ischemia; multiple fistulas were present in two

coronary arteries. Similar to our case, a patient with coronary-to-left ventricle fistulas in all coronary arteries was reported in literature.<sup>5</sup> Even though that patient had severe fistulization and angina pectoris, authors reported a nondiagnostic stress ECG and only minimal perfusion defect in the anterior-apex on myocardial perfusion scintigraphy.

Medical management is the most common treatment modality due to diffuse nature and atypical localization of fistulas.<sup>4,8</sup> Contrary to solitary fistulas, micro-fistulas mostly arise from distal parts of coronaries and tend to be multiple.<sup>4</sup> Beta blockers were used effectively for symptom control in some patients and it is thought that they counterbalance the 'steal phenomenon' by decreasing myocardial oxygen demand.<sup>3,5</sup> Besides, antihypertensive effect of beta blocker probably contributes to symptom control in some patients who has an uncontrolled hypertension.

CA-LVMMFs can be easily overlooked, so all physicians performing coronary angiography

should be careful about these fistulas. As in our case, effective medical treatment may alleviate symptoms and prevent future cardiac events.

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