Case report - Pulmonary
Pulmonary artery dissection associated with multiple coronary–pulmonary artery fistulae

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Received 26 January 2010; received in revised form 4 April 2010; accepted 13 April 2010

Abstract

We experienced the surgical repair of an extremely rare pulmonary artery dissection without pulmonary hypertension. The patient had multiple coronary–pulmonary artery fistulae, which presumably caused pulmonary artery dissection. The surgical treatment included the closure of the multiple coronary fistulae and the resection of the intimal flap in the main pulmonary artery. The patient recovered uneventfully.

Keywords: Pulmonary artery dissection; Coronary artery fistula

1. Introduction

Pulmonary artery dissection is an extremely rare pathology. It usually develops in a pulmonary artery aneurysm/dilatation associated with underlying pulmonary hypertension [1–6]. The condition manifests as cardiogenic shock or sudden death. It is then usually diagnosed postmortem. In this report, we first present an extremely rare case developing pulmonary artery dissection without pulmonary hypertension, presumably caused by multiple coronary–pulmonary artery fistulae.

2. Case report

A 56-year-old woman having palpitation was referred to our institution with a diagnosis of multiple coronary artery fistulae. Enhanced computed tomography (CT)-scans demonstrated multiple fistulae drained from the both coronary arteries, the ascending aorta, and the left subclavian artery into the main pulmonary artery (Fig. 1). Pulmonary artery dissection extending from the main pulmonary artery to the left pulmonary artery was also demonstrated (Fig. 1). Echocardiography also demonstrated that blood flow in the dissecting lumen of the pulmonary artery originated from multiple fistulae. Dipyridamole myocardial scintigraphy did not show any ischemic findings. Cardiac catheterization did not reveal pulmonary hypertension. Pulmonary to systemic blood flow ratio (Qp/Qs) was 1.53. Coronary artery angiography showed multiple huge coronary–pulmonary artery fistulae originating from the both coronary arteries.

At surgery, the heart was exposed through a median sternotomy. Huge coronary artery fistulae which drained to the main pulmonary artery were found. The origins of the fistulae on the ascending aorta and the left subclavian artery were also identified easily and ligated by a suturing technique, after the establishment of a cardiopulmonary bypass. The fistula from the right coronary artery was also ligated at the origin, although the origins of the left coronary artery fistulae were not identified from the outside. After the cardiac arrest with antegrade and retrograde cardioplegic infusion, the main pulmonary artery was longitudinally incised, and the intimal flap was found. The dissection was extended to the proximal part of the bilateral pulmonary arteries, and ‘re-entries’ were seen on each side. There were also three small holes of 5 mm in diameter on the intimal flap, in accordance with three orifices of the multiple fistulae (Fig. 2). Two orifices of fistulae from the left coronary artery, and one from the right coronary artery were identified in the dissected lumen. All of them were closed by 5-0 mattress sutures. After resection of the intimal flap, another orifice of fistula was also seen in the true lumen, which was closed. Finally, the pulmonary artery was closed with a 5-0 running suture. Weaning from the cardiopulmonary bypass was uneventful. The patient was transferred to the intensive care unit in a stable condition. CT-scans on postoperative day 5th showed neither residual dissection nor coronary artery fistulae. However, as there were some thrombi in the left coronary artery, oral administration of aspirin and coumadin were started.

3. Discussion

Pulmonary artery dissection is usually diagnosed in patients having pulmonary hypertension including Eisenmenger syndrome and/or pulmonary artery aneurysm [1–6]. In this case, however, pulmonary hypertension was not
Fig. 1. Enhanced CT-scans demonstrated multiple coronary artery fistulae drained from the bilateral coronary arteries, the ascending aorta, and the left subclavian artery into the main pulmonary artery (left). Pulmonary artery dissection extending from the main pulmonary artery to the left pulmonary artery was also demonstrated (right). CT, computed tomography.

demonstrated. There was only one report on two cases developing pulmonary artery dissection in the settings without pulmonary hypertension [7]. Connective tissue disease related to fragility of the pulmonary arterial wall was also dismissed from the postoperative pathological findings in the present case. However, this patient had multiple huge fistulae from the both coronary arteries connecting to the dissecting (false) lumen of the main pulmonary artery. We believe that the systemic pressure from these multiple fistulae made intimal degeneration of the pulmonary artery around the orifices of fistulae, which may cause dissection. There is an interesting report on pulmonary artery dissection associated with aorto-pulmonary window [8]. However, there have been no similar cases with pulmonary artery dissection due to coronary artery fistulae.

The surgical treatment for pulmonary artery dissection was a simple resection of the intimal flap. It was rather difficult to close the coronary artery fistulae because of the multiple inflow sites. The postoperative CT-scans revealed thrombus formation in the left antero-descending artery due to flow-reduction caused by the closure of the outflow orifice of fistulae. Fortunately, the patient had no ischemic findings on the following myocardial scintigraphy and has had an uneventful postoperative course with administration of coumadin and aspirin. Close observation is required in the future.

References