



## Case Report

# Multiple Coronary Artery Fistulas Combined with Giant Coronary Aneurysms with a Brain Occupying Lesion: A Case Report

Yi-Rui Zang<sup>1</sup>, Ji-Wang Zhang<sup>2</sup>, Ze-An Fu<sup>3</sup>, Jia-Hui Wang<sup>1</sup>, Zhi-Gang Liu<sup>1\*</sup>

<sup>1</sup>Department of Cardiac Surgery, TEDA International Cardiovascular Hospital, Chinese Academy of Medical Sciences, and Peking Union Medical College, Tianjin, China

<sup>2</sup>Department of Radiology, TEDA International Cardiovascular Hospital, Tianjin, China

<sup>3</sup>Department of Cardiac Surgery, TEDA International Cardiovascular Hospital, Tianjin, China

\*Corresponding author: Zhi-Gang Liu, Department of Cardiac Surgery, TEDA International Cardiovascular Hospital, Chinese Academy of Medical Sciences, and Peking Union Medical College, Tianjin, China.

**Citation:** Yi-Rui Z, Ji-Wang Z, Ze-An F, Jia-Hui W, Zhi-Gang L (2023) Multiple Coronary Artery Fistulas Combined with Giant Coronary Aneurysms with a Brain Occupying Lesion: A Case Report. Ann Case Report 8: 1518. DOI: 10.29011/2574-7754.101518

**Received:** 14 November 2023; **Accepted:** 17 November 2023; **Published:** 21 November 2023

### Abstract

The coronary artery fistula (CAF) is an abnormal connection between the coronary artery and a major vessel or cardiac chamber. Coronary artery aneurysm (CAA) is defined as coronary dilatation that exceeds the diameter of normal adjacent segments or the diameter of the patient's largest coronary vessel by 1.5~2.0 times. The intracranial space-occupying lesion is a group of diseases that occupy a particular space within the cranium, bearing many causes, such as trauma, tumors, infections, cerebrovascular disease, etc. The coexistence of these three diseases is rare and can lead to fatal complications, including heart failure, angina pectoris, acute myocardial infarction, and increased intracranial pressure. In this article, we report a case of a young patient who was diagnosed with multiple coronary artery fistula with a giant coronary aneurysm concomitant with right cerebellar occupancy and underwent complex surgical treatment at our institution. At follow-up, the patient recovered good myocardial perfusion. In this article, we discuss and analyze the disease characteristics of this patient and summarize the perioperative problems and decisions as a way to improve the prognosis and survival of CAF patients with multiple disease combinations.

**Keywords:** Coronary Artery Fistula; Coronary Artery Aneurysm; Intracranial Occupying Lesion; Heart Failure ; Cardiac Surgery

### Introduction

Coronary artery fistula (CAF) is a coronary artery malformation with abnormal traffic between the main trunk of the coronary artery or its branches and the atria, ventricles, pulmonary arteries, coronary sinuses, and superior vena cava, and is a rare anatomical abnormality of the coronary arteries, with a prevalence of 0.002%

among the general population, and only 0.05% to 0.9% among patients who underwent coronary arteriography or coronary artery CTA [1]. Aneurysm of the coronary arteries, also known as coronary artery aneurysm (CAA), is an enlargement of the coronary arteries beyond their normal diameter or 1.5 to 2.0 times the diameter of the patient's largest coronary artery. It is rare for CAA to occur, and coronary angiography only detects 0.3% to 4.9% of patients [2]. Giant coronary aneurysms refer to arteries with a diameter exceeding four times that of a normal artery, with an incidence rate of only 0.02% to 0.20% [3]. Due to this, it is rare for coronary fistulas and giant coronary aneurysms to occur

simultaneously with cerebellar lesions occupying cerebellar space. In this case report, we present our hospital's diagnosis and treatment of a patient with multiple coronary artery fistulas, a giant coronary aneurysm, and right cerebellar space.

## Case Report

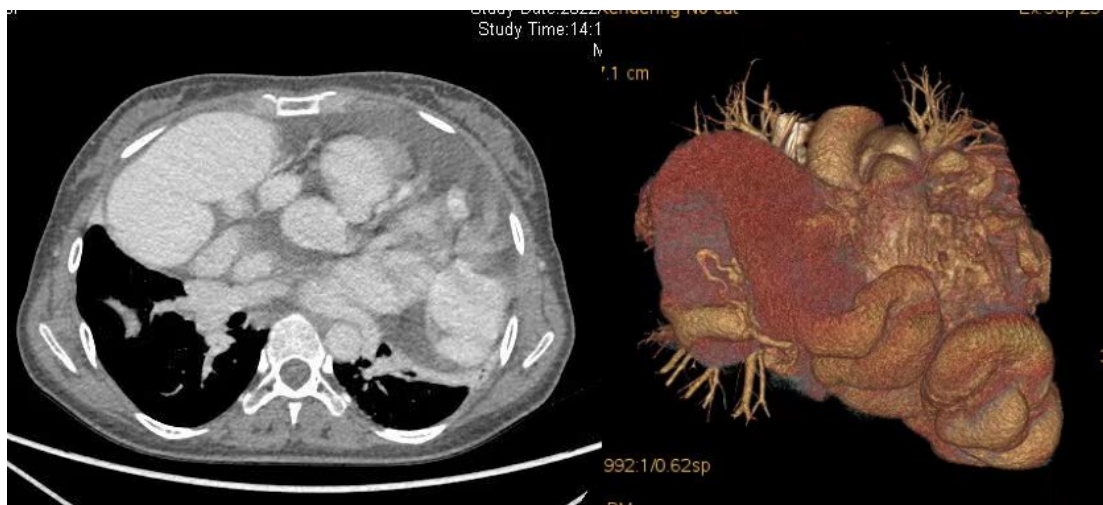
### Overview of the patient

A 33-year-old female patient experienced intermittent dyspnea for one year, which worsened after moving or lying down and was relieved after resting or changing positions. She occasionally complained of chest pain, as well as edema of the lower limbs and eyelids. The patient was treated at another hospital, where a cardiac ultrasound revealed coronary artery right atrial fistula, left ventricular fistula, and mitral valve anterior leaflet prolapse with pericardial effusion. Further treatment was provided to her at our hospital. It is critical to note that the patient has no hypertension, diabetes, or coronary heart disease. She has no history of smoking or drinking alcohol, and no history of congenital or acquired heart disease in her family. A preoperative chest radiograph showed pulmonary vascular congestion and cardiomegaly (Figure 1). A coronary CT showed multiple fistulas with a maximum diameter of 18mm, diffuse aneurysmal dilation of the coronary artery with a maximum diameter of 60mm, multiple

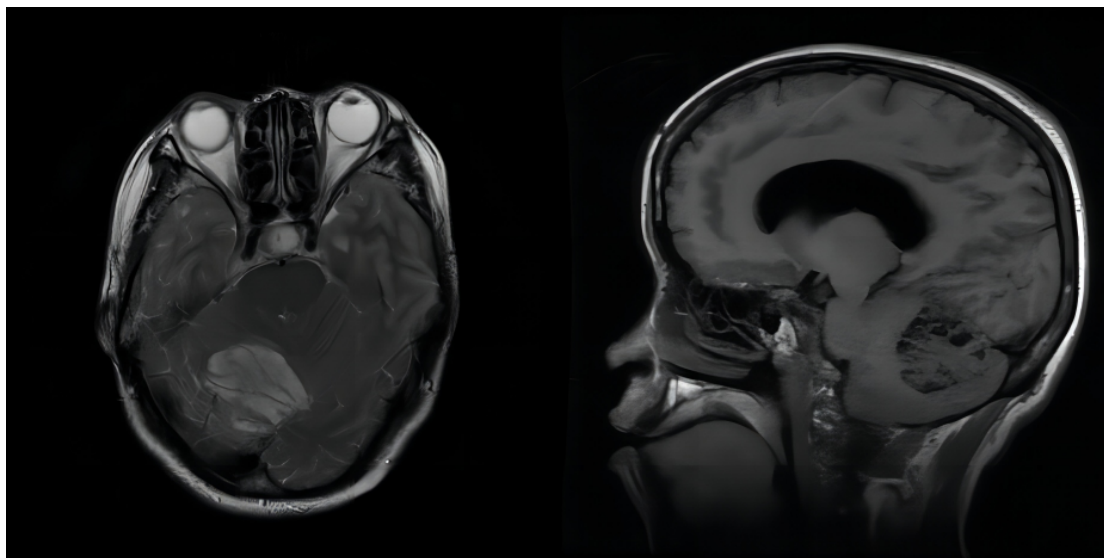
side branches between the left and right coronary arteries, and a large amount of fluid in the pericardium. Three-dimensional CT reconstruction for a more intuitive result (Figure 2). A head MRI revealed a space-occupying lesion in the right cerebellum with obstructive hydrocephalus (Figure 3).



**Figure 1:** Chest X-ray



**Figure 2:** Coronary CT and Three-dimensional CT reconstruction



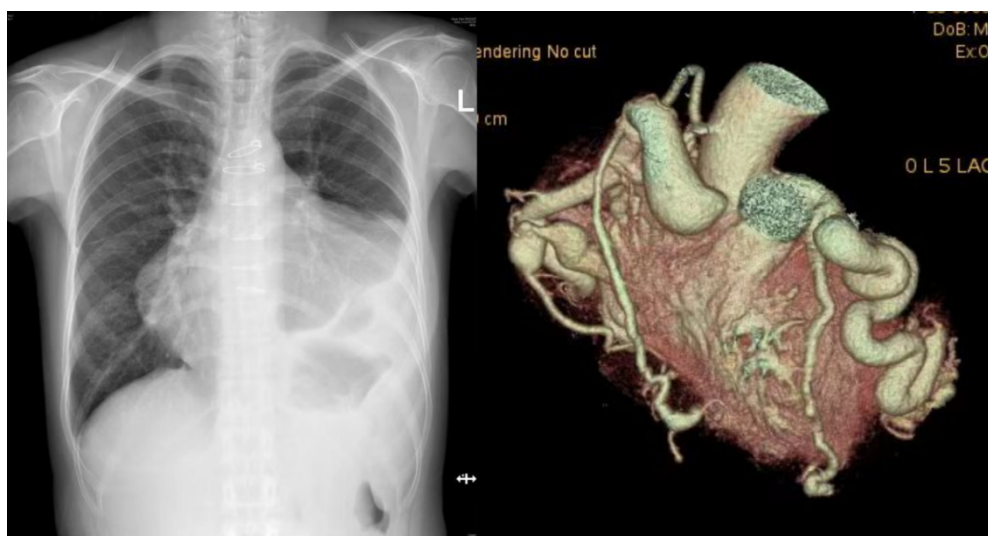
**Figure 3:** Head MRI

Based on examination and clinical symptoms, it can be diagnosed as multiple coronary artery fistulas, giant coronary aneurysms, chronic heart failure insufficiency, cardiac function class III (NYHA), pericardial effusion; and right cerebellar space-occupying lesion. Due to the severity of the patient's symptoms and the imaging examination findings, surgery was recommended.

Following a transesophageal echocardiography (TEE) examination before surgery, the surgical operation was officially initiated. Under general anesthesia, the patient underwent a median thoracotomy. CPB is established and maintained in 349 minutes. The diagonal branch was initially blocked and the right coronary artery was temporarily blocked, as well as the ascending aorta. It took 211 minutes to block horizontally. The aortic root was filled with cardiac arrest fluid, but cardiac arrest was not satisfied, so the right atrium was cut open, and the reverse infusion of arrest fluid was directed directly to the heart. To minimize the risk of perioperative myocardial infarction, the ideal myocardial

protection must be maximized. To expose the external opening of the coronary fistula, the anterior wall of the dilated coronary artery was incised longitudinally, and the coronary fistula was sutured through the incision. 15 small coronary artery traffic branch openings and fistulas were discovered along the inner wall of the tortuous dilated coronary artery, each of which was sutured precisely. The right coronary artery was tortuous, it was not possible to determine its exact location. To prevent postoperative myocardial ischemia, coronary artery bypass grafting was performed. After the successful operation, the patient returned to the ICU, recovered well, and was discharged after 25 days.

The patient adopted a healthy lifestyle after discharge, and underwent coronary CTA in the outpatient clinic four months later, with her heart condition satisfied, the symptoms of heart failure subsided, and adequate myocardial perfusion was achieved (Figure 4). Afterward, the patient was treated for the cerebellar mass by neurosurgery.



**Figure 4:** Post-operative Images

## Discussion

Currently, CAF may result from trauma, transcatheter coronary angiography, coronary artery bypass grafting, atherosclerosis, or a combination of these factors. Despite the fact that the pathophysiology of CAA is currently unclear, its causes include atherosclerosis, coronary fistula, vasculitis, etc. About 25% of giant coronary aneurysms are caused by coronary fistulas [4,5]. A diffuse tumor-like expansion was present throughout the patient's main coronary artery, with its inner diameter reaching about 57 mm at its widest point. There was no obvious atherosclerosis found, which may be directly related to the patient's multiple coronary artery fistulas.

The main pathophysiological problem of CAF is coronary artery steal. Its clinical manifestations and signs are directly related to the amount of blood stolen. Most patients have no abnormalities when they are young. However, as they age, they may experience fatigue, respiratory problems, and fatigue as well as angina and congestive heart failure. There are currently three treatment methods for coronary artery fistulas: (1) conservative medical treatment; (2) interventional treatment; (3) surgical closure and filling. When the patient was admitted to the hospital, she had suffered intermittent dyspnea for one year. She also suffered from heart failure and had a grade III (NYHA) heart function. She was recommended surgery following a variety of examinations and literature research [6,7]. For patients with multiple diseases, preoperative examinations should be completed to the extent possible. Ensure that the surgery goes smoothly. The mortality rate and complication rate of surgery will increase as the patient's age increases. Possible accidents and complications

should be explained to the patient's family before surgery. Surgical procedures include: (1) closure of a fistula through the thoracic or pulmonary artery under extracorporeal circulation; (2) repair of an incisional fistula through the coronary artery under extracorporeal circulation; (3) ligation of a fistulous coronary artery; (4) resect the coronary aneurysm; (5) reconstruct the entire coronary artery. To avoid abnormal blind ends of coronary arteries and missing small fistulas caused by trans-cardiac repair, incised and repaired coronary arteries should be used for existing right atrial fistulas, left ventricular fistulas, and coronary arteries shaped like tumors, with large fistula openings, and long travel distances. It is imperative to perform coronary artery bypass grafting decisively when coronary arteries cannot be accurately detected due to their tortuous course, to restore the coronary blood supply to the maximum extent possible. It's worth mentioning that dilatation of the proximal and middle segments of the right coronary artery is the most common site, while dilatation of the left main coronary artery is rare, occurring in only 0.1% of patients [2]. This patient had both left and right main coronary artery dilatation and simultaneous occupancy of the right cerebellar space was an unusual occurrence.

Since the patient was undergoing surgery under cardiopulmonary bypass (CPB), the impact of CPB on the patient's cerebellar lesions, coronary steal blood phenomenon, and subsequent perioperative myocardial infarction cannot be ignored. Several mechanisms contribute to brain injury caused by CPB, including ischemia, hypoxia, reperfusion injury, hypothermia, inflammation, changes in blood pressure, and so on. It has been shown in recent relevant studies that intraoperative anesthesia induction, aortic clamp closure, CPB time, and other factors can



affect cranial stability and lead to an increased occurrence of adverse events [8]. Additionally, the reperfusion of blood flow after the end of CPB can aggravate cerebral herniation, cause brain edema, and cause cranial reperfusion injury. In this regard, the blood dilution strategy of extracorporeal circulation should be adjusted in order to shorten the operation time as much as possible. Myocardial protection is a routine requirement in cardiac surgery. CPB increases the likelihood of intraoperative coronary artery steal, requiring a retrograde infusion of cardioplegia to obtain optimal myocardial protection and prevent perioperative myocardial infarction. The management of this rare and complex disease presents a challenge to surgeons who need to make accurate predictions about the surgical process, manage the surgical team, select surgical techniques, and control the length of surgery.

### Conclusion

According to the experience summary of this case, we should pay particular attention to the protection of important organs during the operation and to the prevention of postoperative complications. To prevent more serious complications, we will increase screening for cardiovascular diseases, early detection, early diagnosis, and early treatment.

**Author Contributions:** Writing, Yi-Rui Zang; Providing images, Ji-Wang Zhang; Proofreading document, Ze-An Fu; Final inspection, Zhi-Gang Liu.

**Funding:** This research received no external funding.

**Institutional Review Board Statement:** Not applicable.

**Informed Consent Statement:** Not applicable.

**Data Availability Statement:** Not applicable.

**Conflicts of Interest:** The authors declare no conflict of interest.

### References

1. Karazisi C, Eriksson P, Dellborg M (2017) Coronary Artery Fistulas: Case Series and Literature Review[J]. *Cardiology* 136(2):93-101.
2. Devabhaktuni S, Mercedes A, Diep J, Ahsan C (2016) Coronary Artery Ectasia-A Review of Current Literature[J]. *Curr Cardiol Rev* 12(4):318-323.
3. Ibarra-Torres A, Cabrera-Leal C, Lopez-Medina G, Soto ME, Vallejo E, et al. (2019) Giant aneurysm of the right coronary artery, report of a case and review of the literature[J]. *Arch Cardiol Mex* 89(3):227-232.
4. Morita H, Ozawa H, Yamazaki S, Yamauchi Y, Tsuji M, et al. (2012) A case of giant coronary artery aneurysm with fistulous connection to the pulmonary artery: a case report and review of the literature[J]. *Intern Med* 51(11):1361-1366.
5. Khouzam MS, Khouzam N (2021) Giant coronary artery aneurysms involving more than one coronary artery: case report[J]. *J Cardiothorac Surg* 16(1):177.
6. Valente AM, Lock JE, Gauvreau K, Rodriguez-Huertas E, Joyce C, et al. (2010) Predictors of long-term adverse outcomes in patients with congenital coronary artery fistulae[J]. *Circ Cardiovasc Interv* 3(2):134-139.
7. Libertini R, Wallbridge D, Jones HR, Gunning M, Satur CMR, et al. (2018) Giant Circumflex Artery Aneurysm with a Coronary Sinus Fistula[J]. *Ann Thorac Surg* 106(5): e223-e225.
8. Severdija EE, Vranken NP, Teerenstra S, Ganushchak YM, Weerwind PW (2015) Impact of Intraoperative Events on Cerebral Tissue Oximetry in Patients Undergoing Cardiopulmonary Bypass[J]. *J Extra Corpor Technol* 47(1):32-37.