A Novel Hybrid Approach to Correct a Giant Aneurysmal Coronary Artery Fistula in An Adult Male Patient

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Glossary of Abbreviations

CAVF: Coronary arteriovenous fistula
CPB: Cardio-pulmonary bypass
CTA: Computed tomography angiography
IVC: Inferior vena cava
RA: Right Atrium
RPA: Right pulmonary artery
SVC: Superior vena cava
TTE: Transthoracic Echocardiography
Central Message:
First reporting of hybrid Amplatz plug coronary fistula closure, followed by surgical excision of gigantic CAVF aneurysm.

Central Picture:
CTA image of the coronary artery fistula aneurysm, compressing all adjacent structures.
Case Presentation

A 37-year-old male presented with supra-ventricular tachycardia, exercise intolerance and lower limb edema. He reported a 2-year history of dyspnea, palpitations and signs of SVC-Syndrome. Transthoracic echocardiography (TTE) and computed tomography angiography (CTA) diagnosed a large coronary artery fistula. It originated from the proximal left circumflex coronary artery travelling through the transverse sinus behind the main pulmonary artery and aorta, into a gigantic aneurysm within the walls of the secundum septum embryonic folding (splaying the left and right atriums) and draining into the RA. (Video 1). A 15cm fistula aneurysm caused almost complete obstruction of the superior vena cava (SVC), inferior vena cava (IVC), right pulmonary veins and artery (RPA).(figure 1.A). The Research Ethic Board (REB) of Nova Scotia Health approved the study protocol and publication of data (1029235, 4/19/2023). The patient provided informed written consent for the publication of the study data.
A hybrid approach was utilized during the index hospitalization by first occluding the fistula (Amplatz® plug) and subsequently resecting the aneurysm on CPB, to ensure adequate myocardial protection. Using a telescoping technique\textsuperscript{1}, the plug was deployed proximal to the fistula aneurysm, angiographically occluding flow through the fistula. This immediately improved left-sided coronary artery filling (Figure 1C, 1D) and decreased the central venous pressure (38 to 24 mmHg).

A sternotomy was immediately performed on CPB via peripheral cannulation, as the aneurysm was adjacent to the sternum. Innominate vein cannulation was added for upper body drainage. Successful antegrade cardioplegic arrest occurred without the need to manually occlude the fistula (plug resistance to flow). The fistula aneurysm was opened between the SVC and aorta. All accessible aneurysmal free wall tissue was resected. The proximal fistula was identified, and the 2cm orifice (Figure 2B) primarily over sewed with the Amplatz® still in situ. The 5mm aneurysm exit site into right atrium (Figure 2C) was primarily closed. The aneurysmal wall attached to the right- and left atrium aspects of the splayed interatrial septum, was re-approximated with interrupted sutures. The patient was weaned off CPB with normal biventricular function. One week later, re-operation was needed to evacuate an 8cm thrombus in the inter-atrial secundum septum. The space was successfully re-obliterated with bio-glue. Due to the large remaining proximal fistula inlet, our institutional approach was to discharge the patient on warfarin and ASA indefinitely for embolic prophylaxis. Eight-month postoperative CTA showed resolution of all obstruction and normalization of heart size. The patient was completely asymptomatic at follow up.
Discussion

To our knowledge this is the first description of hybrid closure of coronary arteriovenous fistula (CAVF) with percutaneous closure to facilitate coronary protection and aneurysm resection during surgical intervention. CAVF is a rare abnormal communication between a coronary artery and adjacent vessels or cardiac chambers with an incidence of 0.2-1.2%. Eventually, CAVF can result in coronary/fistula aneurysm formation due to ongoing left-to-right shunting. Therapeutic interventions are indicated once presence of symptoms, significant left-to-right shunt or coronary ischemia. Management goals are to stop the left-to-right shunting proximal to fistula aneurysms, while assuring unobstructed TIMI 3 coronary flow. Surgical ligation has been the standard for the treatment of CAVF. However, with technological advancements, interventional closure of fistulas that do not affect distal coronary perfusion has become the preferred strategy. Transcatheter therapy won’t relieve compressive effects of coronary aneurysms.

The Achilles heel of surgical CAVF ligation, is myocardial protection. Once CPB is initiated, the venous drainage results in negative pressure in the right sided chambers, potentially accentuating coronary steal and myocardial ischemia. Unoccluded, the fistula will also steal cardioplegia resulting in ineffective myocardial protection, even with retrograde cardioplegia. Therefore, we elected to occlude the proximal fistula percutaneously with the Amplatz® plug before embarking on the surgical aneurysm resection in our patient, without the need of coronary artery bypass graft surgery as described by Iacona et al. Once the plug was deployed, coronary flow to the left coronary artery normalized (Figure 1D), allowing effective cardioplegic arrest. The plug was left in-situ creating a shorter remaining proximal fistula stump communicating with the left coronary system.


Figure Legends
Figure 1. A. CTA image of the coronary artery fistula aneurysm. The large structure occupying most of the right side of the thorax is the giant aneurysm arising from the left circumflex coronary artery fistula and it is compressing the adjacent structures. B. Follow-up CTA Eight-month postoperatively shows resolution of all obstruction and normalization of heart size. C and D. Images of the coronary catheterization pre- (C) and post (D) Amplatz device implantation. The perfusion of left coronary branches improved post-Amplatz device implantation. The device is visualized occluding the fistula.

Figure 2. A. Image showing the Aneurysm between the ascending aorta and the right atrium. B. Image of the coronary fistula inlet occluded by the Amplatz device. C. Image shows the coronary fistula outlet into the right atrium.