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ABOUT COVER

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Intermittent appearance of right coronary fistula and collateral circulation: A case report

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Abstract

BACKGROUND

Congenital coronary artery fistula can lead to symptoms of chest tightness, chest pain, or exertional dyspnea, which is a congenital vascular malformation that should not be ignored. Patients who have such malformations are frequently observed with different concurrent abnormal anatomic structures. Collateral circulation may have a positive effect on improving the patients' symptoms.

CASE SUMMARY

A 53-year-old female experienced episodic chest discomfort for the past month with symptoms manifesting when she was agitated or overexerted. After a positive treadmill test, the patient underwent coronary angiography. "Ghostlike" intermittent appearance of coronary ventricular fistula and collateral branching were observed. The patient was diagnosed with a right coronary ventricular fistula and collateral circulation.

CONCLUSION

This case shows the likelihood of collateral circulation in patients with coronary

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artery fistula. This may provide medical staff with novel solutions to treat insufficiency of myocardial blood supply induced by cardiovascular malformations.

Key Words: Coronary fistula; Collateral circulation; Congenital coronary artery; Case report

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Core Tip: Congenital coronary artery fistula is a congenital vascular malformation that should not be overlooked. It can lead to symptoms of myocardial ischemia. In the report, we describe a patient who had episodic chest discomfort. Coronary angiography demonstrated a “ghostlike” intermittent appearance of coronary ventricular fistula and collateral branching. The patient was diagnosed with a right coronary ventricular fistula and collateral circulation. This case shows the likelihood of collateral circulation in patients with coronary artery fistula and may provide us with novel strategies for solving insufficiency of myocardial blood supply induced by cardiovascular malformations.

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INTRODUCTION

Congenital coronary artery fistula has an incidence of about 0.002% in the general population and is a rare coronary anomaly. Due to the coronary steal phenomenon, this disease often leads to myocardial ischemia, causing patients to experience chest tightness, chest pain, or exertional dyspnea. The severity of symptoms is closely associated with the size of the fistula. In severe cases, intervention or surgery is required[1-4].

The establishment of collateral circulation may have a positive influence on symptoms in these patients[5]. However, collateral circulation is more frequent in patients with severe coronary stenosis and severe myocardial ischemia but is rarely observed in patients with coronary artery fistula[5,6].

We present herewith an adult female who presented with episodic chest discomfort. In addition to the presence of a right coronary artery fistula, collateral circulation was also observed. The patient was treated with Chinese medicine and her symptoms resolved with no recurrence.

CASE PRESENTATION

Chief complaints

A 53-year-old woman presented to the Cardiology Department of our hospital complaining of episodic chest discomfort.

History of present illness

She had experienced episodic chest discomfort for the past month with symptoms arising when she was agitated or when she overexerted herself.

History of past illness

The patient had a medical history of asthma.

Physical examination

There was no obvious abnormality in the patient's physical examination.

Laboratory examinations

Routine blood tests, routine urine tests, routine fecal tests, blood biochemistry, immune indexes, and infection indexes were normal. Chest radiography and echocardiography were unremarkable. However, the electrocardiogram showed occasional premature ventricular beats, and the treadmill test was positive.

Imaging examinations

Coronary angiography showed that no stenosis was observed in the left or right coronary artery (Figure 1A-C, see Video 1). However, “ghostlike” intermittent appearance of coronary ventricular fistula and collateral branching was observed. The right coronary ventricular fistula and collateral circulation appeared abruptly (Figure 1D and E, Video 2), and then was undetectable (Figure 1F, Video 3). Nevertheless, the patient refused the computed tomography angiography because of her medical insurance.

FINAL DIAGNOSIS

A final diagnosis of coronary heart disease with right coronary artery fistula and the presence of collateral circulation was made.

TREATMENT

The patient was initiated with 23.75-mg metoprolol controlled release, zero order kinetics once a day.

OUTCOME AND FOLLOW-UP

After treatment, the patient reported no symptoms during the follow-up period. It is not necessary for this patient to undergo coronary angiography again.

DISCUSSION

Congenital coronary artery fistula has several types and includes right coronary artery-right ventricle fistula, right coronary artery-left ventricle fistula, left coronary artery-right ventricle fistula, left circumflex artery-right atrium fistula, bilateral coronary artery-cardiac chamber fistula, and cardiopulmonary bypass[7].

Congenital coronary artery fistula is frequently observed concurrently with artery aneurysm or congenital atresia of the main coronary artery. These abnormal anatomic structures significantly increase the risk of thrombosis, myocardial ischemia, ruptured aneurysms, or other life-threatening complications in these patients[8,9]. In this report, our patient had symptoms of myocardial ischemia due to the presence of a right coronary artery fistula.

Coronary angiography performed on this patient showed intermittent signs of collateral circulation of the coronary artery. This may have had a positive effect on relieving chest tightness in the patient. Due to the adverse effect of coronary artery fistula on the normal myocardial blood supply, new collateral circulation may manifest to supply blood to the ischemic regions when chronic myocardial ischemia occurs. This explains why the patient only had symptoms of episodic chest discomfort [10].

Ilhan *et al*[11] presented a case of coronary superior vena cava fistula and variant angina in a patient. This demonstrates the ability of coronary fistulas to develop collateral vessels, such as coronary arteries.

In this case, the patient had collateral circulation from left to right when the left and right blood vessels were unobstructed, *i.e.* blood flow from the arterial fistula side to the normal blood vessel, which has rarely been observed. Because of fluid shear stress, collateral circulation often flows from the normal blood vessels to the occluded blood vessels to relieve insufficient blood supply at the occluded blood vessel[10]. This suggests that collateral circulation of the coronary arteries may be mended, but how this happens is to be deciphered.

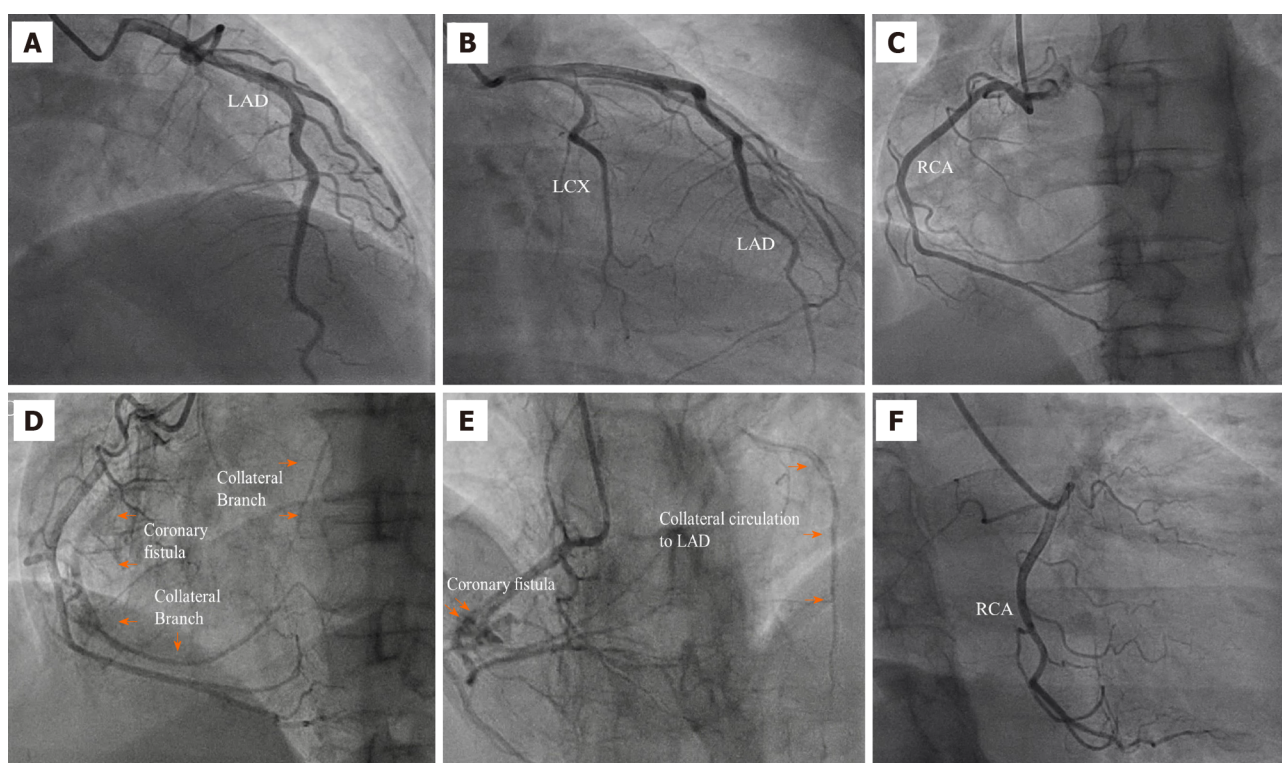


Figure 1 Coronary angiography. A-C: No stenosis was observed in the left or right coronary artery; D and E: The right coronary ventricular fistula and collateral circulation appeared abruptly; F: Then was undetectable. Orange arrows in panel D-E depict the coronary fistula and collateral vessels. LAD: Left anterior descending artery; LCX: Left circumflex; RCA: Right coronary artery.

Heil and Schaper[12] suggested that the generation of collateral circulation arteries requires two stages, *i.e.* the activation of vascular endothelium and the release of growth factors by white blood cells to stimulate the proliferation of vascular cells.

Previous studies have suggested that granulocyte colony-stimulating factor therapy, physical exercise, and external counterpulsation may stimulate the formation of collateral circulation[10].

Concerning small-sized fistulae without the presence of clinical symptoms, regular monitoring can be embraced. For symptomatic and large-sized or giant fistulae, percutaneous coronary intervention or coronary artery bypass graft is recommended [13-15].

CONCLUSION

Only a few reports on congenital coronary artery fistula with concurrent collateral circulation have been reported in patients. Since coronary artery fistulas have been associated with several complications, early detection and treatment are critical.

After performing coronary angiography in this patient who had mild symptoms, we were pleasantly surprised to observe collateral circulation in this patient who was diagnosed with coronary artery fistula. Is the appearance of this structure a congenital coincidence or a compensation mechanism induced by insufficient blood supply? Additional clinical observations and studies are necessary to determine this. Furthermore, the intermittent collateral circulation that was observed in our patient induced a dynamic shunt of the coronary blood flow and hence reveals the complexity of coronary microcirculation.

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