Endobronchial stenting in a two-month-old infant with bronchial compression secondary to tetralogy of Fallot and absent pulmonary valve

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Bronchial compression due to pulmonary artery dilation is an important problem in infants with congenital heart disease, and can complicate the postoperative course. In recent years, airway stenting has become a popular treatment for these cases. We achieved success with endobronchial stenting in a two-month-old infant with bronchial compression caused by a dilated pulmonary artery.

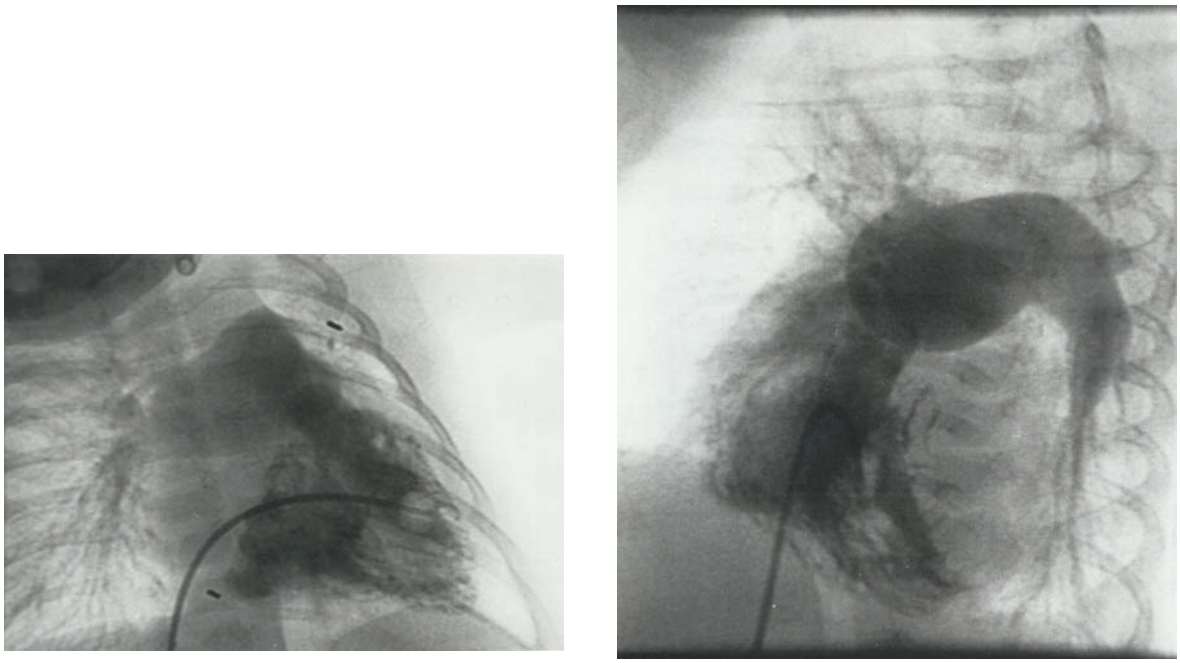
Key words: bronchial stenosis, congenital heart disease, stents and prostheses, treatment.

Respiratory problems secondary to external vascular compression may complicate the course in infants who have undergone repair of congenital heart disease. One major issue is bronchial obstruction in patients with airway compression due to a pulmonary artery dilated by tetralogy of Fallot and absent pulmonary valve[1]. Management of this problem usually involves prolonged ventilatory support; additional high-risk surgical procedures may also be necessary. Endobronchial placement of metallic stents is now a realistic and safe treatment option for these patients[2-4]. We report a case of successful endobronchial stent placement in a two-month-old boy.

Case Report

A two-month-old male infant with tetralogy of Fallot and absent pulmonary valve was referred to our hospital for surgical treatment. On admission, he was cyanotic, tachycardic and tachypneic, and exhibited marked costal retraction. He had a typical "to-and-fro" systolic murmur at the left sternal edge, and his liver was palpable at 3 cm below the costal margin. A chest radiograph showed significant cardiomegaly, pulmonary artery dilatation, and total left pulmonary atelectasis. Echocardiography and angiography revealed concordant connections; right ventricular and atrial dilation, and right ventricular outflow consistent with absent pulmonary valve; anomalous dilation of the left and right pulmonary arteries; and pulmonary valve insufficiency and moderate stenosis (Fig. 1). On catheterization descending aorta O_2 saturation was 95%. Ventricular septal defect (VSDR) and overriding of aorta were noted. In addition, we also noted a large ostium secundum atrial septal defect (ASD) and bi-directional shunting. The patient was resuscitated with an infusion of positive inotropic support and mechanical ventilation. Once he was stable surgery was performed. The tetralogy of Fallot and absent pulmonary valve were repaired with pulmonary artery reconstruction using a xenograft conduit (Cryolife-Ross) and pulmonary branch angioplasty. The VSD and ASD were also closed in the same session.

Postoperative echocardiography showed good repair of the tetralogy of Fallot and the absent pulmonary valve. However, several attempts at extubation failed due to respiratory distress associated with total left lung atelectasis. He remained connected to the ventilator. At 22 days' post-surgery, chest radiograph revealed increased density of the left lung (Fig. 2) and thoracic



(a) (b)

Fig. 1. Right ventriculogram in 15° right anterior oblique and 15° cranial projection (A) and a four chamber view (B) show makedly dilated pulmonary arteries and narrow pulmonary annulus.

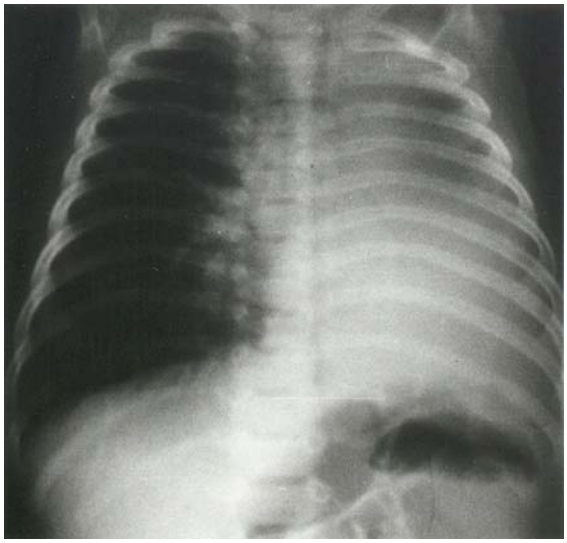


Fig. 2. Pre-procedure chest radiograph showing atelectasis of the left lung.

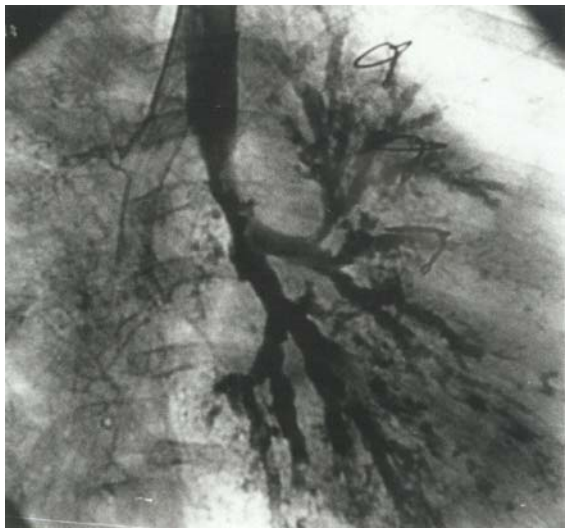
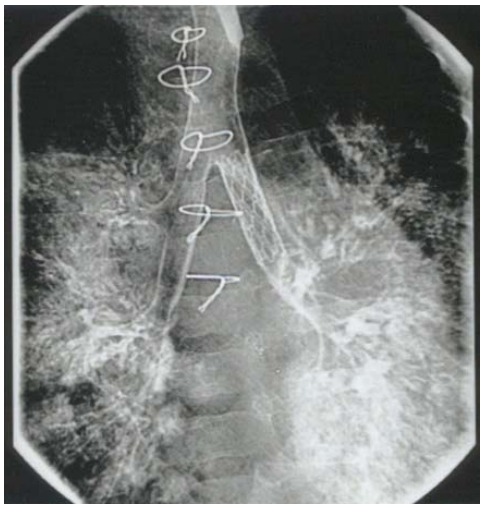


Fig. 3. A bronchogram obtained before stent placement showing the stenosis of left mainbronchus.

computed tomography (CT) demonstrated localized obstruction and collapse of the left main bronchus. Dilute contrast was injected into the trachea, and a tracheogram and bronchogram were obtained. The films confirmed stenosis of the left main bronchus (Fig. 3).

To treat the problem, we used fluoroscopic guidance to introduce a SF multipurpose catheter on a guide-wire (Terumo, Tokyo, Japan) through the patient's endotracheal tube and into the left main bronchus. A 5-mm diameter and 15-mm long metallic stent (Palmaz,

balloon-expandable stent, Johnson and Johnson Interventional Systems Co, Warren, NJ, USA) was then passed through the left bronchus via the catheter, and inserted into the stenotic segment of the airway. A post-interventional bronchogram showed that the proximal end of the stent was within 3 mm of the carina, and confirmed patency of the orifices leading to the left upper and lower lung globes. Stent placement had opened the entire stenotic segment (Fig. 4).



Fi.g. 4. Bronchography, after the stent placment, showing relief of the stenosis.

After the procedure, the patient's left upper lobe expanded normally (Fig. 5). He was able to be extubated five days later, and was discharged eight days after extubation. At 18 months of follow-up, the baby remained well and the stent patency was confirmed with thoracic CT (Fig. 6).

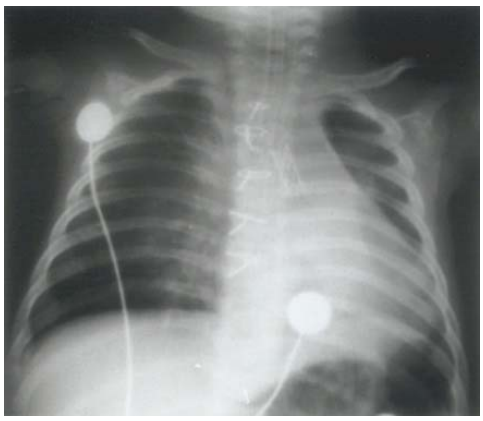


Fig. 5. Post-procedure chest radiograph showing that the lung fields have normal radiographic appearance; the stent is located within the left main bronchus.

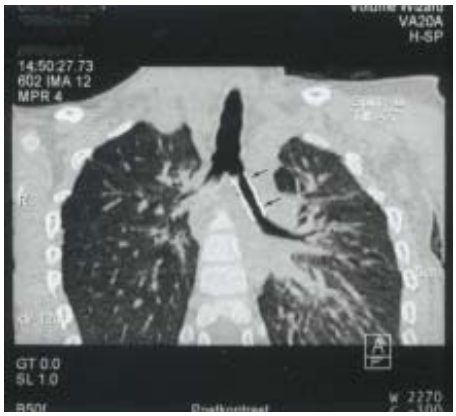


Fig. 6. At 18-months of follow-up, a coronal reformatted CT image shows the patency of the stent (arrows).

Discussion

In infants, the tracheobronchial tree is more susceptible to extrinsic compression because the cartilaginous, muscular, and elastic supports of the airways are weak[1]. As mentioned above, bronchial compression by dilated pulmonary arteries may complicate a baby's postoperative course after repair of congenital heart disease[2-4]. In patients with tetralogy of Fallot and pulmonic insufficiency, the main and left pulmonary arteries are enlarged. Since the left main bronchus is located between the pulmonary artery and the descending aorta, dilation of the pulmonary arteries can compress the bronchus. As a mild or moderately symptomatic infant grows, the diameter and rigidity of the bronchial cartilages increase, and the pulmonary arteries may shift, making intervention unnecessary. However, in some cases, the compression is so severe that the patient cannot be weaned off mechanic ventilation after their heart operation.

In recent years, endobronchial stent placement has been adopted as an alternative to high-risk surgical procedures in pediatric patients with bronchial stenosis. One major advantage is that this can be successfully carried out in infants who are in poor medical condition. The possible complications include infection, re-stenosis, stent migration, erosion and perforation, but this type of stent placement is generally low-risk, easy to perform, and clinical improvement is usually evident within a short time. The long-term outcome of endobronchial stent placement in infants is not yet known. It is true that stent-bronchus size mismatch with growth may lead to obstruction in adult life; however, when the stent becomes loose, it can be removed with relative ease[2].

This case demonstrates that airway stenting for bronchial stenosis due to extrinsic compression in infants is easy to perform, carries low risk, and leads to successful extubation and recovery.

REFERENCES

1. Fischer DR, Neches WH, Beerman LB, et al. Tetralogy of Fallot and absent pulmonic valve: analysis of 17 patients. Am J Cardiol 1984; 53: 1433-1437.
2. Subramanian V, Anstead M, Cotrill CM, Kanga J, Gurley J Tetralogy of Fallot with absent pulmonary valve and bronchial compression: treatment with endobronchial stents. Pediatr Cardiol 1997; 18: 237-239.
3. Hon JK, Jahangiri M, Pastorino U. Endobronchial stenting for extrinsic compression caused by pulsatile pulmonary artery in a 4-week-old infant. Chest 1999; 115: 897-899.
4. Goldman S, Rimell FL, Meza MP, Newman B. Diagnosis and management of left main stem bronchus compression. Ann Otol Rhinol Lryngol 1997; 106: 461-465.