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Case Report Open Access

A simple technique for reimplantation of an anomalous origin of the left coronary artery from the pulmonary artery in an adult

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ABSTRACT

An anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), which may cause the left coronary artery to grow with an anomalous origin from the pulmonary artery, is a rare disease associated with sudden cardiac death and ventricular arrhythmias in adults. Herein, we report a 43-year-old female case of ALCAPA syndrome as assessed by cardiac computed tomography angiography with three-dimensional reconstruction and echocardiography which was corrected by a simple surgical technique. We present this case due to its unusual presentation in an adult.

Keywords: ALCAPA; coronary artery; correction; surgery.

The anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA), which was first described in 1866, is a rare congenital cardiac malformation affecting one of every 300,000 live births.[1] The first clinical description, in conjunction with autopsy findings, was described by Bland et al.[2] in 1933 and the rare congenital anomaly is also called as the Bland-White-Garland syndrome in the literature.

There are two types of ALCAPA syndrome: adult type characterized by well-established collaterals and infantile type characterized by no collaterals. Nearly 90% of infants who experience myocardial infarction and congestive heart failure die within the first year of life.[3] In the adult type, which is seen only 10 to 15% of patients, it may be an important cause of sudden cardiac death and ventricular arrhythmias.[3]

Herein, we present an unusual case of ALCAPA in an adult which was corrected by a simple surgical technique.

CASE REPORT

A 43-year-old woman was referred to our hospital with clinical continuous heart murmur. She had no previous history of cardiac diseases. On admission, she complained about palpitation during heavy exercises.

Physical examination revealed continuous murmur in the left side of the sternum. Chest X-ray showed cardiomegaly and transthoracic echocardiography demonstrated a normal right ventricle with a mildly dilated left ventricle. Left ventricular ejection fraction, measured in M mode, was 65%. No septal defects were found. Color Doppler imaging revealed only mild mitral regurgitation, which was likely to originate from the ventral side of the aorta with prominent laminar diastolic flow compatible with coronary artery flow. Conventional angiography demonstrated no coronary artery stenosis. A written informed consent was obtained from the patient.

Cardiac computed tomography angiography with three-dimensional reconstruction (Figure 1) and echocardiography demonstrated an anomalous origin of the left coronary artery (LCA) from the posterior pulmonary sinus of the main pulmonary artery (PA) with diffusely enlarged and tortuous coronary arteries.

Surgical technique

The heart was approached through median sternotomy. Cardiopulmonary bypass was established with standard aortic and bicaval venous cannulation. Moderate hypothermia (26 °C) was used. After both great arteries were clamped, cold blood cardioplegia

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was injected directly into the ascending aorta as well as into the PA. After both great arteries were detached and the PA was transected above the level of commissures, the anomalous LCA ostium was identified arising from the lateral aspect of the posterior-facing sinus. The anomalous LCA with a button of PA was excised. Despite significant mobilization, critical tension on the anomalous LCA persisted, as the distance between it and the aorta was much higher than anticipated. The PA is re-anastomosed at the side of the transection. The aortic cross-clamp was off. We prepared a 5 cm saphenous vein graft for the proximal portion of the saphenous vein anastomosis into the coronary button. We passed the saphenous vein graft through transverse sinus to the lateral aspect of the aorta. We put side-clamp and performed proximal anastomosis of the saphenous vein graft (Figure 2). The postoperative period was uneventful and the patient was discharged in the fifth postoperative day with anticoagulant regimen.

At the final follow-up visit, she was in New York Heart Association class 1. The postoperative left ventricular ejection fraction was around 60%.

DISCUSSION

A variety of surgical approaches have been proposed in adults with ALCAPA, including ligation of the LCA,

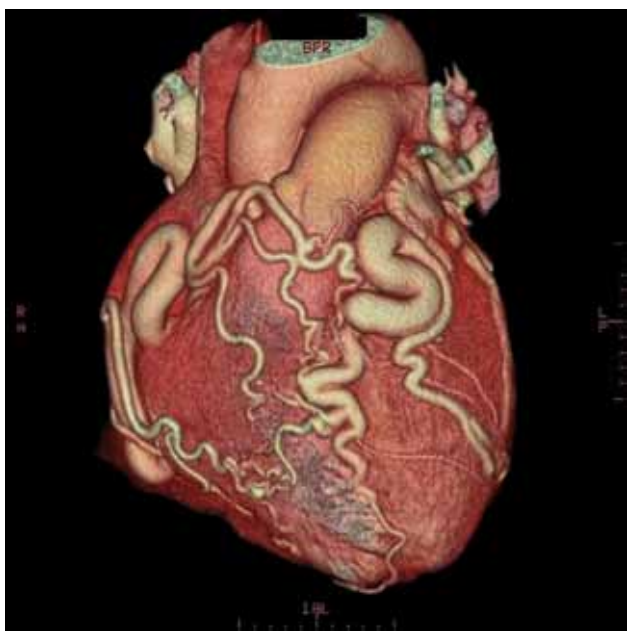

Figure 1. Cardiac computed tomography angiography with three-dimensional reconstruction showing the origins of coronary arteries. The left coronary artery originates from the pulmonary artery.

reimplantation of the LCA to the aorta, creation of an aorta pulmonary window with an intrapulmonary buffle (Takeuchi procedure), and a combination of LCA ligation and coronary artery bypass grafting.[4,5]

Recently, the first case of a giant aneurysm formation after Takeuchi procedure was reported.[6] In this case, a saphenous vein graft into the LCA was performed. The reported complications of Takeuchi repair include the development of PA stenosis at the intrapulmonary baffle, baffle leak, decreased left ventricular function, and mitral regurgitation.[6] Thus, late complications of the Takeuchi procedure are common.

In 1974, Neches et al.[7] were the first to describe the direct reimplantation of the anomalous LCA into the aorta by transferring it with a button of PA. However, direct reimplantation of the LCA may be technically more challenging and hazardous in adults due to its distant, less elastic, and friable nature of the coronary arteries.[8] Creative methods such as direct transfer, tubular reconstruction, and in situ transfer can be used in adults, regardless of the site of the LCA orifice.[9]

It is well known that ALCAPA in elderly is not suitable for direct reimplantation, unless the orifice of the LCA really close to the inner curvature of the aorta, which is extremely small in the minority of patients.[8] In this case, reimplantation of the LCA into the aorta was considered unfeasible due to the distance between the insertion site of the LCA on the PA and the aorta. The main advantage of this method is that the graft run in the anatomical groove behind the PA through transverse sinus.

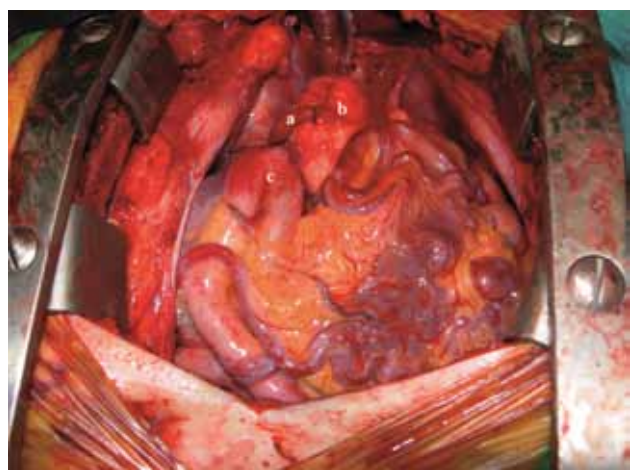

Figure 2. Surgical technique of direct implantation of left coronary artery with a saphenous vein graft.
a: Saphenous vein graft; b: Aorta; c: Right coronary artery.

As we are aware of that the saphenous vein may not be fully patent like the internal thoracic artery in coronary artery bypass grafting, we used the possibly largest conduit - a saphenous vein graft - to match the size of the LCA in our case.

In conclusion, this technique provides tension-free venous graft for the transfer of anomalous origin of the left coronary artery from the pulmonary artery into the aorta, when the anomalous left coronary artery ostium is located at a distance from the aorta in adults.

Declaration of conflicting interests

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