

# Dual-source CT in Diagnosis of Coronary Artery Fistula and its Value

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## Research article

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# Abstract

**Background:** Coronary artery fistula refers to the symptom that the coronary artery trunk or branch connects with the heart cavity or the pulmonary circulation blood vessels in a congenital or an acquired manner. As early detection and diagnosis of CAF was necessary for the prevention and management of late symptoms and complications, we are supposed to master the imaging performance of CAFs so as to make a correct diagnosis. This paper seeks to investigate the imaging manifestations and application value of CT diagnosis of coronary fistula and its application value.

**Methods:** To retrospectively analyze the clinical data and DSCT image data of 62 patients with coronary fistula, and summarize, analyze and summarize their image manifestations and types.

**Results:** 62 CAFs originate from 58 LCA vessels, 32 RCA vessels. The distribution frequency of drainage site from high to low was as follows: 40 cases (63%) of the main pulmonary artery, 10 cases (16%) of the right atrium, 7 cases (11%) of the right ventricle, 3 cases (5%) of the left ventricle, 1 case (2%) of the left atrium, and 1 case of coronary sinus (2%), 1 case of the right lower pulmonary vein (2%). There were 39 cases (63%) of single fistula vessel, and 23 cases (37%) of multiple fistula vessel, including 21 cases of left and right coronary arteries. Coronary artery-pulmonary fistulae were the most common with a total of 40 cases.

**Conclusion:** The popularization of CT improved the detection rate of CAFs. Coronary pulmonary-artery fistula was probably the most common seen type of CAFs. ECG-gating coronary artery CTA can accurately assess the origin of CAFs, the course of fistula vessels, and the drainage site, thereby providing important information for clinicians to treat coronary fistula.

**Trial registration:** retrospectively registered

## Background

Coronary artery fistula or coronary arteriovenous fistula (CAFs) refers to the symptom that the coronary artery trunk or branch connects with the heart cavity or the pulmonary circulation blood vessels in a congenital or an acquired manner, which is a rare coronary artery anomaly. In fact, the widespread application of ECG-gating DSCT in the recent years greatly improved the detection rate of CAFs. Although many cases of CAFs were asymptomatic, some cases of CAFs can display symptoms of ischemic heart disease and heart failure, so early detection and diagnosis of CAF was necessary for the prevention and management of late symptoms and complications. We ought to master the imaging performance of CAFs, especially the imaging performance of DSCT, so as to make a correct diagnosis. This article retrospectively, based on the clinical data and DSCT image data of 61 cases of CAFs, analyzed and summarized their image manifestations and types, and briefly summed up the pathophysiology and clinical manifestations of CAFs.

## Methods

# 1. Cases and clinical data (Patients)

We reviewed the DSCT examination records of cardiac large blood vessels and coronary arteries from January 2006 to July 2019. Since this study was retrospective, the ethics review agency of our hospital abandoned the requirement of informed consent. 62 CAFs were found, including 30 males and 32 females. The age range was 1–84 years old, with a median age of 43 years old. The causes and symptoms of the first MDCT diagnosis in 62 cases were as follows: 33 cases (53%) of cardiothoracic discomfort (including palpitations, chest pain, chest tightness, shortness of breath, and 16 cases (26%) accidentally found in other systemic diseases examination and preoperative examination, 7 cases (11%) of cardiac murmur, 4 cases (6%) of abnormal cardiac structure found in echocardiography, 1 case (2%) with exertional anhelation, lip and finger nail cyanosis, and 1 case (2%) of health examination. Among 62 cases, 13 cases were confirmed as CAFs by coronary angiography (CAG) and 1 case confirmed by CAG in the outpatient hospital; 4 cases were treated with coronary artery fistula closer with coronary angiography in our hospital with 3 cases successful treated and 1 case failed; 6 cases were successfully treated with coronary fistula correction in cardiac surgery of our hospital; 3 cases were followed up by regular visits and confirmed as CAFs in other hospitals.

## 2. Scanning equipments and inspection methods (CT Protocols)

The inspection equipments were Siemens Somatom Definition 64-layer dual-source CT and Siemens third-generation dual-source CT (Somatom Definition and Force; Siemens Healthcare, Forchheim, Germany). Patients were trained to breathe and hold their breaths before scanning so as to minimize breathing movements interference during scanning. Sedatives were applied to pediatric patients. Patients were instructed to take nitroglycerin to dilate the coronary arteries 3–5 minutes before the test. A coronary calcification score scan first and then a retrospective ECG-gating coronary artery enhanced scan were performed, ranging from the tracheal bifurcation to 1cm below the septum of the heart. Non-ionic contrast agent iodoparol was used (370mg/ml / ml, Shanghai Bracco Sine Pharmaceutical Co., Ltd.) with a total amount of 65–85 ml and a rate of 5.0–6.0 ml / sec, after injection finished an additional injection of 40 ml saline at the same rate was performed to wash the superior vena cava and right heart system. Bolus tracking technology was used with the area of interest set at the root of the ascending aorta and the scan was triggered when CT threshold reached 100HU (scanning started after a delay of 6–8 s). Scan parameters were as follows: collimation 0.6 mm, pitch 0.25 to 0.30, tube pressure 120 kV, tube current per rotation: 380 to 438mAs, rotation time: 0.33 s / week, check scan time: 6.5 ~ 9.8 s; reconstruction convolution kernel: kernelB30f, reconstruction layer thickness: 0.75 mm, reconstruction interval: 0.7 mm, reconstruction field of view (FOV): 146 mm, matrix: 512 × 512.

## 3. Image Reconstruction

After reconstruction, an optimal systolic and diastolic image were selected, and the obtained data was transferred to the workstation (Aquarius Ver. 4.4.6) for post-processing such as volume rendering (VR), multi-planar reconstruction (MPR), surface reconstruction (CPR), and maximum density projection (MIP), and then two senior cardiothoracic physicians evaluated the image quality and made a diagnosis, focusing on observing the origin of orificium fistulae, the course of fistula vessels, and the drainage position.

## Results

Among 62 CAFs, there were 91 fistula vessels involved, including 58 LCA-originated vessels (64%), 32 RCA-originated vessels (35%), and 1 descending aorta-originated vessel (1%). The specific origin distribution frequency of fistulas vessels was as follows: 44 left anterior descending branches (48%), 32 right coronary arteries (35%), 7 main branches (8%), and 6 left circumflex branches (7%), 1 diagonal branch (1%), 1 descending aorta (1%). There were 63 drainage sites, and the distribution frequency was as follows: 40 cases (63%) with the main pulmonary artery, 10 cases (16%) with right atrium, 7 cases (11%) with right ventricle, and 3 cases with left ventricle (5%), 1 case with the left atrium (2%), 1 case with the coronary sinus (2%), and 1 case with the right lower pulmonary vein (2%). Among 62 cases of CAFs, 39 cases (63%) were related to single fistula vessel, 23 cases (37%) related to multiple fistula vessels, including 21 cases simultaneously with left and right coronary arteries. The specific types of CAFs are shown in Table 1. The main cardiovascular and cardiac diseases, symptoms, and complications associated with 62 CAFs are shown in Table 2.

Table 1

<b>Origin</b>	<b>Orificium fistulae</b>	<b>Case/Total</b>
Left anterior descending branch (LAD)	Main pulmonary artery	18/62
Right corus artery + LAD	Main pulmonary artery	14/62
Right coronary artery + LAD	Right ventricle	3/62
Left main coronary artery (LM)	Right atrium	3/62
Right coronary artery	Right atrium	3/62
Right coronary artery	Right ventricle	2/62
Left circumflex branch	Right atrium	2/62
LM + LAD + right corus artery	Main pulmonary artery	2/62
Right corus artery	Main pulmonary artery	2/62
Left circuflex branch(LCX)	Coronary sinus	1/62
Left anterior descending branch	Right ventricle	1/62
Left diagonal branch	Main pulmonary artery	1/62
Left circumflex branch	Right lower pulmonary vein	1/62
Right coronary artery + left coronary artery	Right atrium + right ventricle	1/62
Left anterior descending branch(LAD)	Left ventricle	1/62
Right coronary artery + LAD	Left ventricle	1/62
Descending aorta + right corus artery + LAD	Main pulmonary artery	1/62
Right coronary artery sinoatrial node branch	Right atrium	1/62
LM + LAD + LCX	Main pulmonary artery	1/62
Right coronary artery	Left ventricle	1/62
LM + LAD	Main pulmonary artery	1/62
LCX	Left atrium	1/62

Table 2

<b>Major cardiology and cardiac diseases, complications, and auxiliary examination abnormalities</b>	<b>Case/Total</b>
CAF vessel ectasia/aneurysm formation	21/62
Coronary atherosclerosis	18/62
Myocardial bridge	10/62
Pulmonary hypertension	5/62
Coronary ectopic origin	5/62
Electrocardiogram abnormalities (ST segment depression)	4/62
Patent ductus arteriosus	3/62
Atrial fibrillation	3/62
Infective endocarditis	2/62
Double superior vena cava malformation	2/62
Pulmonary artery bicuspid valve and insufficiency	2/62
ECG suggests anterior myocardial ischemia	1/62
Atrial septal defect	1/62
Tetralogy of Fallot	1/62
Tracheal bronchus	1/62
Patent foramen ovale	1/62
Rheumatic heart disease with valvular disease	1/62
Arrhythmia (atrial premature beats)	1/62
Rupture of left anterior cerebral artery pseudoaneurysm with hemorrhagic cerebral infarction	1/62
Acute myocardial infarction (anterior descending subtotal closure)	1/62
Coronary artery fistula after coronary stent placement	1/62
Active secondary tuberculosis	1/62
Left lung small cell lung cancer	1/62

## Discussion

### 1. Causes, pathophysiology and clinical manifestations of CAFs

CAFs were either congenital or acquired (1). More than 90% of CAFs were congenital or sporadic (2). The embryological mechanism of congenital CAF in the heart cavity was the degeneration failure of the peritrabecular sinus system in the myocardium (3). Acquired CAFs were often iatrogenic complications (4–7) of deceleration accidents, percutaneous transluminal coronary angioplasty, endomyocardial biopsy, implantation of permanent ventricular pacing leads, coronary artery bypass graft surgery or stent placement, mitral valve replacement, septal muscle resection or acute myocardial infarction, trauma, chest irradiation and etc. Among 62 CAFs, a 59-year-old male patient with coronary heart disease who underwent coronary stent implantation in 2012 was found no CAFs during CT follow-up in 2015, while he was diagnosed of left circumflex coronary artery fistula to right ventricle by DSCT when re-admitted to hospital due to chest pain in 2017. It was suggested that the CAFs of this patient was secondary.

The basic pathological mechanism of CAFs was that high-pressure blood flow in normal coronary arteries bypassed myocardial arterioles and capillaries, entered low-pressure heart cavities or cardiopulmonary vessels through fistula vessels, and caused myocardial stealing. Myocardial stealing made these patients more prone to symptoms of cardiothoracic discomfort than normal people under conditions of increased oxygen demand such as doing slightly more intense sports, becoming pregnancy and other special physiological conditions. Among 62 CAFs, a 25-year-old female patient suffered the first attack of palpitation with no obvious cause during pregnancy and suffered palpitation accompanied by lip cyanosis again three years later as diagnosed of left artery fistula to right ventricle. It was believed that her increased oxygen demand during pregnancy made the symptoms of coronary artery fistula appear earlier. In fact, nearly half of the clinical cases of CAFs were asymptomatic, and those with symptoms often came to the clinic with chief complaints such as chest pain, chest tightness, and palpitations and etc. It was suggested that the symptoms or severity of CAFs depended not only on the drainage site of the fistula (type of fistula), the diameter of the fistula vessels, the size of the fistula (the amount of shunts) and etc., but also on the patient's age, physical tolerance, complication of other cardiopulmonary diseases and the duration of the diseases. For instance, coronary artery-cardiac fistula was usually a single bulky fistula vessel with a large vascular shunt, and continuous murmurs during diastole and systole in clinic can be easily heard on a patient with this fistula, so patients often came to the clinic at the early stage. Among 62 cases of CAFs there were 19 cases of coronary artery-cardiac fistula including 12 patients (63%) under 30 years old. While coronary artery-pulmonary fistula, due to its small fistula and small shun, was generally asymptomatic or became symptomatic late. For example, among 62 CAFs, there were 31 cases of Coronary pulmonary-artery fistula including 4 patients under 50 years of age and 27 patients (87%) over 50 years old. Other patients came to the clinic with symptoms of myocardial theft, such as arrhythmia, angina pectoris, and myocardial infarction (8–12). Complications of congestive heart failure, endocarditis, thrombosis, pulmonary hypertension, or embolic events may not occur until late in the course of the disease.

## **2. Comparison of image inspection methods of CAFs**

### **Traditional Coronary Angiography (cag)**

Before the emergence or widespread use of cardiac CT, CAG was considered a diagnostic reference for CAFs. CAG itself was also one of the treatments for CAFs. Generally, CAG can well show the origin of the fistula vessels and the proximal movement, but sometimes cannot show clearly the distal fistula vessels and orificium fistulae due to the dilution of the contrast agent. In addition, CAG only provided two-dimensional projection images, making it limited to diagnose CAFs with complex drainage sites and small orificium fistulae. It was reported that only 35% -50% of CAFs were correctly diagnosed by CAG (13, 14).

## **Transthoracic Or Transesophageal Echocardiography**

ECHO has no ionizing radiation. Although the use of microbubbles can help determine more precisely the location and extent of CAFs, ECHO has poor evaluation of CAFs with multiple fistula vessels, complex drainage sites and relatively small orificium fistulae. For example, as the left ventricular systolic pressure was the same as the aortic pressure, color Doppler was not sensitive to the abnormal flow of the fistula-entered pulmonary artery or the left ventricle, reducing the detection rate of coronary artery-pulmonary fistula. Furthermore, ECHO often depended on the operator's experience and technique.

## **Mr Angiography**

MR angiography has no ionizing radiation and no iodine-containing contrast agents, and can sometimes replace conventional CT angiography. For CAFs, however, the biggest drawback of MR angiography was its limited effectiveness in showing the distal and extracardiac structures of fistula vessels (14, 15).

## **CTA**

Compared with other imaging methods, CTA, especially dual-source CT, has the advantages of shorter examination time, higher temporal, spatial, density resolution, and it can better show origin of CAFs, course of fistula vessels, and drainage sites when combined with post-processing such as later volume rendering (VR), multiple Planar reorganization (MPR), curved surface reorganization (CPR). CTA is especially important in diagnosing CTAs with multiple fistula vessels, complex drainage sites, and small orificium fistulae (such as coronary-pulmonary fistula). This information is very important for surgeons and intervention doctors before surgery. These advantages of CTA also explained why the number of occasionally diagnosed CAFs increased with the popularity of CT (16–20). The obvious disadvantage of CT was its radiation exposure, which can be reduced by low-dose technologies such as prospective ECG gating and iterative reconstruction. Research results showed that the combination of tube voltage reduction and dose reduction technology can greatly reduce the radiation dose to 0.1 mSv (21, 22).

## **3. DSCT findings of CAF**

The CT images of CAFs can be categorized mainly into two types. The first type was CAFs with a single origin, a single fistula vessel, and a single orificium fistulae. The affected coronary arteries dilated to varying degrees (typically the expansion and thickening show a "sausage-like" change), took a tortuous course and finally connected directly with the heart cavity or chest blood vessels. CT can clearly show the position and size of the orificium fistulae, and sometimes the contrast agent "jet sign" can be directly observed at the orificium fistulae (Fig. 1, Fig. 4). The second type was CAFs with multiple small or tortuous dilated malformed vessels from the left coronary artery, or right coronary artery or bilateral coronary artery. These vessels can communicate each other forming a reticular or diffuse disordered vessel cluster, and finally entangle and fistula into the main pulmonary artery or heart cavity and etc(Fig. 2). CAFs of this type were most common found in coronary-aortic fistulae. Orificium fistulae of this type was often very small, not often directly visible on DSCT, but in some cases the invisibility of orificium fistulae did not affect the qualitative diagnosis (23), and in some occasions the temporary differences in contrast agent concentration on both sides of orificium fistulae can indirectly indicate the presence of orificium fistulae (Fig. 3).

CAF vessels can originate from any of the three branches of the coronary artery, and the drainage site was more important than the origin in the clinic. We observed that among 62 cases of CAFs, the singly-originated left coronary artery accounted for the highest proportion with a total of 31 cases (50%), followed by 21 cases (34%) originating simultaneously from the left coronary artery and the right coronary artery, singly-originated right coronary arteries totaling 9 cases (15%), and the pulmonary artery has the largest number of drainage site (40/62), consistent with 66 CASs reported by Yang Shan (24), followed by right atrium (10/62) and right ventricle (7 / 62), which validated the view that the pulmonary artery with lower pressure and right heart system are the most common fistula access points (25–28). We observed that the most common type of coronary fistula was the coronary artery-pulmonary fistula, totaling 40 cases (65%), which contradicted the literature (3, 23, 29), though, we have obtained support from a relatively recent literature (30–32). We also noted that the left anterior descending coronary artery-pulmonary fistula was the most common Coronary pulmonary-artery fistula (18/40), followed by the right corus branch-aortic fistula and left anterior descending coronary artery-aortic fistula (14/40). We hold that this is because on the one hand, previous CAG is an invasive test, some patients with asymptomatic coronary fistula will not be examined, and the dilution of CAG contrast agent may cause poor display of some small fistulae, so that some coronary artery-pulmonary fistulae are missed; on the other hand, with the popularization of CT, the detection rate of asymptomatic patients with Coronary pulmonary-artery fistula was getting higher and higher.

## 4. Management of CAFs

The clinical management strategy for patients with CAFs depended on the size and anatomy of the fistula vessel, presence or absence of symptoms, age of the patient, and presence of other relevant cardiovascular abnormalities (33). Asymptomatic CAFs with small orificium fistulae were usually treated with antiplatelet therapy and antibiotics, monitoring of complications and no need of intervention (2).

According to the guidelines recommendation of American College of Cardiology and American Heart Association (34), treatment interventions can be considered in the following situations. First, large CAFs regardless of presence or absence of symptoms; Second, small to moderate-size fistulas (CAFs) with presence of symptoms. These symptoms included myocardial ischemia, arrhythmia, ventricular dysfunction, and endarteritis. Treatment options included surgical ligation and percutaneous transcatheter closure. Surgical ligation was generally recommended for the treatment of patients with large symptomatic CAFs, multiple communications, tortuous communications with aneurysmal arteries, and other heart diseases that require surgery treatment (13, 28). Percutaneous catheterization was a non-invasive treatment with a lower incidence of complications than surgical treatment (35). It was particularly suitable for patients who have surgery-benefited fistula anatomic location, single narrow communication, proximal end-originated fistula vessel, no multiple fistula vessels or large collateral vessels, or (and) no other associated heart disease (23). Many studies have shown that transcatheter closure was feasible, safe and effective, the results of which were almost the same as the surgical results in these cases (36, 37).

The shortcomings of this study were as follows. First the research sample was small, secondly this research was a single-center experience, so it was difficult to estimate the actual prevalence of CAFs in the general population and thirdly the design of this research was retrospective, thus some cases lacked clinical and CAG correlation.

## Conclusion

CAFs was a rare coronary artery anomaly. With the popularization of CT in recent years, the examination rate of asymptomatic CAFs was increasing. ECG-gating CTA (especially dual-source CT) can accurately evaluate the origin of CAFs, the course of fistula vessels and the drainage sites in shorter examination time and with higher time, space, and density resolution, combined with some subsequent image processing. It especially played an important role in diagnosing CAFs with multiple fistula vessels, complex drainage sites and smaller orificium fistulae (such as coronary artery-pulmonary fistula). This information was very important for surgeons and intervention doctors before surgery. Therefore, radiologists should be acquainted with the CT manifestations of CAFs. As the detection rate of asymptomatic CAFs increased, Coronary pulmonary-artery fistula may be the most widely seen type of CAFs. Surely this surely needed to be verified by more subsequent and larger sample reports.

## Abbreviations

DSCT Dual source computed tomography

CAF Coronary arterial fistula

CAG Coronary angiography

ECHO Echocardiography

RCA Right coronary artery

LCA Left coronary artery

## **Declarations**

### **Ethics approval and consent to participate**

Since this study was retrospective, the ethics review agency of our hospital abandoned the requirement of informed consent.

### **Consent to publish**

Not applicable

### **Availability of data and materials**

All data generated or analysed during this study are included in this published article.

### **Competing interests**

The authors declare that they have no competing interests.

### **Funding**

Not applicable

### **Authors' Contributions**

WC conceived and designed the study

LYY contributed to the analysis and manuscript preparation

LJY performed the data analysis and wrote the manuscript.

all authors have read and approved the manuscript

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## Ethics and informed consent:

This study was submitted to and approved by ethics review agency of Guizhou Provincial People's Hospital. Since this study was retrospective, the ethics review agency of Guizhou Provincial People's Hospital abandoned the requirement of informed consent.

We declare that there is no conflict of interest.

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## Figures



Figure 1a



Figure 1b

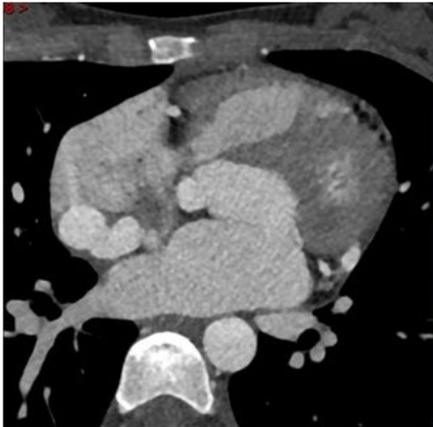


Figure 1c

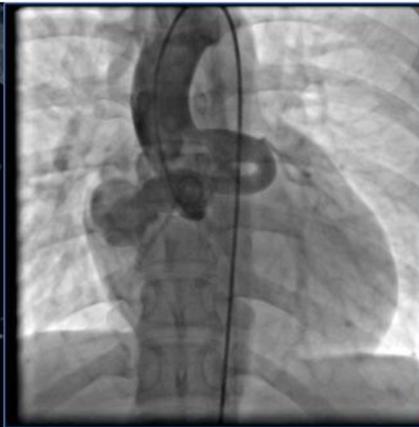


Figure 1d

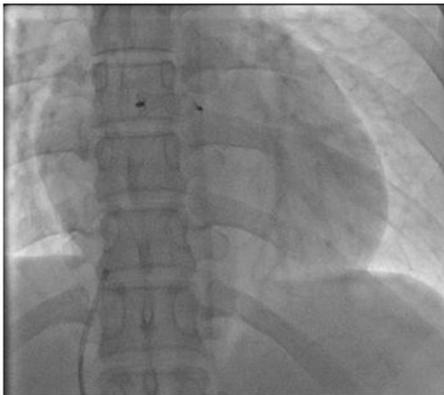


Figure 1e

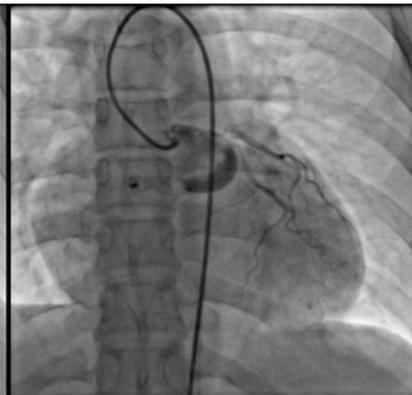


Figure 1f

## Figure 1

A 25-year-old woman suffered heart palpitations for 3 years and her state of illness aggravated for 1 month. DSCT showed that she has the left main coronary artery fistula to right ventricle. It can be seen that her left main coronary artery is thickened and tortuous, and the drainage site is the right atrium (Figure 1a, b). A local aneurysm was formed in the fistula vasculature, and the contrast agent "spray sign" can be seen in the fistula communication (Figure 1a, c arrow). The patient underwent CAG examination

which validated that the left main coronary artery fistula to right ventricle (white arrow in Figure 1d) was in good agreement with DSCT. An occluder was placed in the fistula vessels (Figure 1e white arrow), and the fistula vessels were successfully blocked (Figure 1f white arrow).

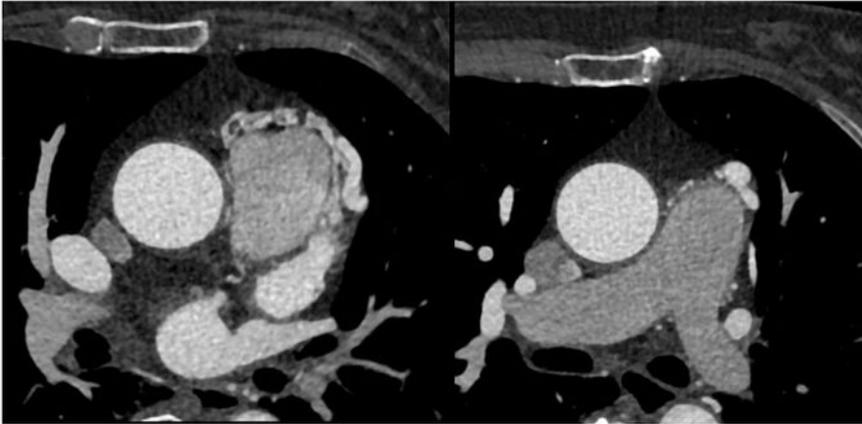


Figure 2a

Figure 2b

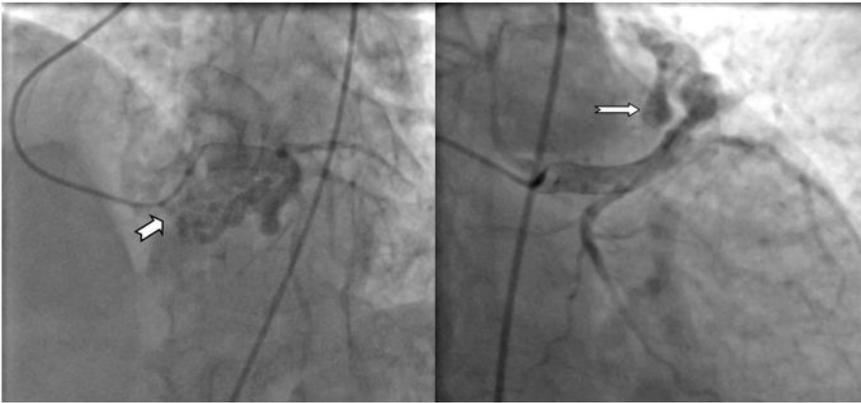


Figure 2c

Figure 2d



Figure 2e

Figure 2f

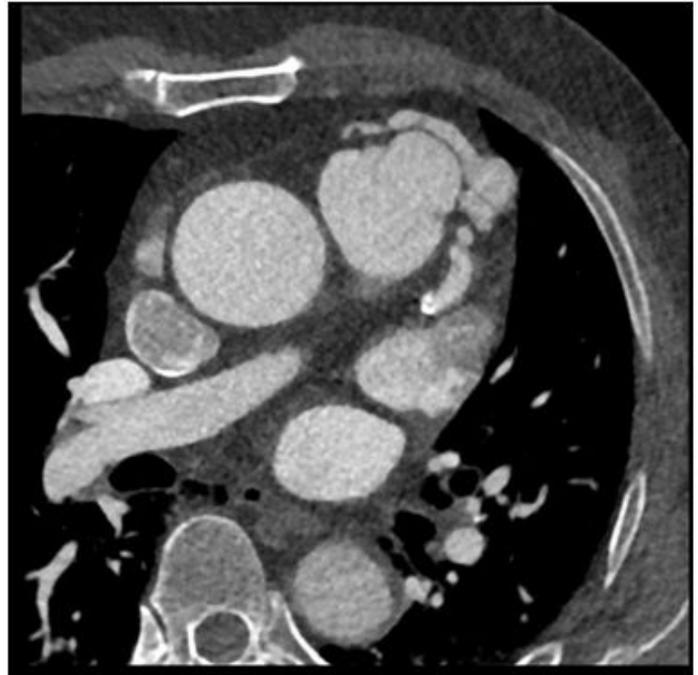
## Figure 2

A 64-year-old woman was sent to hospital after having suffered repeated chest discomfort for 20 days. DSCT before operation showed that the left and right coronary arteries sent tortuous disordered blood

vessels, and the distal anastomosis showed a vascular network tightly wrapping around the main pulmonary artery (Figure 2a white arrow). DSCT preoperatively diagnosed that the right coronary artery and left anterior descending branch-main pulmonary fistula with local aneurysm in the fistula vessels has been formed (Figure 2b white arrow). CAG examination confirmed that the anterior descending branch sent thick disordered fistula vessels (Figure 2c white arrow), aneurysm was locally visible (Figure 2d white arrow), the right coronary artery sent a thickened disordered corus branch (Figure 2e white arrow), and the distal fistula entered into the pulmonary artery (black arrow in Figure 2e). Later, the patient underwent off-pump Coronary pulmonary-artery fistula repair, and a review of CT showed that the number of fistula vessels around the main pulmonary artery decreased obviously and disappeared (Figure 2f).



**Figure 3a**



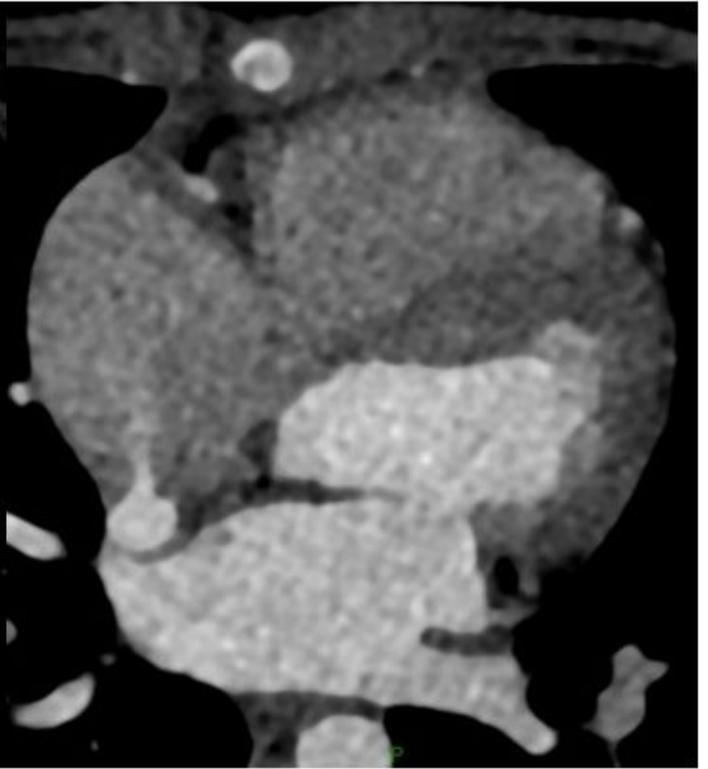
**Figure 3b**

### Figure 3

A 60-year-old man was hospitalized for 20 days with chest tightness and palpitations. The ECG indicated the existence of atrial fibrillation. He underwent a DSCT examination and the left anterior descending branch sent a thick and tortuous blood vessel wrapping around the main pulmonary artery. The orificium fistulae was unclear, but Figure 3a (at the white arrow) showed that the contrast medium leaking to the pulmonary artery early resulted in a concentration difference and caused a density difference with the lower density pulmonary arteries displayed on CT, and eventually the difference disappeared with time prolonged (Figure 3b, white arrow).



**Figure 4a**



**Figure 4b**

#### Figure 4

Figure 4a. - A 71-year-old woman suffered chest tightness and shortness of breath for six months. She was diagnosed as right conic branch and left anterior descending branch-aortic pulmonary fistula. White arrows show jet signs of orificium fistulae. Figure 4b. - A 9-year-old woman suffered cardiac murmur. She was diagnosed as left main coronary artery fistula to right ventricle. White arrows show jet signs of orificium fistulae.