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Surgical intervention of coronary-pulmonary artery fistula with multiple coronary aneurysms and Vieussens' arterial ring formation

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Abstract

Background To our knowledge, there have been extremely few clinical reports on coronary-pulmonary artery fistula (CPAF) accompanied by coronary artery aneurysm (CAA) and Vieussens' arterial ring (VAR), and few reports on related surgical treatments.

Case presentation A 61-year-old female patient was admitted with dyspnea and fatigue after exertion. Coronary CTA, echocardiography and coronary angiography revealed multiple CPAF, along with formation of multiple CAAs and VAR. The patient underwent successful surgical intervention under general anesthesia and cardiopulmonary bypass and experienced an uneventful recovery.

Conclusion Hereby we reported this clinically unusual case of CPAFs with multiple CAAs and VAR and also the details of a successful surgical procedure.

Keywords Coronary-pulmonary artery fistula, Coronary artery aneurysm, Vieussens' arterial ring, Surgical intervention

Background

To our knowledge, clinical reports of coronary-pulmonary artery fistula (CPAF) associated with coronary artery aneurysm (CAA) and Vieussens' arterial ring (VAR) are exceedingly rare, with limited documentation of related surgical treatments. Coronary artery fistulas are rare abnormal connections between a coronary artery and a cardiac chamber or great vessel, potentially diverting blood flow from the myocardial capillary network. VAR is typically described as a collateral pathway between the left anterior descending (LAD) and right coronary arteries, with pathological cases being exceedingly rare [1]. In this case, we reported a case of a 61-year-old female patient who presented with exertional dyspnea and fatigue. The patient was diagnosed with

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CPAFs combined with CAAs and VAR formation and subsequently underwent surgical treatment.

Presentation

A 61-year-old female patient was admitted to the hospital with the chief complaint of dyspnea and fatigue after exertion. She had a history of hypertension for over 20 years which has been controlled well with regular oral medication. Physical examination revealed no abnormalities. Upon admission, a coronary CTA revealed a tortuous and dilated vessel originating from the proximal segment of the LAD artery with an aneurysmal expansion in the mid-segment, measuring approximately $1.8 \times 1.9 \times 1.8$ cm (Fig. 1A-F asterisk). The vessel had two branches with one communicating with the left wall of the pulmonary artery (PA) trunk (Fig. 1E and F upper triangle). While another branch connected with the right coronary sinus and formed VAR (Fig. 1C-F arrow), where an aneurysmal dilation measuring approximately $2.7 \times 2.9 \times 3.6$ cm was noted (Fig. 1A-F star). A conus branch artery from the proximal segment of the left main artery (LM) exhibited an aneurysmal dilation measuring approximately $1.0 \times 0.7 \times 0.7$ cm, extending distally to communicate with the PA. Besides, a vessel originating from the superior wall of the aortic arch displayed localized aneurysmal dilation proximally, with a tortuous distal segment communicating with the PA trunk (Fig. 1E and F lower triangle). Echocardiography revealed multiple CAAs, CPAFs with VAR formation. Coronary angiography showed similar findings with coronary CTA and 30% narrowing in the LAD artery.

The patient was diagnosed with CPAFs with CAAs and VAR formation (Figs. 1 and 2 asterisk, star and arrow). The surgical procedure was performed under general anesthesia and cardiopulmonary bypass was established. After the pulmonary artery fistula was clamped, cold cardioplegia solution was infused from the aortic root. The aneurysms in the left and right sinus region were incised and the entrance to the right aneurysm, approximately 0.4 cm in diameter, was sutured with a biological patch. The exit of the right aneurysm was directly closed with suture. Both the entrance and exit of the left aneurysm were also closed by direct suture. The patient experienced an uneventful recovery and postoperative follow-up coronary CTA showed complete disappearance of aneurysms and VAR (Fig. 2F).

Discussion and conclusions

A coronary artery fistula (CAF) is an abnormal connection that forms between the coronary arteries and the heart chambers, or between the coronary arteries and any part of the systemic or pulmonary circulation near the heart [2]. The prevalence of CAFs in the general population is estimated to be approximately 0.002% [3]. With

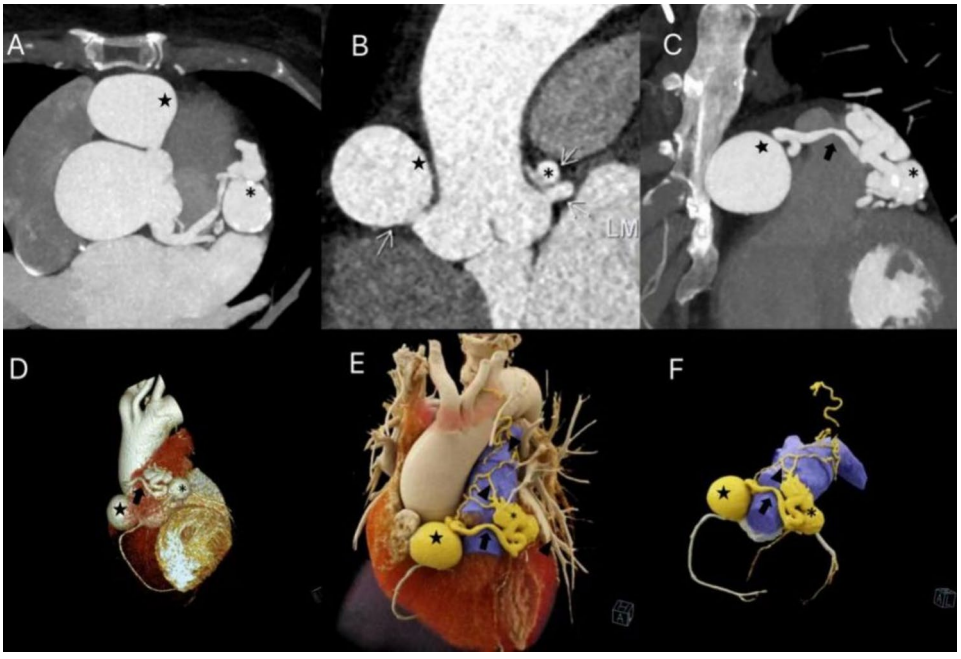

Fig. 1 CTA images in a 61-year-old female patient with coronary-pulmonary artery fistulas (CPAFs) with multiple coronary artery aneurysms (CAAs) and Vieussens' arterial ring (VAR). Transverse section (**A**), coronal section (**B**) and sagittal section (**C**) of CTA showing CPAFs with multiple CAAs and formation of VAR. (**D**-**F**) Reconstruction images of CTA showing CPAFs, CAAs and VAR.
★: Right aneurysm ✻ : Left aneurysm ➡ : Vieussens' arterial ring ▲ : A vessel had two branches with one communicating with the left wall of the pulmonary artery (PA) trunk. ▼ : A vessel originating from the upper wall of the aortic arch showed localized aneurysmal dilatation proximally and tortuous distal connection to the PA trunk

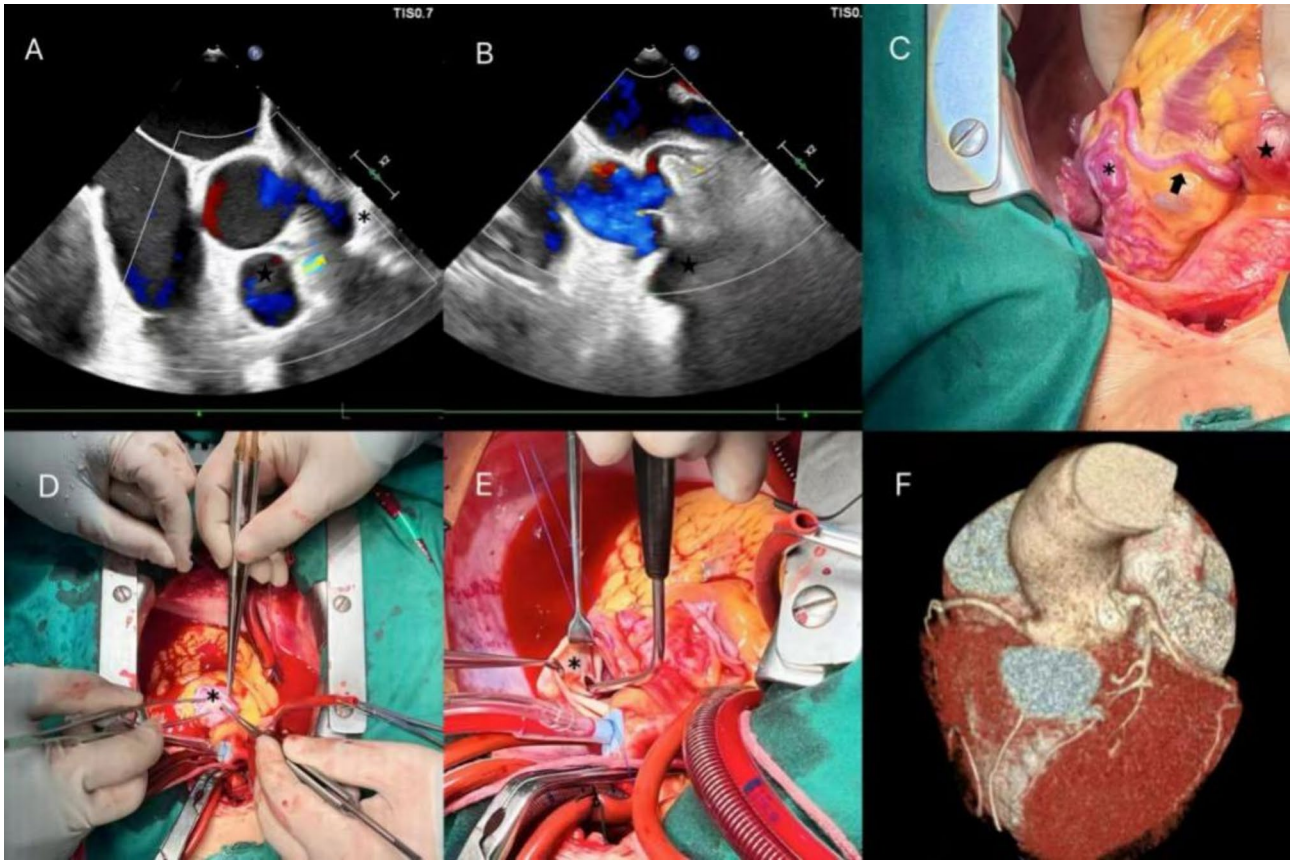

Fig. 2 Echocardiographic, intraoperative and postoperative CTA images in the above-mentioned patients. Echocardiographic images (**A** and **B**) showing the right and left coronary artery aneurysms (CAAs). Intraoperative imaging (**C**-**E**) showing resection of the right and left CAAs. Reconstruction images of postoperative CTA (**F**) showing the complete resection of CAAs.
★ : Right aneurysm ✻ : Left aneurysm ➡ : Vieussens' arterial ring

the widespread use of cardiac CTA, the reported incidence of CAF has increased but remains relatively rare [4]. While most patients remain asymptomatic, some may experience symptoms such as myocardial ischemia, congestive heart failure, or even sudden cardiac death, particularly if coronary aneurysms, thrombosis, infectious carditis, or other congenital heart defects coexist [5]. A CAA is defined as a condition in which the diameter of a dilated segment of the coronary artery is greater than 1.5 times the diameter of an adjacent healthy segment [6]. Aneurysm formation is a potential complication of CAF, and rupture of the aneurysm could be life-threatening. VAR is considered a remnant of the embryonic conotruncal circle [7]. VAR is a common collateral circulation where the left conus branch from the anterior descending artery anastomoses with the right conus branch from the right coronary artery, forming an arterial ring. When an aneurysm occurs secondary to a CPAF, the associated aneurysmal dilation is typically cystic in nature, resulting in a large tumor-like structure. In such cases, the affected blood vessels are often thickened and tortuous, with the aneurysms primarily located near, rather than at, the fistula orifice [8]. The most common clinical symptoms in patients with VAR-pulmonary artery fistula with or without CAAs are chest pain and dyspnea[8]. Currently, there are two main approaches for treating CPAFs: percutaneous or surgical intervention. The primary objective is to close the fistula and block the shunt between the coronary artery and the pulmonary artery. The treatment principle is to achieve fistula closure while preserving normal coronary circulation. Surgical correction is generally recommended for the treatment of large symptomatic CAFs, especially those with multiple communications, tortuous or aneurysmal characteristics, and when CAFs require surgical management of other clinically significant cardiac conditions. A comprehensive coronary CTA should be conducted preoperatively to analyze the opening of the CAA and the blood flow direction of the CAF, enabling the formulation of a detailed and optimal surgical plan to ensure smooth surgical progress. The patient discussed in this study had CPAFs with large CAAs and was therefore treated surgically.

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Author contributions
All authors have substantially contributed to manuscript drafting. HZ, and XM edited the manuscript. All authors have read and approved the final manuscript.

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate
This project has been approved by the Ethics Committee of the Shandong Provincial Hospital Affiliated to Shandong First Medical University, and has obtained written consent from the patient.

Consent for publication
Written informed consent has been obtained from the patient and their family members for publication of relevant clinical information by the authors in pertinent medical publications.

Competing interests
The authors declare no competing interests.

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