Pseudoaneurysm Following Modified Blalock-Taussig Shunt : A Rare Complication Mimicking Pulmonary Disease

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Abstract

The authors report a pseudoaneurysm in a 2-year-old boy presenting with fever, increasing cyanosis and right upper lung shadowing on a chest radiograph at six weeks following modified Blalock-Taussig shunt surgery. Echocardiography and a CT scan of the chest revealed a large pseudoaneurysm originating from the right subclavian artery at the proximal insertion of modified Blalock-Taussig shunt. The patient underwent aneurysmal resection, Blalock-Taussig shunt removal, right subclavian artery ligation and the creation of a central shunt between the ascending aorta and main pulmonary artery. Unfortunately, the patient died 3 hours after the operation.

Key word : Pseudoaneurysm, Blalock-Taussig Shunt

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The modified Blalock-Taussig (B-T) shunt, created by interposing a polytetrafluoroethylene (PTFE) graft between the subclavian and pulmonary arteries, has been used widely as a palliative shunt in neonates and infants with cyanotic congenital heart malformations[1]. Although the modified B-T shunt is associated with a low incidence of failure, complications include serous fluid leakage, diaphragmatic

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paralysis, chylopericardium and excessive pulmonary blood flow[2]. Pseudoaneurysm formation is a rare complication that can lead to death[3]. In this report, the authors describe a patient with pseudoaneurysm formation referred to our hospital 6 weeks after modified B-T shunt creation.

CASE REPORT

A 25-month-old boy was referred with fever, dyspnea, increasing cyanosis and disappearance of shunt murmur. He was diagnosed as having tetralogy of Fallot at 13 months old and underwent modified right Blalock-Taussig shunt creation (PTFE graft) at 24 months of age, at Chiang Mai University Hospital, due to hypoxic spells that were unresponsive to oral propranolol therapy. The operation was uneventful and the patient was discharged on the third day post-operatively on 4 mg/kg of aspirin once daily. Four weeks later he presented with fever and increasing cyanosis at a local hospital. His chest radiograph revealed right upper lobe shadowing. In spite of having two weeks of appropriate antibiotic treatment, he remained pyrexic and his lung shadow persisted on a chest radiograph. An ultrasound of the chest revealed a large right upper lung mass with collapsed surrounding lung tissue and a small amount of fluid in the right pleural space. The patient developed progressive cyanosis and dyspnea which eventually required endotracheal intubation. Upon arrival at our hospital, echocardiography was performed immediately. The study revealed a pseudoaneurysm of at least 5 cm in diameter originating from the right subclavian artery at the site of the shunt (Fig. 1). A color Doppler turbulent flow was demonstrated at the opening of the aneurysm but not in the B-T shunt. Chest radiograph showed dense opacity in the right upper lung field (Fig. 2). The aneurysm was also shown on a CT scan of the chest (Fig. 3). The operation was performed through a right thoracotomy. There was a large pseudoaneurysm occupying the entire right upper and mid-chest. The graft shunt was partially dehiscent from the subclavian artery and displaced anteriorly (Fig. 4). The aneurysm was resected, the PTFE graft removed, the subclavian artery ligated,

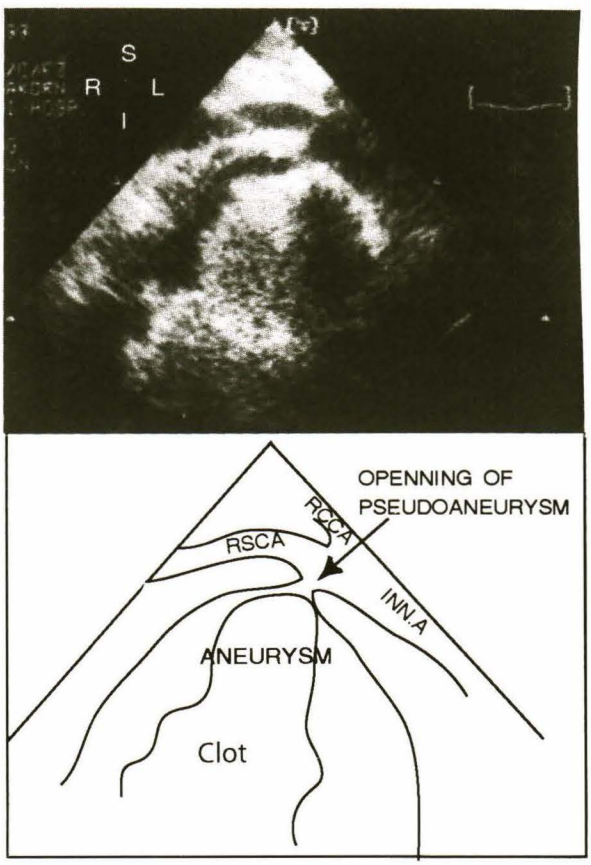

Fig. 1. Echocardiogram in suprasternal notch view showing a large pseudoaneurysm with its opening from the right subclavian artery (RSCA). A large clot in the aneurysm is demonstrated as a hyperechoic mass. The BT shunt is not visualized in this frontal plane because it was partially dehiscent and no flow across the shunt. RCCA = right common carotid artery; INN.A = innominate artery.

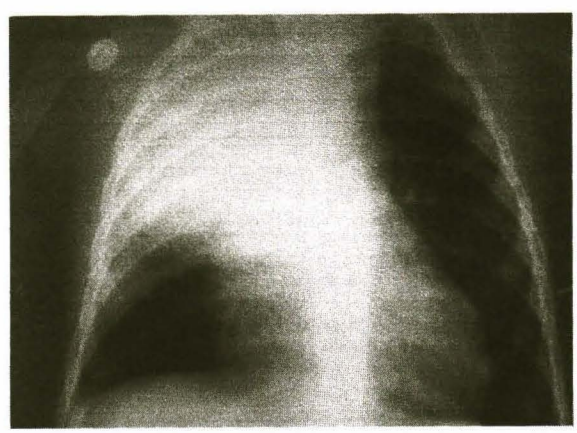

Fig. 2. Chest radiograph demonstrates a haziness that occupies the entire right upper chest.

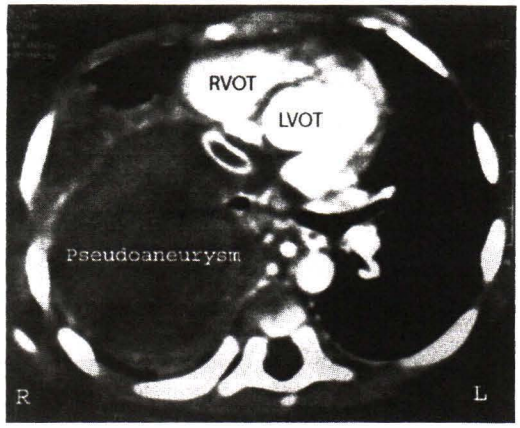

Fig. 3. **A pseudoaneurysmal mass, measuring 6 x 9 cm, occupies the upper right chest cavity on CT scan of the chest. RVOT = right ventricular outflow tract of the heart; LVOT = left ventricular outflow tract of the heart.**

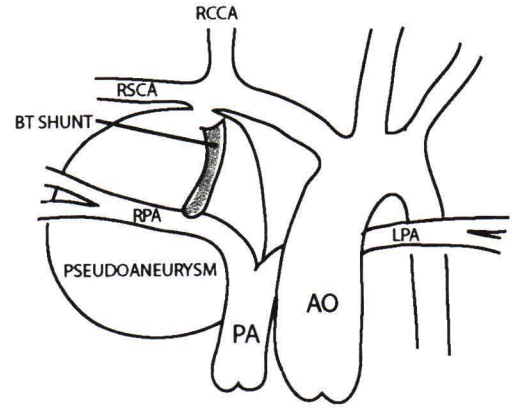

Fig. 4. **Diagram showing a large pseudoaneurysm arising from the right subclavian artery (RSCA) and displacing the shunt graft anteriorly. The upper end of the BT shunt was partially dehiscent from the RSCA. AO = aorta; RPA = right pulmonary artery; LPA = left pulmonary artery; RSCA = right subclavian artery; RCCA = right common carotid artery.**

and a central shunt created between the ascending aorta and the main pulmonary artery. Unfortunately, 3 hours after the operation, the patient developed severe hypotension and cardiac arrest from overshunting and pulmonary edema, and could not be resuscitated. Tissue culture and hemoculture were negative.

DISCUSSION

Pseudoaneurysm formation after modified B-T shunt is a rare complication. The etiologies of the aneurysm may be infection[4] or mechanical stress at the shunt anastomosis[3]. Type of the suture material, especially silk, was once thought to be the cause of the non-infected anastomotic aneurysm[5,6], rarely used in this era, due to better synthetic suture material. Infection possibly contributed to the aneurysmal formation in this patient because of the associated fever, eventhough tissue and graft cultures were sterile. However, the dehiscence of the graft may be evidence of mechanical stress at the proximal shunt anastomosis. Apart from the aneurysm that originated from modified B-T shunt anastomosis, an aneurysm formation can arise from other types of vascular shunts such as the aortopulmonary PTFE shunt[7].

Signs and symptoms of pseudoaneurysm after a modified B-T shunt, as previously reported, were cough, stridor, dypsnea, and acute hemoptysis with lung opacity shown on a chest radiograph that resembled pulmonary illness[3,4,6,8]. Patients with a modified B-T shunt presenting with acute hemoptysis or ipsilateral signs, either clinically or on a chest radiograph, should raise the suspicion of pseudoaneurysm associated with the shunt. Echocardiography, which is a non-invasive investigation, is helpful in demonstrating the size and opening site of the pseudoaneurysm related to the B-T shunt.

There has been a significant number of children who have undergone a modified B-T shunt as a first-stage of palliative treatment for cyanotic heart disease[2]. The authors presented a child with the unusual complication of a modified B-T shunt, which should alert clinicians to be aware of pseudoaneurysm formation in a patient presenting with lung opacity ipsilateral to a modified B-T shunt.

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หลอดเลือดแดงโป่งลวงที่เกิดจากการผ่าตัด modified Blalock–Taussig shunt : ภาวะแทรกซ้อนที่พบน้อยและอาการนำมาคล้ายกับโรคระบบหายใจ

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รายงานผู้ป่วยเด็กชายไทยอายุ 2 ปี ไปโรงพยาบาลใกล้บ้านด้วยอาการไข้ หอบเหนื่อยและเขียวมากขึ้น ผู้ป่วยเป็นโรคหัวใจ tetralogy of Fallot ได้รับการผ่าตัด modified Blalock–Taussig shunt ข้างขวาไปเมื่อหกสัปดาห์ก่อนมาโรงพยาบาล เนื่องจากมีภาวะ hypoxic spells บ่อย การตรวจครั้งนี้ภาพรังสีทรวงอกพบเงาทึบที่ปอดขวาบน ได้ให้การรักษาด้วยยาปฏิชีวนะ สองสัปดาห์อาการไม่ดีขึ้นจึงส่งต่อมายังโรงพยาบาลมหาราชนครเชียงใหม่ การตรวจด้วยคลื่นสะท้อนความถี่สูงของหัวใจและ CT scan ทรวงอกพบว่ามี pseudoaneurysm ขนาดอย่างน้อย 4 x 5 เซ็นติเมตรซึ่งมีรูเปิดมาจากรอยต่อระหว่าง subclavian artery และส่วนต้นของ modified Blalock–Taussig shunt ผู้ป่วยได้รับการผ่าตัดเอา aneurysm และ shunt ออก พร้อมกับผูก–เส้นเลือด subclavian artery และต่อ shunt ใหม่ระหว่าง ascending aorta และเส้นเลือดแดงปอด (main pulmonary artery) หลังผ่าตัดสามชั่วโมงผู้ป่วยมีความดันโลหิตต่ำ ไม่ตอบสนองต่อการรักษาและเสียชีวิตในที่สุด

คำสำคัญ : หลอดเลือดแดงโป่งลวง, ท่อ Blalock–Taussig

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