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Case Report

A Rare Involvement of Left Main Coronary Artery Due to Woven Coronary Artery in a Patient with Behçet's Disease

Sefa Tatar, Yakup Alsancak, Ahmet Seyfeddin Gürbüz, Abdullah İçli

Department of Cardiology, Necmettin Erbakan University Meram School of Medicine, Konya, Turkey

ABSTRACT

In general, woven coronary artery (WCA) is a benign congenital pathology; ocassionally, it may result in adverse cardiovascular events owing to myocardial ischemia. Though all coronary arteries may be affected, the right coronary artery is the most affected. This paper presents an extremely rare WCA effecting the left main coronary artery concurrent with Behçet's disease.

Keywords: Behçet's disease, left main coronary artery, woven coronary artery

INTRODUCTION

Woven coronary artery (WCA) is an extremely rare congenital anomaly with unexplained etiology (1). In this malformation, epicardial coronary arteries divide into long and thin channels. Thereafter, these channels merge to form an artery at the distal vascular bed (2). Normal blood flow after the abnormal coronary segment secures the relevant region, considered to be a good nature of coronary artery anomaly. Although this condition is considered to be benign, it occasionally causes angina pectoris, acute coronary syndromes, or possible sudden cardiac death owing to myocardial ischemia (3-5). Moreover, a few case reports have claimed no adverse cardiovascular events during long-term follow up (6, 7). This paper presents an extremely rare WCA effecting the left main coronary artery (LMCA) concurrent with Behçet's disease (BD).

CASE PRESENTATION

A 53-year-old male patient with a diagnosis of BD for 20 years was admitted to our department for unstable angina pectoris. The patient had a history of coronary artery bypass surgery 10 years ago. Resting electrocardiogram indicated q waves on D3 and aVF as well as nonspecific ST segment changes in precordial derivations with ventricular extrasystoles. Echocardiography indicated the systolic regional wall motion impairment. Coronary angiography was planned due to recurrent angina pectoris. Coronary angiography revealed a rudimentary right coronary artery (RCA) without a significant stenosis. Left coronary system angiography demonstrated a WCA of LMCA proceeding to the left anterior descending coronary artery (LAD) and circumflex artery (Cx). Furthermore, we observed a functional and well-developed left internal mammary artery (LIMA) to LAD anastomosis that associated dense collaterals with peripheral structures. Moreover, angiography revealed a collateral development between the left coronary system and RCA (Figure 1, 2). However, no information exists about the patient's coronary anatomy before his coronary artery bypass surgery. We believed that the patient's anginal complaints maybe associated with coronary steal owing to the dense collateral flow of LIMA. Furthermore, WCA of LMCA may have resulted from the decreased blood flow of Cx artery. So, we decided to optimal antianginal medical treatment to control of patient's symptoms. Written informed consent was obtained from the patient.

DISCUSSION

The etiology of WCA is unclear and is incidentally detected during coronary angiography. The literature indicates a male predominance (10:1) and reveals that RCA is the most affected (1, 4, 6). The recanalized thrombus, antegrade coronary collateral flow, or spontaneous coronary artery dissection should be considered for differential diagnosis (6). In particular, publications have indicated the effectiveness of optical coherence tomography (OCT) in differential diagnosis (8). We believed that the use of intravascular ultrasound or OCT for this patient is inappropriate owing to the diffuse involvement of LMCA. Depending on the affected segment of the coronary system, pharmacological treatment, percutaneous coronary intervention, or coronary artery bypass grafting may be the possible treatme/nt options.

The frequency of vascular involvement among BD patients ranges from 7.7% to 38% and is referred to as vascular BD (9). Males seem to be affected with arterial involvement than females. Vascular involvement more commonly affects the veins than the arteries, and coronary arterial involvement is extremely rare. Cardiovascular involvement in BD patients is estimated to range from 3% to 6%. This may result in pericarditis, myocarditis, coronary artery disease, valvular heart disease or intracardiac thrombus, endocarditis with valvular regurgitation, aneurysms of the coronary arteries or sinus of

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ORCID IDs of the authors: S.T. 0000-0001-8703-5078; Y.A. 0000-0001-5230-2180; A.S.G. 0000-0002-9225-925X; A.İ. 0000-0002-7047-811X.

Corresponding Author: Yakup Alsancak **E-mail:** dryakupalsancak@gmail.com

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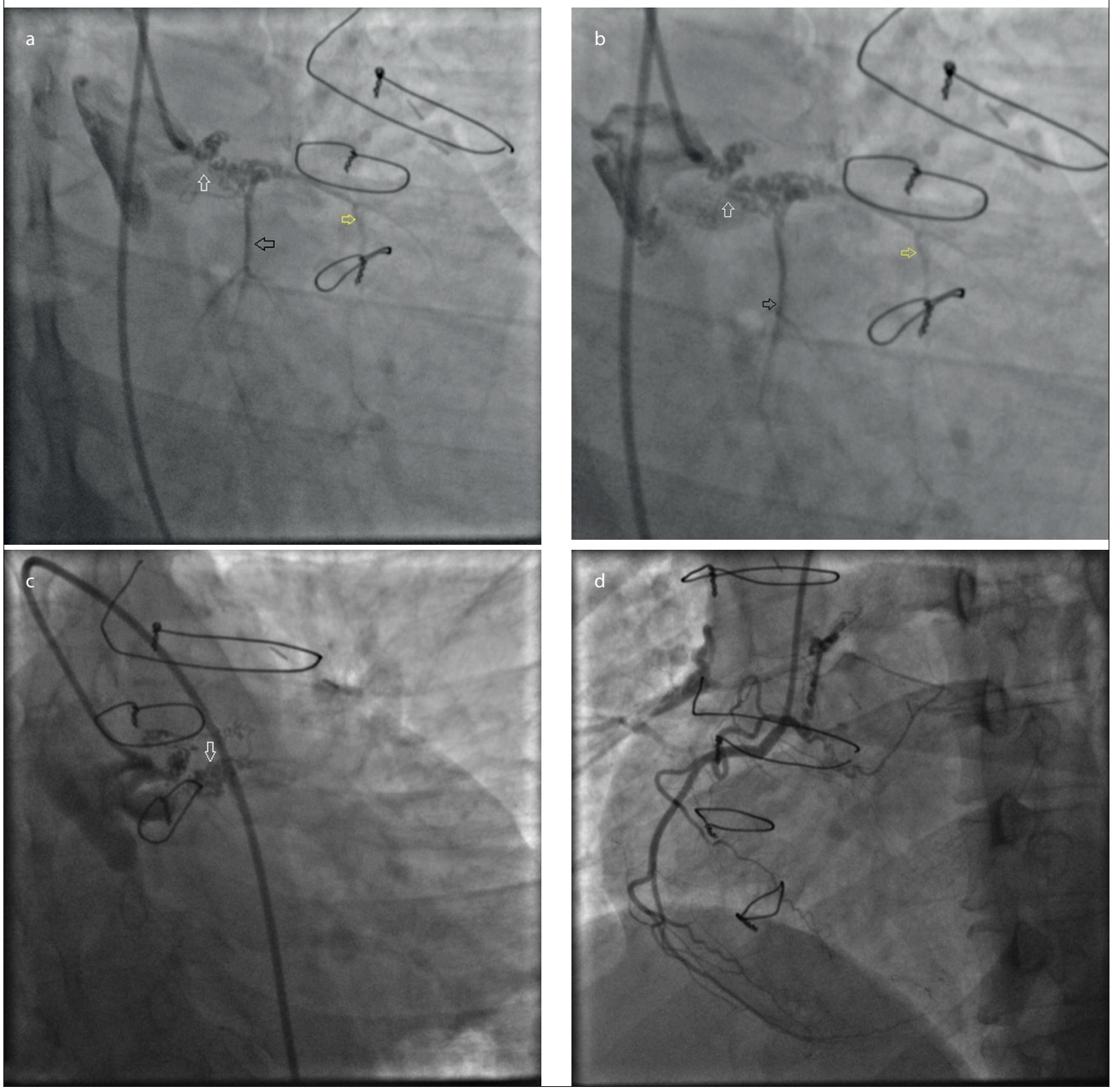
Figure 1. a-d. Coronary angiography depicts woven coronary artery of the left main coronary artery (white arrow), totally occluded left anterior descending coronary artery, dominant septal artery (yellow arrow), and circumflex coronary artery (black arrow) (a). Anteroposterior and caudal angiographic image of vessels [left main coronary artery (white arrow), septal artery (yellow arrow), and circumflex coronary artery (black arrow)] (b). Left anterior oblique caudal projection (spider view) demonstrating woven coronary artery of the left main coronary artery (white arrow) (c). Nondominant right coronary artery in a left anterior oblique view (d)

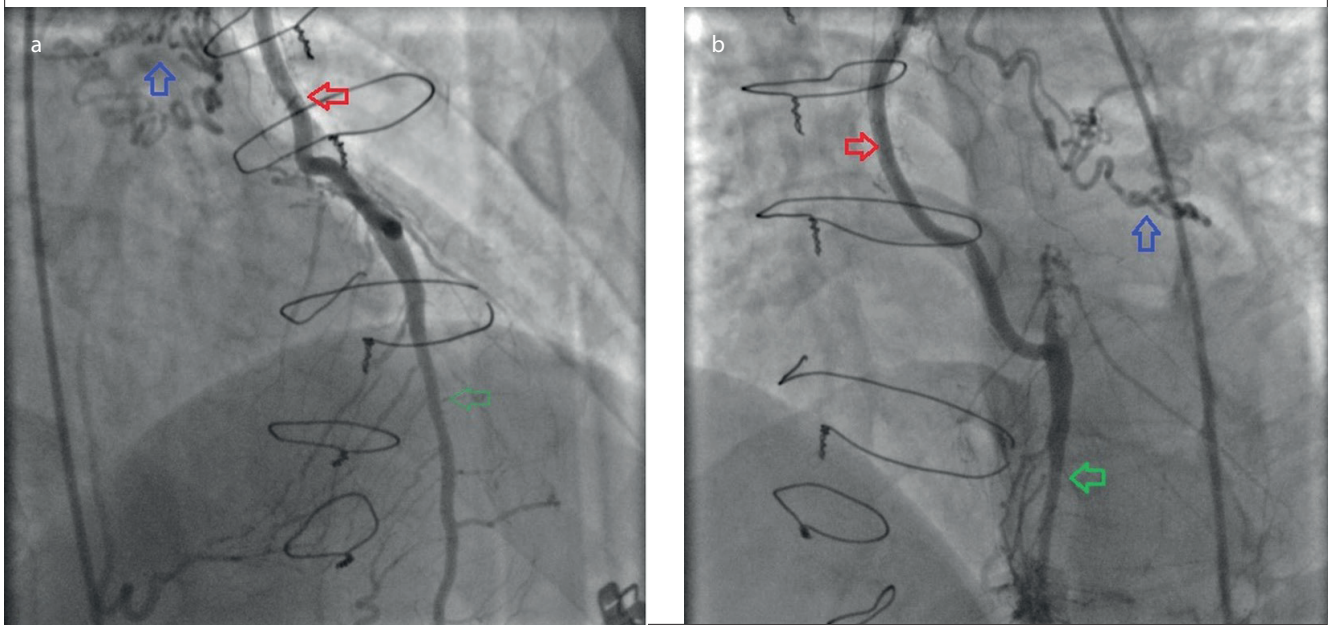


Main Points:

- The etiology of the woven coronary artery is still controversial.
- Involvement in the left main coronary artery is rarely reported.
- In this case, the presence of connective tissue disease (Behçet's Disease) with woven coronary artery may provide a different perspective for etiology.

valsalva, and advanced heart failure (10, 11). Lesions of coronary arteries include stenosis, occlusion, aneurysm, and pseudoaneurysm with or without myocardial infarction. A previously published study has demonstrated that silent myocardial ischemia rate was higher among BD patients compared with that of healthy controls at 19.5% and 2.9%, respectively (12). Peripheral arterial involvement in BD patients may range from 1.5% to 7% (9, 13). When a coronary artery pathology is detected in BD patients, the entire arterial or venous

Figure 2. a, b. Left internal mammary artery (red arrows) to left anterior descending coronary artery (green arrows) and anastomosis and well-developed collaterals (blue arrows) to the peripheral structures (a). Another image of left internal mammary artery (red arrows) to left anterior descending coronary artery (green arrows) and anastomosis and well-developed collaterals (blue arrows) (b)



vascular system must be evaluated for involvement. Doppler ultrasound scan of carotid arteries and iliofemoral arterial and venous system was observed to be in the normal range for this patient.

This case may be essential for two reasons. First, WCA can effect the isolated LMCA. To the best of our knowledge, this report is the first case to focus on the condition of WCA in BD patients in the literature. Since the patient's coronary anatomy before his coronary artery bypass surgery was unknown, commenting on congenital or the acquired appearance of LMCA seems impossible. As for the patient in this case, the LMCA lesion may have mortal consequences; thus, it is acceptable to explain this view through a chronic pathology. Furthermore, LMCA thrombosis may have resulted in coronary artery bypass grafting for this patient. Accordingly, this angiographical image may have resulted from chronic thrombosis recanalization. Second, the appearance of BD as a chronic vasculitics syndrome may result from chronic inflammation.

CONCLUSION

Finally, histopathological examination continues to be the gold standard technique for the diagnosis or differential diagnosis of WCA. This case indicates that the screening of connective tissue diseases may be beneficial in patients with WCA.

Informed Consent: Written informed consent was obtained from patient who participated in this study.

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