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Brief Report

Successful release of recurrent pulmonary venous obstruction after repair of totally anomalous pulmonary venous connection by transcatheter implantation of stents

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Abstract We report a 3-month-old female infant, in whom pulmonary venous obstruction occurred after repair of totally anomalous pulmonary venous connection, and which was successfully released by a transcatheter implantation of a stent using the transseptal approach. Close follow-up is required, since the long-term outcome is still unclear. Nevertheless, transcatheter implantation of stents is a promising option for treating this challenging lesion.

Keywords: Catheter intervention; infant; transseptal approach

PULMONARY VENOUS OBSTRUCTION AFTER THE surgical repair of totally anomalous pulmonary venous connection is a serious complication, with a high risk of recurrence, and a high mortality rate despite aggressive surgical re-intervention and catheter intervention.[1–4]

Implantation of stents into the pulmonary veins has been reported,[1–6] but with only transient and disappointing effects. Recently, Tomita et al.[7] reported the efficacy of such implantations of stents in treating recurrent pulmonary venous stenosis when using larger stents. We report successful release of recurrent pulmonary venous stenosis occurring after repair of totally anomalous pulmonary venous connection by implanting a stent using the transseptal approach.

Case report

A term female newborn presented with profound cyanosis and severe respiratory distress immediately following birth. She was intubated and referred to our institute. The echocardiogram revealed that the common pulmonary venous chamber drained into the superior caval vein through a severely stenotic vertical vein. Emergency surgical repair, performed on the first day of life, was tolerated well. Four weeks later, she developed dyspnoea. Echocardiography showed a stenosis at the site of anastomosis between the common pulmonary venous chamber and the left atrium. On the 29th day of life, the pulmonary venous obstruction was released by surgically resecting the hypertrophic endothelium in the left atrium. A small atrial septal defect was created for a future catheter intervention. The postoperative course was good, and she was discharged in good condition. One month later, however, she developed tachypnoea and loss of appetite. An echocardiogram showed bilateral pulmonary venous stenosis. We decided to place stents to relieve the recurrent pulmonary venous obstruction, not only because we suspected that surgical intervention would likely result in recurrent stenosis at the distal pulmonary vein, but also because balloon angioplasty had previously ended in failure in our experience.

Transcatheter implantation of stents

Informed consent was obtained from the parents. The procedure was performed under general anaesthesia

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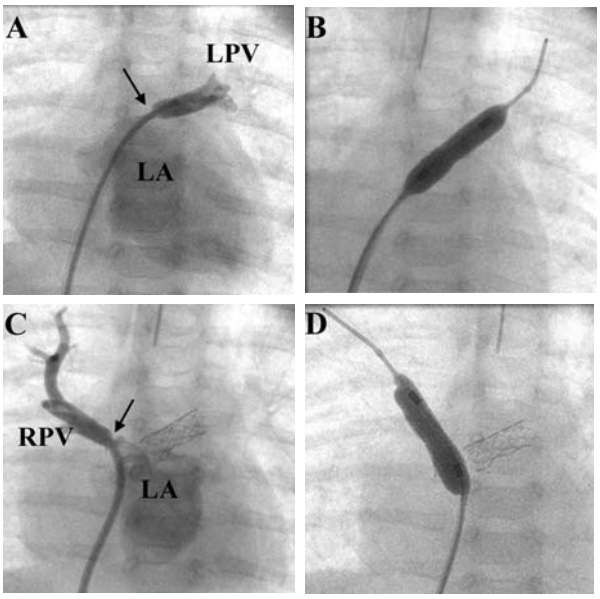

Figure 1.
A. *Left pulmonary vein angiography showed severe localized stenosis (1.7 mm diameter;* arrow*) at its bifurcation.* ***B.*** *A Corinthian IQ stent (Palmaz, 6 mm diameter, 13 mm length) mounted on a balloon catheter (6 mm diameter) was placed and successfully dilated.* ***C.*** *Right pulmonary vein angiography shows the severe localized stenosis (2.0 mm diameter;* arrow*) at the bifurcation.* ***D.*** *A Corinthian IQ stent (Palmaz, 8 mm diameter, 11 mm length) mounted on a balloon catheter (8 millimeters diameter) was introduced and dilated. LPV: left pulmonary vein; LA: left atrium; RPV: right pulmonary vein.*

because of her critical condition. Her body weight was 3.8 kilogram. The left femoral vein was catheterized with a 6 French sheath. The pressure study showed severe pulmonary hypertension, with the ratio of the pulmonary arterial pressure being 0.84 relative to systemic arterial pressure, and with high pulmonary wedge pressures, 16 millimetres of mercury in the right and 10 millimetres of mercury in the left pulmonary arteries.

A transseptal puncture was performed to gain entry into the left atrium, since the atrial septal defect had closed spontaneously. Left pulmonary venous angiography revealed severe localized stenosis, 1.7 millimetres in diameter, at the bifurcation and 4.7 millimetres in diameter at the distal site (Fig. 1A). A balloon catheter (FOX, Abbott, 5 millimetres diameter, 20 millimetres length) was advanced, and balloon dilation was done at 10 atmospheres. A Corinthian IQ stent, of 6 millimetres diameter and 13 millimetres length, mounted on a balloon catheter of 6 millimetres diameter, was placed and successfully dilated. Angiography showed sufficient release of the left pulmonary venous stenosis (Fig. 1B). Right pulmonary venous angiography also showed severe localized stenosis, 2.0 millimetres in diameter at its bifurcation and 4.0 millimetres in diameter at the

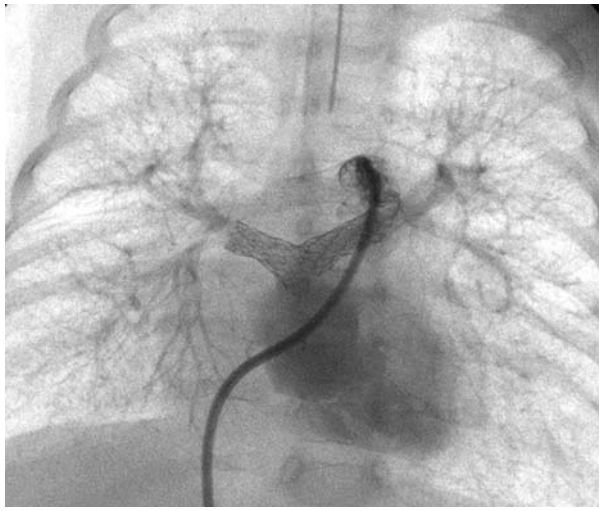

Figure 2.
Pulmonary angiography immediately after implantation of the stents. The pulmonary venous phase showed sufficient release of the bilateral pulmonary venous stenosis.

distal site (Fig. 1C). A Corinthian IQ stent of 8 millimetres diameter and 11 millimetres length, mounted on a balloon catheter of 8 millimetres diameter, was placed and dilated as described above (Fig. 1D). Pulmonary angiography after implantation showed almost complete release of the bilateral pulmonary venous stenosis (Fig. 2). Following extubation in the catheterization room, the respiratory condition was good. She was discharged 13 days later in stable condition.

Redilation of the implanted stents was done using a 7 millimetre diameter balloon at 6 months of age, and using an 8 millimetre balloon at 13 months of age. Pulmonary angiography at 27 months of age showed restenosis in the right pulmonary vein. Balloon angioplasty to the right pulmonary vein was done using an 8 millimetre diameter angioplasty balloon. The pressure study showed that the pulmonary hypertension had improved, with the ratio of pulmonary arterial to systemic arterial pressure now measured at 0.32. At the age of 43 months, the patient was thriving well without respiratory distress under administration of low dose aspirin as an anticoagulation therapy. On echocardiography, the tricuspid regurgitation was mild and the peak velocity was 2.2 metres per second.

Discussion

Several reports have described implantation of stents for pulmonary venous stenosis occurring after surgical repair of totally anomalous pulmonary venous connection, albeit that the results have been disappointing or transient.[1–6] Recently, Tomita et al.[7] reported that such implantation had a promising

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acute effect, and long-term patency was achieved in 2 lesions dilated up to 8.4 and 5.6 millimetres using repeated redilations. We considered that it was necessary to keep the lumen of the vein at least more than 4 millimetres, even in such a small patient as a neonate or an early infant, and that the stents should be dilated up to at least 6 millimetres, expecting the narrowing due to neointimal proliferation of at least 1 millimetre from both sides of the stent. Furthermore, repeated redilation would be needed every few months, because restenosis is likely.[8] In the previous reports, stents of 6 to 10 millimetres were used, however, the expanded diameter of the stents might be insufficient for the size of the body, because our patient is the smallest of those reported thus far. Furthermore, stenosis had previously occurred only at the proximal site, not at the distal site as in our patient. It is another reason why our patient may have had an unusually good outcome from pulmonary venous stenting.

It is of great interest that in the patient we have presented, recurrent stenosis occurred only in the stent within the right pulmonary vein. No restenosis developed in the stent placed within the left pulmonary vein. The reason for this discrepancy is unclear. One possible explanation is that, during the first procedure, there was a slight difference between the right and left pulmonary veins in the disappearance of the waist on dilation. Though the difference was very small, it could have led to shear stress causing endothelial growth on the right. To obtain long-term patency, complete dilation without any residual stenosis might be necessary. This point should be taken into consideration when the stent implantation is performed.

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