Gen Thorac Cardiovasc Surg (2012) 60:355–358
DOI 10.1007/s11748-012-0003-9

CASE REPORT

Bidirectional Glenn procedure for an infected modified Blalock–Taussig shunt

Yuki Sasaki · Tsukasa Ozawa · Tomoyuki Katayanagi · Hiroyuki Matsuura · Tsutomu Saji · Katsunori Yoshihara · Nobuya Koyama · Yoshinori Watanabe

Received: 29 November 2010 / Accepted: 5 May 2011 / Published online: 8 May 2012
© The Japanese Association for Thoracic Surgery 2012

Abstract A 1-month-old girl underwent right modified Blalock–Taussig shunt (mBTS) for pulmonary atresia with hypoplastic right ventricle. Five months after palliation, she suffered from sepsis and progressive desaturation following otitis media. Computed tomography and angiography revealed a pseudoaneurysm surrounding the mBTS graft. After stabilization of the infection, we performed pseudoaneurysm resection, shunt-graft removal, and the bidirectional Glenn (BDG) procedure under cardiopulmonary bypass. Her condition improved, and she was discharged on the 17th day after surgery. When parameters for the partial right heart bypass should permit, the BDG procedure can be a beneficial recovery procedure for the cases of infected pseudoaneurysm after mBTS in Fontan candidates.

Keywords Bidirectional Glenn · Infected pseudoaneurysm · Blalock–Taussig shunt · *Streptococcus pneumoniae* · Pulmonary hypertension

Y. Sasaki · T. Ozawa (✉) · T. Katayanagi · N. Koyama · Y. Watanabe
Department of Cardiovascular Surgery, Toho University Omori Medical Center, Ohmori-nishi 6-11-1, Ohta-ku 143-8541, Tokyo, Japan
e-mail: cbc02537@nifty.com
URL: http://www.ctsnet.org/home/tozawa

H. Matsuura · T. Saji
First Department of Pediatrics, Toho University Omori Medical Center, Ohmori-nishi 6-11-1, Ohta-ku, Tokyo 143-8541, Japan

K. Yoshihara
Department of Critical Care Emergency Center, Toho University Omori Medical Center, Ohmori-nishi 6-11-1, Ohta-ku, Tokyo 143-8541, Japan

Introduction

Infected pseudoaneurysm after a modified Blalock–Taussig shunt (mBTS) is a rare but fatal complication [1–3]. Revision of systemic–pulmonary shunt is required in patients presenting with desaturation due to troubles with mBTS [3, 4]. In addition, use of artificial materials should be avoided in redo procedures for patients with infected pseudoaneurysm after mBTS [5]. We describe an experience of a bidirectional Glenn procedure (BDG) for the successful repair of an infected pseudoaneurysm after a right mBTS.

Case

A female neonate with pulmonary atresia with intact ventricular septum and a hypoplastic right ventricle underwent palliative percutaneous balloon pulmonary valvuloplasty to perforate the atretic pulmonary valve at our institute. However, at the age of 1 month, she underwent a right mBTS using an expanded polytetrafluoroethylene graft (EPTFE; Gore-Tex, W.L. Gore and Assoc, Flagstaff, AZ) via right lateral thoracotomy because of restenosis of the opened pulmonary valve. After the palliation, arterial saturation obviously went up and postoperative course was stable. Echocardiography also showed sufficient shunt flow at 3 weeks and 4 months after the initial surgery.

Her condition remained satisfactory until 5 months after palliation, when she was re-admitted with a high fever of unknown origin. Diagnosis of otitis media was made, and *Streptococcus pneumonia* was isolated in her blood and pharynx. Antibiotic therapy proved effective: subsequent cultures were negative. However, she gradually developed systemic desaturation, and a large aneurysm surrounding

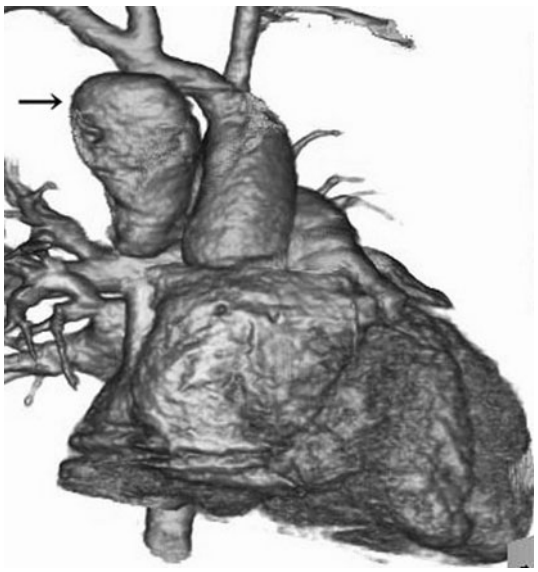

Fig. 1 An image of multidetector-row computed tomography before surgery. A large aneurysm surrounding the modified Blalock–Taussig shunt (*arrow*) was demonstrated

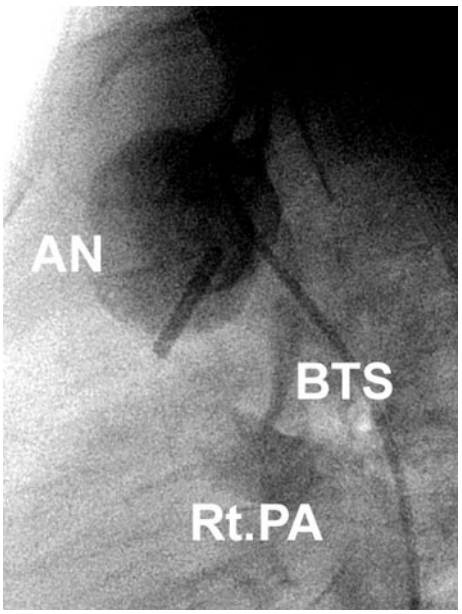

Fig. 2 Lateral angiographic view of right modified Blalock–Taussig shunt. At the proximal anastomosis, leakage of contrast medium visualized a pseudoaneurysm (20 mm in diameter) compressing the modified Blalock–Taussig shunt. The patency of the Blalock–Taussig shunt was confirmed. Aortic pressure was 77/56 mmHg, pulmonary artery pressure was 17/11/14 mmHg, and the pulmonary artery index was 257 mm^2/m^2. (*AN* aneurysm, *BTS* Blalock–Taussig shunt, *Rt. PA* right main pulmonary artery)

the mBTS was identified on echocardiography and computed tomography (CT) (Fig. 1). Although cardiac catheterization confirmed the patency of the mBTS, leakage of medium through the proximal anastomosis demonstrated a pseudoaneurysm compressing the shunt (Fig. 2). After stabilizing the infection and inflammatory response in her laboratory data by thorough antibiotics administration, she underwent a reoperation at the age of 8 months.

Regarding the revision procedure, her sternum untouched so far was easily divided in the midline without any dissecting procedure for adhesion. After cardiopulmonary bypass was established, the aneurysm was incised and opened. The mBTS graft was identified after a dark red thrombi filling the pseudoaneurysm was aspirated (Fig. 3a). The graft had partially detached from the native tissues on both the proximal and distal ends. After complete removal of the pseudoaneurysm and the graft, the orifice of brachiocephalic artery was closed using interrupted stitches. The BDG anastomosis was then performed (Fig. 3b). Due to an increase in mean pulmonary artery (PA) pressure to 30 mmHg after weaning from cardiopulmonary bypass, nitric oxide inhalation therapy was started.

Sildenafil citrate was administered via nasogastric tube, and pulmonary arterial hypertension abated; nitric oxide inhalation was discontinued on postoperative day (POD) 3. She was extubated on POD 5, and her mean PA pressure decreased to 11 mmHg. She was discharged on POD 17 in good condition. Cultures of the mBTS graft, wall of the aneurysm, and thrombi were all negative for causative bacteria. One year later, angiography confirmed good flow in the BDG and sufficient pulmonary vascularity (Fig. 4). Now, she is awaiting completion total cavopulmonary connection.

Discussion

Infected pseudoaneurysm after a mBTS is a fatal complication [1–3]. Its symptoms may include fever [1, 2], cyanosis following shunt obstruction [3, 5], tracheal compression [2, 4], and an upper mediastinal mass on chest X-ray [2, 4].

A number of authors have confirmed positive blood culture before pseudoaneurysm formation around mBTS [1, 2, 5]. In these cases, previous blood stream infection triggered off the subsequent pseudoaneurysm formation surrounding the graft as our case showed.

In patients received insufficient antibiotic therapy due to urgent and premature surgery for pseudoaneurysm, bacteria have frequently been identified in intraoperative cultures [3, 4]. In contrast, intraoperative cultures may be negative in the cases of optimal antibiotic therapy before elective reoperation for pseudoaneurysm [1, 2].

Matsuhisa et al. [1] have also reported a case of infected pseudoaneurysm after mBTS despite negative cultures in the revision. In that case, infected pseudoaneurysm after mBTS has been diagnosed by echocardiography, enhanced CT and cardiac catheterization following positive blood

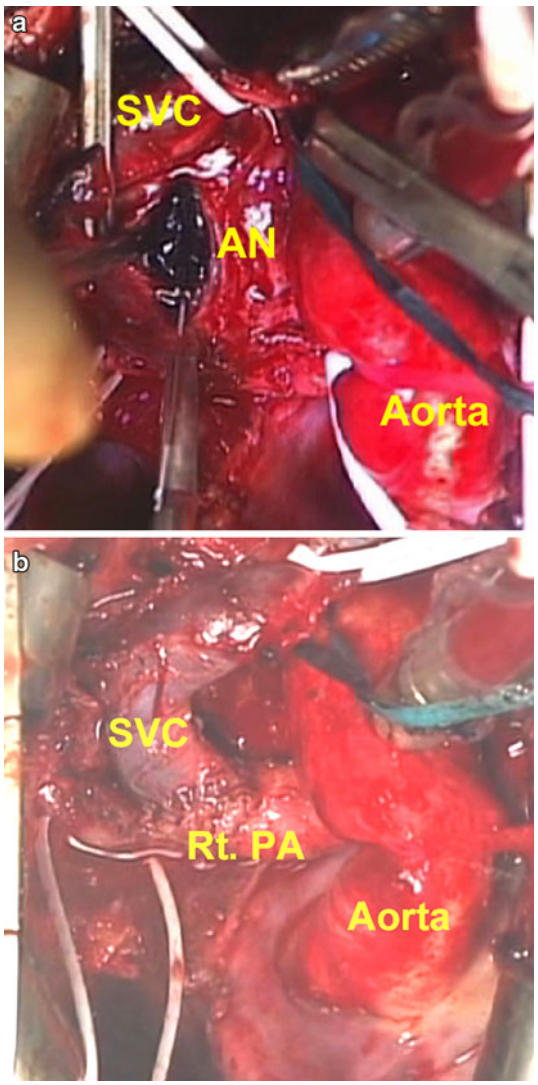

Fig. 3 **a** Operative view of aneurysm incision. The modified Blalock–Taussig shunt graft was surrounded by a dark red thrombi, which filled the aneurysm (*Aorta* ascending aorta, *AN* aneurysm, *SVC* superior vena cava). **b** Operative view after completion of bidirectional Glenn procedure. After complete removal of the aneurysm and artificial graft, a bidirectional Glenn procedure was performed (*Aorta* ascending aorta, *Rt. PA* right main pulmonary artery, *SVC* superior vena cava)

culture after events of high fever and desaturation [1]. The other investigators also emphasized that the late dehiscence of the graft anastomosis should be attributed to infection even if intraoperative culture-negative [6, 7]. Therefore, infected pseudoaneurysm was highly suspected in our case by clinical findings and its progress, although we could not prove positive culture in the intraoperative materials in the revision surgery.

To prevent recurrent infection, targeted antibiotic therapy of sufficient duration is indicated before reoperation, except in cases of rapid progress of arterial desaturation or enlargement of aneurysm [1, 2].

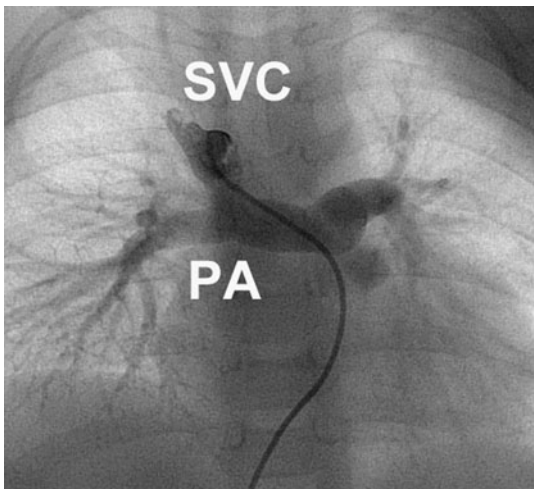

Fig. 4 Angiographic view at 1-year after revision surgery. The catheter was inserted into the superior vena cava through the pulmonary artery. The bidirectional Glenn procedure resulted in sufficient pulmonary vascularity. Aortic pressure was 96/48 mmHg and pulmonary artery pressure was 12/10/12 mmHg. The findings confirmed that the patient was a suitable candidate for a Fontan-type procedure (*SVC* superior vena cava, *PA* pulmonary artery)

If systemic saturation remains stable, the recommended procedure is simple removal of the infected pseudoaneurysm and graft and delaying of the subsequent procedure [1, 6]. However, in our case, the construction of another systemic–pulmonary shunt or alternative surgical maneuver was mandatory.

Carvalho et al. [5] cautioned that the risk of recurrent infection might be increased when the artificial materials used in the construction of another mBTS or in the repair of an intracardiac anomaly. We preferred the construction of BDG anastomosis because of the surgical strategy based on this patient's anatomy and physiology and simultaneously of the unnecessity of the foreign material in such procedure to improve hypoxemia.

Dimas et al. [4] described a case where a right bidirectional cavopulmonary shunt was constructed after removal of an infected pseudoaneurysm. In their report, they noted that patient was doing well 1-year after surgical intervention without recurrence of infection. Their patient's course resembles that of our case and supports our approach in this difficult situation.

BDG was beneficial in regard both to elimination of the foreign material in the field of possible infection and to the staging of the Fontan track as the interim procedure.

Recently, the safety and the feasibility of BDG for early infants have been gradually recognized [8]. BDG might be the most feasible option for infected pseudoaneurysm after mBTS even in the early infants who will undergo total cavopulmonary connection.

Conclusion

We experienced an infant case of infected pseudoaneurysm after mBTS.

BDG was effective surgery in controlling serious infection from the patients' own and vital tissue procedure and simultaneously in achieving one steady forward step in the staged approach of univentricular repair.

References

1. Matsuhisa H, Yoshimura N, Kitahara J, Otaka S, Ichida F, Funada H, et al. An infected pseudoaneurysm following a modified Blalock–Taussig shunt. Interact Cardiovasc Thorac Surg. 2009;8:108–10.
2. Sundararaghavan S, Khalid O, Suarez WA, Bove EL. Single-stage repair of tetralogy of Fallot with pseudoaneurysm: a unique approach. Ann Thorac Surg. 2004;77:2183–4.
3. Parvathy U, Balakrishnan KR, Ranjith MS, Moorthy JSN. False aneurysm following modified Blalock–Taussig shunt. Pediatr Cardiol. 2002;23:178–81.
4. Dimas VV, Kovalchin JP, McKenzie ED, Kung GC. Infected pseudoaneurysm of a right modified Blalock–Taussig shunt. Pediatr Cardiol. 2004;25:700–1.
5. Carvalho JS, Belcher P, Knight WB. Infection of modified Blalock shunts. Br Heart J. 1987;58:287–90.
6. Valliattu J, Jairaj P, Delamie T, Subramanyam R, Menon S, Vyas H. False aneurysm following modified Blalock–Taussig shunt. Thorax. 1994;49:383–4.
7. Hörer J, Malcic I, Schreiber C, Lange R. False aneurysm origination from the proximal anastomosis of a right ventricular to pulmonary artery shunt following staged repair of hypoplastic left heart syndrome. Interact Cardiovasc Thorac Surg. 2011;12:487–9.
8. Petrucci O, Khoury PR, Manning PB, Eghtesady P. Outcomes of the bidirectional Glenn procedure in patients less than 3 months of age. J Thorac Cardiovasc Surg. 2010;139:562–8.