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Complex coronary pulmonary artery fistulae with a large aneurysm: a rare anomaly treated with surgery

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A 77-year-old man presented with an asymptomatic mediastinal mass, incidentally detected by computed tomography. Chest auscultation revealed no murmur. Multidetector computed tomography (MDCT) revealed bilateral coronary pulmonary artery fistulae (CPAFs). The right CPAF connected the conus branch to main pulmonary artery (MPA) through a small aneurysm. The left CPAF

contained a large aneurysm with multiple fistulae from the left anterior descending artery, MPA, and aortic arch (Figure 1). Coronary angiography also showed bilateral CPAFs (Figure 2, Videos 1 and 2). Cardiac catheterization determined the pulmonary to systemic blood flow ratio of 1.2, and single-photon emission computed tomography revealed a normal coronary flow reserve. CPAFs are rare and

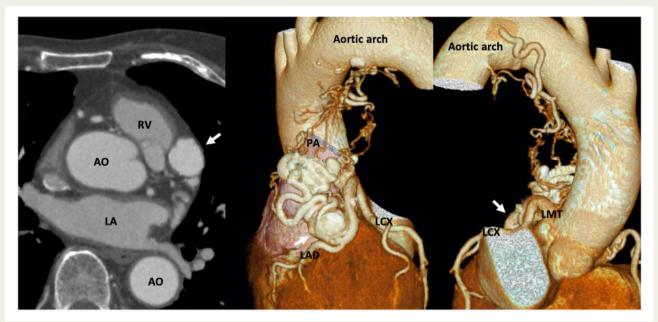


Figure I Preoperative multidetector computed tomography. Left coronary pulmonary artery fistula contained a large aneurysm (white arrows) with multiple fistulae from left anterior descending artery (LAD), main pulmonary artery (PA), and aortic arch. AO, aorta; LA, left atrium; LCX, left circumflex artery; LMT, left main coronary trunk; RV, right ventricle.

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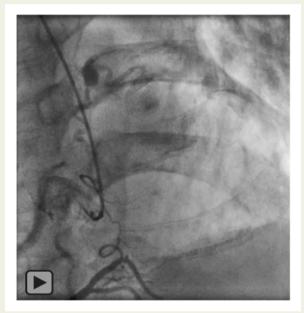
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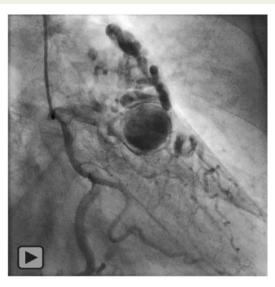


Figure 2 Coronary angiography showing bilateral coronary pulmonary artery fistulae (CPAFs). Right CPAF connecting conus branch to main pulmonary artery (PA) through a small aneurysm. Left aneurysm derived from left anterior descending artery (LAD). LCX, left circumflex artery; RCA, right coronary artery.



Video I Coronary angiography of right coronary artery. Right coronary pulmonary artery fistulae connecting conus branch to main pulmonary artery through a small aneurysm.

generally congenital anomalies: multiple CPAFs account for 45% and coronary artery aneurysms coexist in 19%.¹ The management of CPAFs; surgery or transcatheter occlusion, is controversial.^{2,3} CPAFs with symptoms, aneurysms, or complex vascular communications tend to be treated with surgery.³ While our patient was asymptomatic and haemodynamically stable, we were concerned about the rupture of the aneurysm leading to sudden cardiac death because of the large size of the aneurysm. Considering the complexity of the CPAFs, we thought that transcatheter repair would not occlude the fistulae totally and therefore, surgery would be a more feasible and effective



Video 2 Coronary angiography of left coronary artery. Left aneurysm deriving from left anterior descending artery.

approach. After discussing the risks and benefits of the surgical and transcatheter approaches with the patient, the decision was made to pursue surgical repair. During operation, observable multiple fistulae draining into MPA, and inflow and outflow tracts of left aneurysm were selectively ligated and clipped. Left aneurysm and MPA were subsequently incised to confirm the drainage sites from the inside. The residual feeding vessels were ligated, and MPA was re-sutured. Finally, we confirmed the disappearance of blood flow into the aneurysm when weaning from cardiopulmonary bypass system (Supplementary material online, Figures \$1 and \$2). Postoperative MDCT showed disappearance of contrast enhancement in the aneurysm (Supplementary material online, Figure \$3). Management should be carefully selected considering the anatomical condition of the CPAFs. MDCT plays an important role in pre-management and post-management evaluation.

Supplementary material

Supplementary material is available at European Heart Journal - Case Reports online.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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