Negative results - Congenital

Infectious false aneurysm of the right ventricular outflow tract after repair of congenital heart defect treated with Freestyle® aortic bioprosthesis

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Abstract

Objective: False aneurysm of the right ventricular outflow tract (RVOT) is one of complications after surgery of congenital heart disease. It is controversial about materials for reconstruction of RVOT in this setting, when associated with pulmonary hypertension or bacterial infection. Pulmonary homograft seems to be the first choice, but its availability is extremely limited in Japan. Methods: Therefore, we used Freestyle® stentless bioprosthesis to reconstruct the RVOT in a 14-year-old boy who developed infectious false aneurysm of RVOT after total correction of pulmonary atresia with ventricular septal defect. Results and discussion: After describing the patient, the prosthesis alternative to the pulmonary homograft was discussed.

Keywords: False aneurysm; Right ventricular outflow tract; Freestyle® stentless bioprosthesis

1. Introduction

False aneurysm of the right ventricular outflow tract (RVOT) has been reported as a complication of RVOT reconstruction using a conduit or a homograft [1–6]. Materials for reconstruction of RVOT in this setting are still controversial especially when associated with pulmonary hypertension (PH) or bacterial infection. Pulmonary homograft seems to be the first choice, but its availability is severely limited in Japan. As Freestyle® stentless bioprosthesis has been reported to have good valvular function and resistance to infection [7], we used this bioprosthesis for the present case that developed an infectious false aneurysm of RVOT after total correction of pulmonary atresia and ventricular septal defect.

2. Case report

A 14-year old boy, with a diagnosis of pulmonary atresia with ventricular septal defect (VSD), atrial septal defect (ASD), and major aortopulmonary collateral arteries, was admitted to our hospital because a pulsatile mass on his left anterior chest wall was found. He underwent a Blalock-Taussig shunt and unifocalization of major aorto-pulmonary collateral arteries of left side at 2 years old, and of right side at 8 years old, followed by a definitive surgery using a handmade valved bovine pericardial roll for RVOT reconstruction at 9 years old. At the definitive surgery, jump graft from RVOT to right pulmonary artery was placed. He underwent redo RVOT reconstruction with the valved pericardial roll at age of 13 years. At this reoperation, hemodynamic study showed PH (right ventricle systolic pressure (RVSP) 56 mmHg, left ventricle systolic pressure (LVSP): 66 mmHg, right ventricle/left ventricle (RV/LV) pressure ratio: 0.84). After this second heart surgery, he was suffered from mediastinitis caused by Methicillin-resistant Staphylococcus epidermidis (MRSE), which was medically treated and the patient was discharged 4 months after operation.

Two months after discharge, he was found to have a pulsatile mass on his left anterior chest wall with inflammatory skin change. His laboratory examinations showed a hemoglobin concentration of 8.6 g/dl, a C-reactive protein concentration of 1.6 mg/ml, and the leukocyte count of 7210/mm³. The chest CT revealed a well-enhanced mass deriving from RVOT, which penetrated the chest wall and extended into the subcutaneous space (Fig. 1a). Infectious
false aneurysm of RVOT was diagnosed, and the patient underwent emergent operation.

At the operation, a midline sternotomy was done. The valved pericardial roll was severely attached to the left anterior chest wall. There was a small hole in the 2nd intercostal space and the false aneurysm penetrated the chest wall and extended to the subcutaneous space. On cardiopulmonary bypass (CPB), the valved pericardial roll was carefully removed, and replaced with the Medtronic Freestyle® aortic bioprosthesis.

As the culture of tissue around the aneurysm revealed MRSE again, he was given intravenous antibiotics (vancomycin, sulfamethoxazole/trimethoprim, and gentamicin) until 90 days after surgery. As there was no recurrence signs of infection, he was given oral sulfamethoxazole/rimethoprim after then. Postoperative echocardiography 1 month after re-operation showed a well-seated prosthesis with no PS/PR and grade three of TR indicating persistent PH. No sign of recurrence of mediastinitis was found during the 1 year of follow-up (Fig. 1b).

3. Discussion

False aneurysm of RVOT is not a rare complication after repair of congenital heart disease particularly when associated with PH. Fifteen patients with false aneurysm after repair of RVOT with conduit or homograft were collected with literature [1–6]. Nine of the 15 patients had systemic or supra-systemic PH (Table 1). Regarding

Table 1
Characteristics of 15 patients with right ventricular outflow tract false aneurysms after conduit or homograft implantation

<table>
<thead>
<tr>
<th>Diagnosis</th>
<th>Age at conduit placement</th>
<th>Postoperative interval</th>
<th>Presentation</th>
<th>RV pressure</th>
<th>Defect origin</th>
<th>Treatment</th>
<th>Bacterial infection</th>
</tr>
</thead>
<tbody>
<tr>
<td>Truncus arteriosus 4</td>
<td></td>
<td></td>
<td>Chest swelling</td>
<td>Supra systemic 2</td>
<td>Between homograft conduit and RV 9</td>
<td>Surgical resection direct suture 9</td>
<td>None 13</td>
</tr>
<tr>
<td>TGA/VSD LVOTO 4</td>
<td></td>
<td></td>
<td>Respiratory distress 3</td>
<td>Systemic 7</td>
<td>Between pericardial augmentation and RV or homograft 2</td>
<td>Surgical resection homograft imp 2</td>
<td>Enterococci and S. epidermides 1</td>
</tr>
<tr>
<td>TOF/PA 6</td>
<td>3 months–13 years (mean: 5.3 years)</td>
<td>2 months–4 years (mean: 17.2 months)</td>
<td>Routine examination 5</td>
<td>2/3 systemic 1</td>
<td>Between Dacron conduit or augmentation and homograft 2</td>
<td>Surgical resection composite conduit imp 2</td>
<td>S. epidermides 1</td>
</tr>
<tr>
<td>TOF/absent pulmonary valve 1</td>
<td>2 months–4 years</td>
<td>1/2 systemic 2</td>
<td>Others 2</td>
<td>Between Gore-Tex augmentation and homograft 1</td>
<td>Surgical resection freestyle bioprosthesis imp 1</td>
<td>Aneurysm origin occluded; clam shell device 1</td>
<td></td>
</tr>
<tr>
<td>PA/VSD MAPCAs 1</td>
<td>Unknown 2</td>
<td>1/3 systemic or below 3</td>
<td>Unknown 2</td>
<td>Between pericardial augmentation and valved pericardial roll 1</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*RV, right ventricle; TGA, transposition of the great arteries; VSD, ventricular septal defect; LVOTO, left ventricle outflow track obstruction; TOF, tetralogy of fallot; PA, pulmonary atresia; MAPCAs, major aorto pulmonary collected arteries.
the procedure, false aneurysm was surgically resected in all of the cases except one, in which was closed with catheter intervention. Ten patients were repaired with direct suture, two underwent re-implantation of homograft, and two underwent re-implantation of composite conduit. Only one patient in these 15 cases developed false aneurysm due to infectious complication. Other than conduit or homograft implanted cases, only two cases described infectious false aneurysm of RVOT in the available literature [8,9]. One patient underwent total correction of tetralogy of Fallot using a Dacron patch and developed endocarditis due to *Enterobacter cloacae*, and the other patient with pulmonary stenosis developed endocarditis due to *Staphylococcus aureus* without previous right heart surgery. Both of them, the aneurysm was surgically resected and the orifice was directly sutured.

Regards to the materials of RVOT reconstruction, an autologous monocusp patch, a conduit with or without a porcine valve, and a handmade valved pericardial roll, have been mainly used if the homograft was not used. But their long-term results are not satisfactory because of poor durability or vulnerability to infection. The pulmonary homograft has been used in this situation because of its several advantages, such as excellent hemodynamic performance and resistance against infection [7]. These advantages may be offset, however, by problems of supply of homografts of a suitable size. As the present case showed systemic PH, we used Freestyle® stentless bioprosthesis to reconstruct the RVOT instead of the pulmonary homograft because of its poor availability in Japan. Although long-term outcome has not been shown, the Freestyle® stentless bioprosthesis have been reported to have comparable characteristics to the pulmonary homograft. Barratt-Boyes [7] reported its good late outcome in the aortic position. Moreover, Chard [10] showed its excellent short-term hemodynamic performance for 13 cases of RVOT reconstruction.

In summary, an infectious false aneurysm of RVOT is a life-threatening complication after surgical repair of RVOT and materials to replace it is still controversial. Although long-term follow-up is necessary for further evaluation, in the setting of difficult availability of cryopreserved allograft, the Freestyle® stentless bioprosthesis seems to represent a valuable option for a repair of false aneurysm of RVOT as an alternative.

References