Asymptomatic anomalous origin of left anterior descending artery from the pulmonary artery and multiple atherosclerotic stenoses revealed by silent ischaemia

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Abstract

Anomalous origin of the left coronary artery from the pulmonary artery is a rare congenital disease responsible for a high mortality rate in childhood. Here, we report for the first time the case of an asymptomatic 61-year old patient showing a combination of anomalous origin of the left anterior descending artery from pulmonary artery and atherosclerotic stenosis in both the right coronary artery and the left circumflex artery without anomalous origin.

Keywords: Anomalous origin of the left coronary artery from the pulmonary artery • Anomalous origin of the left anterior descending artery from pulmonary artery • Atherosclerosis • Congenital heart disease • Coronary bypass

INTRODUCTION

Bland–Garland–White syndrome, or anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) syndrome, is a rare congenital heart disease with an incidence of 1 in 300 000 live births and found in 0.5% of children with congenital heart disease [1]. There is usually no manifestation before birth because systemic and pulmonary artery pressures are equal. Clinical presentation depends on coronary artery topography. In cases with a dominant left coronary artery, ischaemic congestive heart failure or mitral insufficiency by annular dilatation reveals this disease early. Without treatment, the mortality rate is very high and death occurs during childhood [2]. When the right coronary artery (RCA) is dominant, sudden death of up to 90% of patients is observed at a mean age of 35 years and results from myocardial ischaemia, heart failure or malignant arrhythmia [3]. Rarely, this disease can be revealed at a later stage.

We report here an original case of an anomalous origin of left anterior descending artery from the pulmonary artery (ALADAPA) associated with atherosclerotic stenosis in the RCA and the left circumflex artery (LCX) revealed by diffuse silent ischaemia before carotid endarterectomy.

CASE DESCRIPTION

A 61-year old patient was admitted to the neurology department for acute ischaemic stroke. His cardiovascular risk factors were type II diabetes mellitus, smoking, arterial hypertension and obesity.

Carotid ultrasound revealed a severe stenosis of the left internal carotid, requiring surgical treatment. The patient underwent a preoperative cardiological evaluation. He did not report any symptom before the stroke and was not taking any medication. Transthoracic echocardiography was normal. Myocardial SPECT revealed five anterior ischaemic segments and the coronary angiography showed severe stenosis of the RCA and the second marginal of the LCX (Fig. 1A and B, white arrows). The left anterior descending artery (LAD) appeared as a chronic total occlusion with collateral filling [Fig. 1B (red arrow) and C], and seemed to arise from the pulmonary artery.

Coronary computed tomography (CT) showed an isolated anomalous origin of the LAD from the pulmonary artery without any atherosclerotic stenosis (Fig. 2A). The heart team’s strategy was to treat the left carotid stenosis first and then to perform myocardial revascularization by coronary artery bypass graft (CABG). In accordance with the latest guidelines, CABG was performed a few weeks following the left carotid endarterectomy, with the left mammary artery to the LAD, right mammary artery to the second posterolateral branch and the saphenous vein to the posterior descending artery. Intra-operative findings confirmed origin of LAD from pulmonary artery (Fig. 2B). The left ventricular ejection fraction remained normal and graft patency was assessed by cardiac CT within days following the surgical procedure (Fig. 2C).

DISCUSSION

The clinical presentation of the ALADAPA syndrome in the adult population varies. Symptoms are related to chronic myocardial
ischaemia leading to ventricular arrhythmia, syncope, dyspnoea, angina or heart failure [4]. CT angiography can detect the ALADAPA. The usual treatment comprises surgical correction by CABG or rerouting through an aorto-pulmonary window (Takeuchi procedure). Here, CABG was required because of the combination of atherosclerotic stenosis and ALADAPA. The long-term outcome after revascularization is excellent [5].

We report an original case of coronary artery disease revealed by silent myocardial ischaemia resulting from an association of ALADAPA and atherosclerosis, with a late manifestation (patient aged over 60 years) and treated by CABG. To date, at the 1-year follow-up, the patient remains asymptomatic without residual myocardial ischaemia.

Conflict of interest: none declared.

REFERENCES


eComment. Combined surgical strategies for anomalous connection of coronary artery to pulmonary artery in adults

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We read with great interest the article by Gurbuz et al. [1]. We congratulate them on their successful effort in the surgical treatment of such a rare case, presenting with both carotid artery stenosis and coronary artery disease accompanied by an anomalous origin of the left coronary artery from pulmonary artery (ALCAPA). In fact, the traditionally-named ALCAPA leads to a left-to-right shunting into pulmonary arteries, resulting in ventricular ischaemia [2]. Although the terminology defines an origin of left anterior descending artery (LAD) from pulmonary artery, the actual flow direction of the blood is from the coronary circulation to main pulmonary artery. This reversed coronary flow leads to a coronary steal phenomenon. In general, in such cases, re-establishment of a dual coronary circulation is the preferred treatment modality [2].