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A rare case of giant right coronary artery aneurysm with coronary sinus fistula

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Introduction: Coronary artery fistula (CAF) is a rare congenital anomaly characterized by abnormal communication between a coronary artery and a cardiac chamber or great vessel. Clinical presentation ranges from an incidental murmur to chest pain, arrhythmias, or heart failure.[1]

Case report: 36-year-old female patient was evaluated in 2010 for an asymptomatic cardiac murmur. Her medical history included a congenital cerebral arteriovenous malformation with aneurysm. Transthoracic echocardiography (TTE) suggested an atrial septal defect with left-to-right (L–R) shunting. Further diagnostic investigation included coronary angiography, which revealed a markedly tortuous, dilated right coronary artery (RCA), measuring approximately 10 mm, with a 5-mm fistula draining into the coronary sinus (CS). Right heart catheterization demonstrated a pulmonary-to-systemic flow ratio (Qp/Qs) of 1.4:1. Radionuclide myocardial perfusion imaging showed no ischemia and confirmed a moderate shunt. Given the absence of symptoms, conservative management with regular follow-up was adopted. The patient remained asymptomatic for 12 years. In 2022, following COVID-19 infection, she developed progressive dyspnea, fatigue, and intermittent chest discomfort. TTE showed preserved biventricular systolic function with a visible RCA and CS dilatation, with turbulent flow. Cardiac computed tomography angiography (CCTA) demonstrated progressive RCA enlargement, which measured 16 mm proximally, while repeat perfusion imaging showed an increased Qp/Qs of 1.9:1. Conservative management was maintained until 2024, when CCTA demonstrated further progression of the proximal RCA aneurysm, which had enlarged to 20 mm. After Heart Team discussion, the patient was referred to a specialized centre in the USA for surgical treatment, due to limited national experience in the treatment of such complex anatomies and the high risk of transcatheter closure. In July 2025, she underwent surgical CAF excision, CS reconstruction, and RCA resection with sextuple saphenous vein graft (SVG) bypass. The graft comprised five sequential anastomoses to acute marginal branches and a distal anastomosis to the posterior descending artery. The procedure resulted in six sequential bypass grafts (SVG–AM–AM–AM–AM–AM–PDA). Postoperative recovery was uneventful. CCTA confirmed graft patency and successful CS reconstruction. Follow-up TTE showed improved right ventricular systolic function and normalized right atrial size. At three- and six-month follow-up, the patient remained clinically stable without recurrent symptoms.

Conclusion: This case highlights the challenges of determining intervention timing in a progressively enlarging RCA aneurysm associated with a coronary sinus fistula. Complex anatomy precluded transcatheter closure, necessitating extensive surgical reconstruction. Multimodality imaging, longitudinal surveillance, and multidisciplinary Heart Team decision-making are essential in managing complex CAFs.

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