Acute Pulmonary Reperfusion Hemorrhage: A Rare Complication After Oversized Percutaneous Balloon Valvuloplasty for Pulmonary Valve Stenosis

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Balloon valvuloplasty became the treatment of choice for valvular pulmonary stenosis following its first description in 1982 by Kan et al, and has almost replaced surgical pulmonary valvotomy in the present day. It is a safe and effective method for children for relief of right ventricular obstruction. The results of the procedure are excellent, without significant complications. This report describes the case of a 12-year-old boy who received successful balloon valvuloplasty for critical pulmonary valve stenosis complicated by an episode of acute pulmonary hemorrhage. Because of cyanosis, hypotension and bradycardia, he received emergent endotracheal intubation with 100% oxygen supplement and the highest infusion rate of inotropic agents. Venoarterial mode extracorporeal membrane oxygenation was indicated for life support due to the persistent high oxygenation index. Extracorporeal membrane oxygenation played a key role in the survival of this patient during the course of treatment. [*J Chin Med Assoc* 2009;72(11):607–610]

Key Words: balloon dilatation, pulmonary hemorrhage, pulmonary stenosis

Case Report

A 12-year-old boy was admitted due to persistent shortness of breath and abdominal fullness for 3 days. He had a past history of pulmonary atresia with intact ventricular septum and had undergone surgical pulmonary valvotomy during infancy. He was then lost to follow-up. However, 1 month prior to this admission, shortness of breath and orthopnea developed progressively, so he was referred to our hospital for further management.

After admission, physical examination revealed signs of right heart failure, including jugular vein engorgement, hepatomegaly and abdominal distension. He had a grade III/VI ejection-type systolic murmur without clicking sound in the left upper sternal border. Chest radiography showed cardiomegaly and decreased lung markings (Figure 1). Twelve-lead surface electrocardiography revealed right atrial (RA) enlargement and right ventricle (RV) hypertrophy. Abdominal sonography demonstrated coarse echogenicity of liver parenchyma accompanied by massive ascites, and echocardiography showed critical pulmonary stenosis with a pressure gradient of 160 mmHg between the RV and pulmonary artery (PA).

During cardiac catheterization, poor RV function, high RA pressure (mean RA pressure, 28 mmHg) and critical pulmonary valve stenosis with a peak-to-peak systolic pressure gradient of 170 mmHg between the RV and PA (PA systolic pressure, 15 mmHg) were found. We used a 10-mm Smash balloon catheter (Boston Scientific Ltd., Watertown, MA, USA) and then an 18-mm Meditech balloon catheter (Boston Scientific Ltd.; pulmonary annulus diameter, 14.7 mm; balloon-to-annulus ratio, 1.2) for pulmonary valvuloplasty. After balloon dilatation, the transvalvular pressure gradient was reduced to 30 mmHg (PA systolic

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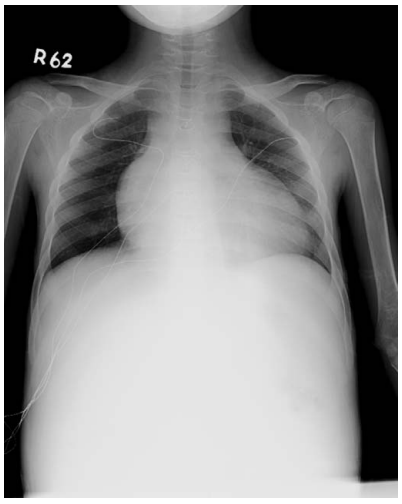

Figure 1. Chest radiography at the time of admission shows decreased lung markings and cardiomegaly with right atrial and ventricular enlargement.

pressure, 45 mmHg). The patient's hemodynamic condition remained stable during the whole procedure (Figure 2).

After the operation, the patient was sent to the pediatric intensive care unit for close observation. However, he developed severe cough with fresh-bloody sputum and dyspnea 10 hours later. Bilateral coarse breathing sounds and respiratory acidosis were noted; emergent chest radiography demonstrated diffuse alveolar pattern over bilateral lung fields, which confirmed the diagnosis of bilateral pulmonary hemorrhage (Figure 3). Significant drops in hemoglobin and hematocrit were also noted, so massive transfusion with packed red blood cells and normal saline were given immediately. Cyanosis, hypotension and bradycardia all occurred in the following 1 hour, and emergent endotracheal intubation was performed. However, the patient's condition remained unstable even under 100% oxygen

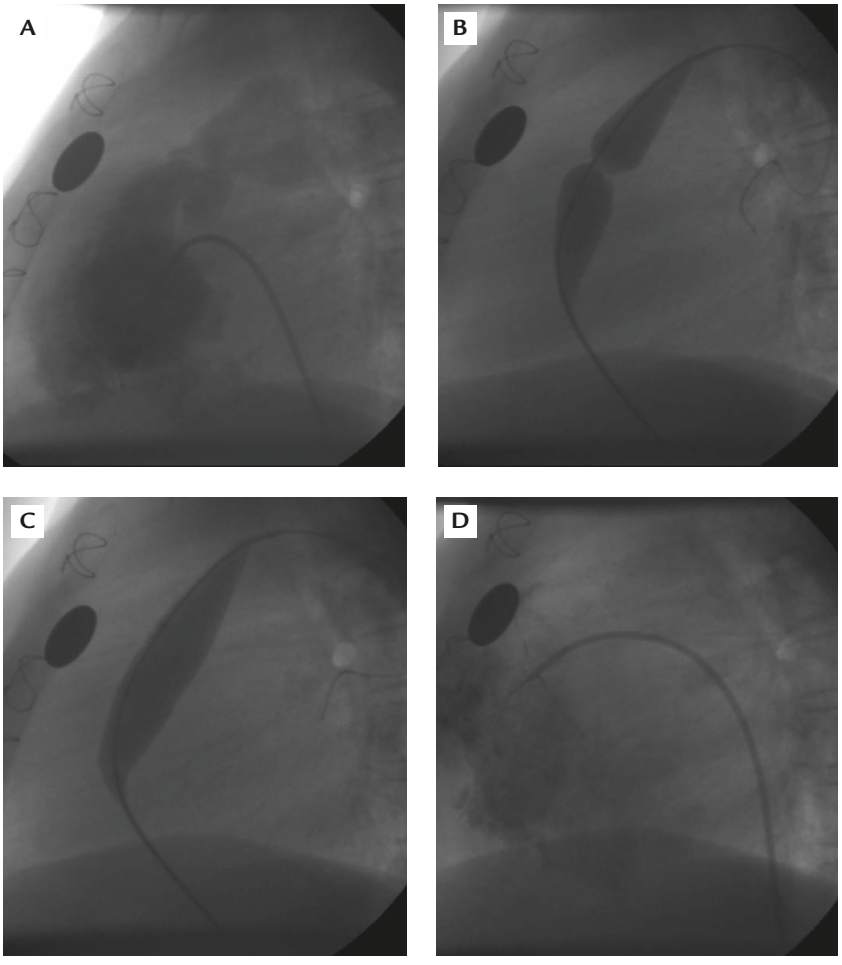

Figure 2. Successful relief of pulmonary stenosis after percutaneous balloon valvuloplasty. (A) Right ventricular angiogram obtained before balloon valvuloplasty shows severe pulmonary stenosis and post-stenotic dilatation. (B) Balloon being inflated after crossing the stenotic valve. (C) The valvular stenosis is relieved after balloon dilatation. (D) Right ventricular angiogram obtained immediately after balloon valvuloplasty shows successful relief of pulmonary stenosis.

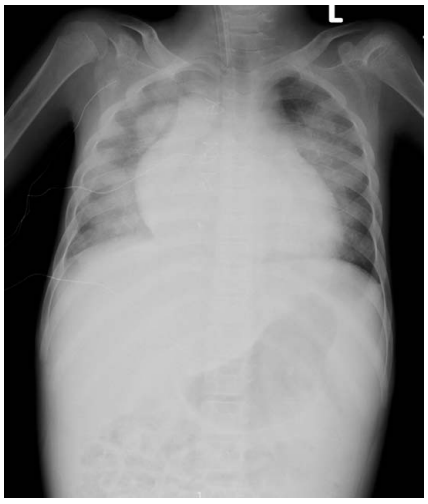

Figure 3. Diffuse alveolar pattern over bilateral lung fields. Pulmonary hemorrhage was suspected.

supplement and the highest infusion rate of inotropic agents (dopamine, dobutamine and epinephrine). Since the oxygenation index was still > 40, venoarterial mode extracorporeal membrane oxygenation (ECMO) was indicated for life support.

After ECMO support for 3 days, his hemodynamic condition became stable. ECMO was successfully removed without any major complications. The patient was extubated the next day. Two weeks later, he was discharged home from the ward smoothly. During clinical follow-up, the pressure gradient between the RV and PA was demonstrated to be only 10 mmHg, and mild pulmonary regurgitation was detected by Doppler echocardiography.

Discussion

Congenital pulmonary valve stenosis comprises 7.5–9.0% of all congenital heart defects.[1] Nowadays, transcatheter balloon valvuloplasty is the first option to manage pulmonary valve stenosis. In addition to its satisfactory short- and long-term results, complications such as arrhythmia, blood transfusion and neurologic problems associated with cardiopulmonary bypass can be safely avoided. In addition, wound infection and prolonged hospitalization are rarely seen because open cardiac surgery is not required. The major complications of balloon valvuloplasty, including cardiac perforation, arrhythmia and tricuspid insufficiency, rarely occur.[1–6]

A 66-year-old man who developed acute pulmonary edema after balloon valvuloplasty under general anesthesia was reported by Walker et al[6] in 2001. They supposed that the acute pulmonary edema might have been caused by the acute increase in pulmonary blood flow after the balloon dilatation procedure. However, information on the complications associated with balloon valvuloplasty in children is limited. In our patient, acute pulmonary hemorrhage was suspected because of the presence of fresh-bloody sputum. The cause of hemorrhage was most likely due to a sudden increase in pulmonary blood flow after valvular dilatation in the context of long-standing stenosis. The probability of acute pulmonary "hemorrhage", rather than "edema", is in direct proportion to the magnitude and severity of the pressure gradient over the pulmonary valve prior to the procedure. A higher initial pressure gradient is associated with a greater severity of pulmonary valve stenosis, and is associated with less blood flow passing through the lung parenchyma. If the reduction in pressure gradient is too drastic and sudden, as in our case, the capillary bed of the lung may not tolerate such a great hemodynamic change. Therefore, transient leakage from the pulmonary capillary bed will occur. The leakage may cause pulmonary "edema" or, in more severe cases, "hemorrhage", depending on the reduction in the pressure gradient and the duration of valvular stenosis. Also, the currently recommended balloon/annulus ratio is around 1.20–1.25. Balloon/annulus ratio < 1.2 is one of the predicting factors of pulmonary restenosis.[7] However, in such a case of long-standing and severe stenosis, it is vital to choose a smaller balloon/annulus ratio while taking gradual steps in the performance of balloon dilatation in order to avoid overflow of the pulmonary artery suddenly.

For children with heart and lung diseases accompanied by failing circulation, ECMO has been established as an important mechanical support. ECMO is also the accepted intervention in a variety of circumstances, including failure to wean from cardiopulmonary bypass, severe low cardiac output syndrome, sepsis, refractory hypoxemia, and pulmonary hypertensive crisis. More recently, the indications for ECMO have been expanded to include patients with acute cardiovascular decompensation, such as unexpected cardiac arrest or intractable arrhythmias.[8–12] Therefore, emergent ECMO plays an important role in life support in patients with associated procedure-related complications before, during and after pediatric catheterization.[2,13] In 2006, Allan et al[14] reported 22 patients who were cannulated emergently for ECMO in the cardiac catheterization laboratory due to catheter-induced complications such as perforation, hypoxemia and severe low cardiac output syndrome. Among those patients, 82% survived and were smoothly discharged. Our patient suffered from acute pulmonary

hemorrhage which induced low cardiac output and hypoxemia after cardiac catheterization. Therefore, ECMO was indicated and led to a satisfactory result. Weaning from ECMO after such a short length of time indicated that the acute pulmonary hemorrhage of this patient was a transient and reversible condition.

In conclusion, we have described a patient with acute pulmonary hemorrhage, a rare complication that was associated with percutaneous balloon valvuloplasty for pulmonary valve stenosis. The short duration of ECMO usage suggested a reversible condition that was possibly associated with a sudden increase in pulmonary blood flow. If the duration of stenosis is long or if the original peak-to-peak systolic pressure gradient over the pulmonary valve is high, it is recommended that a smaller balloon/annulus ratio be considered and the balloon valvuloplasty be performed in a stepwise fashion.

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