A rare case of coronary fistula evaluated by first-pass radionuclide angiography

Byung Wook Choi MD,
Hae Won Kim MD, PhD,
Kyoung Sook Won MD, PhD,
Bong-Il Song MD, PhD

Department of Nuclear Medicine, Keimyung University, School of Medicine, Republic of Korea

Keywords: - Coronary artery fistula
-First-pass radionuclide angiography
-Cardiac multidetector computed tomography
-Coronary angiography

Corresponding author:
Kim Hae Won MD, PhD
Department of Nuclear Medicine, Keimyung University, School of Medicine,
56 Dalseong-ro, Jung-gu, Daegu, 700-712, Republic of Korea
Tel: 82-53-250-7068, Fax: 82-53-250-8128,
hwkim.nm@gmail.com

Received:
16 March 2016
Accepted revised:
8 June 2016

Abstract
Coronary artery fistula (CAF) is a rare anomaly that originates from the coronary artery and drains into the cardiac chamber or the adjacent vasculature. We report a case of CAF in a 77 years old woman with dyspnea on exertion. Using coronary angiography and cardiac multidetector computed tomography, this patient was diagnosed with CAF draining into the left bronchial arteries. First-pass radionuclide angiography (FPRNA) showed early pulmonary recirculation through a left to right shunt. The pulmonary to systemic blood flow ratio was 1.24. The patient received supportive care with vasodilator and antiplatelet therapy. First-pass radionuclide angiography was used to provide physiologic informations, to plan the treatment course for this patient.

Hell J Nucl Med 2016; 19(2): 167-169 *Epub ahead of print: 22 June 2016* *Published online: 2 August 2016*

Introduction

Coronary artery fistula (CAF) is a rare anomaly in which communication exists between a coronary artery and either a cardiac chamber or another vascular structure. The CAF arises more often from the right coronary artery and left anterior descending artery than from the left circumflex artery [1]. Clinical presentation of CAF may be variable, depending on the anatomy, size, and complications. Patients with a small fistula are asymptomatic; these patients are incidentally identified [2]. However, a large fistula can cause symptoms or complications such as chest pain, palpitations, murmurs, dyspnea on exertion, congestive heart failure, arrhythmias, and myocardial infarction [2, 3].

First-pass radionuclide angiography (FPRNA) is a noninvasive technique that provides information regarding a variety of cardiovascular variables and is usually used to document and quantitate left to right shunting cardiac defects [4]. Here, we report a case of CAF in a 77 years old woman with dyspnea on exertion. To the best of our knowledge, this is the first case report of FPRNA in a patient with CAF draining into the left bronchial arteries.

Case Report

A 77 years old woman with a history of dyspnea on exertion and pulmonary hypertension for 3 years was admitted to our hospital. An electrocardiogram showed left ventricular hypertrophy with sinus arrhythmia. Levels of cardiac enzymes were within the normal range (creatinine kinase MB<0.5ng/mL and troponin I=0.10ng/mL). Coronary angiography and cardiac multidetector computed tomography (MDCT) revealed right CAF draining into the left bronchial arteries (Figure 1). To evaluate ischemic insult from CAF, technetium-99m-methoxyisobutyl isonitrile (^{99m}Tc-MIBI) myocardial perfusion single photon emission tomography (SPET) was conducted; however, no significant perfusion defect was observed even after adenosine stress test (Figure 2). We performed FPRNA with a bolus injection of 800MBq ^{99m}Tc-pertechnetate into the antecubital vein and showed delayed and decreased radioactivity of the left lung compared with that of the right lung (Figure 3). The time activity curves obtained from a region of

interest placed over the right lung revealed early pulmonary recirculation of the radiopharmaceutical through a left to right shunt. The calculated pulmonary to systemic blood flow ratio (Qp/Qs) using the gamma variate method was 1.24. The patient had supportive care with vasodilator and antiplatelet therapy and is asymptomatic until now.

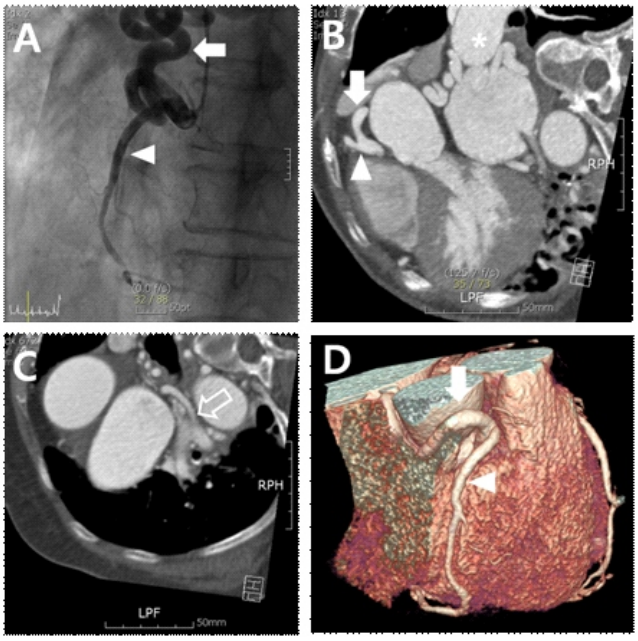

Figure 1. Coronary angiography (A) shows a coronary artery fistula (arrow) originating from the proximal right coronary artery (arrowhead). Cardiac MDCT images (B, C) show that a tortuous vessel (arrow) from the proximal right coronary artery (arrowhead) passes between the ascending aorta and the main pulmonary trunk (asterisk) and drains into the left bronchial artery (blanked arrow). Volume-rendered image (D) shows a coronary artery fistula (arrow) originating from the proximal right coronary artery (arrowhead).

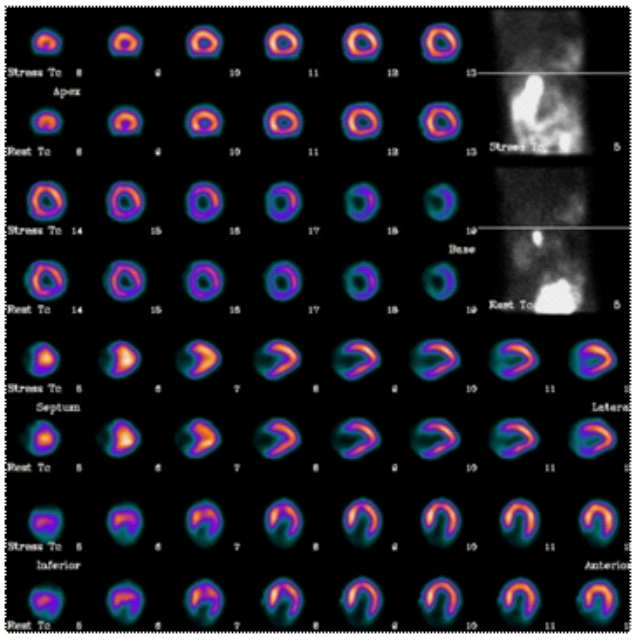

Figure 2. ^{99m}Tc-MIBI myocardial perfusion SPET shows no significant perfusion defect at stress and at rest.

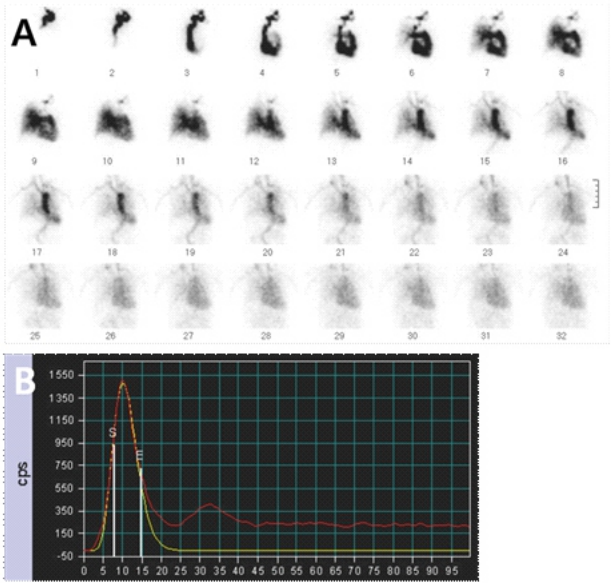

Figure 3. First-pass radionuclide angiography (A) reveals decreased radioactivity in the left lung compared to that in the right lung. After the advent of radioactivity in the left ventricle and thoracic aorta, persistent radioactivity is seen faintly in both lungs, indicating a small amount of pulmonary recirculation through the left to right shunt. The time activity curve (B) reveals early pulmonary recirculation of the radiopharmaceutical through a left to right shunt, evidenced by a small second peak. The calculated pulmonary to systemic blood flow ratio using the gamma variate method was 1.24.

Discussion

Coronary artery fistula is an anomalous connection between a coronary artery and another vessel or between a coronary artery and the cardiac chamber. The resultant physiologic derangement from CAF depends on the site of origin and termination of the abnormal connection and the size of the connection [5]. The major sites of origin are the right coronary artery (55%), the left coronary artery system (35%), and both coronary arteries (5%). The major termination sites of the right and left coronary artery origins are the right ventricle (40%), right atrium (26%), and pulmonary arteries (17%) [1, 6]. The volume of the shunt varies with the size of the fistula and there are differences between the systemic resistance and the resistance in the terminating vessel. In the present case, the patient had a right CAF draining into the left bronchial arteries. The left to right shunt from the CAF in the patient resulted in abnormally high blood flow and pressure directed to the right heart circulation, gradually leading to maladaptive changes that ultimately resulted in pulmonary hypertension. Eventually, due to increased resistance and decreased compliance of the pulmonary vessels, elevated pulmonary pressure resulted in decreased perfusion of the left lung and small amount of pulmonary recirculation, which was seen on the FPRNA image.

A hemodynamically significant fistula with a left to right shunt may lead to congestive heart failure, pulmonary artery hypertension, and myocardial ischemia [7, 8]. The increased blood flow over the systemic to pulmonary artery fistula may reduce the distal intracoronary diastolic pressure and produce ischemia by the coronary steal phenomenon [9]. There is a recognized relationship between myocardial ischemia and infarction in the presence of CAF, even when significant atherosclerotic coronary artery stenosis is lacking. Myocardial perfusion SPET has been used to detect myocardial ischemia and to stratify risk with regard to cardiac events in patients with CAF [10]. In the present case, there was no evidence of myocardial ischemia on the electrocardiogram or by testing cardiac enzyme and myocardial perfusion SPET revealed no significant perfusion defect.

Coronary angiography can show the origin of CAF and is regarded as the gold standard for the diagnosis of CAF [5]. However, the course and drainage site of the CAF may not be seen with coronary angiography because of its imaging pattern and significant dilution of the contrast media [11]. Multiple detector computed tomography (MDCT) provides anatomic images with high spatial resolution, thus allowing assessment of atherosclerotic coronary disease and congenital coronary anomalies. Although conventional coronary angiography and cardiac MDCT could provide accurate anatomic information for CAF, hemodynamic significance is still unremarkable with these modalities. First-pass radionuclide angiography can demonstrate sequential identification of the right ventricle, pulmonary artery, lungs, and left ventricle, and can provide the Qp/Qs ratio, which is needed to diagnose and quantify the left to right shunt [12]. When using the gamma variate analysis of FPRNA, a Qp/Qs ratio of 1.2 is considered evidence of left to right shunt. In the present case, FPRNA showed early pulmonary recirculation through a left to right shunt, and Qp/Qs ratio of the patient was relatively low. The patient received supportive care without undergoing any invasive procedure. Coronary artery fistula should be treated in the presence of large fistulas and progressive left to right shunt, left ventricle volume loading findings, myocardial ischemia, left ventricle dysfunction, and congestive heart failure and in order to prevent endocarditis [13]. Generally, clinical follow-up is recommended for fistulas that do not cause significant shunt.

In conclusion, we report a rare case of CAF draining into the bronchial arteries, which was evaluated by FPRNA. First-pass radionuclide angiography was used to provide physiologic information, to plan the treatment course for the patient with CAF.

Funding

This work was supported by the National Research Foundation of Korea(NRF) Grant funded by the Korea Government(MSIP) (No. 2014R1A5A2010008).

The authors declare that they have no conflicts of interest.

Bibliography

1. Lim JJ, Jung JI, Lee BY et al. Prevalence and types of coronary artery fistulas detected with coronary CT angiography. *Am J Roentgenol* 2014; 203: W237-43.
2. Latson LA. Coronary artery fistulas: How to manage them. *Catheter Cardiovasc Interv* 2007; 70: 110-6.
3. Tiritilli A, Iaria P, Viard P et al. Coronary artery fistulas, a current problem: Clinical and therapeutic considerations. *Ann Cardiol Angeiol* 2016; 65: 31-7.
4. Alazraki NP, Ashburn WL, Hagan A et al. Detection of left-to-right cardiac shunts with the scintillation camera pulmonary dilution curve. *J Nucl Med* 1972; 13: 142-7.
5. Yamanaka O, Hobbs RE. Coronary artery anomalies in 126,595 patients undergoing coronary arteriography. *Cathet Cardiovasc Diagn* 1990; 21: 28-40.
6. Said SA, van der Werf T. Acquired coronary cameral fistulas: are these collaterals losing their destination? *Clin Cardiol* 1999; 22: 297-302.
7. Kiuchi K, Nejima J, Kikuchi A et al. Left coronary artery-left ventricular fistula with acute myocardial infarction, representing the coronary steal phenomenon: a case report. *J Cardiol* 1999; 34: 279-84.
8. Harle T, Kronberg K, Elsasser A. Coronary artery fistula with myocardial infarction due to steal syndrome. *Clin Res Cardiol* 2012; 101: 313-5.
9. Theman TE, Crosby DR. Coronary artery steal secondary to coronary arteriovenous fistula. *Can J Surg* 1981; 24(6): 231-3.
10. Chen ML, Lo HS, Su HY et al. Coronary artery fistula: assessment with multidetector computed tomography and stress myocardial single photon emission computed tomography. *Clin Nucl Med* 2009; 34: 96-8.
11. Schmitt R, Froehner S, Brunn J et al. Congenital anomalies of the coronary arteries: imaging with contrast-enhanced, multidetector computed tomography. *Eur Radiol* 2005; 15: 1110-21.
12. Eterovic D, Dujic Z, Popovic S et al. Gated versus first-pass radioangiography in the evaluation of left-to-right shunts. *Clin Nucl Med* 1995; 20: 534-7.
13. Canga Y, Ozcan KS, Emre A et al. Coronary artery fistula: review of 54 cases from single center experience. *Cardiol J* 2012; 19: 278-86.