

» **Case Report** «

Coronary-Pulmonary Artery Fistula Provides Collateral Flow to an Occluded Left Anterior Descending Artery

Baku Takahashi , MD, PhD, Hideyuki Fumoto, MD, PhD, and Yoshihiro Nakayama, MD, PhD

A 59-year-old man presented with angina. Coronary angiography revealed an occlusion in the proximal left anterior descending artery (LAD), the distal segment of which was supplied by the collateral flow of a coronary-pulmonary arterial fistula (CPAF), originating from the right coronary artery and left sinus of Valsalva. Myocardial scintigraphy revealed ischemia in the anteroseptal region. Coronary artery bypass surgery was performed on the LAD, and the CPAF drains were closed. The CPAF may serve as collateral circulation. Even when CPAF serves as collateral circulation, open surgery could be indicated if the collateral flow is insufficient and the structure is complicated.

Keywords: coronary-pulmonary arterial fistula, coronary artery bypass grafting, collateral flow

Introduction

Coronary-pulmonary arterial fistula (CPAF) is typically considered to be a harmful disease that causes ischemia because it shunts blood away from the native artery, a process termed the “steal phenomenon.”¹⁾ However, two studies have reported that CPAF can function as an endogenous bypass graft for occluded coronary arteries.^{2,3)} These cases were managed conservatively and with endovascular therapy. In the case presented here, CPAF served as a collateral supply to an occluded left anterior descending artery (LAD). Stress myocardial scintigraphy was performed to diagnose ischemia, and surgical revascularization and fistula closure were performed. To our knowledge, this is the


first report of a surgical intervention for a CPAF that supplies circulatory flow to the occluded coronary artery.

Case Report

A 59-year-old man presented at our hospital with month-long symptoms of chest pain and discomfort during exercise. He had hypertension but no family history of heart disease. The patient had not undergone coronary angiography previously. Physical examination revealed no remarkable findings, and laboratory test results did not reveal any abnormalities. Electrocardiography showed ST-segment depression in leads V3–V6. Echocardiography revealed normal left ventricular movement. Coronary computed tomography (CT) revealed a completely occluded proximal segment of LAD. In addition, we observed a CPAF originating from the conus branch of the right coronary artery (RCA) and the left sinus of Valsalva near the ostium of the left coronary artery, which was connected to the distal portion of the LAD (Fig. 1A). Left coronary angiography revealed LAD occlusion in the proximal segment while the distal segment was supplied by the left sinus of Valsalva via the pulmonary artery (PA). Right coronary angiography demonstrated that the apical LAD filled the conus branch through the PA (Fig. 1B). The exact number and location of fistulas could not be identified preoperatively. The ratio of pulmonary to systemic blood flow was 1.32. Thallium-201 myocardial perfusion scintigraphy revealed ischemia in the anteroseptal region. Surgery was performed to achieve revascularization and complete fistula closure simultaneously. Through median sternotomy, the left internal thoracic artery was bypassed to the distal site of the LAD after a cardiopulmonary bypass was established with aortic cannulation in the ascending aorta and bicaval drainage. The main PA was longitudinally incised, and two fistula drains from the CPAF were identified above the pulmonary valve. The drains were closed directly using 5-0 polypropylene inside the PA. The anomalous vessels from the conus branch of the RCA, and the sinus of Valsalva leading to the coronary aneurysm on the PA, were ligated with 2-0 silk near the orifice to

Department of Cardiovascular Surgery, Osumi-Kanoya Hospital, Kanoya, Kagoshima, Japan

Received: December 4, 2023; Accepted: June 5, 2024
Corresponding author: Baku Takahashi, MD, PhD. Department of Cardiovascular Surgery, Osumi-Kanoya Hospital, 6081-1 Shinkawa-cho, Kanoya, Kagoshima 893-0015, Japan
Tel: +81-99-440-1111
E-mail: bakutakahashi@yahoo.co.jp

 This work is licensed under the Creative Commons Attribution-NonCommercial-ShareAlike International license.
©2024 The Editorial Committee of Annals of Vascular Diseases.

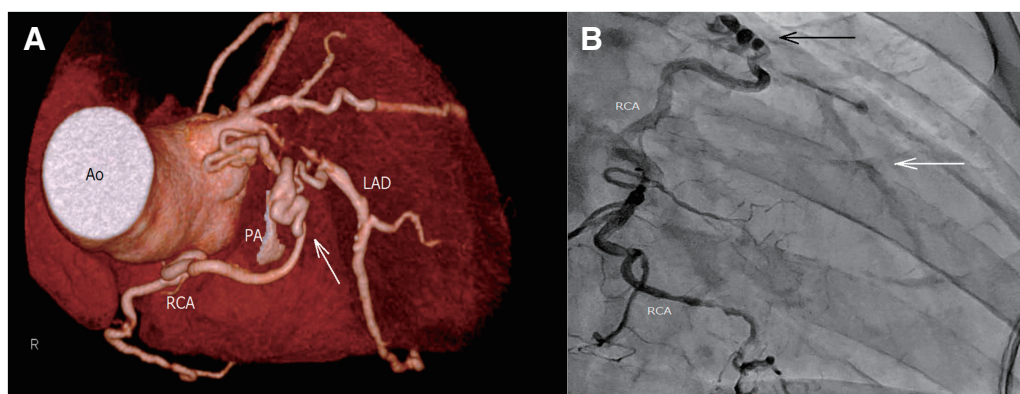


Fig. 1 (A) Coronal CT angiography showing occlusion of the proximal LAD. Abnormal, tortuous vessels appear to be connected to a conus branch of the RCA, the LAD distal to the occlusion, and the left sinus of Valsalva (white arrow). (B) Right coronary angiography shows that the fistula (black arrow) originates from the conus branch of the RCA to the main pulmonary artery, supplying collateral blood flow to the distal segment of the LAD (white arrow). CT: computed tomography; RCA: right coronary artery; LAD: left anterior descending artery

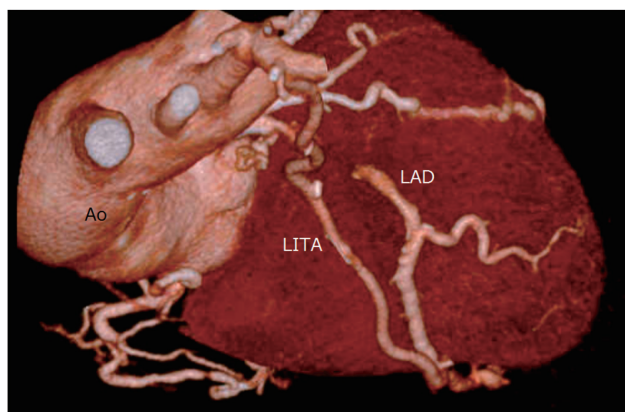


Fig. 2 Postoperative CT showing no signs of fistula and demonstrating a patent bypass graft.
CT: computed tomography; LAD: left anterior descending artery; LITA: left internal thoracic artery

the aneurysm. The patient showed uneventful postoperative recovery. Postoperative coronary CT revealed that the bypass graft was patent, and there were no fistulas or anomalous vessels (Fig. 2). The patient was discharged on postoperative day 16 and has remained asymptomatic for 2 years after surgery.

Discussion

We report a very rare condition in which a CPAF originated from the RCA and left sinus of Valsalva, supplying collateral blood flow to the occluded LAD. Conventionally, CPAF is thought to negatively affect coronary arteries by shunting blood away from the native artery, causing ischemia.¹⁾ However, two cases wherein CPAF functioned as a source of collateral blood circulation to prevent ischemia have been previously reported.^{2,3)}

This suggests the possibility of CPAF behaving as an endogenous bypass graft in cases of significant ischemia of the native coronaries, indicating that CPAF may not necessarily be unprofitable.³⁾ In our case, CPAF may have played a crucial role as collateral circulation to relieve heart damage when the LAD was occluded in this patient.

Guidelines on the indications for a coronary artery fistula (CAF), including CPAF, are lacking due to the rarity of these cases. Al-Hijji et al. recommended that intervention may be considered for symptomatic large or medium CAFs with myocardial ischemia, arrhythmia, rupture, cardiac chamber enlargement, ventricular dysfunction, or endarteritis.⁴⁾ However, the therapeutic indications for cases in which CPAF serves as collateral blood circulation remain unclear. One patient was managed conservatively as there were no signs of ischemia, suggesting sufficient flow from the CPAF to the occluded coronary artery.²⁾ The other case was treated with percutaneous coronary intervention due to insufficient circulation from the CPAF to the occluded LAD.³⁾ From these viewpoints, it is important to evaluate whether collateral flow from the CPAF is sufficient, as well as to determine the indications for the usual CAF. As our case demonstrated CPAF with a small shunt without a steal phenomenon or arrhythmia, stress myocardial scintigraphy was performed to evaluate the collateral supply from the CPAF. This test revealed myocardial ischemia in the anterior wall of the left ventricle, indicating insufficient collateral blood flow from the CPAF to the LAD. Therefore, we concluded that this was an indication of intervention. Thallium-201 myocardial perfusion scintigraphy is known to be a useful method for detecting and locating coronary artery disease.⁵⁾ Scintigraphy is useful, even in unique cases such as this one.

Treatment for CPAF involves fistula closure either surgically or percutaneously.^{4,6)} In addition, revascularization is also required in cases where collateral circulation is insufficient from the CPAF, as in this case. In one case, Yi et al. performed recanalization for LAD occlusion by placing a stent percutaneously in a patient with a CPAF originating from the left circumflex coronary artery (LCx) via the PA, which provided collateral flow to the occluded LAD, subsequently reporting that the collateral channels from the fistula disappeared.³⁾ However, the fistula from the LCx to the main PA remained. The long-term efficacy of this treatment remains unknown, as post-treatment conditions have not been sufficiently described. In this case, even if the occluded LAD was opened percutaneously, the CPAF from the RCA and left sinus of Valsalva would remain. Furthermore, the vessels were extremely tortuous, and multiple fistulas were suspected. Therefore, surgical closure and revascularization were performed. In fact, fistulas that were not detected on preoperative examination were identified intraoperatively, the origins and drains of all fistulas were closed, and LAD was successfully revascularized.

Conclusion

CPAF can function as an endogenous bypass graft for occluded coronary arteries. However, revascularization should be considered when sufficient blood flow is unavailable. Even for the unique CPAF presented here, stress myocardial scintigraphy is useful for the detection of ischemia, and surgical intervention should be effective.

Author Contributions

Study conception: BT

Data collection: BT

Manuscript preparation: BT

Funding acquisition: N/A

Critical review and revision: all authors

Final approval of the article: all authors

Accountability for all aspects of the work: all authors.

Disclosure Statement

All authors have no conflicts of interest to declare.

References

- 1) Mangukia CV. Coronary artery fistula. *Ann Thorac Surg* 2012; **93**: 2084–92.
- 2) Komatsu S, Hirayama A, Sato Y, et al. Coronary-pulmonary fistula serving as a collateral source to the occluded coronary artery in a patient with myocardial infarction. *Int J Cardiol* 2007; **115**: 408–9.
- 3) Yi HT, Lai HC, Hsu H, et al. Left circumflex coronary-to-pulmonary artery fistula as the exclusive collateral to the occluded left anterior descending artery. *Heart Lung Circ* 2014; **23**: e1–3.
- 4) Al-Hijji M, El Sabbagh A, El Hajj S, et al. Coronary artery fistulas: indications, techniques, outcomes, and complications of transcatheter fistula closure. *JACC Cardiovasc Interv* 2021; **14**: 1393–406.
- 5) Korkeila P, Hietanen E, Parviainen S, et al. Exercise thallium-201 scintigraphy in the localization of myocardial ischaemia. *Clin Physiol* 1989; **9**: 555–65.
- 6) Challoumas D, Pericleous A, Dimitrakaki IA, et al. Coronary arteriovenous fistulae: a review. *Int J Angiol* 2014; **23**: 1–10.