A 13-year-old girl, who was suffering complications with bilateral pulmonary artery stenosis after intracardiac repair for tetralogy of Fallot, suffered life-threatening left pulmonary bleeding and edema following inadvertent unilateral stent implantation for a left pulmonary stenosis. Pulmonary edema and subsequent hypoxia persisted despite intensive medical treatment; however, contralateral stent deployment resolved her symptoms quickly.

Life-Threatening Pulmonary Edema Following Unilateral Stent Implantation for Bilateral Branch Pulmonary Stenosis

--- Recovery After Contralateral Stent Implantation ---

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Key Words: Pulmonary edema; Stent implantation

Intravascular stents are being employed increasingly to dilate pulmonary artery stenosis.1–4 Complications of this procedure include ventricular arrhythmia, thrombus formation, compromise of pulmonary artery side branches, intimal flaps, pulmonary artery rupture, and misplacement and migration of stents.1 We report a case complicated by persistent and life-threatening unilateral pulmonary edema.

Case Report

A 13-year-old girl, who had had previous repair of tetralogy of Fallot at 2 years of age at another center, was scheduled to undergo stent implantation for residual bilateral pulmonary branch stenosis. Her body-weight was 29.0 kg and height was 129.6 cm. Although cardiac catheterization at the National Cardiovascular Center 2 years previously revealed oversystemic right ventricular pressure with severe bifurcation stenosis, surgical treatment was contraindicated because of seriously impaired right ventricular pump function (ejection fraction, 0.25).

Doppler echocardiography demonstrated marked dilatation of the right ventricle with pressure overload. Pump function was considerably impaired. Tight pulmonary stenosis was documented at the bifurcation and in the left pulmonary artery. Pulmonary perfusion scan revealed severe hypoperfusion of the left lung, with a right to left ratio of 6.21. Main, left, and right pulmonary artery pressure was 91/14 mmHg (mean, 30), 13/9 (11) mmHg, and 34/10 (19) mmHg, respectively, at cardiac catheterization under intubated general anesthesia. There was no systolic pressure gradient between the right ventricle and the main pulmonary artery. Right and left ventricular pressure was 92/edp = 14 mmHg and 81/edp = 12 mmHg, respectively. Cardiac output by Fick's method was decreased to 1.3 L·min⁻¹·m⁻². A left pulmonary angiogram demonstrated stenosis of the left pulmonary artery and bilaterally at the bifurcation (Fig 1A,B). The narrowest diameter of the left pulmonary artery was 2 mm, while distally it was 9 mm.

We decided to implant a Palmaz type, P1808 stent (Johnson & Johnson Cordis, Miami, FL, USA) in the left pulmonary artery lesion for the following reasons. First, simultaneous deployment of 2 stents in a Y-shape would be
necessary to dilate the bifurcation stenosis, whereas a single P3008 stent could not cover both the left pulmonary artery lesion and the left bifurcation stenosis. Second, obstruction of the right ventricular outflow or even just the right pulmonary artery was risky, both because the right ventricular pump function was seriously impaired and because her pulmonary circulation depended mostly on the right lung. Written informed consent for stent implantation was obtained from her parents.

After predilatation using an Ultra-thin diamond (Boston Scientific, Natick, MA, USA), with a balloon diameter of 6 mm and a length of 2 cm, the P1808 stent was front-loaded on a Power Flex (Johnson & Johnson Cordis), with a balloon diameter of 10 mm and a length of 2 cm, and was deployed through a 9F long sheath (Brite tip; Johnson & Johnson Cordis) without difficulty. Although the left pulmonary artery was dilated to 7 mm, stenosis at the bifurcation remained (Fig 1C,D). Subsequently we decided to deploy two P3008 stents in a Y-shape simultaneously in the bifurcation. After positioning the 9F long sheaths to the distal left and right pulmonary arteries, the stents, which were loaded on the Power Flex with a balloon diameter of 10 mm and a length of 4 cm, were delivered simultaneously over the Medi-tech super stiff guide wire (Boston Scientific). There was great difficulty in negotiating the right ventricular outflow tract, and the patient developed pulmonary bleeding that was probably related to guide wire manipulation. Left pulmonary angiogram documented perforation of a small branch of the left lower pulmonary artery (Fig 2A). Because she suffered progressive hypoxia despite intensive ventilation, we embolized the perforated vessel using a Gianturco coil (MWCE-38-4-3, Cook, Bloomington, IN, USA) (Fig 2B) to control the bleeding, and terminated the procedure.

A chest X-ray showed massive bleeding in the left lung that spread into the right (Fig 3). In spite of complete hemostasis of the perforated vessel, pulmonary bleeding and hypoxia persisted, in which the PaO\textsubscript{2} was approximately 30 mmHg. She deteriorated critically over the following several days. Extremely high positive endexpiratory pressure (20–25 mmHg) ventilation and intensive hemostatic treatment with fresh frozen plasma and platelet-rich plasma failed to improve her condition. Chest X-ray at that time demonstrated a congested left lung, whereas the right lung was almost clear with decreased pulmonary vasculature (Fig 4A). We concluded that relief of the stenosis in the left pulmonary artery resulted in severe pulmonary edema of the left lung, which had a hypoplastic pulmonary vasculature. Right pulmonary stenosis in the absence of the tight left pulmonary stenosis restricted blood flow to the right lung resulting in hypoxia because of ventilation–perfusion mismatch.

After 13 days of intensive medical treatment, we recatheterized her. Pressure measurements revealed left pulmonary artery hypertension, 71/42 (49) mmHg, although the right and main pulmonary pressures were basically similar to the previous catheterization (ie, 38/18 (24) mmHg and 86/17 (37) mmHg, respectively). A pulmonary angiogram showed no focal bleeding. As mild left bifurcation stenosis was still present, the simultaneous deployment of 2 stents in a Y-shape was the most desirable procedure. However, we chose to deploy one P3008 stent to the right bifurcation through a 9F Super Arrow-Flex sheath\textsuperscript{TM} (Arrow, PA, USA) because of concern about her poor condition and the difficulty of simultaneous deployment at the previous procedure. The narrowest segment of the right bifurcation was 6 mm, whereas distally it was 12 mm. Loaded on the Ultra-thin diamond, with a balloon diameter of 12 mm and a length of 4 mm, a P3008 stent was successfully delivered to the right pulmonary artery stenosis (Fig 5A,B). Right pulmonary pressure rose to 64/17 (34) mmHg, while the main pulmonary artery pressure remained unchanged at 90/19 (42) mmHg. Arterial oxygen saturation under con-
controlled ventilation with FiO₂=1.0 increased from 89% to 98%. Complete hemostasis of the pulmonary bleeding and weaning from mechanical ventilation was achieved a few days and 1 week after the procedure, respectively. There was no left pulmonary artery congestion, while the right pulmonary vasculature increased on chest X-ray 10 days after the second stent implantation (Fig 4B). Doppler echocardiography still showed almost equivalent right ventricular pressure overload, while a lung perfusion scan documented recovery of the right to left ratio, which was 0.85.

**Discussion**

Pulmonary edema occasionally complicates successful balloon dilatation or endovascular stent dilatation of peripheral pulmonary stenoses. Most reports suggest that this complication is transient and that conservative medical management results in a resolution of symptoms and radiographic pulmonary edema within a few days. Its mechanism is believed to be an acute increase in capillary perfusion pressure. Arnold et al reported that an increase in vessel diameter of >70%, a >170% increase in distal pulmonary artery pressure, and a mean distal pulmonary artery pressure after dilatation of >20mmHg were risk factors for pulmonary edema. In the present case, pulmonary bleeding was initiated by the perforation of a hypoplastic peripheral pulmonary artery, while subsequent persistent bleeding was caused by left pulmonary edema. Changes in vessel diameter and distal pulmonary artery pressure clearly met the proposed risk criteria. Residual right pulmonary stenosis aggravated the left pulmonary hypertension and edema. After the first stent implantation, we concluded that the cause of her condition was the relief by the stenosis in the left pulmonary artery, resulting in severe pulmonary edema of the left lung, which had a hypoplastic pulmonary vasculature. Right pulmonary stenosis in the absence of a tight left pulmonary stenosis restricted blood flow to the right lung resulting in hypoxia because of ventilation–perfusion mismatch. Consequently, we decided to deploy another stent in the right pulmonary stenosis to resolve these problems. Although significant pulmonary hypertension remained, the second stent deployment resolved her symptoms quickly.

In conclusion, unilateral pulmonary edema in the presence of contralateral pulmonary artery stenosis is a life-threatening situation and necessitates dilatation of the contralateral lesion. A second stent deployment should be performed early if the simultaneous delivery of 2 stents is unsuccessful.

**Acknowledgment**

We thank Dr Peter M. Olley, Professor of Pediatrics, University of Alberta, for his assistance with the manuscript.

**References**