

# Dynamic Compression of the Left Coronary Artery by a Left Ventricular Pseudoaneurysm after Myocarditis

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A 69-year-old man with a recent diagnosis of suspected leptospirosis infection-related myocarditis presented with antecedent arthralgia, myalgia, fever, intermittent anterior chest pain, yellowish sclera, yellowish skin and shortness of breath. His symptoms improved after antibiotic treatment with penicillin for 14 days. However, recurrent chest pain and progressive dyspnea upon exertion developed 2 months later. A newly developed left ventricular outflow tract pseudoaneurysm was identified by cardiac sonography and multi-detector computed tomography of the heart. A subsequent coronary arteriogram demonstrated an left ventricular (LV) pseudoaneurysm causing compression to both the left circumflex coronary artery and the left anterior descending coronary artery with significant stenosis. To the best of our knowledge, this is the first reported case of a LV pseudoaneurysm developing after a clinical course of suspected leptospirosis-related myocarditis causing dynamic compression of the left coronary artery.

**Key Words:** Dynamic compression of coronary artery • Left ventricular pseudoaneurysm • Leptospirosis • Myocarditis

## INTRODUCTION

Pseudoaneurysm of the left ventricle (LV) is a rare cardiac disease that mostly occurs after myocardial infarction, cardiac surgery or trauma. And although LV pseudoaneurysm caused by infection is extremely rare, it has an associated 50% mortality rate. When left untreated, pseudoaneurysms carry a 30-45% risk of rupture.<sup>1</sup> We herein describe an unusual case of LV pseu-

doaneurysm that developed after a clinical course of suspected leptospirosis-related myocarditis causing dynamic compression of the left coronary artery. For clinical physicians, early detection of LV pseudoaneurysm is important because it is prone to rupture. Early surgical intervention is the main therapeutic strategy after confirmed diagnosis.

## CASE REPORT

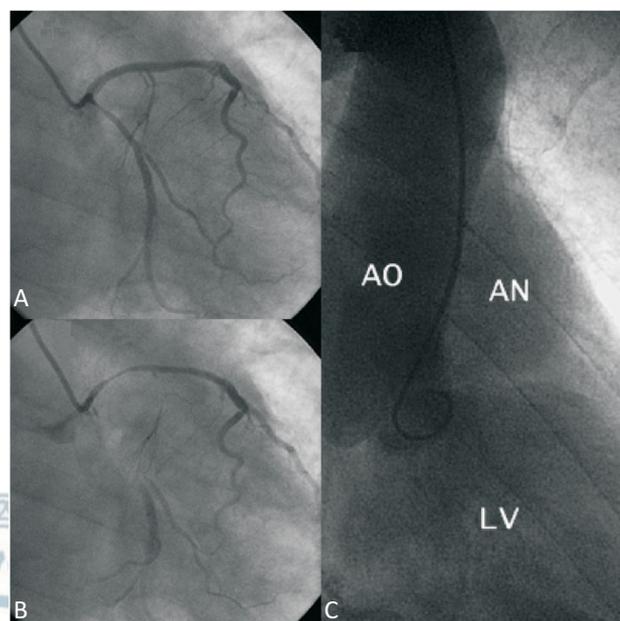
We present the case of a 69-year-old male who worked as a trash collector in an open-air market where rodents, swine, chicken and dogs were kept. He complained of antecedent arthralgia, myalgia, fever, intermittent anterior chest pain, yellowish sclera, yellowish skin and shortness of breath when walking during the past two weeks. On physical examination, his jugular venous pressure was elevated and he had pitting edema on his legs. Bibasilar inspiratory crackles and S3 gallop were also detected. An electrocardiogram of the patient

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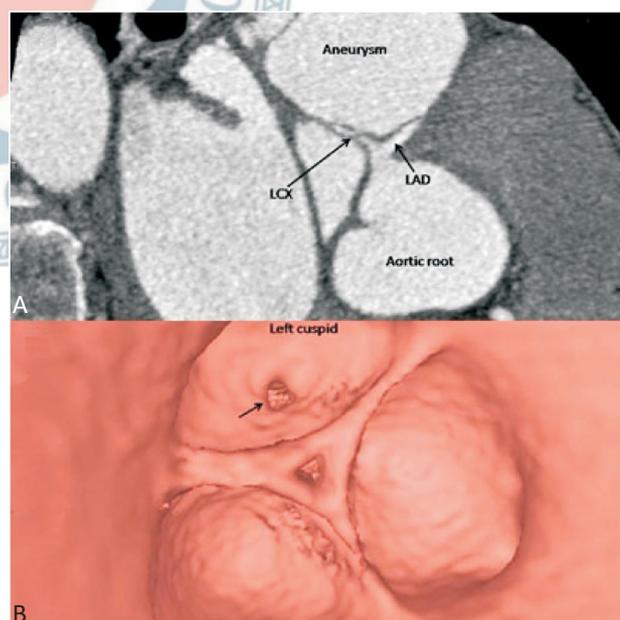
revealed only sinus tachycardia, though chest radiograph demonstrated pulmonary edema. Laboratory data reported a white blood count is  $15800/\text{mm}^3$ , with 92% neutrophils, and 3% bands, a platelet count of  $80000/\text{mm}^3$ , total bilirubin: 4.8 mg/dl with a direct fraction of 2.5 mg/dl, alkaline phosphate: 168 IU/L, aspartate aminotransferase: 44 IU/L, blood urea nitrogen: 44 mg/dl, creatinine: 2.0 mg/dl, cardiac troponin I: 3.56 ng/ml, creatine kinase (CK): 425 U/L and CK-MB: 40 U/L. Echocardiography demonstrated moderate pericardial effusion, with impaired left LV systolic function with LV ejection fraction = 40%, but no aneurysm was visible at that time. A tentative diagnosis of suspected leptospirosis infection-related myocarditis was made according to the patient's occupational exposure, clinical presentation and multiple organ involvement. After he was given a complete antibiotic treatment with penicillin, fever and jaundice were resolved. Additionally, his pulmonary edema and edema in the legs showed improvement after optimal medical treatment for congestive heart failure. Thereafter, the patient was discharged home in stable condition.

However, recurrent chest pain and progressive dyspnea on exertion developed 2 months later. After the patient was admitted for the second time, the cardiac biomarkers including CK, CK-MB, and Troponin-I, were within normal range and electrocardiography (EKG) showed a right bundle branch block (RBBB) pattern. Repeated echocardiography showed LV ejection fraction = 56%, minimal pericardial effusion, moderate mitral regurgitation and moderate to severe aortic regurgitation with a left ventricular outflow tract (LVOT) pseudoaneurysm. Coronary angiography (CAG) showed dynamic external compression to both the left anterior descending (LAD) and left circumflex (LCX) proximal vessel segments (Figure 1A and B). Aortogram revealed an LV pseudoaneurysm (Figure 1C). Subsequent cardiac multidetector computed tomography (MDCT) also demonstrated the presence of an LV pseudoaneurysm causing compression to both the LAD and LCX with significant stenosis (Figure 2A), and a 5 mm defect over the left coronary cusp of the aortic valve (Figure 2B). Surgical intervention was performed and it showed perforation of the left cuspid of the aortic valve, an LVOT defect below the left coronary cusp with pseudoaneurysm, and pericarditis with adhesion of the pericardium. The pa-

tient received patch repair of the LVOT defect, aortic valve replacement and mitral repair. He was discharged



**Figure 1.** (A) External compression of the left circumflex coronary artery (LCX) proximal vessel segment with significant stenosis during diastolic phase. (B) Both the left anterior descending coronary artery (LAD) and the LCX show significant stenosis during systolic phase. (C) Aortogram demonstrated a left ventricular pseudoaneurysm (AN, aneurysm; AO, aorta; LV, left ventricle).



**Figure 2.** (A) An left ventricular pseudoaneurysm causing compression of the left anterior descending coronary artery (LAD) and the left circumflex coronary artery (LCX) with significant stenosis. (B) A 5 mm defect over the left coronary cusp of the aortic valve.

home 14 days after the procedure. At the patient's 12-month clinical follow-up, no symptom recurrence was observed. The pathological report revealed chronic inflammation with plasma cells and histiocyte infiltration compatible with chronic myocarditis.

## DISCUSSION

LV pseudoaneurysm is a rare clinical condition that may involve life-threatening complications including rupture of the aneurysm, LV failure, LV thrombus formation and distal embolization. This potentially dangerous condition has been associated with nearly 50% mortality.<sup>1</sup> Myocardial infarction (MI) is the most common cause of LV pseudoaneurysms, followed by cardiac surgery, trauma, and infection.<sup>2</sup> In a literature review of 253 patients with a pseudoaneurysm in whom the cause was reported, LV pseudoaneurysm caused by infection was extremely rare, accounting for less than 5% of all cases.<sup>2</sup>

In our case, the patient presented with congestive heart failure and suspected leptospirosis-related sepsis at first admission and chest pain at re-admission. A newly developed LVOT pseudoaneurysm was proven by cardiac sonography and MDCT of the heart. CAG demonstrated an LV pseudoaneurysm causing compression to both the LCX and LAD arteries with significant stenosis. To the best of our knowledge, this is the first reported case of an LV pseudoaneurysm developing after a clinical course of suspected leptospirosis-related myocarditis causing dynamic compression of the left coronary artery.

Leptospirosis is an emerging infectious disease with recent large outbreaks in India, Southeast Asia, Malaysia, Nicaragua and Brazil.<sup>3</sup> It is a zoonotic disease, which can cause myocarditis and focal myocyte necrosis in humans.<sup>4</sup> LV pseudoaneurysm may develop after focal myocyte necrosis. However, both the frequency and extent of cardiac involvement in leptospirosis are under-reported and poorly understood. Myocarditis was the cause of death in 4% of the cases in the study by Areen.<sup>5</sup> Evidence suggests that direct myocardial damage occurs in patients with severe leptospirosis, but further studies are needed to elucidate its pathophysiology.<sup>4</sup> The study by de Brito et al. emphasized that involvement of the

conduction system correlates significantly with myocarditis.<sup>6</sup> ECG evidence of conduction system abnormalities such as first-degree atrioventricular block, sinus block and RBBB has been reported.<sup>7-9</sup> In our case, an EKG also demonstrated a RBBB pattern.

In clinical practice, early diagnosis is crucial to avoid complications and to reduce mortality, since pseudoaneurysms are prone to rupture. When left untreated, pseudoaneurysms carry a 30-45% risk of rupture.<sup>1</sup> Early surgical intervention is the preferred therapeutic strategy after a confirmed diagnosis. However, the diagnosis of pseudoaneurysm is complicated because the symptoms of acute pseudoaneurysm are similar to the symptoms of an MI, and chronic pseudoaneurysms have symptoms similar to those of congestive heart failure. The most common clinical presentations of pseudoaneurysm are congestive heart failure (36%), chest pain (30%), and dyspnea (25%), whereas the incidence of sudden death as a presenting symptom is 3%.<sup>10</sup> We describe an unusual case of LV pseudoaneurysm that developed after a clinical course of suspected leptospirosis-related myocarditis causing dynamic compression of the left coronary artery. This diagnosis was made based upon patient exposure history, clinical suspicions and echocardiography follow-up. Thus, it is important for physicians to be on the alert for newly developed chest pain and symptoms of congestive heart failure after an infectious process, LV pseudoaneurysm should also be considered in the differential diagnosis.

The mechanism of pseudoaneurysm-induced chest pain has previously been reported. We proposed four kinds of mechanisms that may cause angina, including the "stealing blood" theory, distal embolization, external compression of coronary arteries and destroyed aortic valve. First, according to the "stealing blood" theory, the pseudoaneurysm "steals" part of the LV stroke volume during systole, hence decreasing the cardiac output and inducing LV volume overload. Both can mismatch the oxygen supply and myocardium demand, which may cause myocardial ischemia and chest pain. The end result of longstanding myocardial ischemia and prolonged LV volume overload is a failing heart. Second, LV thrombus formation may lead to a distal thromboembolism, including cerebrovascular accident and acute MI, causing chest pain.<sup>1,10</sup> Third, if the LV pseudoaneurysm is in the left part of the aorta and superior to the LVOT, as in

our case, it could cause external compression of the left coronary artery, especially in systolic phase, leading to myocardial ischemia. Lastly, as in our case, the aortic valve may be destroyed due to previous infection, causing aortic valve regurgitation with blood flowing in the reverse direction during ventricular diastole, from the aorta into the LV. Hence, coronary artery blood flow is decreased, causing chest pain.

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