Case report

External compression of bronchus by aneurysm from divided major aortopulmonary collateral artery after unifocalization

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Abstract

The right ventricle to pulmonary artery connection with an extracardiac conduit, left pulmonary artery reconstruction, ligation of patent ductus arteriosus, and take-down of right Blalock–Taussig shunt were performed on a 1-year-8-month-old boy who had pulmonary atresia, ventricular septal defect, patent ductus arteriosus, and major aortopulmonary collateral arteries. He previously underwent the unifocalization and right modified Blalock–Taussig shunt at 9 months of age. He repeatedly had a difficulty in weaning from the mechanical ventilator. After removing the aneurysm from the divided major aortopulmonary collateral artery that compressed the left main bronchus externally, it was possible to wean him from the mechanical ventilator. © 2001 Elsevier Science B.V. All rights reserved.

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1. Case report

A 1-year-8-month-old boy who had a tachypnea since 1 week of age was diagnosed as pulmonary atresia (PA) with ventricular septal defect (VSD), patent ductus arteriosus (PDA), and two major aortopulmonary collateral arteries (MAPCAs). One MAPCA from aortic arch communicated with right upper lobe, and the other from descending thoracic aorta communicated with right lower lobe.

First operation was performed at 9 months of age. Unifocalization of a MAPCA from aortic arch, ligation of a MAPCA from descending aorta, and right modified Blalock–Taussig (B–T) shunt through right posterolateral thoracotomy were performed. In angiography prior to second operation, an aneurysm on the stump site of MAPCA from aortic arch was found (Fig. 1). Nine months after the first stage operation, complete correction of PA and VSD using an extracardiac conduit, left pulmonary artery reconstruction, ligation of PDA, and take-down of B-T shunt were performed. However, trial to wean from ventilator failed due to desaturation and carbon dioxide retention, which induced tachypnea and bowel ileus. He was stable under synchronized intermittent mandatory ventilation mode, but the above situation repeated under continuous positive airway pressure or T-piece trials. Ultrastuff computerized tomographic scan revealed that both pulmonary arteries were in normal sizes without stenosis but the left main bronchus had stenosis by external compression. Fibroptic bronchoscopy was performed, which showed an external pulsating mass compressing the posterior portion of the left main bronchus. In cardiac catheterization and angiography, previous aneurysm was not found. On postoperative 22nd day, aneurysmectomy was performed via redo median sternotomy. The operative findings showed that the aneurysm was located between posterior portion of the origin of aortic arch and the upper portion of pulmonary artery. After the ligation of feeding vessel, the wall of the aneurysm was incised and about 3 cc of thrombi were removed. Postoperative bronchoscopy showed that the stenosis still remained but bronchoscope passed without resistance, and there was no pulsation. It was possible to wean him from mechanical ventilation on 6th day after aneurysmectomy.

2. Discussion

Complications of unifocalization were reported as hemothorax, hemoptysis, cardiac tamponade, pulmonary emboli, esophageal bleeding, and tracheal bleeding [1]. In the absence of intersegmental cross circulation, the potential for pulmonary infarction exists [2]. Puga and associates reported that one patient died 3 months after operation of erosion of the central unifocalization graft into the right main bronchus, resulting in catastrophic hemoptysis [3]. We report for the first time that the aneurysm originated...
from divided MAPCA from aortic arch after unifocalization and compressed the left main bronchus.

To prevent this complication, the method of surgical approach should be considered. The central approach, usually performed through a median sternotomy, emphasizes growth of the central pulmonary arteries. The peripheral surgical approach, usually performed through a lateral thoracotomy, emphasizes rationalization of pulmonary blood flow [4]. In this case, the MAPCA from aortic arch had been ligated through right posterolateral thoracotomy as the peripheral approach. Through this approach, the ligation of the MAPCA at its origin had been impossible. It is supposed that the MAPCA from aortic arch should be ligated at the origin of MAPCA through the central approach or contralateral thoracotomy.

Secondly, whether the remnant of MAPCA after ligation or unifocalization may cause the compression over surrounding organs should be considered. The aneurysm was found in angiography before second operation. As it didn’t induce any problem, we had decided not to remove the aneurysm. Retrospectively, if it had been removed simultaneously during second stage operation, this complication would have been prevented.

Incidence of MAPCAs from aortic arch was reported as 1.7% [5]. By acquiring the exact information of pulmonary circulation before unifocalization, it is important to decide the most appropriate surgical approach. The ligation of MAPCA originating from aortic arch needs more meticulous attention.

References