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Case Report

Bilateral coronary-to-pulmonary artery fistulas associated with giant aneurysm in an elderly woman: Case report and literature review

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ABSTRACT

Coronary artery fistula is an abnormal vascular communication of coronary artery with cardiac chambers or any segment of the systemic or pulmonary circulation. The prevalence is 0.9% of all coronary anomalies. Coronary artery fistula arises from the right coronary artery in approximately 50.0% of patients, from left coronary artery in approximately 42.0% of patients, and from both in approximately 5.0% of patients. Low-pressure structures are the most common sites of drainage of the coronary fistulas. If a large left-to-right shunt exists, it can be associated with potential complications, such as arterial aneurysm. Here we report an extremely rare case of a 76-year-old woman with bilateral coronary-to-pulmonary artery fistulas associated with giant aneurysms, detected by coronary angiography and confirmed with coronary computed tomography.

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Introduction

Coronary artery fistula (CAF) or coronary arteriovenous fistula is an abnormal vascular communication of coronary arteries with cardiac chambers or any segment of the systemic or pulmonary circulation, without an intervening capillary network [1]. The causes of CAFs are either congenital or acquired. More than 90% of CAFs are congenital. Acquired CAFs result from iatrogenic events such as coronary stent placement, coronary bypass surgery, trauma, and chest irradiation [2,3]. The prevalence of CAFs seen at coronary computed tomography angiog-

raphy (CCT) is reported to be 0.9% [4,5]. According to the number of fistulous connections, single coronary fistulas, accounting for more than 90.0% of CAFs, are far more common than multiple coronary fistulas, which account for 10.0%-16.0% of CAFs [6]. CAF arises from the right coronary artery (RCA) in approximately 50.0% of patients, from the left coronary artery in approximately 42.0% of patients, and from both in approximately 5% of patients [7]. Low-pressure structures are the most common sites of drainage of CAFs. The incidence of coronary-to-pulmonary artery fistulas accounting for 15.0%-30.0% of all CAFs [8,9]. If a large left-to-right shunt exists, potential complications are pulmonary hypertension, congestive

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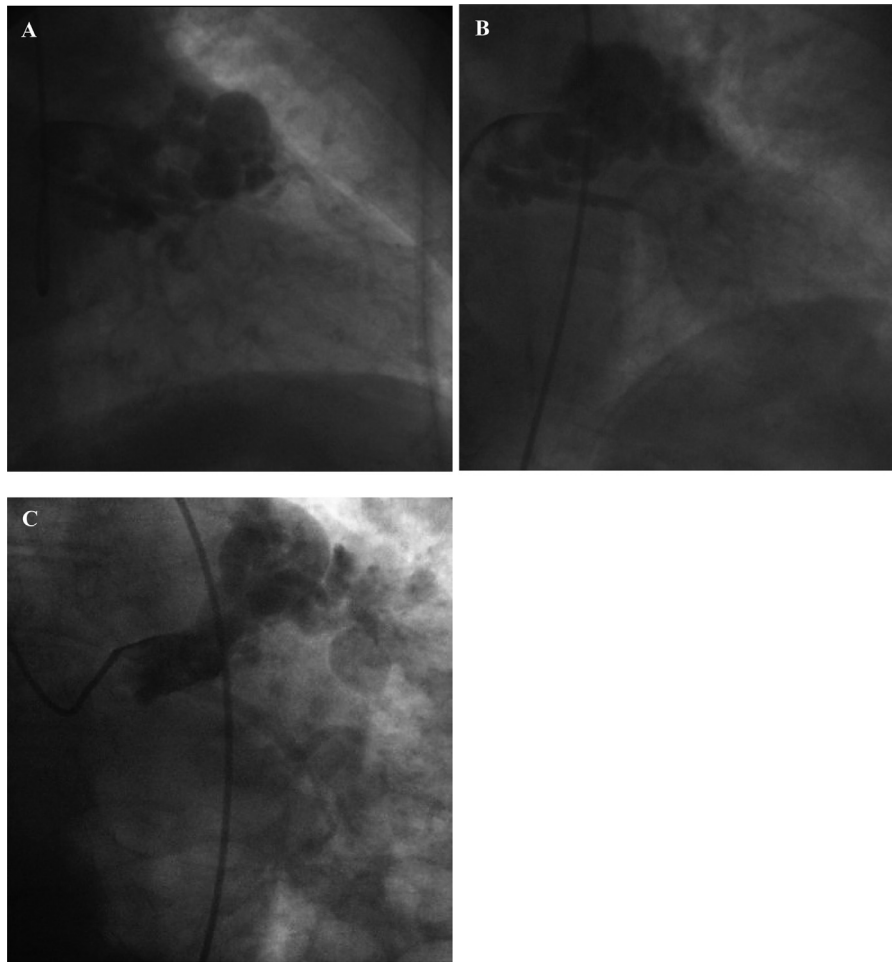


Fig. 1 – CXA of the left coronary-to-pulmonary artery fistula with saccular aneurysm.

CXA showed coronary-to-pulmonary artery fistula arising from proximal tract of left anterior descending artery with a giant saccular aneurysm in latero-lateral (A–B) and in antero-posterior (C) projections.

heart failure, and rupture or thrombosis of the fistula or associated arterial aneurysm [10].

Here we report an extremely rare case of 76-year-old woman with bilateral coronary-to-pulmonary artery fistulas associated with giant aneurysms, detected by coronary angiography and confirmed with CCT.

Case report

Patient's history

A 76-year-old Italian woman (weight 75.1 kg and height 165.8 cm) was referred to the Radiology Department of our Institution for fatigue, orthopnea, and chest pain. The symptoms had started 3 days before. She reported smoking a half pack per day of tobacco and suffered of metabolic syndrome. She denied alcohol or illicit drug use. She had family history for cardio-vascular death. She had a surgical history of cholecystectomy for lithiasis.

Physical exam

The patient's temperature was 34.3°C, heart rate was 90 bpm, respiratory rate was 16 breaths per minute, blood pressure was 150/90 mmHg, and oxygen saturation in room air was 99%. Chest inspection and excursion were normal. Lung auscultation was normal without any added noises. Cardiac auscultation revealed a continuous heart murmur.

Laboratory values

Blood analysis revealed normal hematocrit and platelet count. The blood biochemistries, as well as urine analysis were normal. Cardiac enzymes (troponin T and troponin I) were elevated. Creatinine value was 0.7 mg/dL and creatinine clearance was 94 mL/min (estimated with Cockcroft-Gault formula).

Imaging findings

Electrocardiography showed ST elevation with correct atrioventricular conduction (QRS 0.09 ms).

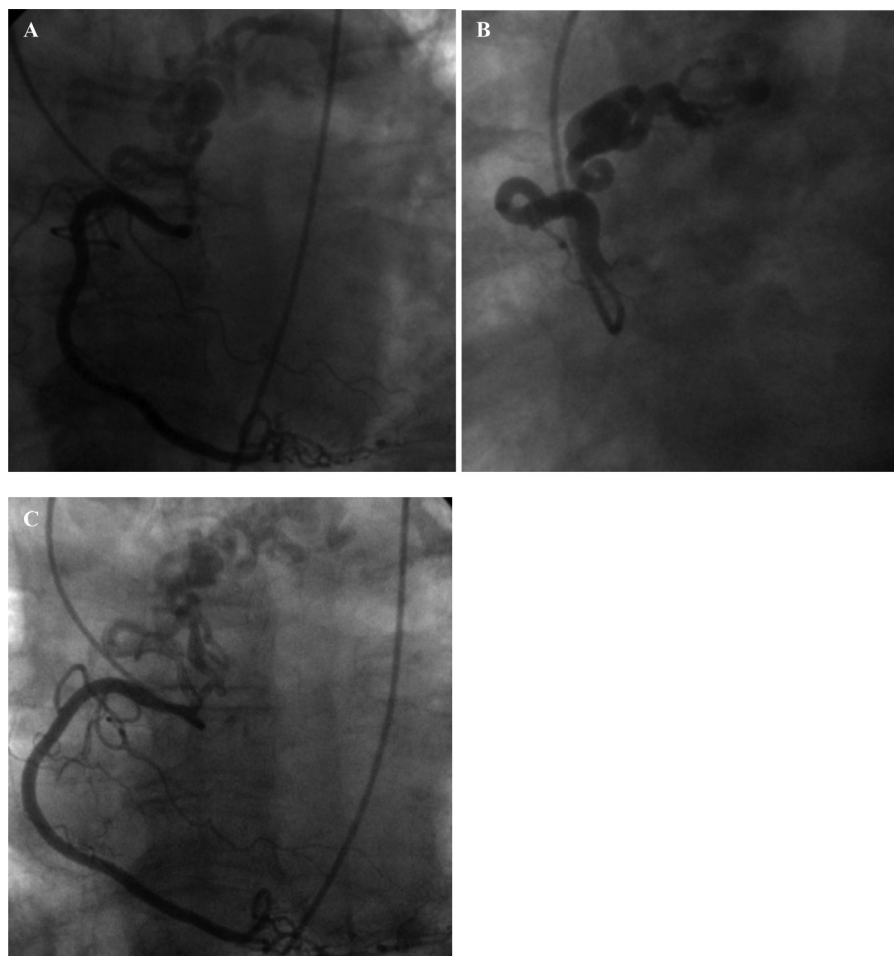


Fig. 2 – CXA of right coronary-to-pulmonary artery fistula. CXA showed coronary-to-pulmonary artery fistula arising from a branch of RCA in latero-lateral (A–B) and in antero-posterior (C) projections.

For this reason, in the suspicion of myocardial infarction, patient performed a coronary angiography that showed 1 coronary-to-pulmonary fistula arising from proximal tract of left anterior descending artery with a giant saccular aneurysm (Fig. 1) and another one arising from a branch of RCA (Fig. 2). To better evaluate the spatial position of the fistulas and aneurysm, patient was scanned using 64-detector CT (Light-speed VCT; GE Healthcare, Milwaukee, USA) with retrospective electrocardiogram (ECG) gating. An upper-extremity 20-gauge intravenous cannula was used for venous access. Heart rate control with a target of 60 beat per minute was achieved using 10-60 mg of propranolol, injected intravenous before data acquisition. Bolus tracking technique was used. A volume of 70 mL of contrast medium (Iopamidolo 370 mgI/mL) was injected, with a flow rate of 5 mL/s followed by 50 mL of saline solution at the same.

CT examination confirmed bilateral coronary-to-pulmonary fistulas from proximal tract of left anterior descending artery with a giant saccular aneurysm and from a branch of RCA. Left circumflex artery had regular course, but was ectasic. Volume rendering reconstructions were performed to help surgical planning (Fig. 3). Then, she performed

ECG-gated single photon emission computed tomography examination with a stress test that excluded akinetic and dyskinetic myocardial regions and confirmed valid ejection fraction (Fig. 4).

Surgical intervention

A median sternotomy was performed. A cardiopulmonary bypass with moderate hypothermia was used. Ligation of the fistulous tracts was done and aneurysm was resected. During the first postoperative day, the patient performed a chest X-ray that showed interstice-alveolar edema, moderate bilateral pleural effusion and very increase cardiac shadow. The patient died during the fourth postoperative day for surgical complications.

Discussion

A CAF is a congenital or acquired abnormal vascular communication of coronary artery with cardiac chambers or

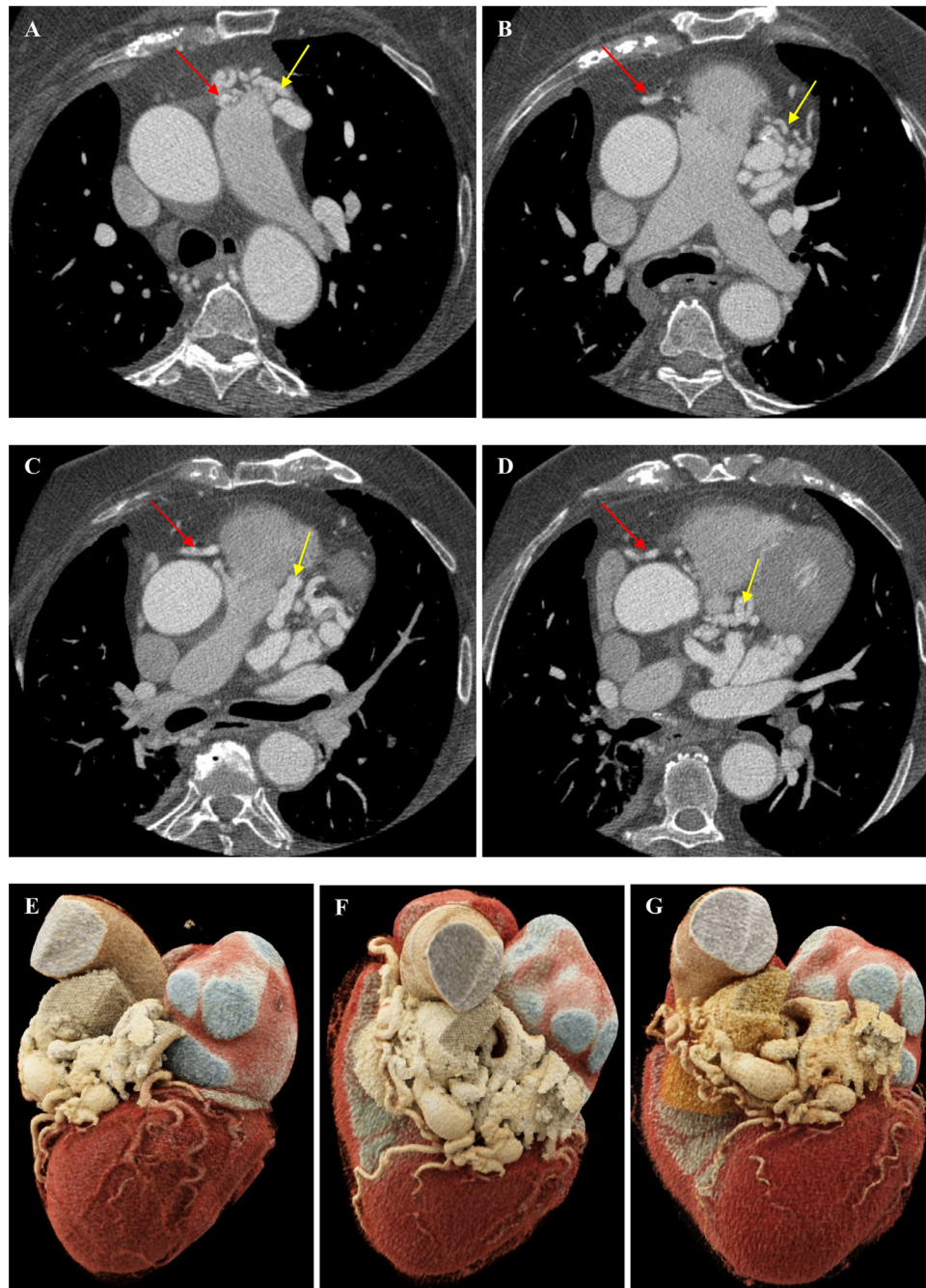


Fig. 3 – CCT of bilateral coronary-to-pulmonary artery fistulas.

CCT axial images go from cranial to caudal (A–D). CCT showed the drainage site (A), course (B–C), and origin (D) of the coronary-to-pulmonary artery fistula arising from left anterior descending artery (yellow arrow) and drainage site (A), course (B–C), and origin (D) of the coronary-to-pulmonary artery fistula arising from a branch of RCA (red arrow). Volume rendering reconstructions were performed to help surgical planning (E–G).

any segment of the systemic or pulmonary circulation. The prevalence of CAFs seen at CCT is reported to be 0.9% [2,3]. According to the number of fistulous connections, multiple coronary fistulas are 10.0%-16.0% of CAFs [4]. In 5% of patients, CAF arises from both RCA and left coronary artery [5]. The incidence of coronary-to-pulmonary artery fistulas accounting for 15.0%-30.0% of all CAFs [6,7]. Nineteen to twenty-five

percent of CAF are associated with arterial aneurysm [8]. The most common clinical presentation of CAF is a continuous heart murmur. If symptoms develop, most patients present later with dyspnea and right ventricular enlargement or dysfunction related to progressive enlargement of the fistula and an increase in the left to right shunting [11]. Selective coronary angiography used to be the reference standard for

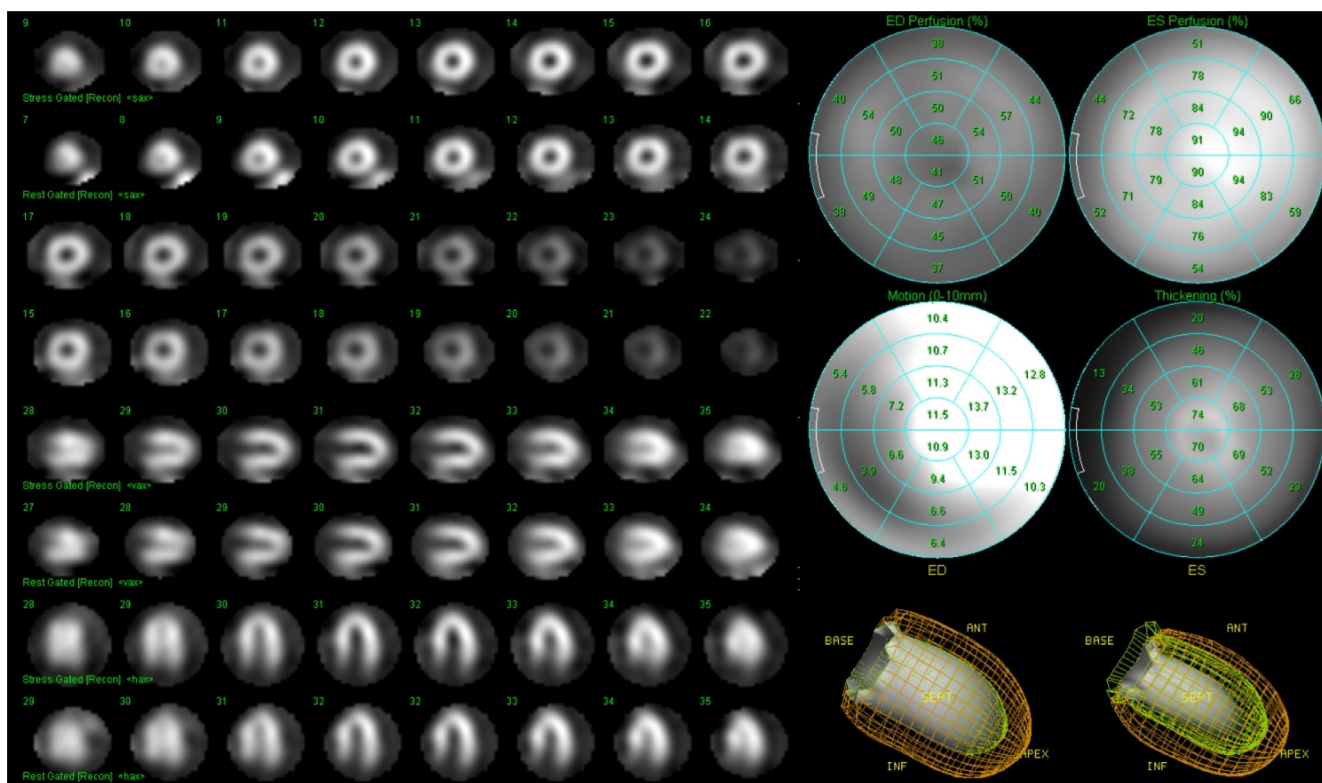


Fig. 4 – ECG-gated SPECT examination.
ECG-gated SPECT excluded akinetic and dyskinetic myocardial regions and confirmed valid ejection fraction.

assessment of CAFs. It enables precise visualization of the anatomy of the CAF, including fine vessels [12]. However, coronary angiography is invasive and involves risks of procedure related complications. Furthermore, it yields 2-dimensional projection images with reported correct diagnosis rates of 35%-50% [13]. CCT is useful for evaluation of CAFs because it involves a shorter acquisition time and yields higher temporal and spatial resolution and allow excellent anatomic information, including the origin, course, and drainage site of CAFs and serve as a basic guide for treatment planning [14]. Surgical ligation of the fistula is safe and effective, with good results. Transcatheter embolization of the fistulous connection is the nonsurgical treatment option [15]. The main indications for embolization are proximal location of the fistulous vessel, single drain site, extra-anatomic termination of the fistula away from the normal coronary arteries, older patient age, and absence of concomitant cardiac disorders requiring surgical intervention [16]. On the other hand, indications for surgery include a large CAF characterized by high fistula flow, multiple communications, very tortuous pathways, multiple terminations, significant aneurysmal formation, need for simultaneous distal bypass, or presence of large vascular branches that can be accidentally embolized [17].

In this work, we report an extremely rare case of a 76-year-old woman with bilateral coronary-to-pulmonary artery fistulas associated with giant aneurysms, detected by coronary angiography and confirmed with CCT.

Lee et al. reported a case of a 75-year-old woman with progressive chest discomfort, with her chest X-ray that showed

cardiomegaly with a bulging contour at right hilar area suspicious of anterior mediastinal mass. CCT showed huge intrapericardial aneurysm in the right atrioventricular groove that received blood from the mid-RCA, which drained into the main pulmonary artery and coronary angiography provided confirmation of RCA aneurysm, showed multiple varicosities involving the left anterior descending artery associated with bilateral coronary artery to the main pulmonary artery fistula [18]. Li et al. reported a case of a 66-year-old woman with mediastinum tumor. CCT showed a huge round mass in the cavum pericardii adjacent to the left ventricular. Coronary angiography revealed bilateral CAFs and a giant coronary aneurysm [19]. Fujimoto et al. presented the cases of an 83-year-old female with bilateral coronary-to-pulmonary fistulas aneurysms associated with a giant coronary aneurysm with calcifications.

Ethical standards

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the Helsinki Declaration of 1975, as revised in 1983. Informed consent was obtained from all individual participants included in the study.

Informed consent

Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Supplementary material

Supplementary material associated with this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.radcr.2019.04.020>.

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