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Giant Coronary Artery Aneurysm Following Takeuchi Repair for Anomalous Left Coronary Artery from the Pulmonary Artery

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Abstract

A 33 year old woman with anomalous left coronary artery arising from the pulmonary artery status post Takeuchi repair at age 7 presented for evaluation. The Takeuchi procedure creates an aortopulmonary window and an intrapulmonary tunnel that baffles the left coronary artery to the aorta. A mediastinal mass was identified as a giant aneurysm of the left coronary artery resulting in compression of the pulmonary artery and left upper pulmonary vein. The patient underwent open repair with patch closure at the aortic entrance of the left coronary Takeuchi repair and resection and evacuation of the aneurysm. A saphenous vein graft to the left anterior descending was performed. Postoperative echocardiography demonstrated normal left ventricular function. This is the first reported case of giant aneurysm formation following Takeuchi repair. Reported complications include the development of pulmonary artery stenosis at the intrapulmonary baffle, baffle leak, decreased left ventricular function, and mitral regurgitation. In conclusion, late complications of the Takeuchi procedure are common, underscoring the importance of lifelong follow-up at a center with experience in treating coronary anomalies.

Keywords

congenital heart disease; coronary anomaly; aneurysm

Anomalous left coronary artery from the pulmonary artery (ALCAPA) is a rare but serious congenital coronary abnormality first reported in 1885 by Brooks¹. It generally presents in infancy with features of cardiac ischemia or heart failure, though it can present as late as adulthood². As ALCAPA is associated with both morbidity and mortality, early surgical repair is recommended. However, given the evolution in surgical strategies over the last several decades, little is known about the types of complications that may occur later in life following ALCAPA repair.

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CASE DESCRIPTION

A 33 year old woman was found to have ALCAPA at age 7 years and underwent repair with the Takeuchi procedure³ without reported complications. Following repair, she was followed regularly as a child and was unaware of any abnormalities. As an adult, she did not receive regular care from a cardiologist. She successfully carried a twin pregnancy to full gestation without complications at age 31. In the year prior to presentation, she began experiencing palpitations and dyspnea on exertion and sought cardiovascular evaluation. Physical examination disclosed a soft precordial systolic ejection murmur. Chest radiograph showed a mediastinal mass abutting the left cardiac contour (Figure 1). Occasional ventricular premature complexes were seen on electrocardiogram and on Holter monitor. Echocardiogram revealed a large vascular mass overlying the main pulmonary artery that was partially thrombosed. Left ventricular size and ejection fraction were normal with no regional wall motion abnormalities. Chest computed tomography identified the mass as a giant (9×8cm) aneurysm of the left main coronary artery (Figure 2). The aneurysm had a small patent lumen but was otherwise thrombosed (Figure 3). It caused external compression of the pulmonary trunk and the left upper pulmonary vein. Chest radiograph at age 17 disclosed prominence of the left heart border.

Coronary angiogram demonstrated a mildly dilated right coronary artery with collaterals supplying most of the circumflex and left anterior descending coronary artery territories. The patient underwent successful open surgical repair with patch closure at the aortic entrance of the left main coronary artery Takeuchi repair and resection and evacuation of the largely thrombosed aneurysm. A single saphenous vein graft to the left anterior descending coronary artery was performed. Postoperative echocardiography demonstrated a left ventricular ejection fraction of 60% with no regional wall motion abnormalities.

COMMENTS

There have been no reported cases of giant aneurysm formation following Takeuchi repair for ALCAPA. Reported complications include the development of pulmonary artery stenosis in the location of the intrapulmonary baffle, baffle leak, decreased left ventricular systolic function, and mitral regurgitation⁴. Late complications of the Takeuchi procedure are common, underscoring the importance of lifelong follow-up at a center with experience in treating coronary anomalies.

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Figure 1. Chest Radiograph

A large mediastinal mass abutting the left cardiac contour is seen.

Figure 2. Three-Dimensional Computed Tomography Reconstruction of Giant Aneurysm

A three-dimensional volume rendered computed tomography image demonstrating the connection between the aorta and the giant left main coronary artery aneurysm is shown (unlabeled white arrow). The right coronary artery (RCA, arrow) was dilated and supplied most of the left coronary circulation territory.

Figure 3. Computed Tomography Images of Giant Aneurysm

The largely thrombosed aneurysm with a small patent lumen (*) is shown in the sagittal plane (panel A). The lumen (*) of the aneurysm communicates (white arrow) with the aorta (Ao, panel B). The giant aneurysm exerts mild compression on the main pulmonary artery (MPA, panel C). LV= left ventricle