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Letter to the Editor

A rare case of anomalous left coronary artery from the pulmonary artery (ALCAPA) presenting congestive heart failure in an adult

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Abstract

A 41-year-old woman with no modifiable coronary risk factors presented with a progressive exertional dyspnea. Chest radiography showed an enlarged cardiac silhouette with reinforced pulmonary vasculature in bilateral lower lung fields and both pleural effusion. Echocardiography revealed a dilated, globally hypokinetic left ventricle with an ejection fraction of 40%. Multislice cardiac computed tomography revealed abnormal origin of LCA from the main pulmonary artery receiving collaterals from a normally originating dilated tortuous right coronary artery. Coronary angiography revealed a single, large, and tortuous RCA arising from the right sinus of Valsalva and giving off extensive collateral vessels coursing over the right ventricular wall, the interventricular septum, and the apex to the left coronary artery that was drained into the proximal main pulmonary. The diagnosis of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) was confirmed. The patient underwent successful reimplantation of LCA to aorta after closure of the ostium (in the main pulmonary artery) of the anomalously originating LCA. After surgical and medical treatment, the patient's symptoms were relieved and both pleural effusions were improved on chest radiography. After then, the patient was discharged on medication.
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Anomalous origin of the left main coronary artery (LCA) from the pulmonary artery (ALCAPA), also known as Bland-White-Garland syndrome, is a rare malformation [1]. Few patients survive past childhood without surgical repair, and up to 90% die suddenly at a mean age of 35 years [2]. We describe a case of a 41-year-old patient who has a congestive heart failure with anomalous origin of the left main coronary artery from the pulmonary artery.

A 41-year-old woman with no modifiable coronary risk factors presented with a progressive exertional dyspnea, weight gain and pitting edema on both legs that had been aggravated for the last several months. On physical examination, a blood pressure of 100/80 mmHg with irregular heart rate of 104/min and a grade 3/6 holosystolic murmur was heard along the left sternal border and apex. Chest radiography showed an enlarged cardiac silhouette with reinforced pulmonary vasculature in bilateral lower lung fields and both pleural effusion (Fig. 1), while his ECG revealed atrial fibrillation with rapid ventricular rate. Echocardiography revealed a dilated, globally hypokinetic left ventricle with an ejection fraction of 40%, moderate MR and mild TR. CT angiogram (64 MDCT) revealed abnormal origin of LCA from the main pulmonary artery receiving collaterals from a normally originating dilated tortuous right coronary artery (Fig. 2A–D). There were no stenotic lesions. Coronary angiography revealed a single, large, and tortuous RCA arising

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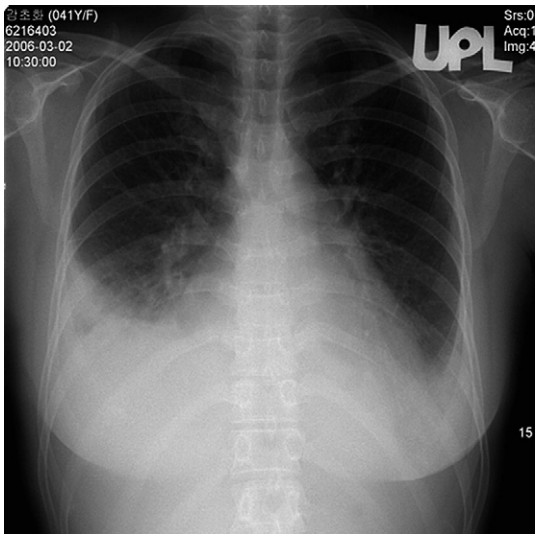

Fig. 1. Chest radiography showed an enlarged cardiac silhouette with reinforced pulmonary vasculature in bilateral lower lung fields with both pleural effusion.

from the right sinus of Valsalva and giving off extensive collateral vessels coursing over the right ventricular wall, the interventricular septum, and the apex to the left coronary artery that was drained into the proximal main pulmonary (Fig. 3A–D). There was no significant coronary artery disease and the left ventricular systolic function was decreased. The calculated left to right shunt (Qp:Qs ratio) was 1.02. The diagnosis of anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) was confirmed. The patient underwent successful reimplantation of LCA to aorta after closure of the ostium (in the main pulmonary artery) of the anomalously originating LCA (Fig. 4). Follow-up CT angiogram showed well positioning of left coronary artery at ascending aorta (Fig. 5A, B). After surgical and medical treatment, the patient's symptoms were relieved and both pleural effusion were improved on chest radiography (Fig. 6). After then, the patient was discharged on medication.

Anomalous left coronary artery from the pulmonary artery is a rare, congenital cardiac anomaly accounting for approximately 0.25–0.5% of all congenital heart disease [1]. It is usually an isolated cardiac anomaly but, in rare incidences, has been described with patent ductus arteriosus, ventricular septal defect, tetralogy of Fallot, and coarctation of the aorta [3]. Collateral circulation between the right and left coronary systems ensues after birth and the left ventricular myocardium is chronically underperfused, since flow is preferentially directed into the pulmonary vascular bed and away from the left ventricular myocardium due to the low pulmonary vascular resistance (coronary steal phenomena). Left untreated, the mortality rate in the first year of life is 90% secondary to myocardial ischemia or infarction and congestive heart failure [2,3]. In rare cases, the clinical presentation of myocardial ischemia may be delayed into early childhood, or even adulthood as in our patient, when intracardiac shunt is minimal.

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