# Cardiology in the Young

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## **Brief Report**

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# An unusual case of coronary artery fistula successfully treated by transcatheter approach

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#### Abstract

Coronary artery fistulas are rare, but one of the most common forms of congenital coronary abnormalities. These patients are often diagnosed incidentally undergoing coronary angiography, but with the advent of novel cardiac imaging tools, there is an increasing rate of detection as well as transcatheter management of these fistulas. Our case is unusual in a way that it involved a combination of two separate coronary artery fistulas arising from both the coronary systems draining into the same site.

Coronary artery fistulas are rare cardiac anomalies. Echocardiography has a key role for the diagnosis of congenital and acquired heart defects, but it has limitations in detail coronary visualisation and shunt assessment of the fistulas. Here, we present an interesting case of two coronary artery fistulas originating, respectively, from right coronary artery and left anterior descending artery, and draining into the right ventricular apex at the same site.

## **Case presentation**

A 9-year-old boy, on incidental detection of murmur, otherwise asymptomatic, presented for outpatient cardiology review. His cardiovascular examination was unremarkable except for grade 3/6 continuous murmur heard at the level of the lower left sternal border.

Transthoracic echocardiography revealed dilated coronaries measuring 5 mm each & a continuous turbulent flow in colour Doppler analysis of left anterior descending artery coursing along the lateral wall of left ventricle and draining into the right ventricular apex. There was another abnormal vessel with continuous turbulent flow around the same drainage site, but it could not be profiled well. Cardiac chambers were normal sized with normal left ventricle systolic function.

CT coronary angiography confirmed two tortuous dilated coronary artery fistulas draining into the right ventricular apex at the same site. First fistulous vessel was originating from the diagonal branch of left anterior descending artery and second one was from the distal segment of right coronary artery.

Patient was taken to the cath lab for haemodynamic assessment of the shunt (Qp:Qs 1.6:1) and percutaneous closure of fistulas, if suitable. We successfully occluded the left-sided fistula with 6 mm Amplatzer vascular plug II and right-sided fistula with 6 mm \* 14 mm MReye (Cook medical) coil (Figs 1 and 2). Check angiogram after 10 minutes showed no residual fistulous flow with no ST-T changes in ECG. He was discharged the following day on dual anti-platelet therapy daily to prevent thrombosis in the proximal cul-de-sac. The patient had been doing well at 6 months follow-up with normal ECG and echocardiography.

## Discussion

Coronary artery fistula is a rare abnormal direct communication between an epicardial coronary artery and a cardiac chamber (cameral) or a major blood vessel (arterio-venous), without intervening myocardial capillary bed.<sup>1</sup> Historically, coronary angiography has been used as a reference standard imaging modality for diagnosis of coronary artery fistulas with a reported incidence of 0.08% to 0.3%<sup>2,3</sup>; but with the advent of novel cardiac imaging techniques, particularly echocardiography and cardiac CT, there is an increasing rate of detection of these fistulas.<sup>4,5</sup>

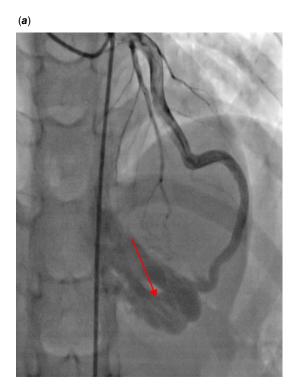
Coronary artery fistulas are mostly solitary; frequently originating from right coronary artery (50–60%) than left side (45%).<sup>6,7</sup> But at times they could be multiple; only 3–5% of coronary artery fistulas are reported to originate from both coronary arteries (as in the case presented above).<sup>8</sup>

Although coronary artery fistulas are mostly asymptomatic; but if untreated, haemodynamically significant fistulas cause clinical symptoms of heart failure, pulmonary hypertension, or angina in 19% of patients <20 years and in 63% of older patients. In general, the indication

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1770 G. Biyani et al.



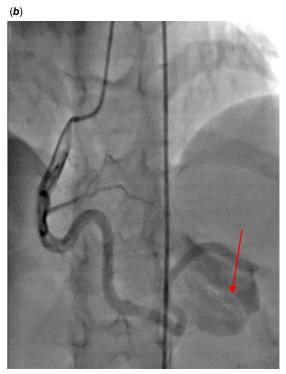
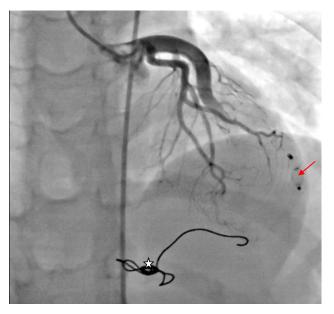


Figure 1. Selective LMCA (a) and right coronary artery (RCA) (b) angiogram images showing dilated tortuous coronary artery fistulas originating, respectively, from diagonal branch of LAD and distal RCA and draining into the right ventricular apex at the same site (red arrows).



**Figure 2.** Fluroscopic image showing occlusion of left-sided coronary artery fistula (CAF) with Amplatzer vascular plug II (red arrow) and right-sided CAF with MReye coil (white star).

for intervention is haemodynamically significant shunt, regardless of symptoms to prevent the occurrence of fistulous-related complications such as aneurysmal formation, infective endocarditis, arrhythmias, premature atherosclerosis, and sudden death,  $^{10}$  and also there is low probability of spontaneous closure of coronary artery fistuls (1–2%).  $^{11-13}$ 

The ACC/AHA Guidelines for the Management of Adults with CHD recommend either surgical ligation or transcatheter closure as both have comparable success rates, morbidity, and mortality (<1%) outcomes. <sup>13–15</sup> Percutaneous approach is recommended over surgery, if anatomy of the fistula is favourable for the transcatheter technique.

## Implications in clinical practice

Our case is unusual in a way that it involved a combination of two separate coronary artery fistulas from both the coronary systems draining into the same site. We stress on the importance of using different multimodality imaging techniques for detailed anatomic delineation of the fistulas to guide the selection of appropriate interventional techniques.

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Conflicts of interest. None.

## References

- Yun G, Nam TH, Chun EJ. Coronary artery fistulas: pathophysiology, imaging findings, and management. Radiographics 2018; 38: 688-703.
- Mangukia CV. Coronary artery fistula. Ann Thorac Surg 2012; 93: 2084–2092.
- 3. Mavroudis C, Backer CL, Rocchini AP, Muster AJ, Gevitz M. Coronary artery fistulas in infants and children: a surgical review and discussion of coil embolization. Ann Thorac Surg 1997; 63: 1235–1242.

Cardiology in the Young 1771

- 4. Yamanaka O, Hobbs RE. Coronary artery anomalies in 126,595 patients undergoing coronary arteriography. Cathet Cardiovasc Diagn. 1990; 21: 28–40.
- Lim JJ, Jung JI, Lee BY, Lee HG. Prevalence and types of coronary artery fistulas detected with coronary CT angiography. Am J Roentgenol 2014; 203: W237–43.
- McNamara JJ, Gross RE. Congenital coronary artery fistula. Surgery 1969; 65: 59–69.
- 7. Qureshi SA. Coronary arterial fístulas. Orphanet J Rare Dis 2006; 1: 51.
- Ata Y, Turk T, Bicer M, Yalcin M, Ata F, Yavuz S. Coronary arteriovenouse fistulas in the adults: natural history and management strategies. J Cardiothoracic Surg 2009; 4: 62.
- 9. Chia BL, Chan AL, Tan LK, Ng RA, Chiang SP. Coronary artery-left ventricular fistula. Cardiology 1981; 68: 167–179.
- 10. Liberthson RR, Sagar K, Berkoben JP, Weintraub RM, Levine FH. Congenital coronary arteriovenous fistula. Report of 13 patients, review

- of the literature and delineation of management. Circulation. 1979; 59: 849–854.
- Cotton JL. Diagnosis of a left coronary artery to right ventricular fístula with progression to spontaneous closure. J Am Soc Echocardiogr. 2000; 13: 225–228.
- Graham DA, Reyes P, Pires LA. Images in cardiology. Coronary artery fístula. Clin Cardiol. 1998; 21: 597–598.
- Buccheri D, Luparelli M, Chirco PR, Piraino D, Andolina G, Assennato P. A call to action for an underestimated entity: our algorithm for diagnosis and management of coronary artery fistula. Int J Cardiol 2016; 221: 1081–1083.
- Buccheri D, Chirco PR, Geraci S, Caramanno G, Cortese B. Coronary artery fistulae: anatomy, diagnosis and management strategies. Heart Lung Circ 2018; 27: 940–951.
- Uyar IS, Akpinar B, Senarslan O, Sahin V, Uc H. Multiple coronary fístulae to left ventricle, with acute myocardial infarction. Asian Cardiovasc Thorac Ann. 2015; 23: 561–563.