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Coronary-pulmonary fistula with common sac: An uncommon variant

Neeraj Jain, Shashidhar Achar, Naveen K. Garg[1], Sunil Kumar

Departments of Radio Diagnosis, [1]Cardiology, SGPGIMS, Lucknow, Uttar Pradesh, India

Correspondence: Dr. Neeraj Jain, Department of Radio Diagnosis, SGPGIMS, Lucknow - 226 014, Uttar Pradesh, India.
E-mail: neerajdmrd@gmail.com

Abstract

A 68-year-old male patient presented with chief complaints of chest pain and dyspnea on exertion. On physical examination, his pulse was regular at 82 bpm and blood pressure was 140/80 mmHg. Resting electrocardiography (ECG) was within normal limit and chest X-ray also did not reveal any significant abnormality. Routine blood investigations were unremarkable; transthoracic echocardiography also did not show any significant abnormality. Catheter coronary angiography revealed severe triple vessel disease and showed possibility of coronary artery fistula. Computed tomography (CT) coronary angiography showed three aberrant branches arising from right and left coronary arteries forming a sac which subsequently opened into the main pulmonary artery.

Key words: Coronary artery fistula; coronary artery pulmonary artery fistula; CT angiography; fistula

Introduction

Coronary artery fistulae are relatively uncommon, prevalence ranging from 0.1% to 0.9% in computed tomography (CT) coronary angiographic studies.[1-3] A majority of studies reported direct communication between coronary artery and pulmonary artery while our case showed three branches arising from two different coronary arteries and forming a sac on the surface of the main pulmonary artery (MPA). Only a few case reports are available in the literature, which show similar sac-like structure.[4]

Case Report

A 68-year-old male patient presented with chief complaints of chest pain and dyspnea on exertion. On physical examination, his pulse was regular at 82 bpm and blood pressure was 140/80 mmHg. Resting electrocardiography (ECG) was within normal limit and chest X-ray did not reveal any significant abnormality. Routine blood investigations were unremarkable; transthoracic echocardiography also did not show any significant abnormality. Catheter coronary angiography revealed severe triple vessel disease and showed possibility of coronary artery fistula. CT coronary angiography was advised for confirmation and better delineation of fistula.

Catheter angiography showed two aberrant branches arising from proximal left anterior descending artery (LAD) running in cranial direction anterior to aorta and one from right coronary artery (RCA), which also coursed cranially toward the MPA. All three branches were seen to open into a sac-like structure from which a jet of contrast was noted in the direction of MPA [Figure 1].

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CT coronary angiography was performed on Philips 64 Slice multidetector CT scanner with retrospective ECG gating, which confirmed the presence of fistulous communication between branches of LAD, RCA, and MPA. Two tortuous aberrant branches from proximal segment of LAD and one tortuous branch from proximal segment of RCA were noted coursing cranially in relation to the MPA and forming a sac-like structure in close relation to antero-superior wall of MPA [Figure 2; Figure 3A], which in turn shows a small communication with MPA, as shown by the presence of a jet of coronary contrast density within MPA (contrast shunt sign) [Figure 3B].

All three coronary arteries showed multi-segmental disease with the presence of soft calcified and mixed density plaques.

Discussion

Coronary artery fistulae are relatively rare, with prevalence ranging from 0.1% to 0.9% in CT coronary angiographic studies.[1-3]

A majority of fistulae are congenital in nature; however, acquired fistulae are not uncommon and occur following iatrogenic trauma, inflammation, and myocardial infarction. Fistulous communication can also be seen between coronary artery and cardiac chambers, which are known as coronary cameral fistula and with great vessels and coronary sinus.

Most fistulous communications are solitary; however, multiple fistulous communications have also been reported in the literature. Approximately 50%-60% of fistula arise from RCA, LAD is involved in 25%-42% of patients, whereas 5% of cases show origin from both right coronary and left anterior descending arteries.[5,6] A majority of coronary fistulae are seen with cardiac chambers with right ventricle as the most common site of drainage (41%). Communication with pulmonary arterial system is relatively rare and reported in approximately 17% of patients with coronary artery fistulae.[5,7] However, according to the study done by Lim *et al.*, coronary to pulmonary fistula is the most common type. Associated congenital cardiac anomalies have been reported in 20%-45% of patients, which include tetralogy of Fallot, atrial septal defect (ASD), ventricular septal defect (VSD), patent ductus artery (PDA), and so on.[8]

A majority of studies reported direct communication between coronary artery and pulmonary artery while our case showed three branches arising from two different coronary arteries and forming a sac on the surface of MPA. Only few case reports are available in the literature showing a similar sac-like structure.[4]

Most of these patients are asymptomatic due to the small size of the fistula, and are detected incidentally on routine

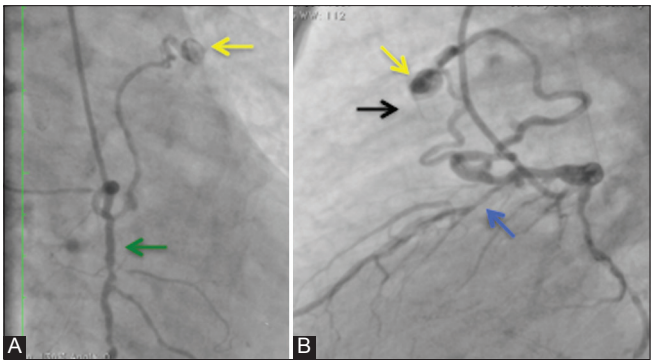

Figure 1 (A and B): Catheter angiography images shows one aberrant branch arising from RCA (green arrow) (A) and two branches from LAD (blue arrow) (B) forming a sac like structure (yellow arrow) from which a small jet of contrast (black arrow) seen in the direction of main pulmonary artery (B)

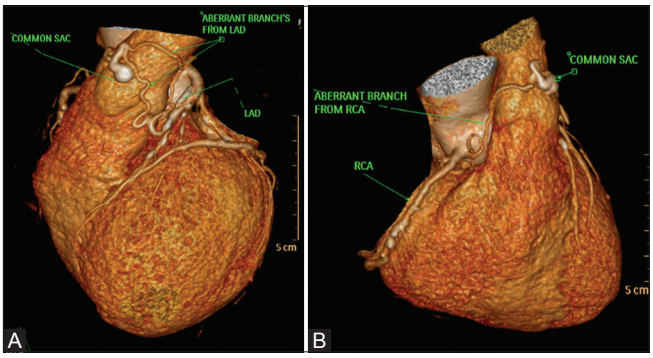

Figure 2 (A and B): Volume rendered image (A) shows two aberrant branches arising from LAD. Volume rendered image (B) shows an aberrant branch arising from right coronary artery and forming a sac like structure along antero-superior surface of main pulmonary artery

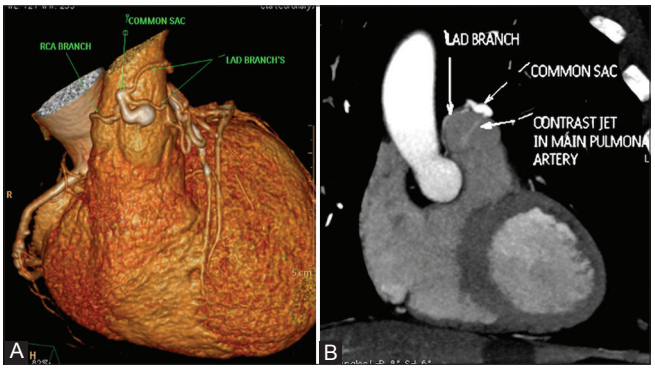

Figure 3 (A and B): Volume rendered image (A) shows sac like structure with supplying branches along antero-superior surface of main pulmonary artery. Oblique coronal image (B) shows a sac like structure and contrast jet in main pulmonary artery

CT coronary angiographic study. Moderate to large fistula may present with symptoms of myocardial ischemia, arrhythmia, stroke, and symptoms of left to right shunting.

Catheter coronary angiography has conventionally been used to diagnose coronary artery fistula; however, accurate course and anatomy can be difficult to assess particularly distal drainage sites due to dilution of contrast agent. It is also

invasive in nature. However, it offers option of therapeutic intervention in the form of percutaneous transcatheter closure.

CT angiography is an excellent modality to detect and delineate the coronary artery fistula due to its multiplanar reconstruction capability, volume rendering technique, and simultaneous evaluation of coronary, non-coronary, and extra cardiac structures. CT angiography can show the exact site of origin from the artery, course, and anatomy of distal vessel, exact site of communication, presence of aneurysmal dilatation, and thrombus formation. The relationship between involved artery and adjacent vital structures can also be delineated in great detail, which helps in precise surgical planning.[5,9] Coronary artery, which is involved in fistulous communication, appears dilated, tortuous, and lacks normal tapering. Fistula can be seen as direct communication between coronary artery and pulmonary artery, cardiac chambers, and other vascular structures such as inferior *vena cava* (IVC), bronchial artery, and coronary sinus. The "contrast shunt sign" occurred when a jet of contrast agent is seen flushing a partially opacified or un-opacified draining cardiac chamber or vessel.[10] The cardiac chambers or great vessels may appear dilated due to volume overload and lung fields may also show changes due to increased pulmonary flow if fistulous communication is fairly large.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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