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Bilateral Coronary Artery-Right Ventricular Fistula with Rare, Large Tortuosity and Common Exit

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Case Report

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Abstract

We present a case study of a complex congenital bilateral coronary artery fistula (CAF) involving the right coronary artery (RCA), left anterior descending artery (LAD), and right ventricle (RV). The angiographic features and imaging data during interventional occlusion are described, and a systematic review of published cases of this disease in the past 50 years is conducted to enhance clinical awareness regarding the diagnosis and treatment of this condition.

TAKE HOME MESSAGES

- This case highlights a rare case of bilateral coronary artery-right ventricular fistula with multiple imaging tests and interventional procedures.
- A systematic review summarized this group of cases worldwide, provide some useful experiences for rare diseases.

HISTORY OF PRESENTATION

A 20-year-old male patient was admitted to the hospital due to exercise-induced shortness of breath. The patient did not report any chest pain, cyanosis, or other symptoms. Upon admission, the patient's blood pressure was measured at 124/56 mmHg (1 mmHg = 0.133 kPa) and heart rate at 77 bpm. Physical examination revealed loud and persistent murmurs between the 3rd and 4th intercostal space adjacent to the sternum.

PAST MEDICAL HISTORY

The patient had no relevant medical history.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis included patent ductus arteriosus, aortic valve stenosis with regurgitation, Kawasaki disease, and Pneumonic dyspnea disease.

INVESTIGATIONS

The electrocardiogram showed sinus rhythm and no abnormal waveforms. Transthoracic Echocardiography (TTE) demonstrated normal anatomical alignment, biventricular size and function, and valvular anatomy. Color and spectral Doppler showed the presence of multiple high-speed (Fig. 1C) and two-phase abnormal blood flow bundles in the RV. These bundles measured approximately 5mm, with a peak velocity of 2.58 m/s and a peak pressure difference of 27mmHg. The left ventricular systolic function was within the normal range (EF = 62%), indicating the presence of a fistula between the RCA and the LAD that extended into the RV (Fig. 1B). Coronary angiography (CAG) showed that the RCA was small, while the left main artery (LMA), LAD, and left circumflex artery (LCX) were not narrowed. The CAF appeared tortuous and thick, and the left coronary artery fistula (LCAF) communicated with the right coronary artery fistula (RCAF)(Fig. 3A). Coronary computed tomography angiography (CCTA) revealed that the lumen of the LCAF and RCAF was thickened and tortuous, with the widest point measuring approximately 12mm. After the distal-end confluence, it communicated with the RV (Fig. 2B).

MANAGEMENT

On September 17, 2023, a CAF interventional procedure was performed under local anesthesia. During the procedure, the right femoral vein (RFV) were punctured, The Qp/Qs was 2.01 as measured by right heart catheterization(**Table 1**).The right femoral artery (RFA) was punctured, and a 6F-pigtail catheter was advanced from the RFA to the ascending aorta (AAO). Non-selective CAG revealed an abnormal shunt of the LCAF and RCAF to the RV, with the fistula being tortuous. No nourishing blood vessel was identified throughout the procedure. The largest diameter of the LCAF was approximately 12mm, with the narrowest diameter measuring about 5mm. The largest diameter of the RCAF was approximately 14mm, and the smallest diameter was about 5mm. Selective CAG using a 6F JL4.0 catheter and a 6F JR4.0 catheter demonstrated that the LCA and RCA initiation and distribution were normal. Compression of the middle of the RCA by the RCAF was observed to result in 50% stenosis, while no significant stenosis or dilation was found in other vessels. A 7F JL3.5 guide catheter was used to create a pathway through the RFA-DAO-AAO-LCAF, and a 7mm AMPLATZER ^{TM} II Vascular Plug (AGA Medical Corporation, USA) was inserted into the distal end of the fistula through the guide catheter. Angiography confirmed complete occlusion without any residual shunt and no impact on coronary blood supply. The vascular plug was securely fixed under TTE and X-ray monitoring, and then released. The pathway was established through the RFA-DAO-AAO-RCAF using a 6F JR3.5 guide catheter, but the catheter could not reach the distal end of the fistula. The reason was that the RCAF was too tortuous, resulting in insufficient support for the catheter. Subsequently, a 5F MPA1 catheter was inserted, and a 7mm AMPLATZER ^{TM} IV Vascular Plug (AGA Medical Corporation, USA) was released at the proximal end of the fistula, and the subsequently released vascular plug followed the blood flow to the narrowest segment distal to the fistula. Angiography revealed incomplete closure and residual shunt. Another 7mm AMPLATZER ^{TM} IV Vascular Plug was then released at the proximal end of the fistula. The released vascular plug did not move too much with the blood flow because the blood flow became slower after the previous blockage. Angiography confirmed complete blockage of the plug, with no residual shunt and no impact on vascular blood supply. The vascular plug was securely fixed under TTE and X-ray monitoring. AAO angiography showed that all three vascular plugs were in good position, and no significant residual shunt was observed (Fig. 3C).

After the interventional therapy, the cardiac murmur disappeared upon auscultation and symptoms related to dyspnea disappeared. Subsequent examination of TTE confirmed the proper position of the vascular plug. CDFI showed no noticeable shunt signal. The patient was prescribed dual antiplatelet drugs, including aspirin (100 mg, once daily) and clopidogrel (75 mg, once daily). The patient's condition remained stable during the 1-month, 3-month, and 6-month follow-up periods, with no medication or medical device complications.

A CCTA at 6-month after the treatment revealed optimal remodeling of the RCAF and suboptimal remodeling of the LCAF. We intend to continue antiplatelet therapy with regular CCTA follow-up every 1–2 years until optimal remodeling of the LCAF has occurred. Should any hemodynamic changes or evidence of sustained coronary artery dilation emerge during the observation period, it then needs to be adapted to long-term anticoagulation and subacute bacterial endocarditis prophylaxis, with surgical treatment if necessary, therefore, long-term outcome needs to be confirmed by further follow-up.

DISCUSSION

CAF is a rare anomaly of the coronary arteries, which can be present at birth or acquired later in life. It can originate from any of the three main coronary arteries and the drainage tube can enter any cardiac cavity or great vessels. CAF accounts for approximately 0.27–0.40% of all congenital heart defects. [1] Based on the number of fistulas, single CAF accounts for over 90% of all cases, making it much more common than bilateral CAF, which accounts for only 10% of cases. CAF occurs in approximately 50% of patients with the RCA, 42% of patients with the LCA, and 5% of patients with both. [2] Small and medium vessels are more common, while large cases are rare. [3] The patient in this case has bilateral huge CAF, which is even rarer. A thorough search was conducted on PubMed for cases of this disease reported between 1974 and 2020, and only 32 cases were found. Clinically, CAF can present with symptoms such as palpitations, shortness of breath, angina pectoris, and heart failure, or it can be completely asymptomatic. Continuous murmurs can also be detected incidentally, with typical murmurs heard above the tip of the sternum and along the left border of the sternum. Based on our analysis of 32 cases of bilateral CAF (**Table 2**), [1,8–38] the most common type was bilateral coronary artery-pulmonary artery fistula, accounting for 28 cases (87%). Among these cases,18 (56%) originated from the LAD and RCA. In 26 cases (81%), the fistulas terminated separately, while in only 6 cases (18%), they terminated together. CAG remained the preferred diagnostic method for most cases. The main symptoms reported were chest pain in 12 cases (37%), dyspnea and tachypnea in 8 cases (25%), angina in 5 cases (15%), and no clinical symptoms in 7 cases (22%). Physical examination revealed persistent murmurs in the precordial area in 16 cases (50%). Overall, 17 patients (53%) opted for conservative treatment, 12 patients (37%) underwent surgery, and only 3 patients (9%) received interventional therapy.

CAFs were classified as proximal or distal based on the site of CAF origin from the conduit coronary artery [4] (in this paper, the two coronary arteries were proximal CAF (Fig. 3B)), and based on this anatomical classification and post-treatment remodeling characteristics to guide the management of CAF and follow up, to facilitate the clinical practice is adopted in this paper. The diagnosis and treatment of bilateral CAF with a common exit has a lot of uniqueness. Regarding interventional indications, the hemodynamic alterations of the CAF opening in the right ventricle in this case are analogous to those observed in congenital heart disease with left-to-right shunting. The LCAF and RCAF were > 2 times the largest diameter of the coronary vessel not feeding the fistula. The Qp/Qs value was 2.01, which can explain the patient's clinical manifestation of obvious dyspnea after activity. Coronary CTA and angiography demonstrate that the fistulas do not emit branching nourishing blood vessels, which aligns with the indications proposed by most scholars for the closure of coronary artery fistulas. [5,6] Closure can be performed either percutaneously or surgically in treatment centers with the experienced expertise. In this case, based on the patient's preference and our center's extensive experience in percutaneous treatment of coronary fistulas, an interventional treatment was initially attempted, with satisfactory results.

Some risks and challenges are associated with interventional procedures that differ from those associated with single or two different opening coronary artery fistulas. In the case of a single or two-variable-opening coronary artery fistula, three routes of interventional occlusion have been demonstrated to be effective: downstream occlusion of the fistula opening via the venous route, retrograde occlusion of the narrowest part of the fistula through the arterial route, and occlusion through the arterial and venous routes by creating a circumferential orbit. There is not a lot of previous experience to draw on for this case. In terms of blocking strategy, should the site of blockage be the narrowest part of the two fistulas separately? Or should the common trunk be blocked? If the fistula is closed separately, what is the sequence of closure? Is it possible that one fistula can be successfully closed while the other still requires surgical treatment? Considering the need to puncture the femoral artery for selective coronary angiography regardless of the specific decision, we first tried the femoral route to close the fistula. During the operation, both coronary fistulas were extremely tortuous, with poor maneuverability of the guidewire after reaching the distal end, and due to the interference of the special anatomical structure of the distal common trunk, the guidewire was made to enter the contralateral fistula after reaching the distal end of CAF, and the right ventricle could not be entered. This ruled out the strategy of establishing a circumferential orbit for blockage by the arterial and venous routes. Given the imaging results, which indicated that the common trunk was relatively short with no stenosis that could be blocked and that there was a high risk of blocker dislodgement from the venous route for blocking the common trunk, the strategy of blocking the narrowest part of the single coronary fistula via the arterial route was selected. Because the LCAF has a longer and more tortuous course than RCAF and is expected to be more difficult to block, the left fistula is blocked first, and if blocking is unsuccessful, the patient is advised to choose surgical treatment. The LCAF catheter was able to reach the distal end of the fistula for distal occlusion, while the RCAF was unable to reach the distal end of the fistula for proximal occlusion. Unintentionally self-contrast results of different blockage closure sites appearing in the same patient. A CCTA at 6 months post-treatment demonstrated optimal remodeling of the RCAF.[4] The demonstration of completion of the fistula segment by thrombus from the point of occlusion without invasion of the proximal epicardial artery (Fig. 1D), suboptimal remodeling of the LCAF, partial thrombosis creating residual cul-de-sac in a proximal fistula segment (Fig. 2D). Unexpectedly confirms the view that the closure of CAF of proximal origin near the point of origin would be more appropriate. [4]

In terms of complications and prognosis, bilateral fistula openings are complex to maneuver and take significantly longer to operate than the simple lesions described above. Operative time has always been an independent risk factor for operative risk and therefore should be performed by experienced operators in centers. Minimizing the operative time may reduce the risk of complications arising from operations such as potential coronary artery spasm, coronary artery entrapment, perforation, and embolization. In cases of bilateral coronary artery fistulae, postoperative attention should be paid to the possibility of bilateral coronary artery thrombosis with proximal extension and persistent coronary artery dilatation, which would be more catastrophic than coronary artery fistulae from a single supplying artery. This complication may occur regardless of whether percutaneous or surgical treatment is employed. However, there is no standardized postoperative medication to which to refer, and the principal medications applied are anticoagulant or antiplatelet. [6,7] Considering

the evidence-based evidence on the use of antithrombotic drugs after many percutaneous coronary stenting procedures, therefore, we chose a dual antiplatelet therapy strategy of aspirin and clopidogrel to avoid acute massive thrombotic events.

In conclusion, the interventional occlusion of bilateral coronary artery fistulas with a common exit presents a unique set of risks and challenges that differ from those encountered in the interventional treatment of single or two different coronary artery fistulas. The experience of successful cases of occlusion can provide a reference point for subsequent percutaneous close of such rare cases in terms of indications for the procedure, interventional technique, complications, and prognosis.

CONCLUSIONS

The rare case of bilateral coronary arteriovenous fistula with common opening has some unique features in terms of the percutaneous closure indications, interventional techniques, complications, postoperative management and follow-up, and long-term anatomical monitoring of the coronary arteriovenous fistula after occlusion and adjustment of the treatment strategy as appropriate is necessary.

Abbreviations

CAF = coronary artery fistula

LCAF = left coronary artery fistula

RCAF = right coronary artery fistula

TTE = transthoracic echocardiography

CAG = coronary angiography

CCTA = coronary computed tomography angiography

MRA = magnetic resonance angiography

Declarations

This study was conducted in accordance with the principles of the Declaration of Helsinki and was approved by the Ethics Committee of Hainan Affiliated Hospital of Hainan Medical University. Written informed consent was obtained from all participants or their legal guardians before inclusion in the study. Written informed consent for publication of the study data and accompanying clinical information was also obtained from the participants or their legal guardians.

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All authors declare no conflict of interest.

Author Contribution

Wei-song Tang wrote the main manuscript text and prepared all figures. All authors reviewed the manuscript.

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Tables

(Table 1) The hemodynamic parameters of cardiac catheterization

Gender	**Age**	**Height (cm)**		**Weight (Kg)**	
male	20	156		52	
-	**Right atrium**	**Right ventricle**	**Left ventricular**	**Main Pulmonary artery**	**Descending aorta**
Systolic blood pressure (mmHg)	7	26	112	22	124
Diastolic blood pressure (mmHg)	3	9	32	11	63
Mean arterial pressure (mmHg)	4	15	58	15	83
Oxygen saturation (%)	73	-	-	87	100
Oxygen Saturation of Mixed Venose Blood (%)		72	**Hemoglobin concentration (g/L)**		132
Heart rate (bpm)	82	**Qp/Qs**		2.01	-
Body surface area (m^2)	1.59	**Left to right shunt (L/min)**		4.69	-
Oxygen consumption (ml/min)	238.5	**Right to Left shunt (L/min)**		0	**Corrected for body surface area**
Cardiac index [L/(min*m2)]	2.90	**Total pulmonary vascular resistance (Wood)**		1.61	2.56
Pulmonary blood flow (L/min)	9.30	**Pulmonary arterial resistance (Wood)**		1.61	2.56
Circulating blood flow (L/min)	4.61	**Systemic vascular resistance (Wood)**		17.12	27.23
Effective pulmonary blood flow (L/min)	4.60	**Rp/Rs**		0.09	-

(Table 2) Data from 32 Cases of Bilateral CAF

Year	Cnt	Sex	Age	Type	Origin	Route	Termination	Symptoms	Inspection	Tre
1974	JP	M	38y	Bilateral CA-PA Fistula	LCX/RCA	Tortuosity	Separate	Persistent murmur	CAG	Co tre
1981	L	M	38y	Bilateral CA-PA Fistula	LAD/LCX/RCA	Tortuosit	Separate	Chest pain	CAG	Co art lig
1998	JP	F	49y	Bilateral CA-PA/RA/GCV Fistula	LAD/CB	Tortuosity	Separate	Persistent murmur	CAG	Co tre
2002	L	M	52y	Bilateral CA-PA Fistula	LCA/RCA	Tortuosit	Separate	Palpitation/Dizziness/Persistent murmur	CAG	Co tre
2006	TR	M	58y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Common	Chest pain/Angina/Persistent murmur	CAG	Co tre
2007	NL	M	39y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Angina/Persistent murmur	CAG	Co art lig
2007	UK	-	40y	Bilateral CA-RA Fistula	LCA/RCA	Tortuosity/Huge	Separate	Palpitation/Persistent murmur	MRA	Co tre
2007	TR	F	60y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Chest pain/Palpitation/Dyspnea/Persistent murmur	CAG	Co art lig
2008	KR	F	75y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Separate	Precordial distress	CAG	Co art lig
2008	FR	M	57y	Bilateral CA-PA Fistula	LAD/RCA	-	Separate	Chest pain	CAG	CA
2008	GR	M	51y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Common	Chest pain/Tachypnea/Nausea	CAG	Co tre
2008	KR	-	64 y	Bilateral CA-RA Fistula	LAD/RCA	Tortuosit	Separate	Dyspnea/Precordial distress	CAG	Co tre
2009	BR	F	49y	Bilateral CA-PA Fistula	LCA/LAD	Tortuosity/Huge	Separate	Persistent murmur	CTA	Co tre
2009	FR	M	58y	Bilateral CA-PA Fistula	LSN/RCA	Tortuosit	Separate	Chest pain	MDCT	Co em
2009	TR	M	55y	Bilateral CA-PA Fistula	LCX/RCA	Tortuosit	Separate	Chest pain	CAG	Co tre
2010	IN	M	45y	Bilateral CA-PA Fistula	LCX/PD	Tortuosity	Common	Angin	CAG	Co tre
2010	AU	F	46y	Bilateral CA-PA Fistula	LCA/RCA	Tortuosity	Separate	Angina/Persistent murmur	CAG	Co art lig
2011	AT	M	34y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate		CAG	Co tre
2011	JP	M	58y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Precordial distress	CAG	CA
2012	JR	-	60y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Chest pain/Dyspnea/Persistent murmur	CAG	Co tre
2014	TR	M	33y	Bilateral CA-PA Fistula	LCX/RCA	Tortuosit	Separate	Angina	CAG	Co em

2014	UK	F	76y	Bilateral CA-LV Fistula	LCA/RCA	Tortuosit	Separate	Chest pain/Persistent murmur	CAG	Co tre
2014	IN	M	65y	Bilateral CA-PA/LV/RV Fistula	LAD/RCA	Tortuosit	Separate	Tachypnea	CAG	Co tre
2015	JP	-	60y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Common	Persistent murmur	CAG	Co art lig
2015	RS	M	23y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Separate	-	CAG	Co art lig
2016	PT	F	70y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Chest pain/Weakness/Dyspnea	CAG	Co tre
2016	SK	F	55y	Bilateral CA-PA Fistula	LCX/RCA	Tortuosit	Separate	Angina/Tachypnea	CAG	Co art lig
2016	JP	M	37w	Bilateral CA-PA Fistula	LCA/RCA	-	Separate	Persistent murmur	CAG	CA
2017	BR	F	59y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity/Huge	Common	Persistent murmur	CAG	Co tre
2017	US	M	71y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Separate	Chest pain	CAG	Co tre
2019	IN	F	76y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosit	Separate	Weakness/Chest pain/Dyspnea/Persistent murmur	CAG	Co art lig
2020	JP	F	52y	Bilateral CA-PA Fistula	LAD/RCA	Tortuosity	Common	Migraine/Persistent murmur	CAG	Co em

Notes: Cnt., Country in ISO country code (JP: Japan, IL: Israel, TR: Turkey, NL: Netherland, UK: United Kingdom, KR: Croatia, IN: India, FR: French, GR: Greece, AU: Australia, BR: Brazil, US: United States, AT: Austria, RS: Serbia, PT: Portugal, SK: Slovakia); Age in, y (years), w (weeks); Sex, F= female, M= male; Type, CA=Coronary Artery, PA=Pulmonary Artery, RA=Right Atrium, LA=Left Atrium, RV=Right Ventricle, LV=Left Ventricle; Origin: CB=Right Conus Branch, LSN=Left Sinus Branch, PD=Posterior Descending Artery; Treatment: CABG=Coronary Artery Bypass Grafting, CSI=Coronary stent.

Figures

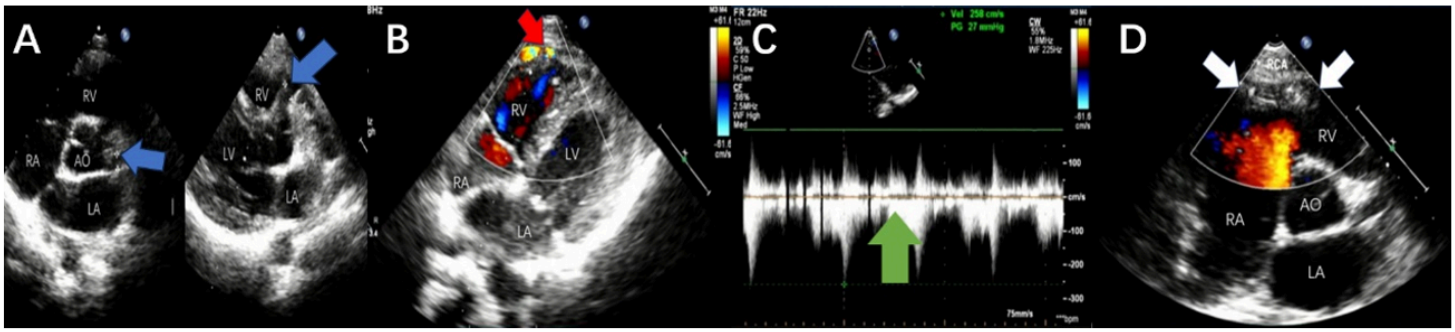


Figure 1

CAF Pre-Closure and Post-Closure TTE

(A) Parasternal long-axis view showing dilated coronary sinus **(blue arrow)**. **(B)** Right ventricle inflow view showing high velocity turbulent flow **(red arrow)**from the coronary sinus into right ventricle. **(C)** Continuous-wave Doppler showing continuous high-velocity flow throughout the cardiac cycle from the coronary sinus into right ventricle **(green arrow)**. **(D)** Post-Closure TTE showing occluders **(white arrow)**, no blood flow signal was observed around the occluders, indicating complete occlusion. AO = ascending aorta; LA = left atrium; LV = left ventricle; RA = right atrium; RV = right ventricle.

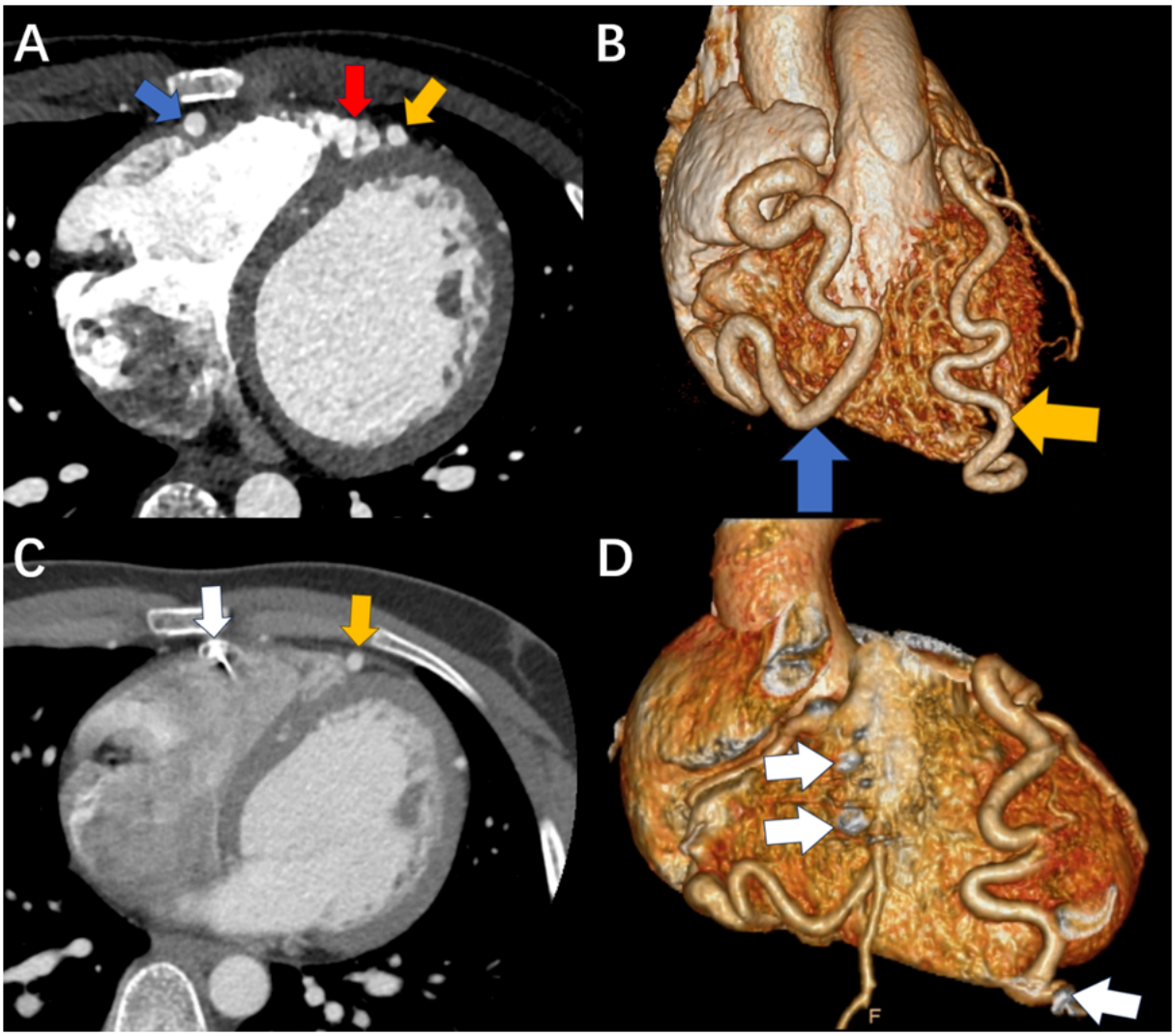


Figure 2

Comparison of preoperative and postoperative CCTA

(A and B) The right coronary artery fistula **(blue arrow)**, The left coronary artery fistula **(yellow arrow)**, common exit into RV **(red arrow)**. **(C)** Distal occluder in RCAF **(white arrow)** and LCAF **(yellow arrow)**. **(D)** Three occluders (white arrows). The imaging revealed complete occlusion of the fistula segment of RCAF by thrombus from the point of closure, with no invasion of the proximal epicardial artery. Suboptimal remodeling of the LCAF, with partial thrombosis creating residual cul-de-sac in a proximal fistula segment.

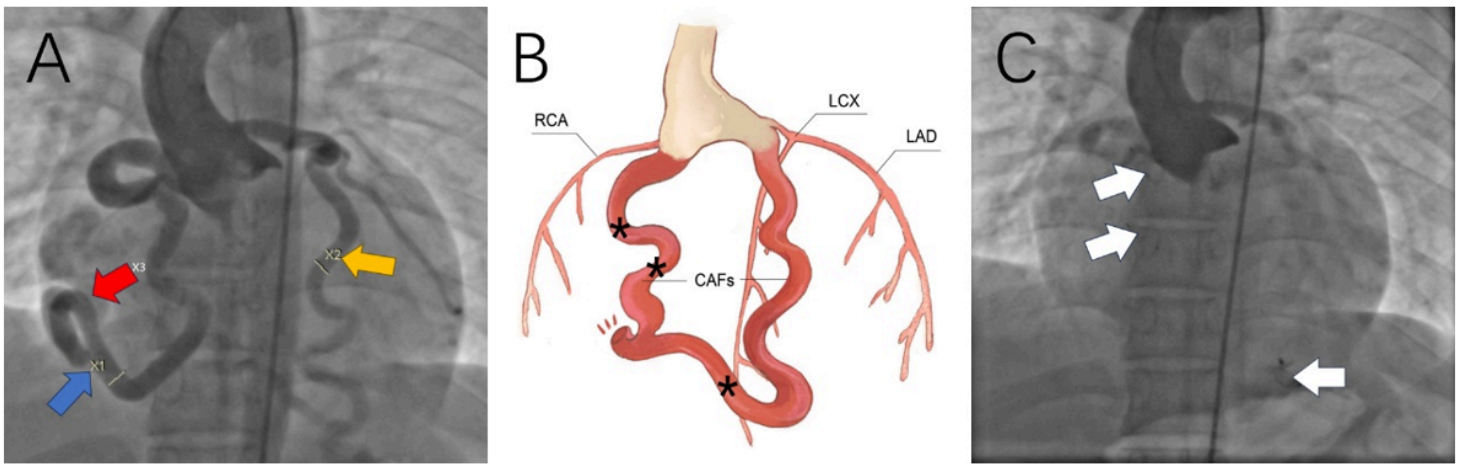


Figure 3

Coronary Angiography During CAF Closure

(A and C) Contrast injection into the coronary system before and after CAF device closure. **(A)** Preoperative angiography imaging shows bilateral CAF originating from the left coronary sinus **(yellow arrow)** and right coronary sinus **(blue arrow)** with a common breach at the RV **(red arrow)**. **(B)** Anatomy of a coronary artery fistula"*" shows three occluders. **(C)** Postoperative angiography imaging shows three occluders**(white arrows)** visualized with no blood filling or residual shunt in the fistula, and the fistula is completely blocked.