CASE REPORT

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A Complicated Case Report of Coronary Artery Fistula

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ABSTRACT

Background: Coronary artery fistulas (CAFs), also, known as coronary arteriovenous malformation, are aberrant connections between coronary arteries and other structures, such as other artery branches or heart chambers. CAFs are infrequent and asymptomatic in young patients, but symptoms and complications become more frequent with age. CAFs can affect hemodynamic parameters and lead to complications, such as myocardial ischemia, heart failure, arrhythmia, and infective endocarditis. **Objective:** The aim of this article was to present a typical CAF case with severe symptoms who underwent successful embolization to resolve their symptoms. **Case presentation:** A 50-year-old Vietnamese male visited our cardiac outpatient clinic (S.I.S General Hospital, Can Tho, Vietnam) because of exertional dyspnea and chest pain. Signs of congestive heart failure and abnormal murmur were not presented on chest auscultation. Diagnostic digital subtraction angiography was performed to determine the detailed angioarchitecture of the CAF, revealing a fistulous connection between the left anterior descending artery (LAD) and the LV chamber through an aneurysm. In addition, the RCA measured 7 mm in diameter with a fistula (16 × 9 mm) draining into an aneurysm and then terminating into the LV chamber. The patient had an RCA aneurysm with a fistula into the LV. It was treated successfully by closing the fistula with a vascular plug. Access to the fistula was complex and difficult because of complications due to the CAF. After the procedure, the patient had no chest pain or shortness of breath and was discharged after three days. After six months, he was taking dual antiplatelet therapy and antihypertensive medications and felt better. We performed contrast computed tomography (CT) to examine the fistula after a year, which showed the successful closure of the fistula without any relevant alteration in the coronary artery. **Conclusion:** CAF closure is indicated if patients have symptoms or secondary complications, and percutaneous closure is a safe and effective method to manage CAF. A CAF is rare and does not have specific symptoms, making it difficult to diagnose. Most patients are asymptomatic and have serious recent complications. Currently, the percutaneous transcatheter method is popular because it is noninvasive and successful in most patients.

Keywords: Coronary artery fistulas, computed tomographic angiography, percutaneous transcatheter closure.

1. BACKGROUND

A coronary artery fistula (CAF) or coronary arteriovenous fistula is an acquired abnormal connection of coronary arteries with cardiac chambers or any segment of the systemic or pulmonary circulation (1-4). In adulthood, symptoms range from mild dyspnea to ischemic events or heart failure, which are related to the left-right shunt (5). Computed tomographic angiography (CTA) with 3D reconstruction can be used to accurately assess the complex anatomy of CAFs: the site and the number of origins, drainage sites, and the associated anomalies (6). Percutaneous transcatheter closure is indicated in patients whose anatomy is favorable for the procedure.

2. OBJECTIVE

We present a CAF case with complicated anatomy and fistula size who was managed successfully using this method.

3. CASE PRESENTATION

A 50-year-old Vietnamese male visited our cardiac outpatient clinic (S.I.S General Hospital, Can Tho, Vietnam) because of exertional dyspnea and chest pain. His past medical history was significant for hypertension and smoking.

His family history was unrevealing. On examination, his blood pressure was 150/70 mmHg, heart rate was regular (about 63 bpm), oxygen saturation was 96% on room air, and respiratory rate was 22 times/minute. Signs of congestive heart failure and abnormal murmur were not presented on chest auscultation. The electrocardiogram showed a sinus rhythm without pathological Q waves, ST segment, or T wave changes. Cardiac biomarkers were negative for myocardial damage. Both his complete blood count and biochemical blood test were normal. The echocardiogram showed normal left ventricular function with an ejection fraction of 62%, a dilated right coronary artery (RCA; about 7.8 mm in diameter), a normal left coronary artery (3.4 mm in diameter), and an abnormal shunt into the left ventricle (LV) near the base of the mitral valve. CTA showed an ostial RCA diameter of 8 mm and a coronary fistula originating from the terminal RCA and draining into the LV chamber.

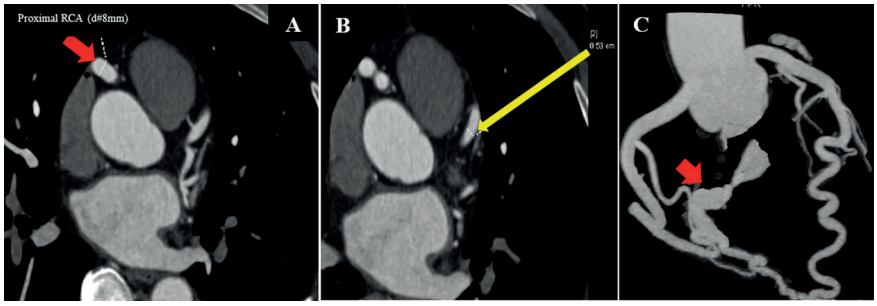

Figure 1. Preoperative CTA: (A) The diameter of the proximal RCA was about 8 mm (red arrow). (B) The diameter of the proximal LAD was about 5 mm (yellow arrow). (C) The connection between the LAD and RCA, fistula from RCA to LV (red arrow). Key: RCA, right coronary artery; LAD, left anterior descending artery; LV, left ventricle.

Diagnostic digital subtraction angiography was performed to determine the detailed angioarchitecture of the CAF, revealing a fistulous connection between the left anterior descending artery (LAD) and the LV chamber through an aneurysm. In addition, the RCA measured 7 mm in diameter with a fistula (16 × 9 mm) draining into an aneurysm and then terminating into the LV chamber. We did not discover any atherosclerosis disease (Figure 1).

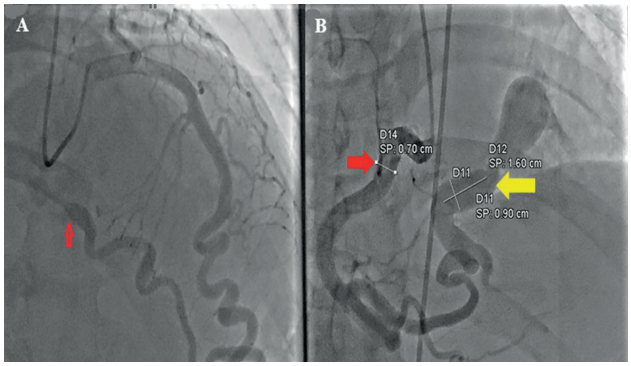

Figure 2. Coronary angiogram: (A) The diameter of the proximal RCA was 7 mm (red arrow). (B) The RCA contained an aneurysm (16 × 9 mm) anterior to the fistula into the LV (yellow arrow).

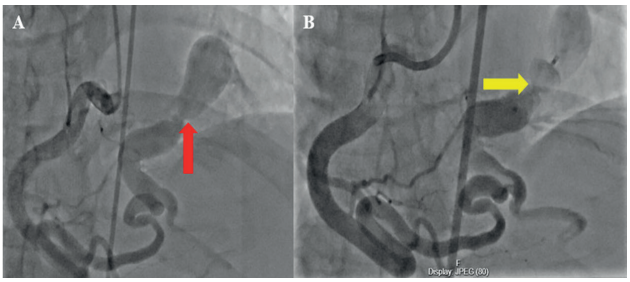

Figure 3. Percutaneous transcatheter closure: (A) The connection between the RCA and LV before the procedure (red arrow). (B) The site of the plug after the procedure (yellow arrow).

The patient had an RCA aneurysm with a fistula into the LV. It was treated successfully by closing the fistula with a vascular plug. Access to the fistula was complex and difficult because of complications due to the CAF. The diameter of the entire RCA was about 7 mm, and an aneurysm (16 × 9 mm) was present in the distal RCA (Figure 2). The RCA was also long and tortuous. Therefore, we created an arteriovenous (AV) wire loop to provide a stable and solid path during device delivery. The ASAHI Hyperion AL 0.75 7F catheter (Asahi Intecc) was inserted into the RCA. We chose the ASAHI CHIKAI 300 cm wire (Asahi Intecc) with the Merit Maestro Microcatheter (Merit Medical) to approach the distal RCA through the fistula into the LV and then the ascending aorta. Then, we inserted the Guide Wire M 150m and ASAHI Hyperion JR 4.0 6F catheter into the right femoral artery to the ascending aorta, using the snare to catch the wire and pull the artery out to the right femoral artery. The 5F SteerEase Introducer (LifeTech Scientific) was guided through the right femoral artery to the aneurysm in the distal RCA. We placed the Lifetech Konar-MF VSD Occluder 6/4 mm (LifeTech Scientific) at the narrowest point of the aneurysm in the distal RCA and before the point draining into the LV chamber. The angiogram recorded the Occluder in the correct position, and the flow through the RCA fistula was reduced. We finished the procedure (Figure 3).

After the procedure, the patient had no chest pain or shortness of breath and was discharged after three days. After six months, he was taking dual antiplatelet therapy and antihypertensive medications and felt better. We performed contrast computed tomography (CT) to examine the fistula after a year, which showed the successful closure of the fistula without any relevant alteration in the coronary artery.

4. DISCUSSION

Several studies have reported that CAFs are uncommon, affecting <1% of the general population (3). Most fistulas are asymptomatic for a long time and can remain during an individual's lifetime. Therefore, most patients

are asymptomatic and diagnosed incidentally through physical examination or for other reasons unrelated to the condition. However, some patients present with signs and symptoms of its complications: congestive heart failure, myocardial ischemia, myocardial infarction, arrhythmias, hemopericardium, cardiac tamponade, endocarditis/endarteritis, pulmonary hypertension, and other cardiopulmonary functional abnormalities (2). We presented a patient with symptomatic CAF when he was mature. He had coronary artery disease symptoms and risk factors (hypertension and smoking). Our previous case report detailed a 35-year-old woman who developed symptoms after her second pregnancy, which made us misunderstand the other disease relating to peripartum cardiomyopathy (7). In all the above cases, CAFs are difficult to diagnose and can be missed. Many signs and symptoms are associated with CAFs, such as chest pain, dyspnea with exertion, and fatigue, all equivocal and nonspecific (5). Chest pain is the most common symptom. Lim et al. showed that angina pectoris occurred in 57% of cases, and the other symptoms included congestive heart failure and pulmonary hypertension (6).

While our patient's electrocardiogram showed sinus rhythm without pathological Q waves, ST segment, or T wave changes, our previous case had tachycardia and block A-V type 1 (7). Generally, electrocardiograms are unhelpful and are used to differentially diagnose other conditions, such as myocardial ischemia, myocardial infarction, and arrhythmias (5). Our cases were discovered by transthoracic echocardiogram. There are many signs in echocardiograms to suggest CAFs. The direct signs are dilation of the coronary artery, aneurysm, and fistula into other structures. In our case, the RCA was dilated to about 7.8 mm, the left coronary artery measured 3.4 mm, and there was an abnormal shunt into the LV near the base of the mitral valve. Our previous case also had the same result: the proximal RCA was dilated to 18.6 mm and accompanied by an aneurysm in the middle RCA (30 mm) and a two-way fistula into the right atrium; the left-main coronary artery was normal with a diameter of about 3 mm (7). Indirect signs in echocardiograms associated with changing heart structure include the ejection fraction, dilated LV, mitral valve regurgitation by annulus dilatation, tricuspid valve regurgitation, and pulmonary arterial pressure increasing (7). Many noninvasive techniques exist to diagnose and follow up CAFs, such as transthoracic echocardiogram, transesophageal echocardiogram, and magnetic resonance imaging. We can use methods to detect and evaluate the CAF's anatomy and hemodynamics. Two-dimensional and pulse Doppler echocardiography can be used to detect the coronary arteries: the dilation and turbulent flow in the fistula and the recipient chamber. Transesophageal echocardiography is used intraoperatively to identify the precise location of the fistula's drainage. Magnetic resonance imaging and multidetector CT have also become alternative methods to evaluate the anatomy, flow, and function of CAFs (5).

The gold standards to diagnose CAFs are cardiac catheterization and coronary angiography, which help to plan appropriate management (8). Cardiac contrast CT is a useful, noninvasive, and accurate imaging technique for detecting major coronary artery anomalies and may be performed as an alternative to coronary angiography in diagnosing (6). The CT scanning protocol assesses the anatomy of CAFs: origin, drainage site, and fistulous tracts (9). In a previous case, the fistula originated from the RCA, the most common origin site of CAFs, accounting for 50%–55% of cases (10). The origin in the remaining cases is the two coronary arteries (the LAD and RCA), called coronary-to-bronchial artery fistulas (9). The prevalence of coronary-to-bronchial artery fistulas in CTA is estimated to be 0.61%. The most commonly involved chamber is the right ventricle (41%), followed by the right atrium (26%) (11) and the left atrium and LV (3%–5%).

Coronary cameral fistulas are classified into arterioluminal (direct connection with the cardiac chamber) and arteriosinusoidal (indirect connection through a cardiac sinusoidal network) subtypes. In the arterioluminal subtype, the constant high blood flow through the coronary arteries may lead to their aneurysmal dilatation. A CAF into the left heart chamber (i.e., left-to-left shunt) led to a large left heart volume and an increased LV pressure in our previous case, with the last consequence being LV hypertrophy (12). In contrast, in another previous case, a CAF into the right heart chamber (i.e., left-to-right shunt) led to increased right heart volume, tricuspid valve regurgitation, and pulmonary artery hypertension (7). Most coronary fistulas are asymptomatic and detected incidentally. However, while large fistulas change hemodynamics, surgery or percutaneous procedures are required to treat the arterioluminal subtype successfully; the arteriosinusoidal type is treated conservatively (5).

The indications for treatment strategy depend on the fistula's size, morphology, and anatomy; the patient's age; and associated cardiac disorders, which are used to select suitable techniques for each individual (13). The options for managing CAFs include surgical ligation alone (with or without cardiopulmonary bypass) or accompanied by coronary artery bypass grafting and transcatheter closure (3, 7).

Based on the recommendation of Al-Hijji M et al., all our cases are suitable for percutaneous transcatheter treatment (3). This noninvasive method is preferred for older patients at risk of perioperative complications. The most suitable option for our patient was transcatheter closure by dual vascular access (right femoral artery and right femoral vein). The case reported here was difficult for us to perform closure in two situations. While the patient's fistula had two origins from the RCA and LAD, we only occluded from the RCA because it was hard to access it through the LAD. The RCA was long and tortuous. Therefore, we created an AV wire loop to provide a stable and solid path during device delivery. This strategy contributed to an increased success rate and reduced procedural time. In contrast, our previous case's fistula had only one origin from the RCA, so we approached

it from the RCA (7). After determining coronary artery and fistula anatomy, fistula size, and the location of the functional RCA, we released an instrument to plug two positions: the distal end of the fistula and the waist of the RCA after the functional coronary artery.

5. CONCLUSIION

A CAF is rare and does not have specific symptoms, making it difficult to diagnose. Most patients are asymptomatic and have serious recent complications. An echocardiogram is useful to suggest the disease, and a CT with 3D reconstruction confirms the diagnosis and assesses the complex anatomy of CAFs. This information is essential for planning treatment therapy. Currently, the percutaneous transcatheter method is popular because it is noninvasive and successful in most patients.

- **Patients Consent Form**: The authors certify that they have obtained all appropriate patient consent forms.
- **Ethics Approval and Consent to Participate**: Ethical approval was not necessary for the preparation of this article. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Informed consent for patient information to be published in this article was obtained.
- **Author's contribution**: Pham Thi Thao Trang and Nguyen Minh Duc contributed to write original draft. Do Nguyen Tin, Nguyen Manh Cuong, Tran Chi Cuong, and Mai Hoang Dil contributed to undergo percutaneous transcatheter closure procedure, collect and interpret the imaging. Pham Thi Thao Trang, Nguyen Manh Cuong, Nguyen Tran Tran, and Le Minh Thang made substantial contributions to collect patient data and clinical data analysis. All authors have read, revised, and approved the final published version of the manuscript. All authors were responsible for submission of our study for publication..
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