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CASE REPORT

Subclavian artery pseudoaneurysm long after the division of modified Blalock–Taussig shunt

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Abstract
A massive but asymptomatic left subclavian artery pseudoaneurysm was diagnosed in a 30-year-old female patient with transposition of the great arteries, ventricular septal defect, and pulmonary atresia. After undergoing bilateral modified Blalock–Taussig shunts at the age of 4 months and 3 years, respectively, she underwent the Rastelli operation and division of both shunts at the age of 6 years of age. The pseudoaneurysm was not discovered at the follow-up investigation at age 14. During the time period from age 18 to 30 years, she was lost to follow-up, she was confirmed to be free from infectious disease, traumatic accident, or vasculitis. Pregnancy-induced hypertension was not associated during her pregnancy. Graft replacement of the left subclavian artery and redo right ventricular outflow tract reconstruction were successfully performed under deep hypothermic circulatory arrest at the age of 33 years.

Keywords Left subclavian artery pseudoaneurysm · Modified Blalock–Taussig shunt · Conotruncal anomaly

Introduction

Although pseudoaneurysm formation on the subclavian artery after placement of a modified Blalock–Taussig shunt (mBTS) was not rare, cases of a subclavian artery pseudoaneurysm that developed more than 10 years interval from mBTS have never been reported, excepting for one literature [1]. In that report, all patients complained of hemoptysis and shuts were not divided but simply ligated. This paper describes a patient with a gigantic but asymptomatic subclavian artery pseudoaneurysm discovered incidentally, after 24 years interval from the division of a BTS, while undergoing perinatal investigations.

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Case report

A 30-year-old female with transposition of the great arteries, ventricular septal defect, pulmonary atresia and juxtaposed right atrial appendage, who had undergone the Rastelli operation at the age of 6 years, was readmitted to our outpatient clinic for perinatal management. She had no history of pregnancy and delivery before admission. She had been followed-up at our outpatient clinic until she was 18 years old, after which she relocated to a different area and follow-up was discontinued. A chest X-ray taken when she was 14 years old showed no specific findings except for a cardiomegaly and a calcified outflow patch (Fig. 1a). She had previously undergone two mBTS; a right BTS with a 4-mm expanded polytetrafluoroethylene graft via right posterolateral thoracotomy at the age of 4 months of age, followed by a left mBTS with a 5-mm knitted Dacron graft via left posterolateral thoracotomy at the age of 3 years. Both shunts were completely divided during the Rastelli operation.

On arrival, echocardiography detected an elevated right ventricular pressure due to right ventricular outflow tract obstruction. Two years later, when she was 32 years old, a massive lesion in the left upper mediastinum was first noticed on a chest X-ray and retrospective review of the patient's chest X-rays revealed that the lesion had been already observed at the previous admission, when she was

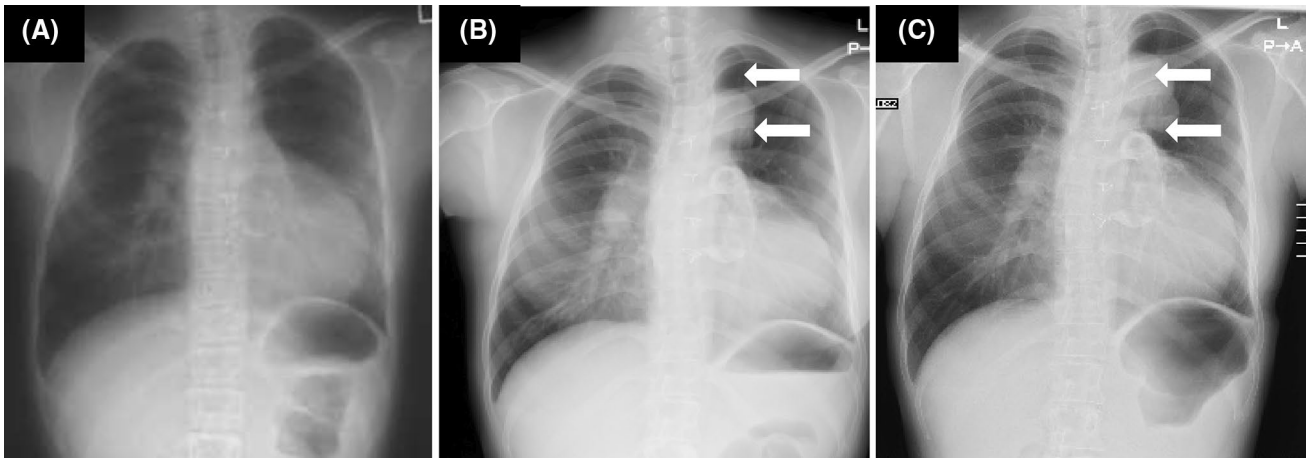

Fig. 1 Chest X-ray (**a**) showed calcified outflow patch and cardiomegaly only when she was 14 years old, but an abnormal mass lesion (white arrows) in the left upper mediastinum was observed when she was 30 years old (**b**), and it increased when she was 32 years old (**c**)

30 years old (Fig. 1b, c). Enhanced three-dimensional (3D) computed tomography showed a pseudoaneurysm (53 mm in diameter) that originated from of the left subclavian artery (Fig. 2a) where the previous mBTS was anastomosed. Subsequent cardiac catheter examination showed that the right ventricular systolic pressure was about 70% of the systemic arterial pressure. The diameter of the ascending aorta was 41 mm. The patient had no pregnancy-induced hypertension, with her systolic blood pressure kept under 100 mmHg in the third trimester of pregnancy.

Pseudoaneurysm exclusion, replacement of left subclavian artery, and right ventricular outflow tract reconstruction were performed when the patient was 33 years of age. Following a femoro-femoral cardiopulmonary bypass, a median full sternotomy was extended above the left clavicle. The sternocleidomastoid was divided to identify the distal subclavian artery. The right ventricular outflow tract, distal aortic arch, and distal left subclavian artery were carefully dissected and successfully encircled. Under the deep hypothermic circulatory arrest, the left pleural space was explored and pseudoaneurysm sac was opened. The previously placed shunt graft was not found in the aneurysm. The aneurysm sac adhered strongly to the left upper lobe and could not be removed. The left subclavian artery was then replaced with a 6-mm ringed expanded polytetrafluoroethylene graft. The right ventricular outflow tract was reconstructed with a 24-mm expanded polytetrafluoroethylene graft with handmade trileaflet valves. Postoperative

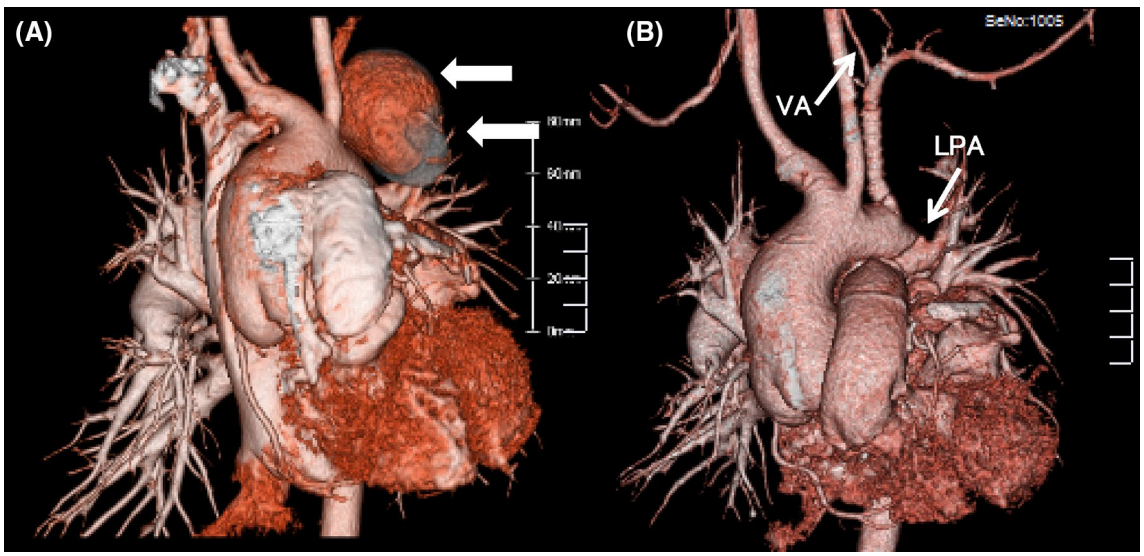

Fig. 2 Enhanced 3D computed tomography findings before (**a**) and after (**b**) the replacement of left subclavian artery. White arrows indicate pseudoaneurysm. *VA* vertebral artery, *LPA* left pulmonary artery

enhanced 3D computed tomography showed a patent subclavian graft, vertebral artery and distal subclavian artery (Fig. 2b).

Comment

As far as we reviewed, pseudoaneurysm that formed after more than 10 years interval from the division of systemic to pulmonary shunt using artificial graft has never been reported. Although the subclavian artery pseudoaneurysm can occur by infection, traumatic accident, weakness of the artery and so on [2]; all of which was not seen with this patient. Relation of pregnancy-induced hypertension was also suspected [3], but she was free from preeclampsia. Previous surgical stress tension caused by the suture shunt graft to the subclavian artery itself might be triggered formation of the pseudoaneurysm. Our case suggests that period check-ups using chest X-rays are recommended after division of the shunts for the patient's entire life.

The indication for the endovascular repair was discussed before surgery. At the time, covered stent grafts were not formally permitted in Japan for small caliber arteries such as the subclavian artery [4]. Occlusion of the left subclavian artery by the thoracic endovascular stent graft and individual reconstruction of the vertebral artery and distal subclavian artery was an alternative option. Loss of vascular elasticity by inserting the endovascular stent graft is known to increase the shear stress on the proximal aortic wall, which can sometimes resulting in retrograde dissection or aneurysm formation [5]. Especially in patients with conotruncal anomalies, aortic root dilatation and/or systemic semilunar valve regurgitation are known to progress naturally [6]. Thoracic endovascular repair was, therefore, thought to be a contraindication.

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Compliance with ethical standards

Conflict of interest The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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