

Percutaneous transcatheter embolization in large tortuous coronary artery fistula patient: a case report

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Abstract

Coronary artery fistula is a rare anomaly involving the coronary artery and a heart chamber or vessel. Percutaneous intervention has been shown to be effective and safe in fistulas that are small and non-tortuous; however, it is not an absolute contraindication in fistulas that are large and tortuous. We report a delayed diagnosis of a single, large-diameter, tortuous coronary artery fistula that manifested as myocardial ischaemia due to the steal phenomenon in a 49 year old male. The undesirable connection was successfully obliterated by percutaneous embolisation, followed by an improvement in symptoms and daily activities. Steal phenomenon is the fundamental mechanism of myocardial ischaemia in coronary artery fistula, as confirmed by improvement in symptoms and coronary artery perfusion following occlusion of the fistula. Percutaneous catheterization is safe and effective for coronary artery fistula closure, and the occlusion site should be precise to achieve complete occlusion and prevent complications.

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Introduction

Coronary artery fistula (CAF) was first described by Krause in 1865.¹ It is an abnormal structure that connects the coronary artery to the heart chamber or thorax blood vessel.² Most CAF is asymptomatic, while other symptoms vary from dyspnoea, fatigue, arrhythmias, heart failure, myocardial ischaemia or infarct mainly due to the steal phenomenon; when fistulas steal feeding coronary perfusion.^{1,3} High-flow, symptomatic, or elderly fistulas are corrected percutaneously or surgically. In recent decades, percutaneous approach is considered effective

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and safe, particularly for small diameter and straight fistula. However, method of closure should be decided by clinicians on an individual basis.⁴

We report percutaneous transcatheter coil embolisation at the proximal part of a large tortuous shaped CAF that successfully occluded fistula flow and improved angina symptoms due to steal phenomenon.

Case Report

A 49-year-old Javanese man was admitted to the Dr. Soetomo general hospital Surabaya, on 8th June 2021. He had recurrent chest pain for the last two years. The pain lasted 5-10 minutes, worsened with activity, and subsided with rest. Previous electrocardiography (ECG) and troponin level were not suggestive of ischaemia or infarct. He had diabetes for ten years and regularly consumed glimepiride and metformin daily. He denied a history of hypertension, smoking, dyslipidaemia, renal impairment, stroke, and coronary artery disease. There was no family history of sudden death or stroke. Vital signs and physical examination were normal.

Electrocardiogram showed sinus rhythm 75x/min, normoaxis, and early repolarization in lead II, III, and aVF. Posteroanterior thorax x-ray showed normal lungs and heart with a cardiothoracic ratio (CTR) of 42%. Echocardiography results showed concentric remodelling of the left ventricle without regional wall motion abnormalities and a 61% ejection fraction by TEICH.

Cineangiography of the left main coronary artery (LMCA) ostium presented a large tortuous fistula from the proximal left anterior descending coronary (LAD) artery (Fig. 1). The coronary fistula was larger than the LAD artery. Contrast from the catheter flowed from the proximal LAD to the PA, while the perfusion of LAD in the distal site of CAF was reduced.

Despite its tortuous and wide diameter, the fistula's proximal site was narrow and straight, making it an embolisation target. He then underwent coil embolisation with Coil Hydrosoft Helical 3mm/4cm, Coil Hydrosoft 10, 3D 3mm/4cm, and Coil Hydrosoft 10 3D

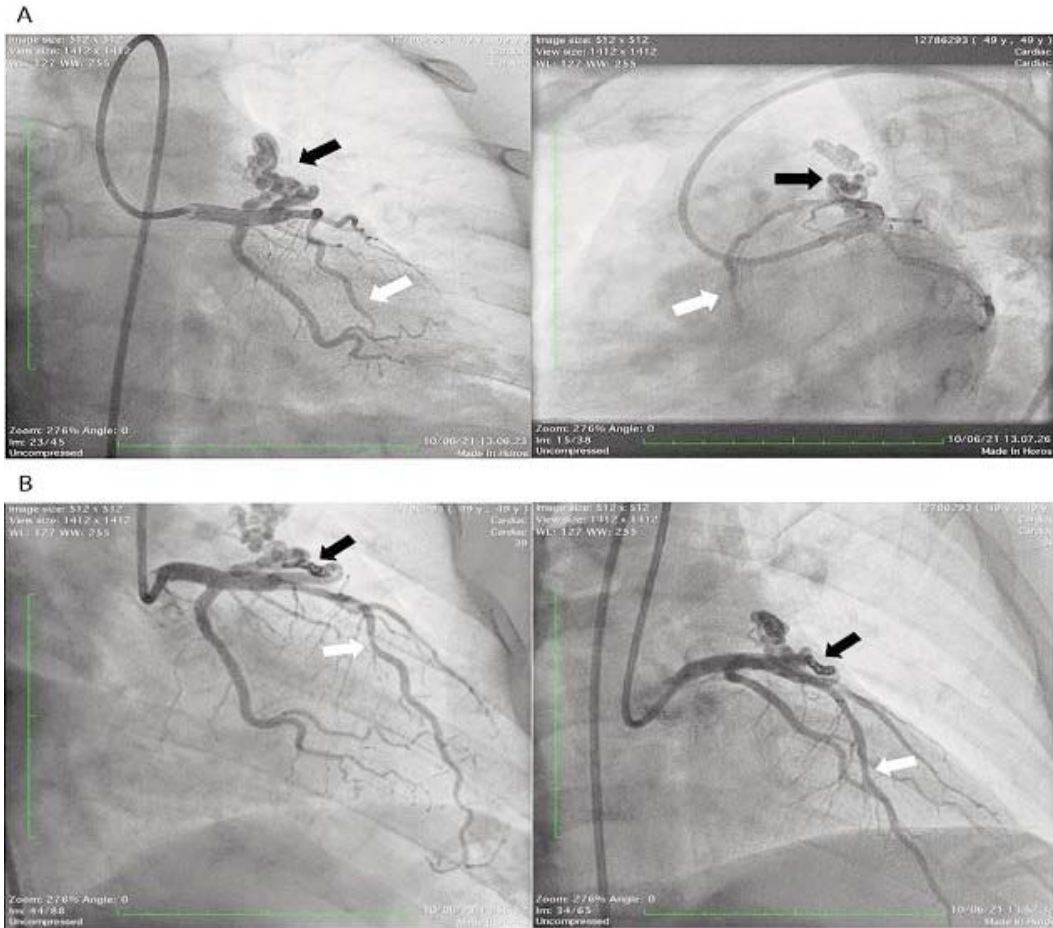


Figure-1: Angiography examination before (A) and after (B) embolization. (A) Coronary angiography showed tortuous shaped fistula (black arrow) between proximal Left Anterior Descending (LAD) and pulmonary artery (PA). Distal LAD artery perfusion was decreased (white arrow). (B) Distal LAD perfusion was improved, and 50-60% stenosis (white arrow) in LAD was uncovered after embolization.



Figure-2: Coronary angiography five months after fistula embolization showed coil was on previous position (white arrow) and obstruct fistula flow (green arrow). Distal LAD artery flow was TIMI 3 flow.

3mm/6cm.

Post-coiling evaluation yielded 50-60% stenosis of LAD artery at the distal end of the fistula. Coil embolisation was performed without any complication during or post-procedure. Subsequently, he was treated with dual antiplatelet therapy for a month (Aspirin 160mg and Clopidogrel 75mg, once a day), followed by single antiplatelet therapy (Aspirin 160mg) for five months.

A cineangiography follow up was done five months later and revealed the coil position on the origin site with minimal residual fistula flow barely visualized (Fig.2). Distal LAD vascularisation improved with TIMI 3 flow, compared to before embolization (TIMI 1 flow). He had no complaint of any daily activity limitation and related symptoms.

Discussion

Despite CAF prevalence being extremely rare and most of the cases being asymptomatic, CAF is the most common coronary anomaly that causes haemodynamic instability and complications, particularly in patients older than 20 years. The possible complications include heart failure, pulmonary hypertension, bacterial endocarditis, and myocardial ischaemia or infarct.¹

Herein, the patient complained of recurrent typical chest pain for two years, however, routine cardiovascular examinations were normal. The disparity between symptoms, risk factors, and routine examinations may lead to misdiagnosis and a delay in treatment. Previous case reports revealed continuous cardiac murmur and vessel dilatation^{5,6} which can be used as valuable diagnostic clues for CFA, but these were not observed in the current case report. This may be due to the fact that the CFA in our case was between the LAD and PA, whereas the CFA in a previous case was between the LAD and coronary sinus (CS)⁵ which notably has much lower pressure than LAD. The significant pressure difference between these two compartments causes murmur and significant vessel dilatation, which can be detected by echocardiography. Meanwhile, Angiography is still the gold standard for diagnosing CAF^{1,3,7} and should be considered in patients with chest pain and normal cardiovascular examinations. Interestingly, CAF was mostly diagnosed through incidental angiography.^{1,3,7}

The most common form of CAF is a single fistula originating from the RCA that drains into the right heart chamber.⁷ While left to right shunt was the basic mechanism of heart failure,⁴ steal phenomenon is the primary pathophysiology of myocardial ischaemia, with prevalence of angina in adolescents ranging between 3

and 7%.¹ Previous case report define the steal phenomenon by ST-elevation and abnormal troponin value,⁴ however, our case defined it by diminished CAF and improved LAD perfusion from TIMI 1 to TIMI 3 as well as symptoms improvement after complete fistula occlusion. In addition, as 60% stenosis of the coronary arteries can cause stable angina,⁸ the 60% stenosis of the LAD may also contribute to myocardial ischaemia in our patient, particularly during an increase in oxygen demand. However, we are convinced that the primary cause of ischaemia in our patient was coronary steal phenomenon as the patient was symptom-free following embolisation.

American College of Cardiology / American Heart Association (ACC/AHA) guidelines recommend percutaneous or surgical closure with class I recommendation for large or small-to-moderate size fistulas with myocardial ischaemia, arrhythmias, endarteritis, ventricular dysfunction, and enlargement.⁹ Moreover, some authors recommended closure even in asymptomatic patients to prevent complications.¹ Some studies demonstrated that percutaneous coronary intervention is safe and effective, but surgery is recommended for fistulas that are particularly tortuous, large, or high-flowing.^{1,3} Previous literature reported percutaneous catheterization for large tortuous fistula was unsuccessful due to significant residual shunt and device instability⁵ or inability to enter into the distal part of fistula.⁴ In favourable fistula anatomy, occlusion as distal as possible near the terminal part of fistula is preferred to avoid the possibility of occluding the feeding artery. However, if no significant narrowing is observed in the fistula's terminal portion, the coil may migrate beyond the fistula and into the pulmonary circulation. Therefore, regardless of the catheter closure technique, the occlusion should be at a precise point.⁷

Despite the difficulty, a large tortuous fistula can still be embolised at the narrow and straight part of the fistula by catheterization. Thrombosis could be one of the embolisation complications in dilated fistulae, however, embolising just right before the narrowest part may minimize coil detachment to the distal site. Other possible catheterization complications include persistent symptoms, myocardial damage, endocarditis, thrombosis, accelerated HF, arrhythmias, severe valvular disease, and coronary aneurysm.³

Based on clinical evaluation, our patient no longer has any symptoms or activity limitations following embolisation. Mid-term follow-up angiography revealed no coil dislodgement and an improvement in LAD vascularisation following embolization.

This case study has certain limitations. First, the flow before and after embolization was not reported quantitatively to identify the steal phenomenon. However, we measured the flow based on the well-known TIMI classification, and the absence of angina after embolisation supported this conclusion. Second, successful embolisation of a large tortuous fistula demonstrated a favourable outcome in short-to-intermediate-term follow-up; however, long-term follow-up is necessary to determine the efficacy of percutaneous embolisation by this method.

Conclusion

Coronary artery fistula is a rare cause of chest pain that may be identified late in the absence of an abnormal standard cardiovascular examination. In favourable fistula anatomy, distal coronary occlusion is recommended, but the precise occlusion site depends on the fistula anatomy of each individual instance. The case of a 49 year old male with a single, large-diameter, tortuous coronary artery fistula manifesting as myocardial ischaemia due to the steal phenomenon, is presented. The undesirable connection was successfully obliterated by percutaneous embolisation leading to an improvement in symptoms and daily activities.

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