Case report

Pseudoaneurysm of subclavian artery 21-years after staged repair of tetralogy of Fallot

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Abstract

Three cases of pseudoaneurysm of subclavian artery in long-term follow-up after staged repair of tetralogy of Fallot are described. The angiographical study revealed that aneurysms were located in the systemic end area of previously ligated modified Blalock–Taussing shunt. Aneurysmectomy with resection of a shunt and segment of subclavian artery was performed. We believe division of a prosthetic shunt should be routinely performed during complete repair of tetralogy to prevent morbidity in the long-term follow-up period. © 2002 Elsevier Science B.V. All rights reserved.

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1. Introduction

Many centres have adopted early primary repair of tetralogy of Fallot (TOF), however, the modified Blalock–Taussing shunt (MBTS) is frequently used as a staged, palliative operation for surgical treatment of TOF [1–3]. A double ligation or clipping of the MBTS is considered a standard surgical procedure during intracardiac repair of tetralogy. Even after repair, the presence of the shunt’s prosthetic material as a restrictive factor, may produce morbidity in the long-term follow-up [4–6]. We present three patients after staged repair of TOF. They had pseudoaneurysm of subclavian artery (SA) in the systemic end of ligated but not divided MBTS more than two decades after radical operation.

2. Case reports

2.1. Patient 1

A 33-year-old man was referred to our hospital with hemoptysis and a history of TOF. Diagnosis during infancy included right-sided talc pleurodesis at 4 years, left MBTS at 8 years and complete repair at 13 years. The chest roentgenogram showed a circumscribed, non-calcified, round lesion in the left upper lobe. Further investigation revealed the absence of destructive lung processes. Selective angiogram showed a pseudoaneurysm of the left SA coinciding with the location of the mass. It was decided to perform an operation. The operative plan was to revise SA and MBTS. After general anesthesia was achieved, a left-side third interspace thoracotomy was employed. The left pleural adhesions were lysed. During attempts to dissect the shunt, profuse bleeding occurred from the left pulmonary artery. Emergency femoral–femoral cardiopulmonary bypass (CPB) was carried out. The shunt was found to be doubly ligated and totally thrombosed. It was lying in a mass of false aneurysm and clots. Corresponding segment of SA had disintegrated with partial disconnection of artery wall and end of the shunt. After debridement of the friable parts of the lung tissue, a MBTS was divided and its pulmonary remnant oversewn. The subclavian end of the shunt was removed together with segment of the left SA artery. The postoperative course was complicated by respiratory failure and gastrointestinal bleeding. Patient died on the 24th postoperative day in ICU because of disseminated intravascular coagulation syndrome.

2.2. Patient 2

A 34-year-old patient was admitted with signs of hemoptysis. Twenty-two years ago he was successfully operated upon for TOF after previous left MBTS. The patient had neither fever, cough, sweating nor dyspnea. A left upper lobe mass on chest X-ray film was investigated and selective
angiogram confirming a pseudoaneurysm of the left SA. The aneurysmectomy with resection of a shunt and segment of SA was performed through the left sided thoracotomy after the left ventricle bypass (cannulation of left atrium appendage and descending aorta) had been established. Histology showed the normal internal elastic layer of the SA artery continuous with the fibrous connective tissue of the pseudoaneurysm. On the 15th postoperative day the patient was discharged. After 11 months of follow-up he remains asymptomatic.

2.3. Patient 3

A 33-year-old patient was presented with intermittent hemoptysis, 25 years following left MBTS and 18 years following complete repair of TOF. He denied a history chest trauma, tuberculosis, lung tumor. Selective angiogram showed a pseudoaneurysm of the left SA (Fig. 1). The left thoracic cavity was entered on the third intercostal space. Careful surgical dissection was undertaken, during which the subclavian portion of the Dacron shunt was found free lying in a massive pseudoaneurysm. Off-pump aneurysmectomy was performed as in case 1–2. Hemoptysis was immediately resolved. The postoperative course was uneventful. There is no evidence of recurrence 7 months after discharging.

3. Discussion

The early and late results after one or two-stage repair of TOF has significantly improved in recent years [2,3], however, the role of the MBTS, initially the cornerstone of a surgical treatment of TOF, is still discussed [4–6]. This method has become controversial in recent years because there is evidence of shunt-related morbidity and mortality. The potential disadvantages of MBTS are volume overload of the left ventricle, shunt obstruction or occlusion, seroma, Horner’s syndrome, shunt endocarditis, pulmonary artery distortion, and development of pulmonary vascular obstructive disease.

Pseudoaneurysm formation is uncommon following a MBTS. To our knowledge, a description of pseudoaneurysm of the SA after MBTS at the long-term follow-up after TOF repair has not been reported. According to definition, a pseudoaneurysm results from a breach in the vascular wall, with the aneurysmal ‘sac’ comprised only of the outer arterial layers or surrounding connective tissue. Extravascular injury such as gunshot, stab wound and blunt trauma or erosion are an etiological factors associated with arterial pseudoaneurysms. The exact pathomechanism of the cases we present is unknown, however, it seems likely that the patients developed a chronic trauma of the SA by the rigid prosthetic shunt with time. Gladman and colleagues demonstrated similar pulmonary artery distortion after implantation of a modified BT shunt in 33% of cases [5]. Angiographically, they showed a tent of the pulmonary artery at the site of a shunt as a result of a prosthetic graft pulling the pulmonary artery upward. These distortions were observed within 1.4 years of palliation time. It is reasonable to assume that prosthetic shunt can’t become longer with the somatic growth of a patient and fixes the distance between anastomosed vessels. This fact was proved by Monro and co-workers [7]. They performed clipping and division of MBTS at the time of corrective procedure. The follow-up study showed that the distance between clips may increase within 3.5 cm following 13 years after correction.

Our cases suggest that SA, like constituent part of systemic end of MBTS, can undergo not only distortion but even injury with subsequent pseudoaneurysm formation, which can have a significant impact on the results of complete repair of TOF more than two decades after operation. Although MBTS is a palliative procedure of choice in some patients with TOF, this shunt does not grow simultaneously with the somatic growth of a patient and it can cause a SA pseudoaneurysm formation in long-term follow-up after corrective operation.

In conclusion, our purpose in describing these patients is to emphasize the importance of division a MBTS at the time of complete repair of TOF to prevent the morbidity in the long-term follow-up period.

References


