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fistula on coronary blood flow*

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## Effects of ligation of a coronary artery fistula on coronary blood flow

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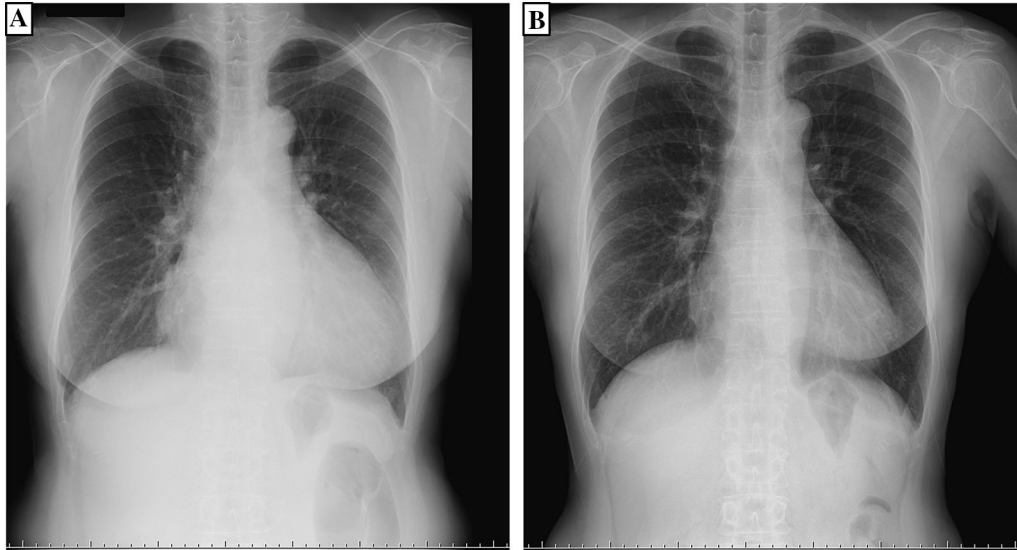
A 60-year-old woman with a known right coronary artery (RCA) to coronary sinus (CS) fistula presented with a 2-month history of dyspnea and chest tightness on exertion and persistent atrial tachycardia. Chest X-ray revealed biventricular dilation, which had not been seen 7 years before (Figure 1). The transthoracic echocardiography showed a normal left ventricular ejection fraction with no wall motion abnormalities, enlargement of all four chambers, and left-to-right shunting with a pulmonary-to-systemic flow ratio (Qp/Qs) of 1.3. Transesophageal echocardiography demonstrated a dilated, tortuous RCA, a fistula between the RCA and dilated CS, and stenosis of CS ostium oppressed by the

dilated RCA (Figure 2). Computed tomography (CT) angiography confirmed the echocardiographic findings: a significantly dilated and tortuous RCA and CS with a single fistula between the terminal RCA and the aneurysmal CS (Figure 3). Additionally, an epicardial collateral vessel was visualized from the distal left anterior descending artery to the distal RCA. <sup>13</sup>N-ammonia positron emission tomography (PET)/CT during adenosine triphosphate (ATP)-induced coronary hyperemia revealed a reversible perfusion abnormality in the inferior and inferolateral regions of the left ventricle. The coronary flow reserve ratio (CFR) was decreased in the RCA territory (Figure 4A). The patient underwent

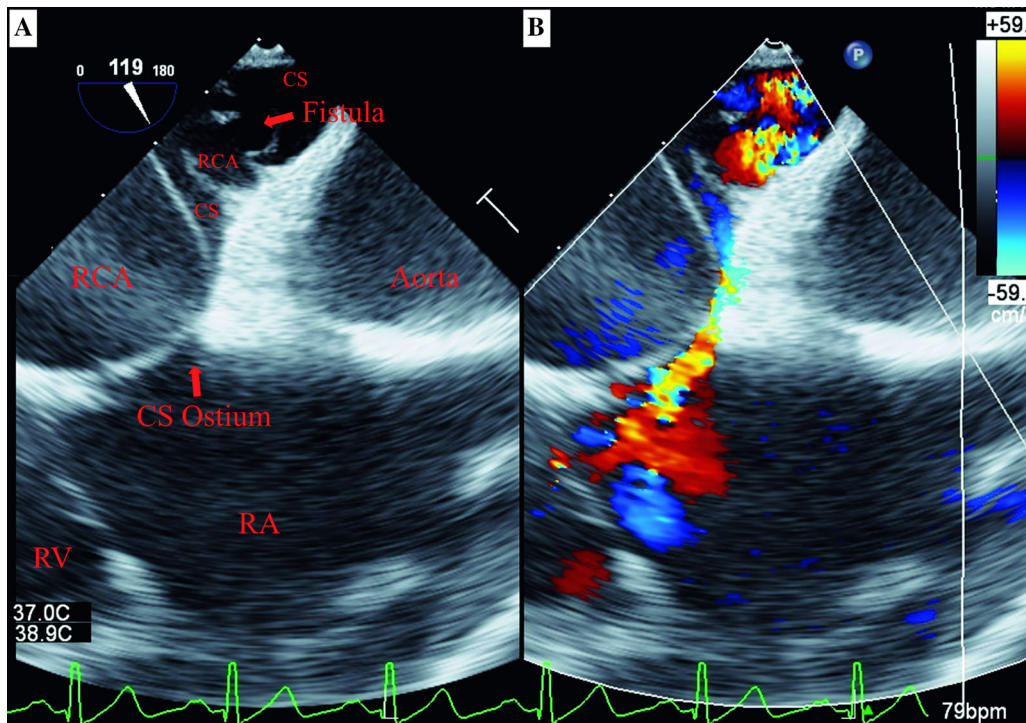
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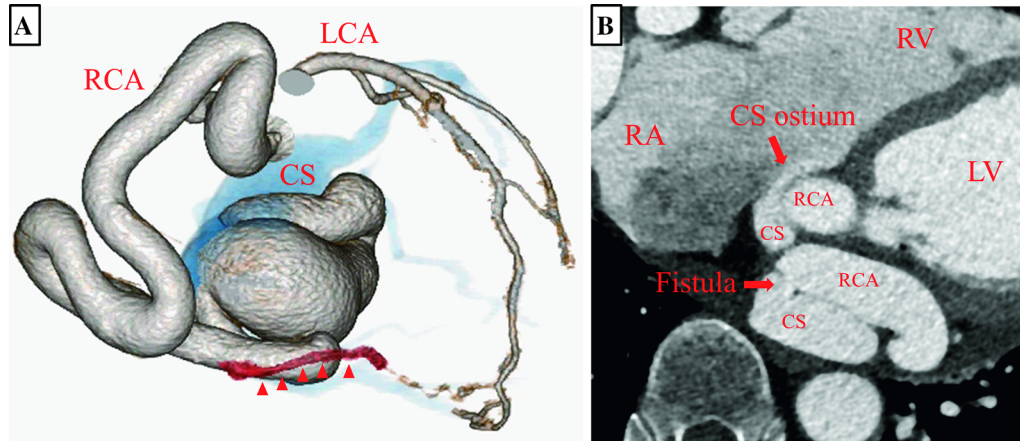


**Figure 1.** Chest X-ray PA view (A) revealed cardiac enlargement at a cardiothoracic ratio of 69% compared with the 49% revealed in the chest X-ray done 7 years ago (B).



**Figure 2.** Two-dimensional transesophageal echocardiography demonstrates the massively dilated distal RCA and aneurysmally dilated proximal CS (A). A turbulent color flow from the distal RCA into the proximal part of the CS was identified as a single fistula by color Doppler flow imaging (B). An enhanced flow into the RA from the narrow CS ostium, which seemed oppressed by the dilated distal RCA with a peak velocity of  $2.8 \text{ m}\cdot\text{s}^{-1}$  at the narrow CS ostium, is visible. RCA, right coronary artery; CS, coronary sinus; RA, right atrium; RV, right ventricle.



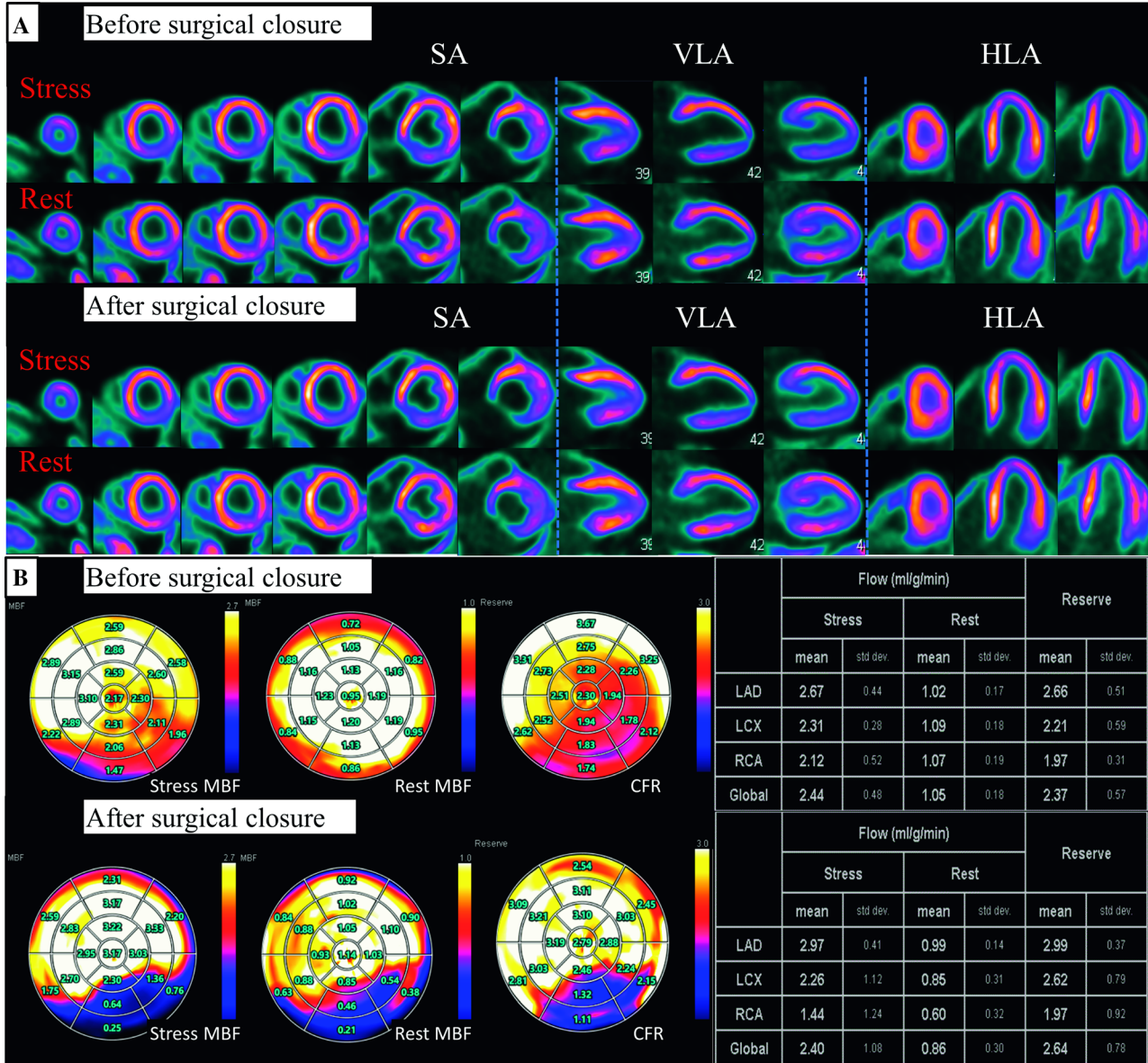


**Figure 3.** CTA (volume-rendered anterior image) demonstrates an extremely enlarged RCA in full length with apparent aneurysmal dilatation in the terminal RCA, an enormously enlarged CS in the proximal portion, and a normal LCA (A). At maximal dilation, the RCA was 36 mm and the CS was 24 mm, highlighting that no epicardial branch from the RCA was observed throughout the entire RCA. Instead, an epicardial collateral vessel was visualized from the distal LAD to the distal RCA (red arrow heads). Axial-enhanced CTA clearly depicted a fistula between the terminal RCA and proximal CS with distinct visualization of the narrow CS ostium compressed by the distal RCA (B). CTA, computed tomographic angiography; RCA, right coronary artery; CS, coronary sinus; LCA, left coronary artery; LAD, left anterior descending coronary artery; RA, right atrium; RV, right ventricle; LV, left ventricle.

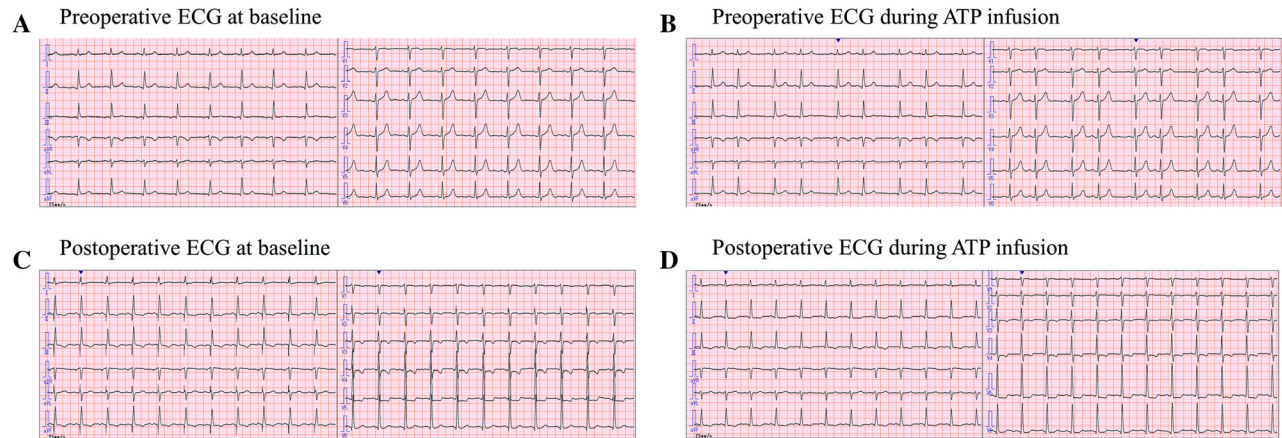
surgical ligation of the fistula under cardiopulmonary bypass without grafting the distal RCA because of small vessel size.<sup>1</sup> PET/CT repeated 16 days after surgery demonstrated a significant reduction in stress/rest myocardial blood flow and CFR in the RCA territory with a larger area of ischemia in the affected area (Figure 4B). For pre and postoperative PET examinations, electrocardiograms were done at baseline and during stress with ATP infusion (Figure 5). Although frequent atrial arrhythmia and symptomatic hypotension

complicated the postoperative course, the patient was discharged 31 days after surgery and the 5-month postoperative follow-up was uneventful.

Herein, we considered through the novel case that the myocardial ischemia before surgery was likely due to coronary steal phenomenon.<sup>2</sup> The worsened ischemia after surgery was likely due to thrombotic occlusion of the RCA aneurysm. Overall, <sup>13</sup>N-ammonia PET/CT can help assess the functional status and quantify coronary blood flow in patients with coronary artery anomalies.<sup>3</sup>



**Figure 4.**  $^{13}\text{N}$ -ammonia PET/MPI demonstrating the preoperative reversible myocardial ischemia on the posterior walls that was significantly expanded postoperatively concerning the location and extent throughout the RCA-supplied territory (A). ATP-induced stress/rest  $^{13}\text{N}$ -ammonia PET/CT was performed to quantify MBF, myocardial perfusion, and CFR (B). The preoperative MBF at rest and stress for the RCA were  $2.12 \text{ mL}\cdot\text{g}^{-1}\cdot\text{min}^{-1}$  and  $1.07 \text{ mL}\cdot\text{g}^{-1}\cdot\text{min}^{-1}$ , respectively, and CFR for the RCA was 1.97 with a localized drop in the RCA territory, which implies the possibility of localized microvascular dysfunction. Postoperatively, rest and stress MBF for the RCA showed a significant decline to  $1.44 \text{ mL}\cdot\text{g}^{-1}\cdot\text{min}^{-1}$ ,  $0.60 \text{ mL}\cdot\text{g}^{-1}\cdot\text{min}^{-1}$ , respectively, with a substantial drop in CFR over the RCA territory, suggesting severe myocardial ischemia potentially provoked by the enhanced turbulence flow effect. *PET/MPI*, positron emission tomography myocardial perfusion imaging; *SA*, short axis; *VLA*, vertical long axis; *HLA*, horizontal long axis; *MBF*, myocardial blood flow; *CFR*, coronary flow reserve; *MBF*, myocardial blood flow.



**Figure 5.** Preoperative electrocardiograms during  $^{13}\text{N}$ -ammonia PET showed within-normal ranges at baseline (**A**) and an increased incidence in supraventricular premature contraction without remarkable ST segment changes during ATP infusions (**B**). In contrast, postoperative electrocardiograms revealed a postoperative temporary epicardial pacing rhythm, a depression of the ST segment in the anterior precordial leads, and a T wave inversion without pathological Q wave both in the anterior precordial and limb leads at baseline (**C**). The findings were similar during ATP perfusion with enhanced own beat at a heart rate of 78 (**D**). *PET*, positron emission tomography; *ATP*, adenosine triphosphate.

## Disclosure

The authors indicate that they have no financial conflict of interest.

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