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Endovascular Treatment of Hemoptysis after Arterial Switch Operation

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ABSTRACT

A 17-month-old patient with transposition of the great arteries and ventricular septal defects presented without pulmonary over-circulation and significant aortopulmonary collaterals before surgery, and subsequently presented with hemoptysis and pulmonary congestion after arterial switch operation. Postoperative angiography revealed one aberrant right bronchial artery and two aortopulmonary collaterals arising from both subclavian arteries. Percutaneous embolization of the abnormal vessels caused dramatical improvement in patient's conditions and rapid weaning from mechanical ventilation support, and he was discharged without any complications. This highlights the importance of cardiac catheterization to detect collateral vessels as a cause for pulmonary over-circulation and hemoptysis even though they seemed trivial and irrlevent before surgery Embolization is an effective and safe method of treatment for hemoptysis in children.

Keywords: Arterial switch operation, Hemoptysis, Bronchial artery, Aortopulmonary Collateral sembolization

CASE PRESENTATION

A 17-month-old boy weighing 9kg, was admitted to our hospital with mild dyspnea and cyanosis. Transthoracic echocardiography revealed D-transposition of the great arteries (d-TGA) with large perimembranous and muscular ventricular septal defect (VSD) and pulmonary hypertension (PH). A frontal chest X-ray showed cardiomegaly with cardiac contours appearing like an egg on string and pulmonary congestion (Figure 1A).

Diagnostic cardiac catheterization and angiography (Figure1B, 1C) was performed to confirm the common coronary pattern and multiple VSDs. Small aortopulmonary collateral arteries were found to arise from branches of the aortic arch, but we took it for granted that these vessels were too trivial to affect hemodynamics. The pulmonary vascular resistance index was 2Wu.U.m² under 21% inspired oxygen, which revealed he still had the chance to undergo primary repair. At the ninth day of admission, he underwent arterial switch operation (ASO) with VSDs closure. At surgery, there was increased pulmonary venous return but no other unusual findings were encountered. He was transferred to the intensive care unit and was cared for by skilled medical staff. Suitable postoperative care, including the use of catecholamine and mechanical ventilation, was provided. However, the recovery of hemodynamic status was slow, with signs of pulmonary congestion and cardiomegaly on chest X-ray. Even worse he suffered from hemoptysis approximately 20 mL of fresh blood 1 day after surgery, and recurrent hemoptysis gradually worsened over the next 2 days despite the use of laser coagulation and hemostatic drugs. Postsurgical echocardiographic evaluation failed to show residual defects and there was no evidence of neo-aortic stenosis or insufficiency, but unique supra-aortic anomalous color doppler signals to the pulmonary circulation was identified, suggesting aortopulmonary collaterals.

Two weeks after surgical repair, the baby was restudied by cardiac catheterization. Right catheterization showed a mean pulmonary arterial pressure of 45 mmHg, and pulmonary angiography ruled out the presence of any pulmonary arteriovenous malformation. On selective aortic arteriography (**Figure 2),** one enlarged aberrant right bronchial artery which followed the normal

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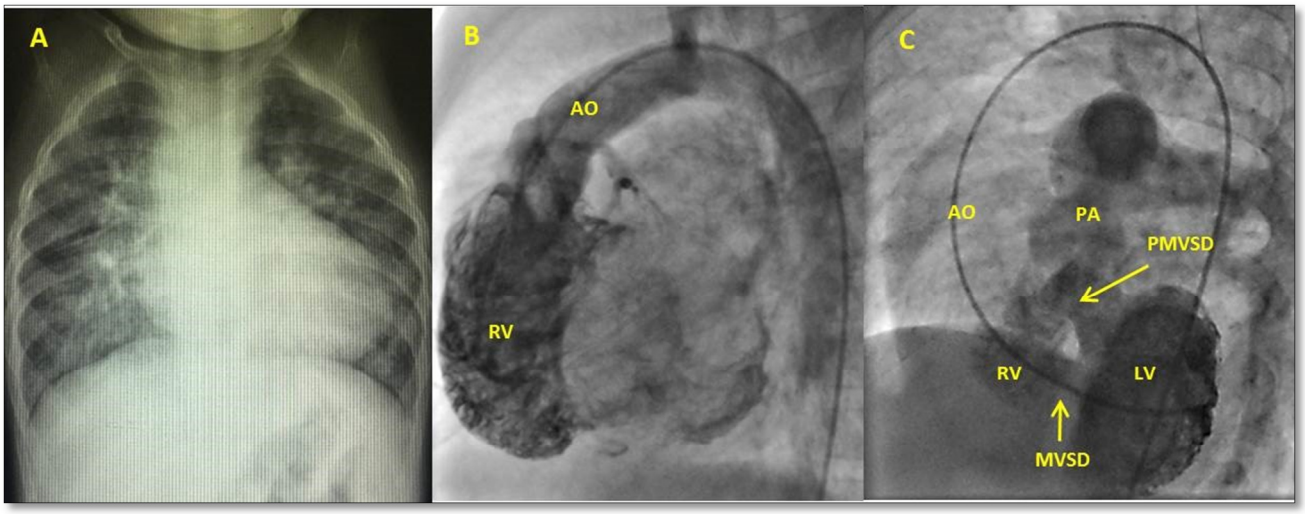

Figure 1. The Chest X-ray (A) and angiography (B and C) before surgery.
RV=right ventricular; LV=left ventricular; AO=aorta; PA=pulmonary artery; MVSD=muscular ventricular septal defect; PMVSD=perimembranous ventricular septal defect.

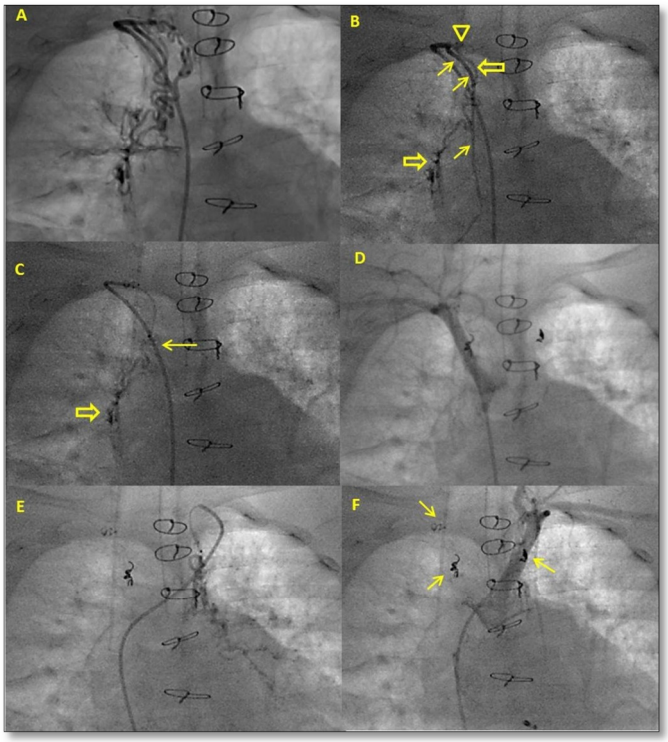

Figure 2. Arterial angiography and embolization.
A. Right internal mammary artery, aberrant right bronchial artery and aortopulmonary collateral artery sequentially originated from right subclavian artery;
B. Right aortopulmonary collateral arteries was occluded by one microcoil (marked by a triangle), and blood flow of right internal mammary artery (thin arrows) was not affected. Aberrant right bronchial artery was marked by a hollow arrow;
C. Distal right bronchial artery (hollow arrow) was embolized by 500-700um Embosphere microsphere and one microcoil was placed in the proximal (thin arrow), using coaxial microcatheter technique;
D. Right subclavian artery angiography showed aortopulmonary collateral artery and right bronchial artery were occluded completely;
E. Left aortopulmonary collateral artery originated from left subclavian artery;
F. Left aortopulmonary collateral artery was occluded completely. Microcoils were marked by fine arrows.

branching pattern of the airways with an area of hypervascularity and neovascularity was identified from the right subclavian artery, two aortopulmonary collateral arteries with segmental parenchymal distribution arised from both subclavian arteries. There were no dilated bronchial arteries originating from the T5-T6 segments. We suspected that these abnormal vessels were responsible for the hemoptysis and were reasonable to be occluded to improve the condition. All of these abnormal vessels were sequentially embolized by 500-700um Embosphere microsphere (Merit Medical, USA) and microcoils (Tornado; Cook Medical, USA), using coaxial microcatheter technique. The closure of these anomalous vessels was associated with significant improvement of the clinical status of the baby, permitting successful extubation 7 days after embolization and decrease of the inotropic support. He was discharged with an oxygen saturation of 93% and no complications happened. There was no recurrent hemoptysis after a follow-up of 12 months.

DISCUSSION

The most common complications after an ASO are the supravalvular pulmonary stenosis, neoaortic valve dysfunction and myocardial ischemia related to coronary artery translocation [1]. However, about half of patients with TGA undergoing ASO present with anomalous aortopulmonary or bronchial collateral arteries detected during post-operative cardiac catheterization studies [2]. Flow through these vessels is trivial to mild in the majority of patients and appears to be of no hemodynamic significance, but a few patients have pulmonary edema and congestive heart failure as well as hemoptys is due to enlarged collateral arteries.

Though pre-operative angiography revealed small aortopulmonary vessels in this patient, its diminutive size may have dissuaded us from further investigation or aggressive treatment. Moreover, the increased pulmonary venous return seen during the operation did not affect our ability to wean him from cardiopulmonary bypass. Unexpectedly, the small collateral arteries became enlarged and distorted after the operation, which caused pulmonary hyperperfusion and airway haemorrhage with hypoxia. The reason for the dilation of the aortopulmonary collaterals remains unclear; some researchers speculated that this may relate to hyperoxia associated with cardiopulmonary bypass and/or lower pulmonary vascular resistance associated with general anaesthesia [3].

Unexplained pulmonary hypercirculation and airway haemorrhage after ASO could be an indicator of anomalous enlarged collateral arteries. The main responsible vessels for bleeding in the present case included one aberrant right BA and two aortopulmonary collaterals, all of them originated from the subclavianarteries. Selective embolization of systemic collaterals or BA is a well-established procedure that has been widely used for the treatment of hemoptysis from a variety of causes, it aims to reduce the perfusion pressure to the fragile pathological collateral arteries that are usually responsible for hemoptysis [4].To achieve this, all abnormal appearing collateral arteries should be embolized if possible.

While there have been no controlled studies comparing embolic agents, reported materials include gelatin sponge, polyvinyl alcohol particles, spherical embolics, coils, and n-butylcyanoacrylate [5]. Shunts to the pulmonary circulation as large as 325 μm have been described, and therefore, the particles used are usually greater than 350 μm in size [4]. In this case, we used Embosphere microsphere (Merit Medical, USA) with a diameter of >500 um to achieve distal embolization to reduce recurrence rate of hemoptysis , and also placed microcoils in the trunk of the responsible vessels to ensure immediate effect of hemostasis.

In conclusion, small aortopulmonary vessels detected preoperatively may dilate during or after surgery, and cause extra-pulmonary blood supply and hemoptysis, adversely affect the postoperative clinical course. It's reasonable to occlude these vessels as much as possible even they seemed trivial and irrlevent before surgery. Earlier investigations such as angiography should be performed when the post-operative course is suggestive of pulmonary over-circulation, embolization can be a treatment option in children with abnormal vasculature bleeding. It is an effective and safe method of treatment for hemoptysis secondary to a variety of causes.

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