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CASE REPORT

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In-stent repeated thrombosis secondary to coronary artery aneurysms percutaneous coronary intervention in Behchet's Disease: A Case report

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Abstract

Coronary artery aneurysm (CAA) is a rare and poor prognostic manifestation of Behcet's disease(BD). Percutaneous treatment approaches frequently failed to ameliorate acute coronary system (ACS). Long-term follow-up is recommended as the prognosis of coronary involvement and the risk of further disease progression with percutaneous intervention in BD are unknown. long-term anticoagulant and antiaggregant therapies should be considered to prevent further thrombosis and/or embolism.

KEYWORDS

anticoagulant and antiaggregant therapies, Behchet's disease, coronary artery aneurysm, re-occlusion

1 | INTRODUCTION

Behcet's disease (BD) is a multisystemic vasculitis of unknown etiology. Giant coronary aneurysms which can lead to myocardial infarction, sudden death, or chronic coronary insufficiency are extremely uncommon but the most severe complications of BD. We present the case of a 34-year-old male who had an acute coronary infarction manifestation and was later found to be suffered with BD. Coronary angiography revealed multiple aneurysms along the left anterior descending, left circumflex artery, and right coronary artery. Percutaneous coronary intervention treated the occlusion and aneurysms of the right coronary artery (RCA), but the coronary angiography clearly revealed an acute stent thrombosis in 24 hr One year later, coronary angiography showed a total occlusion of the right coronary artery and no progression of coronary artery aneurysms treated with anticoagulant and antiaggregant therapies. Coronary artery

aneurysm (CAA) is a rare and poor prognostic manifestation of BD. Percutaneous treatment approaches frequently failed to ameliorate acute coronary system (ACS). Long-term follow-up is recommended as the prognosis of coronary involvement, and the risk of further disease progression with percutaneous intervention in BD is unknown. Long-term antiaggregant therapies seemed not working well in preventing thrombosis and/or embolism.

Behchet's disease (BD) is known as a chronic inflammatory process with recurrent oral and genital aphthous ulcerations, skin eruptions, eye inflammation, and neurological manifestations.[1] Cardiovascular involvement is a serious complication, although involvement of coronary arteries is extremely rare and often associated with poor prognosis.[2-4] We describe a BD patient with acute myocardial infarction (AMI) complicated with coronary artery aneurysm (CAA) and progressive in-stent thrombosis observed after percutaneous coronary intervention treatment.

2 | CASE PRESENTATION

A 34-year-old Chinese man with 1-hour persisting chest pain was admitted to our hospital. The electrocardiogram (ECG) showed evidence of ST segment elevation at the inferior and right ventricular leads. Coronary angiography (CAG) revealed multiple aneurysms along the left anterior descending, left circumflex artery, and right coronary artery with a thrombotic occlusion following the distal aneurysm (Figure 1). Consequently, two drug eluting stents (DES, 3.0 33mm; DES, 4.0 18mm) were implanted successfully in the RCA followed by thrombus aspiration. In the cardiology ward, the patient was treated with antiaggregant and anticoagulant agents, including aspirin, ticagrelor, tirofiban, and enoxaparin. One day later, the patient complained with chest pain and the ECG was found elevated ST segment in precordial and right ventricular leads. Emergency CAG revealed an acute stent thrombosis of the RCA (Figure 2). Balloon dilatation was performed to achieve TIMI 0-I flow with unsatisfactory result. Conservative management with antiaggregant and anticoagulant therapies was continued in the all. On the basis of recurrent myocardial infarction, the history of aneurysm formation, and recurrent oral ulcers, the diagnosis of BD was suspected. The rheumatology team confirmed diagnosis of BD with positive pathergy test. In consideration that immunosuppressive treatments may interfere with the ventricular healing process of AMI and increase the risk of scar thinning and myocardial rupture, immunosuppressive treatments were not prescribed for him. In the patient with BD, venous, arterial, pulmonary, and other cardiovascular complications were evaluated carefully by duplex ultrasonography, thoracic CT, echocardiography examination, and magnetic resonance imaging. No other cardiovascular complications associated with BD such as vein thrombosis, systemic arterial aneurysms, pulmonary artery aneurysms and cardiac involvement including pericarditis, cardiomyopathy, congestive cardiac failure, valvular diseases, sinus of Valsalva aneurysms, ventricular aneurysms were seen. The patient was subsequently discharged on triple therapy, including rivaroxaban, aspirin, and ticagrelor. The patient remains asymptomatic after the procedure. One year later, coronary angiography showed a total occlusion of the right coronary artery and collaterals

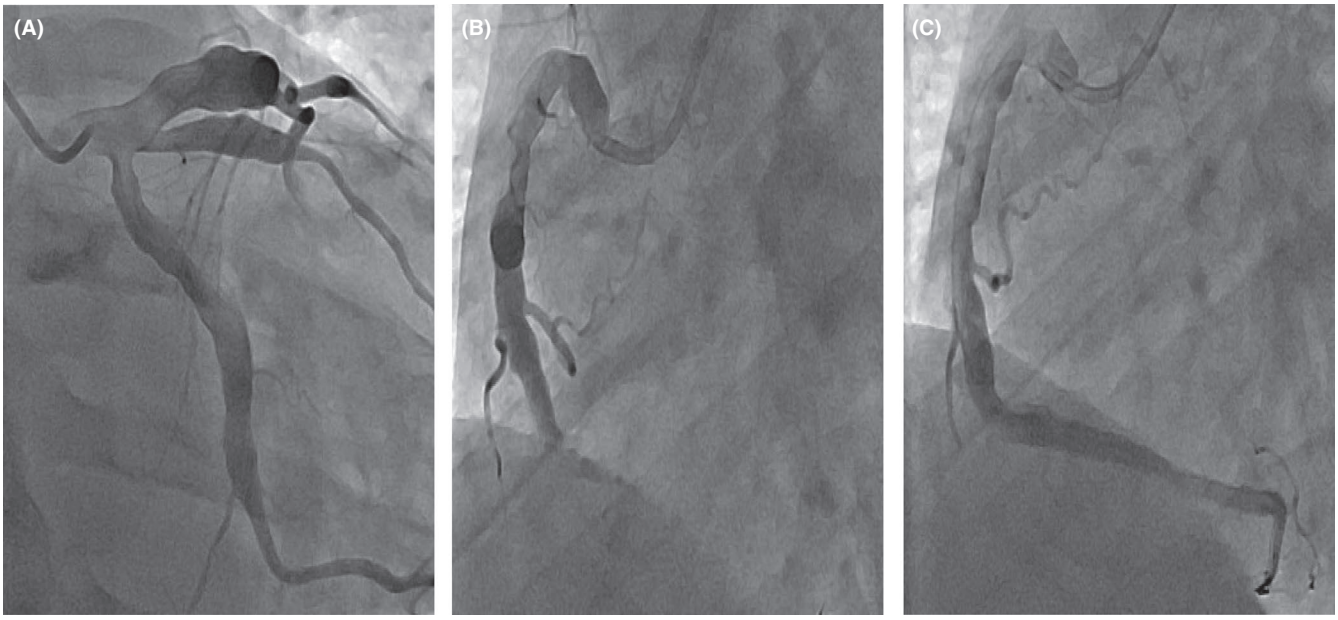

FIGURE 1 coronary angiography. A, Left anterior descending and left circumflex artery aneurysms. B, right coronary artery aneurysm with a thrombotic occlusion following the distal aneurysm. C, Coronary angiography after drug-eluting stent implantation

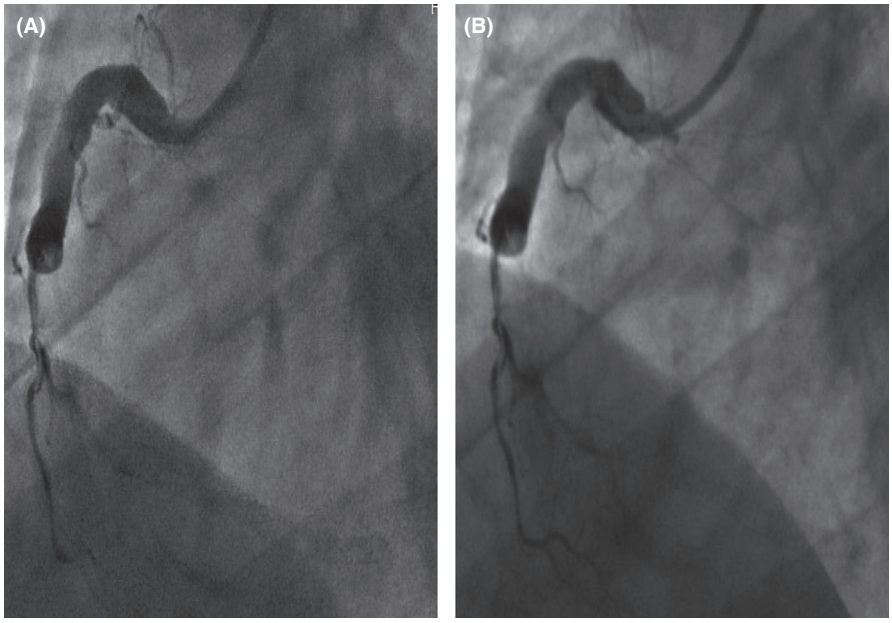

FIGURE 2 One day later, coronary angiography. A, An acute stent thrombosis of the right coronary artery. B, The right coronary artery with TIMI 0-I flow after balloon dilatation

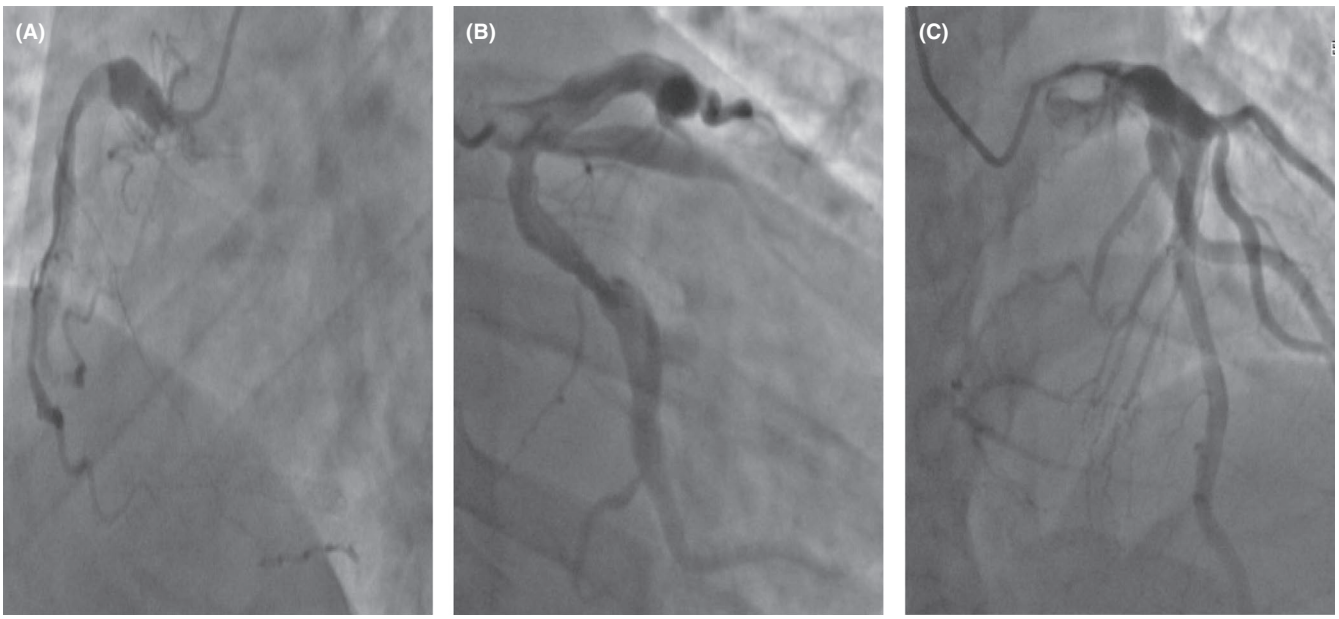

FIGURE 3 One year later, coronary angiography. A, A total occlusion of the right coronary artery. B, Left anterior descending and left circumflex artery with no progression of coronary artery aneurysms. C, The right coronary artery collaterals from LAD

from LAD, left anterior descending, and left circumflex artery with no progression of coronary artery aneurysms treated with anticoagulant and antiaggregant therapies (Figure 3). The patient had been treated with triple therapy, including rivaroxaban, aspirin, and ticagrelor for one year. One year later, he received only long-term anticoagulation therapy with rivaroxaban. Noninvasive methods such as doppler ultrasonography, CT, or MRI had been preferred in evaluation and follow-up of arterial lesions not only when clinically indicated but also routinely once every year. No significant changes had been observed during the second- and third-year follow-up.

3 | DISCUSSION

Arteries in BD are affected by the perivascular and endovascular inflammatory process, resulting in a unique spectrum of stenosis, thrombotic obstruction, bleeding, and aneurysms.[5,6] Cardiac involvement is showed mostly as coronary arteritis, myocardial infarction, and aortic insufficiency. Among patients with BD, coronary artery disease has a prevalence of 0.5% by the Silk Route[6-8]Trial.

Thrombosis is one of the primary complications of giant aneurysm. Although acute myocardial infarction can be treated with percutaneous coronary intervention or surgical

revascularization,[9] there are no long-term results on its complications and outcomes.[10] In our case, coronary angiography clearly revealed an acute stent thrombosis in one day. One year later, coronary angiography still showed a total occlusion of the right coronary artery. In BD, increased thromboembolism is triggered due to endothelial dysfunction, von Willebrand factor release, platelet activation, increased thrombin and fibrin release, and antithrombin deficiency.[11,12] It is believed that such impaired microvascular function is the cause of coronary events in these patients.[13] Thrombus management is determined based on the mobility of the thrombus. If the thrombosis seems relatively mobile and apt to cause emboli, anticoagulant and antiaggregant therapies and immunosuppressive agents may be used. Combination therapy of low-dose aspirin and warfarin has been shown to reduce risk of myocardial infarction compared to aspirin alone.[14] In our case, the patient received aggressive dual antiplatelet and anti-coagulation therapy, but still could not stop the in-stent acute thrombosis. Triple therapy including rivaroxaban, aspirin, and ticagrelor, after discharge seemed to stop the progressing of thrombosis growing up further.

4 | CONCLUSION

Coronary artery aneurysm (CAA) is a rare and poor prognostic manifestation of BD. Percutaneous treatment approaches are frequently used to ameliorate the acute presentation of the disease, but have a great challenge of acute in-stent thrombosis. Long-term follow-up is recommended as the prognosis of coronary involvement and the risk of further disease progression with percutaneous intervention in BD is unknown. Long-term anticoagulant and antiaggregant therapies should be considered to prevent further thrombosis and/or embolism.

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CONFLICT OF INTEREST

The authors report no conflict of interest.

AUTHOR CONTRIBUTIONS

YM and J-HL: studied the conception and designed the study. M-LG: involved in acquisition of data and analysis and interpretation of data (eg, statistical analysis and computational analysis). M-LG, YM, and J-HL: wrote, reviewed, and/or revised the manuscript. Yi Mao and J-HL: supervised the study.

ETHICAL APPROVAL

The study was approved by the institutional review board.

DATA AVAILABILITY STATEMENT

Data in the current study are available from the corresponding author on reasonable request.

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