Radiology Case Reports 18 (2023) 3140–3144

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Cinematic rendering of the coronary-pulmonary arterial fistula ☆,☆☆

*Qiang Liu, MD**, *Jicheng Jiang, MD*

Department of Radiology, The First Affiliated Hospital of Jishou University, Shijidadao Rd 26, Jishou, Hunan Province, 416000, China

ARTICLE INFO

Article history:
Received 26 April 2023
Revised 15 June 2023
Accepted 16 June 2023

Keywords:
Coronary-pulmonary arterial fistula
Computed tomography
Cinematic rendering

ABSTRACT

Coronary-pulmonary arterial fistula is a rare coronary artery abnormality disease, which refers to a coronary fistula that terminates at the pulmonary artery. Coronary-pulmonary fistulas are much less common in children than in adults, and the small fistulas are easily missed. We report the case of a 9-year-old girl who presented with coronary-pulmonary arterial fistula. She underwent multimodal imaging, including chest X-ray, echocardiography, computed tomography with 3-dimensional cinematic rendering. We found that the cinematic rendering images clearly showed the small-caliber fistulous connections. The combination of CT and echocardiography can effectively help doctors understand the anatomical details and hemodynamic information.

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Introduction

Coronary fistula (CAF) is a rare coronary artery abnormality in which abnormal blood vessels originating in the coronary arteries bypass the normal capillary and have abnormal communication between the ventricles or large vessels [1]. CAF is mostly congenital and partly acquired, which has been reported to occur in approximately 0.1%-0.2% of all patients undergoing invasive coronary angiography [2]. In addition, the prevalence of CAF has been reported to be as high as 0.9% when incidentally detected in coronary computed tomography angiography (CCTA), which has been widely used in recent years [3].

Coronary-pulmonary arterial fistula (CPAF) is a type of CAF, which refers to a coronary fistula that terminates at the pulmonary artery. In adults, CPAF is the most common type of CAF, accounting for 76.8%-89.5% of cases [4,5]. In contrast, coronary-pulmonary fistulas are rare in children, and only 7.5% of patients with coronary-pulmonary fistulas are younger than 18 years of age [6]. Some CPAFs are incidental findings with no clinical significance, but there are some cases with significant hemodynamic effects that require surgical intervention. CPAF has been reported to be associated with serious complications such as ruptured aneurysms, ischemic heart disease, sudden cardiac death, pulmonary hypertension, and congestive heart failure in some cases [3].

☆ Acknowledgments: Software development and access provided in cooperation with Siemens Healthineers.
☆☆ Competing Interests: The authors have declared that no competing interests exist.
* Corresponding author.
E-mail address: 363944395@qq.com (Q. Liu).
https://doi.org/10.1016/j.radcr.2023.06.033
1930-0433/© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/)

Case description

A 9-year-old girl was admitted to hospital with cough and phlegm for 4 days without obvious inducement, with a maximum body temperature of 39°C. After oral antipyretic treatment with ibuprofen, her body temperature could be reduced to normal, but it was easy to repeat, without chills and convulsions, accompanied by paroxymorous cough, with phlegm not easy to cough up, and vomiting twice when coughing violently. The girl had a normal physique, normal development, medium nutrition, and no history of surgery, trauma, blood transfusion or infection. The parents were healthy, nonconsanguineous, no similar medical history in the family, no history of infection or genetic history in the family. No abnormalities in thoracic symmetry, bilateral symmetry in respiratory movement, no pleural friction, thick breathing sound in both lungs, no rales heard. There was no eminence in the precardiac area, and the apex beat was normal, pulsating 1 cm outside the left mammary line of the V intercostal space, and no tremor was palpable. No enlargement of the heart boundary, auscultation: heart rate 95 beats/min, rhythm, normal heart sound, no noise heard in the auscultation area of each heart valve. Body temperature: 36.5°C, heart rate: 95 beats/min, respiration: 24 beats/min, body weight: 29.5 Kg, blood pressure: 85/65 mm Hg.

Blood routine: white blood cell 3.79*109/L, neutrophil 43.70%, lymphocyte 43.90%, red blood cell 4.78*1012/L, platelet count 235*109/L. Seven blood clotting items: antithrombin III 123.51%. Procalcitonin test - fluorescent quantitative addition: procalcitonin test <0.05 ng/mL. Esr: 16.00 mm/h. Chest X-ray of the patient showed clear bilateral lung fields, little hilar shadow, and no obvious abnormality of cardiophrenic (Fig. 1).

Routine liver function test, routine myocardial enzyme profile test and routine renal function test showed creatine kinase isoenzyme 40.0 U/L, lactate dehydrogenase 336 U/L, creatinine 36.0 μmol/L, uric acid 160 umol/L. Three rheumatism tests and immune function tests showed that antistreptolysin "O" was 129.00 IU/mL. Group A Streptococcus testing was negative. Epstein–Barr virus (EBV) antibody test showed EBV nuclear antigen IgG >600.00 U/mL, EBV capsid antigen IgG 171.00 U/mL, EBV early antigen IgG 11.40 U/mL, and 25-hydroxytotal vitamin D test 20.10 ng/mL. Serological tests for Mycoplasma pneumonia were positive 1:160. The doctor thought that the patient had mycoplasma pneumonia, and the patient was given ceftazidime for anti-infection, interferon and vidarabine for anti-virus, beclomethasone propionate and compound ipratropium bromide for atomization, Shensu granules for cough, ambroxol for phlegm, deoxynucleotide for immune regulation, and fluid infusion.

Echocardiography of the heart revealed turbulence or eddy flow in the lumen of the main pulmonary artery, and a small left coronary-pulmonary artery fistula was suspected (Fig. 2). Coronary CT angiography (CTA) is recommended after cardiothoracic consultation. Axial CTA images reveal anatomic ambiguousness of the blood vessels adjacent to the left wall of the pulmonary trunk, and clear small tortuous vessels visible in CT MPR images (Fig. 3). Cinematic rendering of the CT data further demonstrated the small caliber fistula connection between the left coronary artery and the main pulmonary artery (Fig. 4).

Discussion

CPAF is a rare disease, when this occurs, part of the blood from the coronary arteries enters the pulmonary artery, resulting in reduced blood supply to the heart, which then causes the patient to experience some symptoms of cardiac insufficiency. Unlike adults, who are usually asymptomatic, children are mostly symptomatic. With increasing patient age, the fistula gradually enlarges with increased shunt flow via the fistula in children [7]. Coronary artery fistulas should be treated if

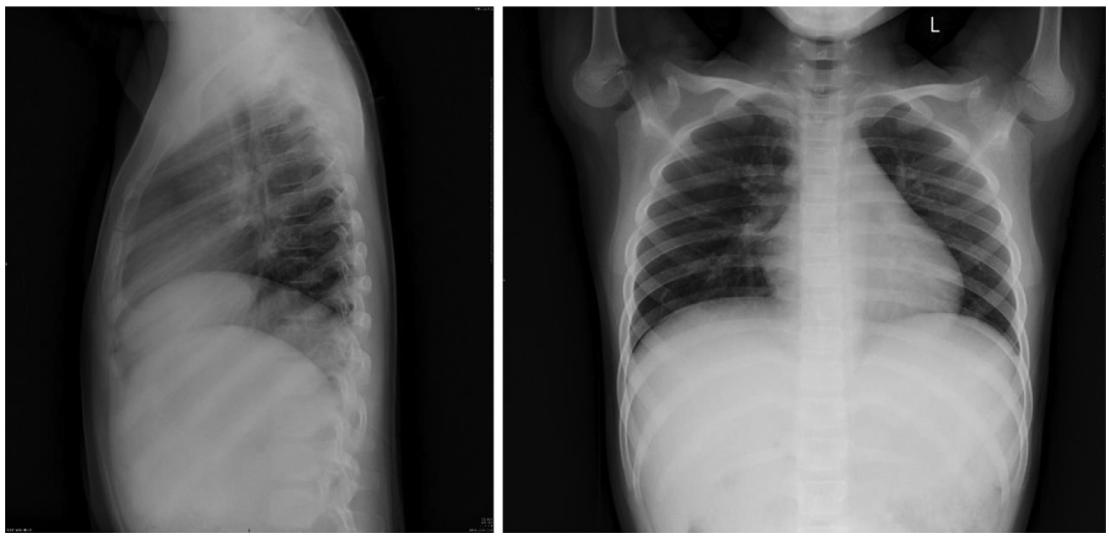

Fig. 1 – Chest X-ray of the patient demonstrates clear bilateral lung fields, little hilar shadow, and no obvious abnormality of cardiophrenic.

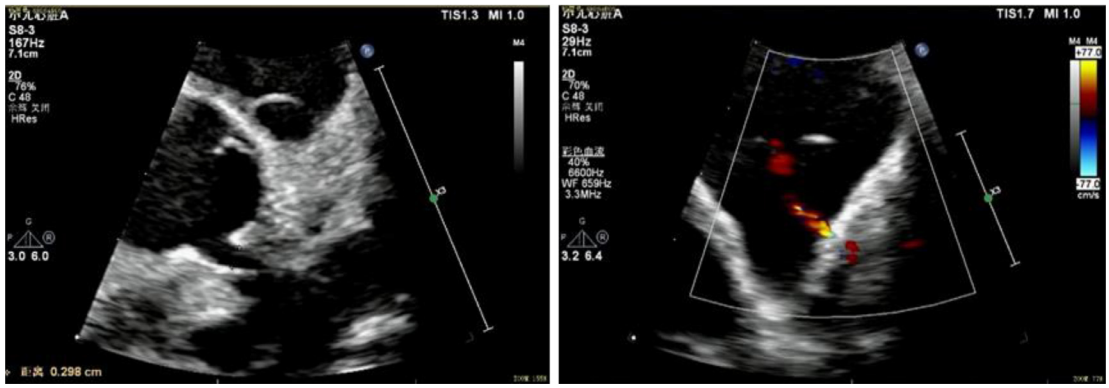

Fig. 2 – Echocardiography of the heart demonstrates turbulence or eddy flow in the lumen of the main pulmonary artery, and a small left coronary-pulmonary artery fistula was suspected.

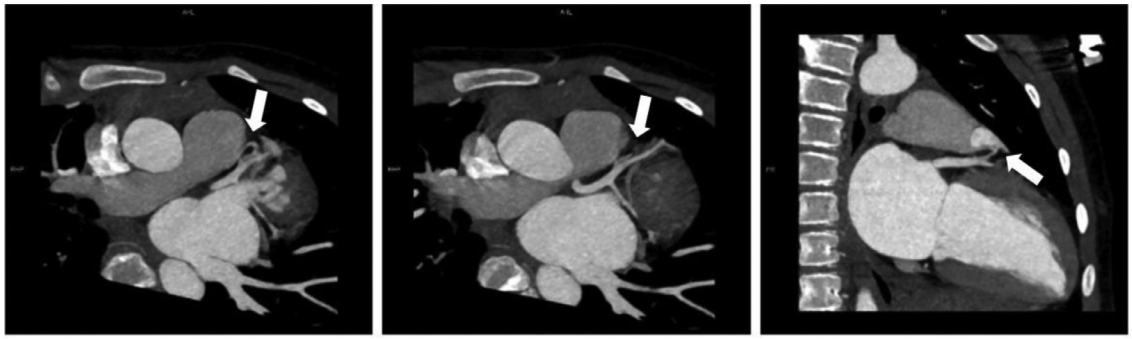

Fig. 3 – MPR images show a left coronary-pulmonary artery fistula vessels draining into the left lateral wall of main pulmonary artery (arrow) with an averaged diameter of 1.3 mm.

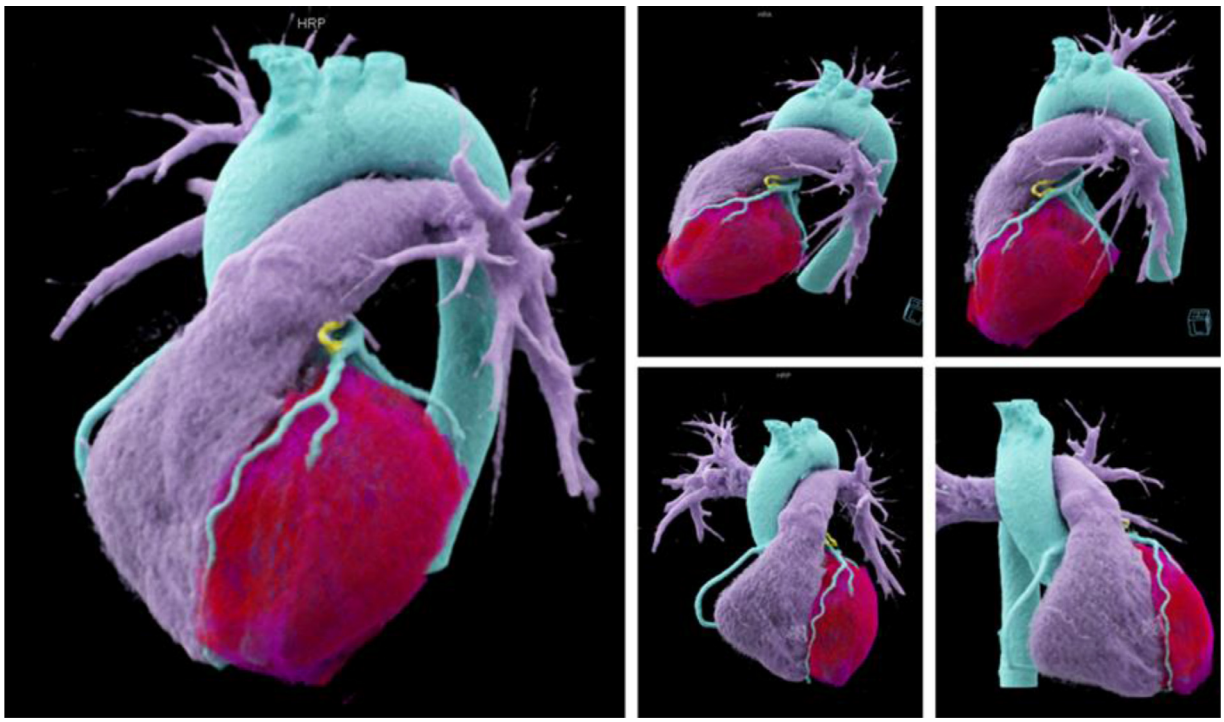

Fig. 4 – The 3D cinematic rendering reconstruction of the CT dataset demonstrates the small caliber fistula connection between the left coronary artery and the main pulmonary artery (yellow vessel) with an averaged diameter of 1.3 mm.

the fistula is large with substantial shunt flow that may lead to myocardial ischemia, ventricular dysfunction, and congestive heart failure, surgical ligation and catheterization are common treatments for coronary artery fistulas [8]. Prompt diagnosis and obtain accurate morphological evaluation of blood vessels are essential for precise surgical treatment [9].

Traditional imaging techniques for detecting CPAF mainly include coronary angiography (CAG), transthoracic echocardiography (TTE), and coronary computed tomography angiography (CCTA) [10]. CAG has limitations in detecting smaller drainage site, mildly expanded fistula vessels and lower drainage into MPA from abnormal fistula vessels because of lower contrast resolution, 2-dimensional fluoroscopic imaging and projection imaging style. TTE can specifically show the drainage site of CPAF, the blood flow into the pulmonary artery and its flow rate, but cannot directly and clearly show the course of the fistula, its origin and adjacent relationship. By combining advanced electrocardiogram (ECG)-gated technology with multiple 3-dimensional reconstruction methods, high-resolution images obtained by CCTA can provide more comprehensive diagnostic information, including accurate anatomical description of fistulas and complex vascular relationships of adjacent structures. CT images can evaluate the origin, number, size, and course (tubular/ worm-like dilation/significant aneurysm formation/ wall attachment sign) of fistula vessels, drainage site, drainage site imaging features (pierced sign, isodensity sign, smoke sign, jet sign), and main pulmonary artery (MPA) [11].

Previous case reports have shown the diagnosis of coronary fistulas using multimodal imaging; however, limited by the poor performance of detail visualization on 3-dimensional CT imaging, only large fistulas can be seen, and tiny connection cannot be shown [7,12]. One unique aspect of our case report is that cinematic renderings of the patient's CT data were also generated as part of her imaging evaluation. Cinematic rendering, a novel 3D visualization technology for postprocessing of CT images, provides photorealistic images with the potential to improve visualization of anatomic details. Compared with traditional volume rendering, cinematic rendering uses more complex lighting models, enhances surface details, produces realistic shadows, increases the depth of 3D visualization, which can have better visualization effects on the details of the heart structure and coronary vessels [13]. In this case, the global lighting model of cinematic rendering takes into account the multiple scattering and reflection of the light by the anatomical structures, and the anatomical details and spatial relationships of small blood vessels are highlighted. On this realistic image in Fig. 4, the relationship between the pulmonary artery, the coronary artery, and the coronary-pulmonary arterial fistula can be well demonstrated. Compared with conventional CT images, cinematic rendering provides more accurate visualization, which can reduce the missed diagnosis of the tiny connection and enables surgeons to understand the anatomy more quickly. However, the rendering optimization of volume data by this technique makes the computational burden of postprocessing significantly increased, and 3D images cannot be obtained quickly. With recent improvements in computation, cinematic rendering can assist general surgeons in preoperative preparation and intraoperative guidance [14].

In conclusion, the cinematic rendering images realistically shows the small-caliber fistula connection. The combination of CT and echocardiography can effectively help doctors understand the anatomical details and hemodynamic information, and then formulate the appropriate clinical treatment plan.

Patient consent

Written and informed consent was taken from the patient's parents. They were informed that no personal details will be revealed in the publication of this case.

REFERENCES

[1] Fernandes ED, Kadivar H, Hallman GL, Reul GJ, Ott DA, Cooley DA. Congenital malformations of the coronary arteries: the Texas Heart Institute experience. Ann Thorac Surg 1992;54(4):732–40.

[2] Yildiz A, Okcun B, Peker T, Arslan C, Olcay A, Bulent Vatan M. Prevalence of coronary artery anomalies in 12,457 adult patients who underwent coronary angiography. Clin Cardiol 2010;33(12):E60–4.

[3] Kim H, Beck KS, Choe YH, Jung JI. Coronary-to-pulmonary artery fistula in adults: natural history and management strategies. Korean J Radiol 2019;20(11):1491–7. doi:10.3348/kjr.2019.0331.

[4] Zhou K, Kong L, Wang Y, Li S, Song L, Wang Z, et al. Coronary artery fistula in adults: evaluation with dual-source CT coronary angiography. Br J Radiol 2015;88(1049):20140754 May. doi:10.1259/bjr.20140754.

[5] Lim JJ, Jung JI, Lee BY, Lee HG. Prevalence and types of coronary artery fistulas detected with coronary CT angiography. AJR Am J Roentgenol 2014;203(3):W237–43. doi:10.2214/AJR.13.11613.

[6] Li N, Zhao P, Wu D, Liang C. Coronary artery fistulas detected with coronary CT angiography: a pictorial review of 73 cases. Br J Radiol 2020;93(1108):20190523. doi:10.1259/bjr.20190523.

[7] Goo HW. Imaging findings of coronary artery fistula in children: a pictorial review. Korean J Radiol 2021;22(12):2062–72. doi:10.3348/kjr.2021.0336.

[8] Hijikata S, Sakurai K, Maeba S, Azegami K. Cardiac tamponade due to ruptured coronary-pulmonary artery fistula aneurysm: a case report. Eur Heart J Case Rep 2020;4(3):1–5. doi:10.1093/ehjcr/ytaa084.

[9] Shibata K, Maeda S, Kawamura M, Nakatsuji H, Ryugo M, Tsutsumi Y, et al. Successful surgical treatment for ruptured aneurysm of coronary-pulmonary artery fistula complicated with cardiac tamponade. JACC Case Rep 2022;4(19):1283–7. doi:10.1016/j.jaccas.2022.08.026.

[10] Hang K, Zhao G, Su W, Bao G, Zhao Q, Jiao Z, et al. Coronary artery-to-pulmonary artery fistula in adults: evaluation with 320-row detector computed tomography coronary angiography. Ann Transl Med 2021;9(18):1434. doi:10.21037/atm-21-4404.

[11] Li JL, Huang L, Zhu W, Ye WT, Yan LF, Zhong XM, et al. The evaluation of coronary artery-to-pulmonary artery fistula in adulthood on 256-slice CT coronary angiography: Comparison with coronary catheter angiography and transthoracic echocardiography. J Cardiovasc Comput Tomogr 2019;13(1):75–80. doi:10.1016/j.jcct.2018.10.013.

[12] Sharifkazemi M, Mohseni-Badalabadi R, Hosseinsabet A, Hajizeinali A. Case report: multimodal imaging diagnosis of a giant coronary artery fistula: a report of two cases. Front Cardiovasc Med 2022;9:986078. doi:10.3389/fcvm.2022.986078.
[13] Rowe SP, Johnson PT, Fishman EK. Cinematic rendering of cardiac CT volumetric data: principles and initial observations. J Cardiovasc Comput Tomogr 2018;12(1):56–9. doi:10.1016/j.jcct.2017.11.013.
[14] Elshafei M, Binder J, Baecker J, Brunner M, Uder M, Weber GF, et al. Comparison of cinematic rendering and computed tomography for speed and comprehension of surgical anatomy. JAMA Surg 2019;154(8):738–44. doi:10.1001/jamasurg.2019.1168.