

Double Stenting Technique for Successfully Closure of Coronary Aneurysm in Behçet's Disease "Case Report of Coronary-Behçet"

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Abstract

Behçet's disease is an immune-mediated vasculitis characterized by oral and genital ulcerations, manifestations affecting the skin, eyes, central nervous system and musculoskeletal system. Arterial involvement is less common than venous involvement. The coronary arteries are rarely affected, only about 20 cases with coronary aneurysm, with still unknown etiopathogenesis and treatment, have been described in the literature. Regarding the high rupture risk, percutaneous coronary intervention is rarely performed with delayed endothelialization due to inflammatory process requiring prolonged anticoagulation. We present the case of 33-years-old man with Behçet's disease in remission presenting to our department with an acute coronary syndrome. Angiography revealed a large coronary aneurysm developed from the circumflex artery, followed by a 1-1-1 Medina lesion at its bifurcation with the first obtuse-marginal. We performed a double stents technique using CULOTTE-technique that helped stabilize the patient with a partial aneurysm thrombosis. Angiography at 5 days confirmed the complete aneurysm thrombosis. He was discharged under Aspirin 100mg, Clopidogrel 75 mg, Rivaroxaban 10mg for 1 month and Prednisone with a coronary CT scan at 3 months. The Culotte-technique enabled to gradually exclude the aneurysm by placing a double struts layer facing its neck. In the event of obstruction failure, the other alternatives are a covered stent placement (combined with DES) or coils deployment in the aneurysm through the stents struts and finally the surgical resection with an aorto-coronary bypass. Coronary-Behçet is rare; most cases are seen in young men. Our case demonstrates the necessity of regular coronary noninvasive check-up in Behçet patients.

Keywords: Behçet • Behçet • Double stenting technique • Culotte technique

List of abbreviations: BMS: Barr Metal Stent • Cx: Circumflex • DES: Drug Eluting Stent • ECG: Electrocardiogram • EF: Ejection Fraction • NC: Non-compliant; OM: Obtuse Marginal • POT: Proximal Optimization Technique • PTFE: Polytetrafluoroethylene • STEMI: ST Elevation Myocardial Infarction • TTE: Transthoracic echocardiography

Introduction

Behçet's disease is an immune-mediated systemic vasculitis characterized by oral and genital ulcerations, manifestations affecting the skin, eyes, central nervous system and musculoskeletal system [1]. Both genders at the third and fourth decade of life can be affected [1]. The diagnosis is based on the occurrence of disease signs and on positive clinical criteria as referred to the International Clinical Criteria for Behçet's Disease [2,3] (Table 1). Arterial involvement is less common than venous involvement [2,4], it consists in accelerated endothelial damage due to platelet overactivity and inflammation leading to thrombotic events and vascular wall destruction giving aneurysmal dilatation and accelerated atherosclerosis as a major contributor to cardiovascular events [1]. The frequently concerned arteries are the abdominal and thoracic aorta, pulmonary, iliac and femoral arteries [1,4]. The coronary arteries are rarely affected, only about 20 cases with coronary aneurysm or accelerated atherosclerosis [1], whose etiopathogenesis and treatment remain unknown, have been described in the literature [5-8]. Regarding the high risk

of rupture, percutaneous coronary intervention has been described in only rare cases with delayed endothelialization due to inflammatory process requiring prolonged double antiplatelet therapy [1,9]. Corticotherapy may reduce in-stent restenosis risk [10,11]. We described here a clinical case concerning the management of a large and compressive coronary aneurysm developed from the circumflex artery in a patient with history of Behçet's disease in remission presenting with an acute coronary syndrome.

Patient Information

De-identified patient specific information

We present the case of 33-years-old man with a history of Behçet's disease in remission (more than 3 episodes of oral aphthous ulcers associated to uveitis and genital aphthous ulcers) presenting to our department, with a successfully thrombolized posterolateral STEMI and EF at 52% on TTE, for coronary angiography 12 hours later. (Figure 1).

Clinical findings

The angiography was performed by 6F right radial access, with Judkins left and right 5F diagnosis catheter. It revealed a large coronary aneurysm developed from the circumflex artery, followed by a 1-1-1 Medina lesion at its bifurcation with the first obtuse marginal (Figure 2). The distance between the left main and the concerned bifurcation was +/- 20 mm.

Diagnostic assessment

Faced to a TIMI 3 flow and pain sedation, we decided to stop the procedure, perform a CT scan and order a covered stent of adequate size

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Table 1. Behçet's disease diagnosis criterion: adapted from the international study group for Behçet's disease [3].

Criterion	Description
Recurrent oral ulcer	Aphthous ulcers with at least 03 episodes during last 12 months
	Associated with two or more of the following signs
Recurrent genital ulcer	Aphthous ulcers or healing
Ocular injury	Uveitis cells in the dilated eye fundus exam or retinal vascularitis
Skin injuries	Erythema nodosum, papulopustular cutaneous injuries or pseudofolliculitis with typical acellular nodules
Pathergy test	Diagnosed after 24-48 hours

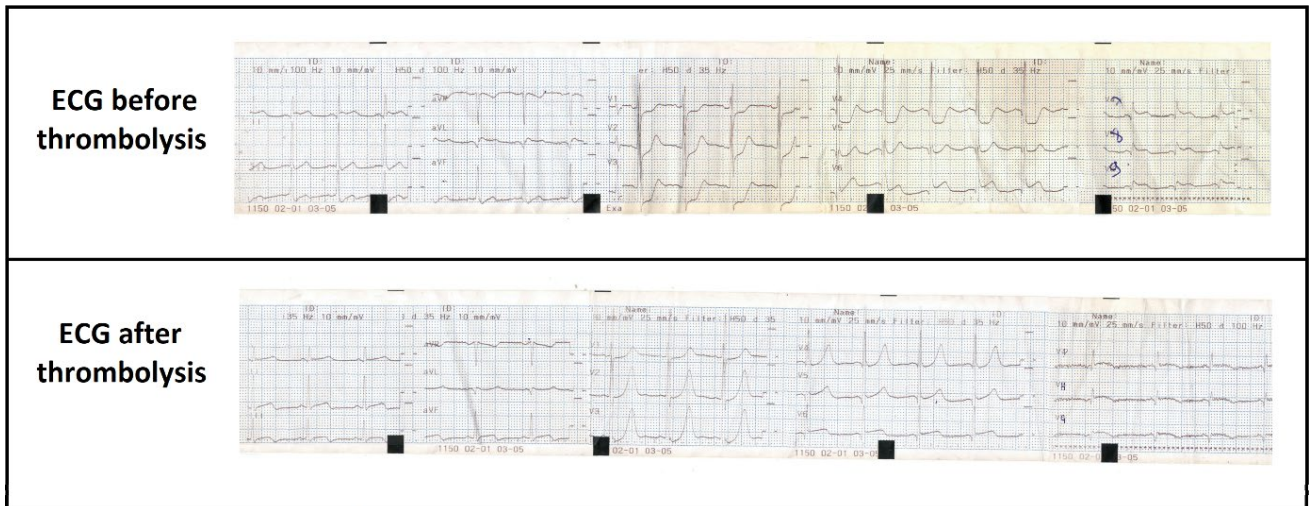


Figure 1. ECG before and after thrombolysis.

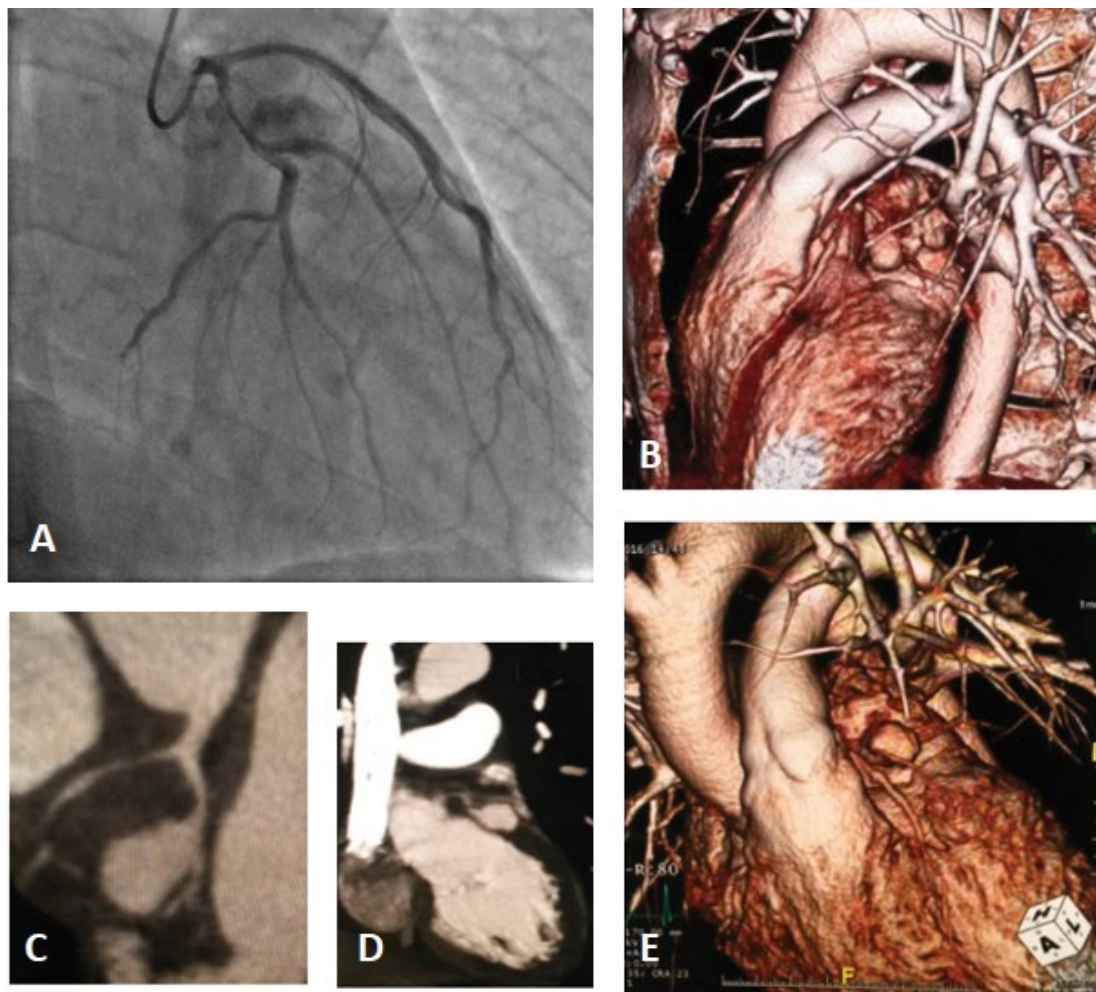


Figure 2. Circumflex artery aneurysm discovered in angiography and CT scan.

(2.80 × 19) in order to exclude the aneurysm. The CT scan done the next day confirmed the existence of a large pseudoaneurysm measuring 15/31 mm with a 4 mm neck developed on the proximal part of the circumflex artery, 14 mm from the left main and 2 mm from the bifurcation circumflex-obtuse marginal which is seat of a soft atherothrombotic plaque type Medina 1-1-1 (Figure 2). Three days after our patient presented a pain recurrence with ST-segment depression and Troponins elevation, the return to the Cath Lab, with right femoral 6F access, revealed an aneurysm worsening with deterioration of blood flow on the circumflex and the obtuse marginal (TIMI 1 Flow) requiring two stents technique.

Therapeutic intervention

We opted for a CULOTTE technique in order to put two layers of struts next to the aneurysm neck and try to exclude it (Figure 3). Otherwise, the angioplasty will be completed by a covered stent placement (ordered, not yet available) or by coils deployment through the stents struts. We started the procedure under Heparin, by placing the 0.014 guidewires in both Cx and the OM, and then we performed a 2.5 × 20 balloon predilatation in both axes.

implanted a first 2.75 × 30 DES in the circumflex-OM axis followed by a POT with a 3.5 × 12 NC balloon, and then we exchanged the guidewires and opened the strut to the distal circumflex with a 2.75 × 15 NC balloon. We implanted a second 3.0 × 30 DES in the Cx-Cx axis, taking the precaution of removing the guidewire from the OM so as not to trap it between two stents. We performed a second POT with the 3.5 × 12 NC balloon then recrossed towards the OM and opened the new stent strut with another 2.75 × 15 NC balloon followed by a Cx-OM kissing balloon with the two 2.75 × 15 NC balloons and we ended with a final POT with the 3.5 × 12 NC balloon. At the end of the procedure (after 40 min and 120 cc of iodinated contrast in total), the TIMI 3 flow was restored on both Cx and the OM with pain sedation and a slowing down of the flow in the aneurysm without a total exclusion (Figure 4).

Follow-up and outcomes

The patient remained stable after the procedure with a normalization of his ECG and EF at 56%.

Five days after his angioplasty and on receipt of the covered stent

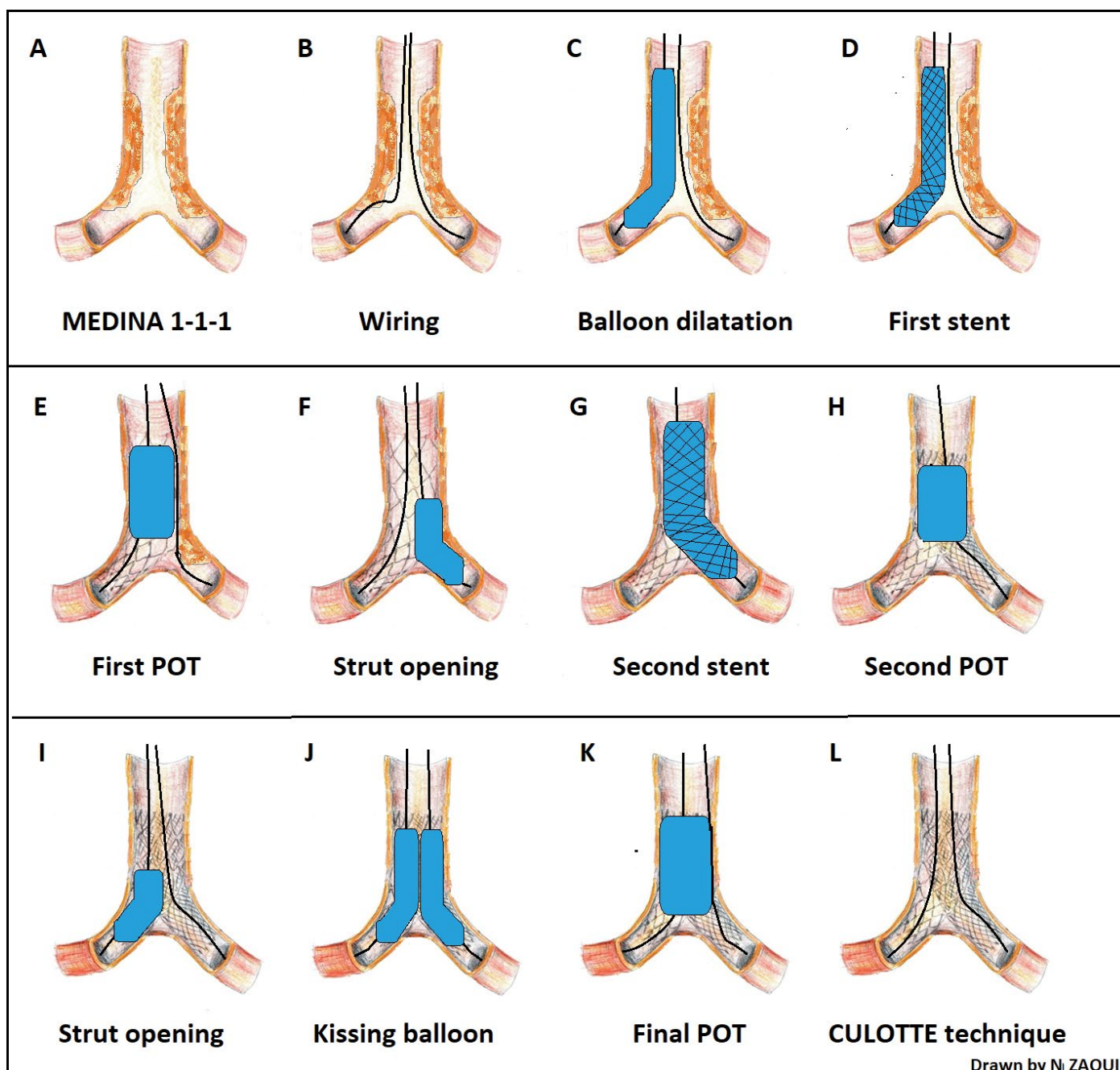


Figure 3. CULOTTE technique.

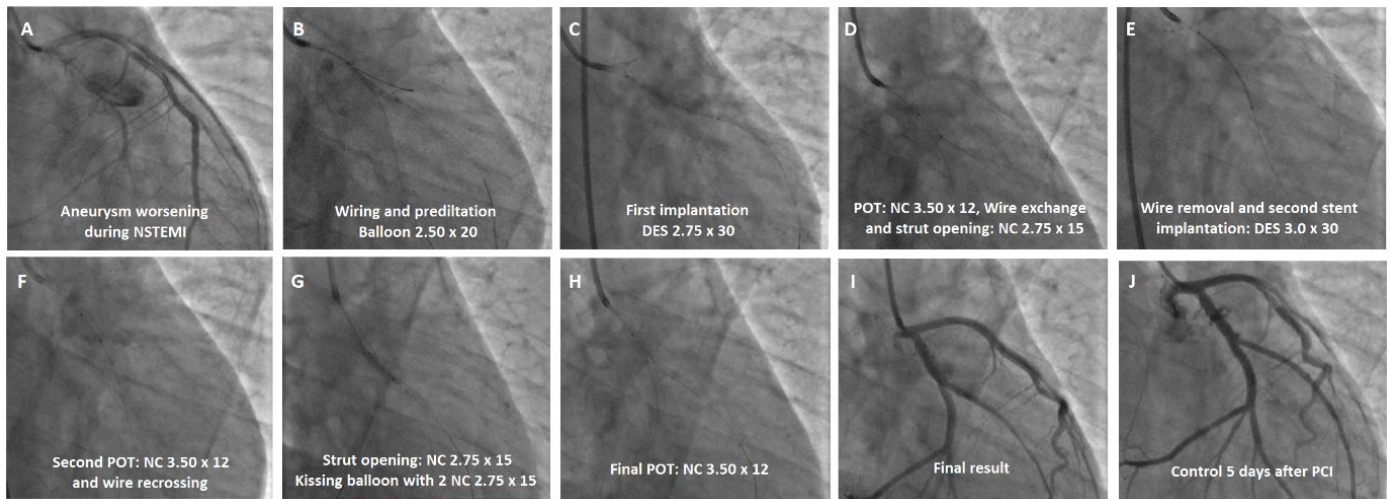


Figure 4. CULOTTE technique and aneurysm exclusion during angiography.

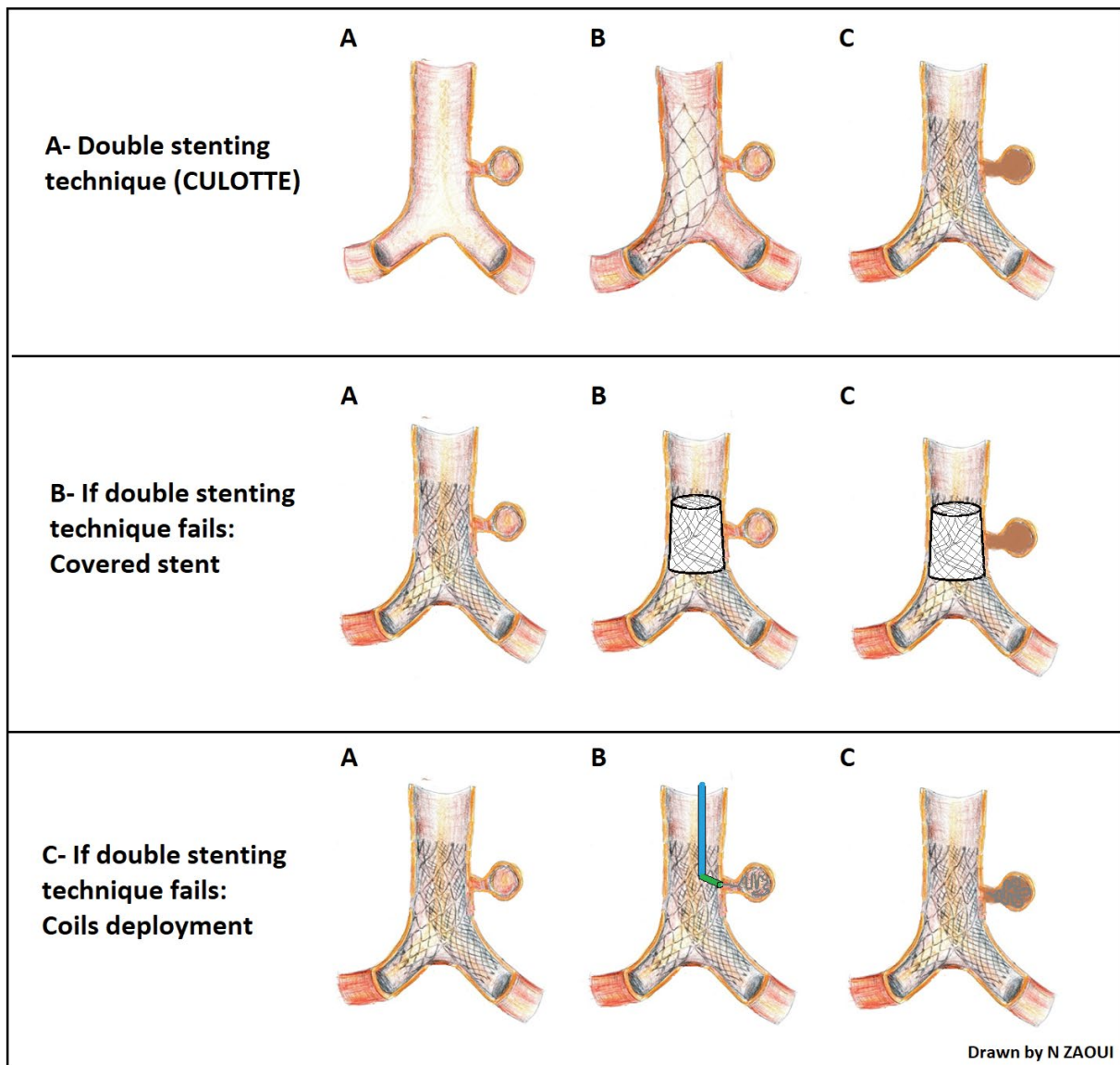


Figure 5. Aneurysm management techniques.

(Graftmaster 2.80 ×19), the return to the catheterization room by the radial access shows the permeability of the CULOTTE and the almost total aneurysm thrombosis (Figure 4) We therefore decided to not implant the covered stent (2 layers of DES in the CULOTTE and 2 layers of BMS on the Graftmaster). The

patient was put on Aspirin 100 mg, Clopidogrel 75 mg, Rivaroxaban 10 mg (to avoid thrombus extension from the aneurysm to the stents) and Prednisone 60 mg as loading dose then 20 mg per day. A CT scan check-up at 3 months was planned (confirming the success of the procedure) as well as an extension

assessment by cerebral and thoraco-abdominal CT (returning negative: no aneurysm).

Discussion

Our patient, first, presented a STEMI responding to thrombolysis testifying to coronary thrombosis by embolization from the aneurysm, by plaque rupture or by mechanical compression. During its second episode (NSTEMI), the worsening of the aneurysm may be linked to a reduction in coronary flow by rethrombosis, the aneurysm in turn compressed and reduced coronary flow. The choice of the angioplasty technique during the recurrence was guided by the need to keep both Cx and OM open and by the possibility of excluding the aneurysm by a double stent layer.

In the case of aneurysm persistence, the options available are (Figure 5) the combination of a covered stent or coils deployment in the aneurysm [12-14]. After having noted the aneurysm exclusion, we decided to not implant the covered stent in order to avoid adding to the double layer of CULOTTE DES another double layer of BMS due to the Graftmaster (made of 'a sheet of PTFE between 2 BMS). We associated anticoagulation with double antiplatelet therapy to prevent the thrombosis spreading from the aneurysm to the stents. This case demonstrates the interest of a non-invasive coronary check-up in Behçet's disease and the difficulty of invasive management of these cases.

Conclusion

Coronary Behçet is rare; most cases are seen in young men. Acute myocardial infarction in these cases may be due to aneurysms that predispose to thrombosis or coronary crushing or due to coexistence of accelerated atherosclerosis. Percutaneous coronary intervention can be dangerous because of the high risk of rupture and the delayed stent endothelialization that requires prolonged anticoagulation (no data about the optimal duration). Corticotherapy may reduce in-stent restenosis risk.

Acknowledgements

We thank our paramedics who participated in the percutaneous procedure.

The Primary Take-away Lesson of this Case

This case demonstrates the interest of a non-invasive coronary check-up in Behçet's disease and the possibility of interventional management of these cases.

Procedure videos are available on our YouTube channel: [Nassime Zaoui]

http://www.youtube.com/channel/UCUps-8DbHFB8EN_ARUCpeZg

Patient Perspective

The patient was very satisfied with the result despite his triple visit to the Cath Lab.

Informed Consent

The patient consented to the sharing and publishing his case and procedure images subject to anonymity

Ethics Approval and Consent to Participate

The patient consented to undergo the procedure.

Consent for Publication

The patient consented to the sharing and publication of data, images and results.

Availability of Data and Material

The images presented during this work are available from the corresponding author on reasonable request.

Competing Interests

The authors declare that they have no competing interests.

Funding

Not applicable.

Authors' Contributions

NZ was responsible for realization of the percutaneous procedure and participated in the writing of the manuscript. AB participated in the percutaneous procedure and the writing of the manuscript. TM participated in the percutaneous procedure. NB participated in the realization of echocardiography and in the writing of the manuscript. AT participated in the realization of echocardiography and the follow-up of the patient during hospitalization. NI participated in the follow-up of the patient after hospitalization. All authors read and approved the final manuscript.

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