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Aneurysmal coronary arteries with a giant coronary sinus fistula resulting in mitral regurgitation

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Short title: Coronary sinus fistula and mitral regurgitation.

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Coronary artery fistula (CAF) originating from a left circumflex coronary artery (LCx) and draining into a coronary sinus (CS) is a rare clinical entity [1-4]. We present an extremely rare case of a patient with an aneurysmatic left main coronary artery (LMCA) and LCx with a fistula of the latter draining into the CS and contributing to mitral regurgitation (MR). The latter is an extraordinary finding in CAF patients.

A 57-year-old man with a diagnosed severe MR and a history of heart failure was admitted due to clinical exacerbation. Hitherto, no other cardiac abnormalities had been detected. Transthoracic echocardiography revealed impaired LV function (EF≈40-45%), a pattern of turbulent flow close to sinuses of Valsalva, an eccentric posterior MR jet and a dilated CS (CSd≈20mm, arrow) (Figure 1A, Video S1). Transesophageal echocardiography confirmed MR, demonstrated a turbulent flow in the LMCA (Figure 1B) and a severely dilated LCx (Figure 1C) affecting the mitral valve. A coronary angiography revealed aneurysmatic LMCA and LCx with a giant CAF draining from the LCx into the CS (Figure 1D, Video S2) and a stenotic lesion in the RCA (Figure S1). A computed tomography confirmed the above-mentioned findings (Figure 1E). LAD and obtuse marginal branch were of normal diameter. The patient was referred for an open-heart surgery (Figure S2). Intraoperative assessment revealed a non-coapting mitral valve with a distorted posterior mitral annulus due to the aneurysmatic LCx and dilated CS secondary to CAF. Due to the valve morphology, the distorted valve was replaced with a bioprosthesis. LMCA and a fistulous connection were ligated, LAD was bypassed with a left internal mammary artery, the origin of the obtuse marginal branch was occluded and bypassed with a saphenous vein graft. The vein graft was also used to bypass the RCA. Left atrial appendage was closed to reduce thromboembolic risk.
The patient was discharged from the hospital on 7th postoperative day. On follow-up three months later, he reported no symptoms, had normal prosthetic valve function, patent grafts and no fistulous communications (Figure 1F).

References
Figure 1. A-Transthoracic echocardiography (parasternal long axis view) demonstrating an eccentric posterior MR jet, a dilated coronary sinus (arrow) and a turbulent flow close to sinuses of Valsalva (Ao–aorta, MR–mitral regurgitation); B-Transesophageal
echocardiography (mid esophageal short axis view) demonstrating a turbulent flow in the LMCA (arrow); C-Transesophageal echocardiography (midesophageal comissural view) demonstrating a severely dilated LCx (arrow) affecting the mitral valve (interrupted arrow); D-Non-selective coronary angiography demonstrating a giant left circumflex coronary artery fistula draining into the coronary sinus; E-Computed tomography demonstrating the giant LCx fistula draining into the CS; F-Post-operative computed tomography demonstrating the patent left internal mammary to the LAD graft (arrow), the saphenous vein to the obtuse marginal graft (interrupted arrow) and no fistulous communications.

Supplementary material:

Video S1. Transthoracic echocardiography (parasternal long axis view) demonstrating an eccentric posterior MR jet and a turbulent flow close to sinuses of Valsalva.

Video S2. Non-selective coronary angiography demonstrating a giant left circumflex coronary artery fistula draining into the coronary sinus.

Figure S1. Right coronary angiography demonstrating a significant stenotic lesion (arrow).

Figure S2. Intraoperative image of a fistula.