Left Internal Mammary Artery-to-Pulmonary Vein Fistula: A Rare Cause of Unstable Angina

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Abstract
The incidence of acquired left internal mammary artery-to-pulmonary vein fistulas has been increasing in the last few decades. This has been attributed to the increase in coronary artery bypass surgery (CABG). The most commonly reported symptoms are angina and dyspnea. The timing of the presentation varies widely from a few months to several years after CABG. Medical management is the treatment of choice and usually controls the symptoms in most patients. Percutaneous intervention is, however, indicated when medical therapy fails. In this case report, a 72-year-old man with a history of CABG presented with progressively worsening chest pain and dyspnea. Troponin was negative and the electrocardiogram showed no acute ischemic changes. He was found to have left internal mammary artery-to-pulmonary vein fistula on coronary angiogram. His symptoms improved upon intensifications of his guideline-directed therapy for coronary artery disease. This represents an unusual cause of unstable angina.

Keywords
coronary artery, bypass graft, pulmonary vein, fistula, angina, coronary angiography, left internal mammary vein

Introduction
Coronary fistula is extremely rare and has a reported overall incidence of 0.08% to 0.4%.1 Etiology can be congenital or acquired. The incidence of congenital cases of internal mammary artery (IMA) fistula has been relatively stable in the last 30 years with a reported incidence ranging from 2 to 4 cases every 10 years.2 The incidence of acquired cases of IMA-to-pulmonary vein (PV) fistula has, however, been increasing within this same time frame. Most recent cases of acquired left internal mammary artery (LIMA)-to-PV fistulas have been linked to coronary artery bypass graft surgery (CABG), inflammatory states, trauma, or malignancy.3 This increase has been attributed to both the improvement in diagnostic imaging and angiographic techniques, and a rise in CABG surgeries. In one review, the most reported etiology was CABG and it accounted for over half of the acquired cases.2 The clinical presentation of LIMA-to-PV fistula varies. Angina has been reported as the most common presenting symptom. This is especially in acquired cases.2 In this report, we present a case of unstable angina resulting from LIMA-to-PV fistulas in a 72-year-old man with coronary artery disease (CAD) status post-CABG.

Case Presentation
A 72-year-old man with a past medical history significant for atrial fibrillation, hypertension, and CAD status post-CABG (which was performed 6 years prior) was transferred from an outside facility for evaluation of chest pain of 3 weeks duration. Chest pain was substernal, radiated to the left arm and jaw, and was associated with dyspnea. His medications were simvastatin, diltiazem, amiodarone, aspirin, and as-needed nitroglycerin. His symptoms were initially exertional and...
relieved by nitroglycerin. It, however, progressed to occurring at rest with nitroglycerin providing minimal to no relief. It was at this point that he presented to the outside facility. There, troponins were reported as negative ($\times 3$) and electrocardiogram (ECG) showed no ischemic changes. Notably, the patient reportedly had an abnormal stress test 2 weeks before this presentation. He continued to have active chest pain and was eventually transferred to our facility.

Upon presentation at our facility, the patient reported ongoing chest pain. His blood pressure was 128/77 mmHg and his pulse was 84 beats/min. Troponin was negative. The ECG showed sinus rhythm, old right bundle branch block (RBBB), premature ventricular complexes (PVCs), and no ischemic changes. This is shown in Figure 1. The echocardiogram was unremarkable with an ejection fraction of 55%. His chest pain persisted. Simvastatin and diltiazem were discontinued. An angiotensin-converting enzyme inhibitor, $\beta$-blocker, high-intensity statin, and long-acting nitrates were added. Based on strong suspicion of acute coronary syndrome, a coronary angiogram was performed. Findings were notable for widely patent saphenous vein graft (SVG) to the posterior descending artery (PDA), SVG to the second diagonal artery, SVG to the obtuse marginal artery, and left internal mammary artery (LIMA) to left descending artery (LAD) graft. A nonobstructive disease was noted in the right coronary artery. There were, however, multiple fistulous communications between LIMA and PVs as shown in Figures 2 and 3. Unstable angina secondary to LIMA-to-PV fistulas was diagnosed. Guideline-directed medical therapy for CAD was intensified and his symptoms subsequently improved. He was eventually discharged to a cardiac rehabilitation program with an appointment to follow-up with cardiothoracic surgery as an outpatient.

**Discussion**

As stated earlier, identified cases of LIMA-to-PV fistulas have been increasing in the last few decades partly due to the increased rate of CABG.6 Six years prior to presentation, our patient had CABG with LIMA graft to LAD performed. The etiology of fistula formation after CABG is not well understood. A few possibilities have been postulated; neovascularization underlies each etiology. For example, inadvertent
trauma from dissection of the LIMA and disruption of visceral pleura during CABG may lead to formation of adhesion, neovascularization, and disruption of normal blood flow, which can ultimately lead to a fistulous communication between the LIMA and the nearby PV.3,4 Surgical techniques have also been linked to the pathogenesis of fistulas after CABG. For instance, compared with surgical clipping, electrocoagulation has been linked to an increase in the risk of neovascularization post-CABG. Furthermore, repeat CABG, minimally invasive CABG, and pericardial flap have also been reported to increase the risk of developing fistulas.4

There is a wide variation in the timing of patient presentation following CABG. It can occur from as early as 2 months to as far as 16 years after surgery.5,6 Our patient presented with the LIMA-to-PV fistulas 6 years after CABG. He presented with angina and dyspnea. This is supported by the literature. Angina is reported as the most common presenting symptom, occurring in as many as 88% of patients with acquired LIMA-to-PV fistula.2,7 Dyspnea was the presenting symptom in about 6% of patients.2

The appropriate treatment of LIMA-to-PV fistulas is again variable and should be individualized.2 The management depends on the presence of comorbid cardiac conditions, anatomy of the fistula, and clinical presentation.8 Conservative medical management is the most common treatment approach and can be used in patients with mild symptoms.2 In one study, over half of patients treated with only anti-anginal medications reported improved symptoms.2 This was the situation with our patient as his symptoms improved upon the optimization of his anti-anginal therapy. He was symptom-free at the time of discharge. Sometimes, the disease may progress leading to further dilation of the fistula and worsening of symptoms.9 When this happens, coil embolization and percutaneous intervention (PCI) techniques using stents can be performed. Apart from failed medical therapy, PCI has also proven effective in patients who are at high risk of surgery or have advanced symptoms.8 Furthermore, endovascular approach is another treatment strategy that has been used successfully to treat these fistulas. In this approach, coil embolization and coil with polytetrafluoroethylene (PTFE)–covered stents have been utilized with great results.5,10 Surgical clipping is implicated in fistula etiology; ironically, it has also successfully been used in the treatment of fistulas when medical therapy fails.11 Our patient may benefit from any of these interventions if medical therapy fails to control his symptoms. Our patient is scheduled for outpatient follow-up with cardiothoracic surgery for further evaluation.

**Conclusion**

The development of fistulous communication between an internal mammary artery and pulmonary vasculature following CABG is extremely rare. It usually presents with angina and dyspnea. Although it is not common, it should be considered as a differential diagnosis in post-CABG patients who experience worsening anginal symptoms. Medical management is the usual initial treatment of choice.

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**Ethical Approval**

Our institution does not require ethical approval for reporting individual cases or case series.

**Informed Consent**

Verbal informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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