

Case Report

Isolated Bilateral Coronary Artery Ostial Stenosis in Aortitis Syndrome

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Isolated bilateral coronary artery ostial stenosis is a rare condition that has been associated with various diseases. We present the case of a 47-year-old male with bilateral coronary ostial stenosis. The patient's history was not significant for systemic diseases. The aorta had prominent signs of aortitis of unknown etiology, possibly due to a chronic mycotic process. The patient underwent emergency surgery and had an uneventful course. The two-year follow up is excellent.

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Isolated stenosis of the coronary ostia associated with normal distal coronary arteries is a rare cause of myocardial ischemia that has been reported sporadically in the literature. Involvement of the coronary arteries in aortitis was first reported in 1951 by Foving and Logan.¹ Although uncommon, this site of involvement has important symptomatic and prognostic implications.

Case presentation

A 47-year-old male, a physician, presented with angina of two-hour onset. He had no conventional risk factors for coronary artery disease. On clinical examination all pulses were normal; blood pressure was 110/60 mmHg in the left upper limb. On presentation the cardiac examination was normal but the patient experienced intense retrosternal pain. On his way to the hospital the patient had received one tablet of aspirin. There was no clinical evidence of involvement of arch vessels or aortic branches. His electrocardiogram at rest was normal. The echocardiogram showed a left ventricular ejection fraction of 60%, normal aortic root and aortic dia-

meter. Laboratory values were normal and the venereal disease research laboratory test was negative. An emergent coronary angiogram revealed bilateral coronary ostial stenosis of 90-95% (Figures 1 & 2). The collateral network was not extensive. On angiography all the other arteries of his body were normal. Emergency coronary artery bypass grafting (CABG) on a heart lung machine was performed two hours later, with anastomosis of the left internal mammary artery to the left anterior descending artery and reversed saphenous vein grafts to the obtuse marginal and right coronary artery. The postoperative period was uneventful and the patient was discharged on the ninth postoperative day. At two-year follow up he is essentially free of cardiac disease and asymptomatic.

A histologic examination of the aortic wall showed diffuse dense chronic plasmocytic inflammation and fibrotic alterations due to non-specific aortitis, possibly resulting from a chronic mycotic process. The microbiological examination of the aortic specimen was negative (Figure 3).

The pericardium and the myocardium had no signs of inflammation, past or active. The patient was subjected to a thor-

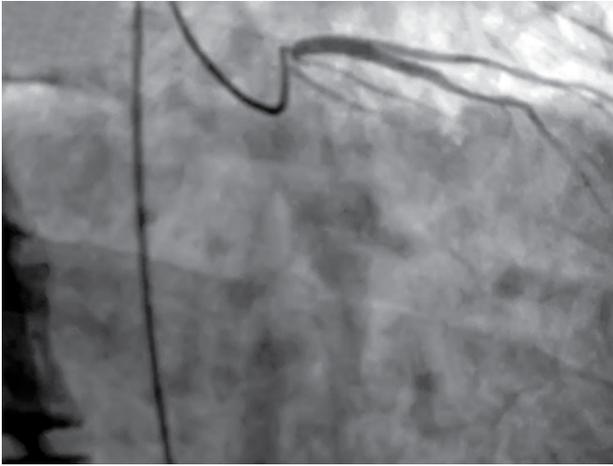


Figure 1. Coronary angiogram shows more than 90% ostial left coronary artery stenosis.

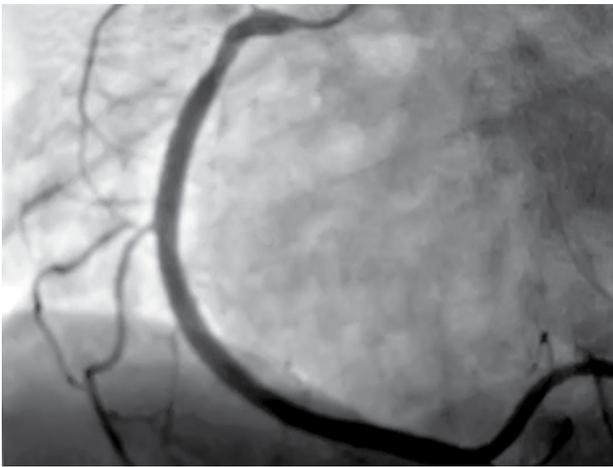


Figure 2. Coronary angiogram shows 90% ostial right coronary artery stenosis.

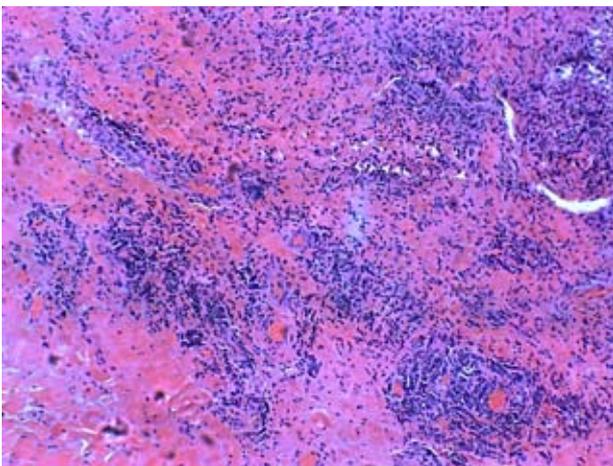


Figure 3. Aortic wall with diffuse dense chronic plasmocytic inflammation and fibrotic alterations.

ough laboratory examination after discharge for systemic and infectious diseases, but the results were negative.

Discussion

Isolated coronary ostial stenosis caused by non-specific aortitis is extremely rare, but is considered as an important risk factor related to mortality. Non-specific arteritis was first reported by Savory and Kussmaul in 1856. Although most cases of isolated ostial stenosis are thought to be atherosclerotic in origin,² early reports have associated this type of coronary disease with congenital³ and syphilitic⁴ causes and diseases that may cause aortitis, mainly Takayasu disease.⁵ Radiation exposure is another condition associated with aortitis with ostial stenosis.⁶ Ostial stenosis of the coronary arteries remains the most common pattern of involvement of coronary arteries in aortoarteritis. Regardless of etiology, aortitis may result in an injury to the aorta or its branches. The inflammatory process that occurs in response to the injury may ultimately result in one of two morphological changes in the affected vessels: 1) stenosis or occlusion from fibrosis; and 2) dilatation or aneurysm formation from destruction and weakening of the arterial wall. All forms of arteritis are capable of producing dilatation or aneurysm formation, but only Takayasu's arteritis can produce narrowing or occlusion of the aorta or its branches, with the rare exception of stenosing arteritis, which sometimes follows radiation therapy in childhood. Clinical symptoms occur, depending upon the extent and location of the vascular disease. Stenotic or occlusive lesions may result in ischemia to limbs or organs distal to the site of obstruction.⁷

A chronic inflammatory infiltrate in which lymphocytes and plasma cells predominate is characteristic of chronic periaortitis. Although periaortitis is the commonest condition associated with inflammatory aneurysms, tuberculosis, non-specific aortitis, Takayasu's disease and Kawasaki's disease may be other causes.

Involvement of the coronary arteries in Takayasu's arteritis was first reported in 1951. Treatment of the coronary disease has not been reported to have good results since arterial inflammation is persistent in over 40% of cases.⁸ At the same time endarterectomy and stenting of ostial stenosis have shown equally bad results.⁹ Studies of surgical series have reported in-hospital mortality of 7.9 to 8.7%. Kang et al¹⁰ reported successful management of left main coronary

artery stenosis using angioplasty and stenting with a drug-eluting stent.

In our patient we hardly had any option apart from CABG. The limitation of the disease to the coronary ostia without more distal lesions is the unusual and important feature of this case. Isolated bilateral coronary ostial stenosis is very uncommon. To the best of our knowledge only about 20 surgically treated cases have been reported in the literature.¹¹ It is noteworthy that there was no other aortic branch or arterial involvement. In such patients, surgical patch angioplasty has been proposed as an alternative to CABG, but due to the rarity of the disease and its sporadic appearance in the literature, there is no evidence that it is superior in terms of efficacy or long-term survival.¹⁰

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