Radiology Case Reports 16 (2021) 1790–1793

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

Case Report

Fistula from right and left coronary arteries to pulmonary artery: Coronary CT angiography and coronary angiography findings ☆,☆☆

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ARTICLE INFO

Article history:
Received 7 February 2021
Revised 3 April 2021
Accepted 6 April 2021

Keywords:
Coronary vessel anomalies
Fistula
Coronary angiography
Computed tomography angiography

ABSTRACT

Coronary artery fistula is an abnormal communication between the coronary artery and either the cardiac chamber or the great vessel. In particular, the coronary-to-pulmonary artery fistula can be supplied by either one or both coronary arteries and drains to the pulmonary trunk. We report a unique case of fistula originating from both coronary arteries and draining into both sinuses of the main pulmonary artery in a 57-year-old female who experienced chronic chest pain and palpitation. Dilated and tortuous fistulas were found in coronary angiography and coronary computed tomography angiography examinations. To aid early diagnosis and clinical management, radiologists should be aware of the characteristic radiologic findings.

Introduction

The coronary artery fistula is the direct precapillary connection between a branch of a coronary artery and the lumen of a cardiac chamber, the coronary sinus or superior vena cava, or a pulmonary artery or pulmonary vein close to the heart. The fistula arises from the right coronary artery in 50%-60% of patients, left anterior descending artery in 25%-42% of patients, and both arteries in approximately 5% of patients [1]. The most common drainage sites in order of decreasing frequency are the right ventricle (14%-40%), right atrium (19%-26%), pulmonary artery (15%-20%), coronary sinus (7%), left atrium (5-6%), left ventricle (2%-19%), and superior vena cava (1%) [2]. Although most patients with coronary artery fistula are usually asymptomatic, some of

☆ Competing Interests: The authors declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

☆☆ Funding: The authors received no financial support for the research, authorship, and/or publication of this article.

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https://doi.org/10.1016/j.radcr.2021.04.014

them present dyspnea, fatigue, orthopnea, chest pain, endocarditis, arrhythmias, stroke, myocardial ischemia, or myocardial infarction [1]. The clinical manifestations of coronary-to-pulmonary artery fistulas (CPAFs) depend on the extent of the left-to-right shunts, which are usually small and lack obvious clinical symptoms [3]. Nonetheless, a large left-to-right shunt may lead to complications such as congestive heart failure and pulmonary hypertension, coronary steal, and fistula rupture or the associated arterial aneurysm [1-3]. Several authors consider that symptomatic patient should receive endovascular treatment in order to reduce symptoms even when left-to-right shunt is within border limits [4,5]. The case reported herein involved a large and tortuous fistula between both the right and left coronary arteries and the main pulmonary artery. The CPAF was visualized using both coronary computed tomography (CT) angiography and coronary angiography.

Case report

A 57-year-old female was admitted to our hospital for evaluation of atypical chest pain and palpitation, 2 months after a traffic accident. The patient had no remarkable medical history of conditions such as hypertension, diabetes, and cardiac disease. Her blood pressure was 109/68 mmHg, and her pulse rate was 73 beats/min. The physical examination was nonspecific, and no significant findings were observed on her complete blood count and blood chemistry. In addition, a 12-lead electrocardiogram showed a normal sinus rhythm, and plain chest radiographs showed normal configuration. Moreover, transthoracic echocardiography showed normal-sized cardiac chambers and normal left ventricular systolic function. The patient underwent cardiac catheterization that showed a pulmonary-systemic flow ratio (Qp/Qs) of 1.1. An outside clinic non-contrast chest computed-tomography (CT) scan showed no abnormalities.

For further evaluation, the patient underwent electrocardiogram-gated coronary CT angiography, which was performed using a dual-source CT scanner (Somatom Definition, Siemens Medical Solutions, Forchheim, Germany). The several feeding arteries of the CPAF arise from the proximal right coronary artery and the proximal left anterior descending artery (Fig. 1A). The patient's CPAF showed several fistulous drainage sites in both the right and left sinuses of the main pulmonary artery (Fig. 1B). Three-dimensional CT volume rendering images provided a fine anatomic assessment.

The coronary angiography demonstrated that the fistula originated from both the right and left coronary arteries, which went upward running toward each sinus of the main pulmonary artery (Fig. 1C). These findings of coronary angiography were consistent with those from the coronary CT angiography (CCTA). No evidence of atherosclerotic stenosis or other diseases was observed on the coronary arteries. The patient was discharged and instructed to be followed up by her cardiologist.

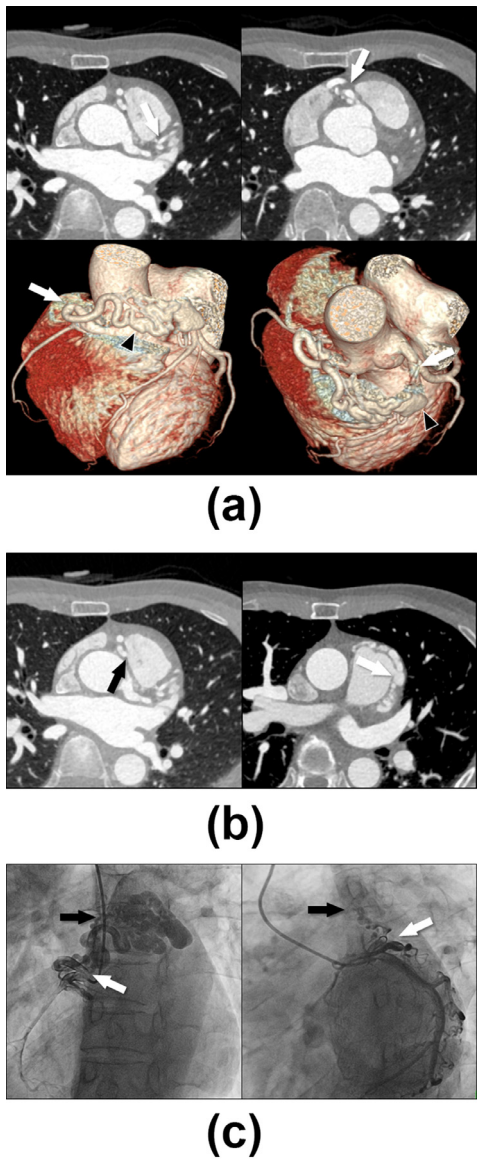

Fig. 1 – A 57-year-old female with both coronary arteries to pulmonary artery fistula detected during evaluation of atypical chest pain and palpitation. (A) The feeding arteries branch from the proximal left anterior descending artery and right coronary artery (white arrows) on axial CT images (upper). The origins of the feeding arteries of the CPAF are the right coronary artery and left anterior descending artery (white arrows) on 3-D volume rendering images. The tortuous arteries of the CPAF are located anterior to the main pulmonary artery (black arrowheads) (lower). (B) The feeding artery from the right coronary artery drains into the right sinus of the main pulmonary artery (black arrow) and the drainage site of the CPAF from the left anterior descending artery is the left sinus of the main pulmonary artery (white arrow). (C) The feeding artery from the right coronary artery (white arrow) communicates with the main pulmonary artery (black arrow) on coronary angiography (left image). The origin of the CPAF from the left anterior descending artery (white arrow) drains into the main pulmonary artery (black arrow) (right image).

Discussion

CPAFs may appear as isolated anomalies in most cases, but they can also be associated with diverse patterns of congenital heart disease possibly related to the development of coronary arteries. During days 33-36 of the embryonic stage, there are 6 coronary enlargements in the embryo. These enlargements normally involute, except for the 2 from the right and left aortic sinuses. CPAF may result from persistence of one or more of the pulmonary arterial enlargements, hence the term accessory coronary artery [6,7]. On the other hand, acquired cases are associated with complications developed after cardiac operation, secondary to percutaneous intervention, endomyocardial biopsy, chest trauma, and myocardial infarction [3,7,8].

The clinical manifestations of CPAF depend on the extent of the left-to-right shunt. Most cases are asymptomatic given the nonsignificant amount of shunting. However, a large pressure difference across the shunt may cause the fistula to become tortuous and dilated. This progress may result in aneurysmal changes with risk of rupture and eventually influence the patients' hemodynamic [3-5]. In addition, coronary steal may occur due to the increased size of the distal portion of the fistula. The symptoms of the patient in this study varied from none to chest pain, including atypical chest pain and dyspnea [7]. The pathophysiologic mechanisms of the symptoms are volume overload as a result of the shunt, coronary steal that causes a decreased myocardial oxygen supply and the lack of capillary formation [5]. It is important to note that the symptoms of CPAF can be similar to those of acute coronary syndrome (eg, aortic dissection), which shows a high mortality rate and requires early treatment.

The role of treatment remains controversial in asymptomatic cases. Commonly, the asymptomatic patients are recommended prophylatic anticoagulation or antibiotics and need constant follow-up [1]. Some authors recommended that symptomatic CPAF should be treated to prevent late complications even when Qp/Qs is within border limits [4,5,8]. Generally, indication for endovascular surgical ligation or embolization includes a pulmonary to systemic blood flow ratio exceeding 1.5-1.7 or right ventricular volume overload, progression of pulmonary hypertension or congestive heart failure, symptoms of ischemia or volume overload in the right ventricle on echocardiography, a history of infective endocarditis, and aneurysm formation [5]. Prognosis from after successful closure of CPAF is relatively excellent. Long-term follow-up is important due to the potentiality of persistent dilation of the coronary artery, postoperative recanalization, calcification, thrombus formation, and myocardial ischemia [5,8].

Identifying the drainage site of a CPAF is clinically more important for surgical closure than identifying its origin. In one study, CPAFs in most patients (54 out of 55, 98%) drained into a single site at the left sinus of the main pulmonary artery, consistent with the results of other studies [6]. In our patient, CCTA showed very unusual imaging features including 2 origins of feeding arteries and multiple fistulous drainage sites in both the left and right sinuses of the main pulmonary artery.

Coronary angiography is one of method for CPAF diagnosis. Nevertheless, coronary angiography is invasive, thus requiring patient hospitalization. In addition, the precise course of the CPAF may be difficult to delineate due to the projectional nature of 2-dimensional coronary angiography images, possibly resulting in diagnostic misinterpretation [9-11]. CCTA is considered the preferred noninvasive imaging methods in terms of efficacy, accuracy, and diagnostic confidence for the detection of anatomic details of congenital anomalies [12,13]. Recent developments of modern CT platforms for dose reduction and image quality improvement can get the higher spatial or temporal resolution, greater anatomic coverage, and quantitative imaging capabilities [12-16]. For example, Dual-source CT scanners can blend or divide raw data acquired from each tube, allowing the generation of images at different radiation doses in a single CT examination [17]. Furthermore, dual-energy CT can enhance both the tissue contrast in virtual monoenergetic imaging (VMI) by using reconstruction algorithms and the visualization of vascular anatomy [12-15]. A VMI enables optimization of the kiloelectron volt (keV) level to evaluate the objective and subjective image quality of coronary vascular anatomy, which also allows for optimization of scan protocols by reducing the necessary amount of contrast material or radiation dose [12-16,18]. Different technical improvement of CCTA can provided higher diagnostic interpretation value of CPAF and can be adopted to safely reduce the radiation dose for effective evaluation and follow-up.

We reviewed imaging findings of CPAF with multiple feeding and draining arteries. To prevent potential CPAF complications, radiologists and clinicians must be aware of the corresponding imaging details.

Authors' contributions

LHJ: Acquisition and analysis of the work, Drafted the work. HJH: Conception of the work and substantively revised it. KJH: Design of the work and substantively revised it. LKH: Writing – review & editing. PS: Writing – review & editing. PSH: Writing – review & editing. All authors have checked the authorship to a submitted version and agreed to the author list and contributions.

Ethical approval

All procedures performed in human participants were in accordance with the Declaration of Helsinki and approved by the Institutional Ethical Committee.

Informed consent

Informed consent was obtained from the individual included in the study.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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