Case Report

Reversing Left Bronchus Obstruction by Compression of a Pulmonary Artery Stent

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We report a case where endovascular stenting as part of the treatment of complex cardiovascular disease led to airway compression. Using a novel technique, this was successfully reversed. Simultaneous inflations of two balloons—one in the obstructed bronchus and one in the aorta combined with external compression of the chest—resulted in compression the stent. This report illustrates that by thinking "out of the box" and bearing in mind spatial relationships inside the chest, it is possible to diminish the anterior–posterior diameters of a stent. This intervention proved to be successful with relief of the left bronchial compression and improvement of chronic airway infection. © 2016 Wiley Periodicals, Inc.

Key words: balloon; angioplasty; compression; stent; obstruction

INTRODUCTION

Tracheobronchial compression by cardiovascular structures can complicate the course of congenital heart disease in children [1]. Airway compression is caused either by anomalies of the configuration of the great vessels or by enlargement of normal structures [2]. We report a case, where endovascular stenting as part of the treatment of complex cardiovascular disease led to airway compression.

CASE REPORT

The patient, a 5-year-old boy, was known with complex univentricular heart disease: double inlet left ventricle, transposition of the great vessels, and an interrupted aortic arch. He required several procedures: he was initially managed by a hybrid procedure consisting of bilateral pulmonary artery banding and a ductal stent shortly after birth. At 6 months of age, a Damus Kaye Stansel with extensive reconstruction of the aortic arch was carried out. This was complicated postoperatively by persistent wheezing and respiratory distress. Computerized tomographic (CT) scanning of the chest at this stage revealed mild narrowing of the main stem left bronchus ascribed to pressure exerted by the neo aortic arch, but the child responded well to inhalation therapy. He then proceeded to a bidirectional Glenn operation at age 15 months. During the surgery, a trimmed down (to 12 mm) Genesis PG 1910 stent (Cordis Europa, N.V.) on a 10 mm balloon was implanted to treat residual stenosis at the site of the previous left pulmonary artery band (hybrid). At the age of 3.5 years, a fenestrated Fontan was performed.

During his first school winter, recurrent chest infections were treated with physio- and inhalation therapy (mucolytics, inhaled steroids, bronchodilators) as well as antibiotics. He became progressively more cyanosed and a diagnostic cardiac catheterization was performed to evaluate pulmonary artery growth, to exclude collaterals, and to do a proof occlusion of the fenestration.

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Conflict of interest: Nothing to report.

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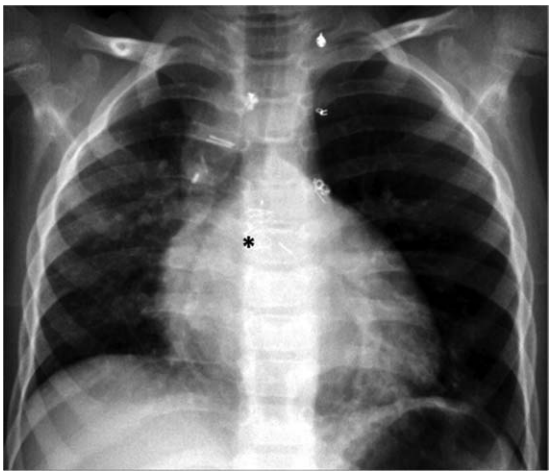

Fig. 1. Chest radiography. Chest radiography shows reduced blood flow to the left lung. Stent in left pulmonary artery (asterisk).

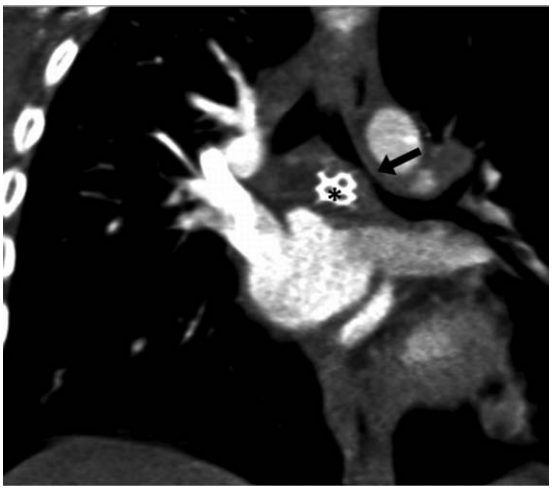

Fig. 2. CT scan. CT scan demonstrates compression left main bronchus (arrow). Asterisk = stent in left pulmonary artery.

Aortic saturations were 85% with acceptable cardiac pressures. Since the child was now older, it was decided to accommodate for future somatic growth by dilating the stent in the left pulmonary artery even though no gradient was measured. The LPA was dilated using a 12 mm Opta Pro® angioplasty balloon (Cordis Europa, N.V.). Following the procedure, angiography showed somewhat sluggish flow in the LPA which we ascribed to probable vascular spasm at that stage. The child was extubated uneventfully and discharged the following day, with the decision to close the fenestration at a later stage.

He was referred for respiratory evaluation because of severe and persistent airway problems in the following 3 months. Despite repeated antibiotic treatment and chronic inhaled steroids, he had a continuous productive cough with sputum expectoration, persistent wheeze, and progressive dyspnea. On clinical examination, the child was cyanotic, underweight (height 50th centile, weight 10th–25th) with chest hyperinflation and in- and expiratory wheezes audible only over the left chest. Saturations in room air were 83%. A chest X-ray showed hyperinflation and hypoperfusion of the left lung with signs of alveolar infection (Fig. 1). Repeat chest CT showed external compression of the left main bronchus by the stent; this was confirmed by bronchoscopy (Fig. 2). On repeat history taking, it was clear that airway symptoms had worsened since the last cardiac catheterization when the left pulmonary artery stent was dilated.

We were faced with a dilemma of how to manage this child: (i) conservative "wait and see" approach to allow for outgrowth, (ii) surgery consisting of removal of the stent, (iii) aortoplasty to make more room in the chest, or (iv) stenting of the left main bronchus. In the end, we decided on a novel strategy to attempt to reduce the external diameter and/or configuration of the stent. Compressing a stent in the midthoracic "open" space would be difficult since all the structures are compliant and therefore give way easily. The challenge here was to exert sufficient pressure directly on the stent in order to deform it. We decided to use two balloons to compress the stent, but simultaneously had to ensure that the forces would be applied to the external surfaces of the stent. An 8 mm Powerflex® Pro(Cordis Europa, N.V.) was placed in the left main stem bronchus by our pulmonologist and a 20 mm Tyshak® balloon (NuMED, NY) in the aorta retrograde over a wire. Both balloons were simultaneously inflated (Fig. 3). At the moment of maximum inflation, quick sharp external chest compression was delivered over the sternum under fluoroscopic control. The configuration of the stent changed to a more oval shape and repeat angiography showed a marked improvement of flow in the LPA (Fig. 4A and B).

Immediately after the intervention, the ventilation of the left lung improved: symptoms of coughing and sputum production improved markedly post-30 months of the procedure.

DISCUSSION

External vascular compression on the airways may complicate the clinical course of children with congenital heart disease [2]. It is estimated that the incidence of this comorbidity is around 1–2% [3].

Although the most common congenital anomalies associated with airway compression are the vascular rings, many other vascular mechanisms of airway compression exist, especially in the context of complex congenital

heart disease such as absent pulmonary valve syndromes and interrupted aortic arch with dilated cardiomyopathy [4]. In our patient, review of the surgery showed that the aorta was retracted more caudally into the chest than usual. Since there is limited space in the chest, one must always bear in mind that, in an enclosed space, "giving" to one structure could be "stealing" space from another. These spatial arrangements should always be taken into consideration when performing procedures. Bronchial compression as a result of pulmonary artery stenting has been reported before [5]. In this report of a patient with transposition of the great vessels, the bilateral pulmonary artery stents were surgically removed.

Stenting of the compressed airway has been reported but this is often associated with complications such as stent dislocation, retention of secretions, formation of granulation tissue, and airway obstruction, which may be fatal [6]. Most authors, therefore, caution against the use of stents in unrelieved external airway compression in children and support their use only if there are no good alternatives [7].

Deformation of stents, although rare, has been reported in children and adults as a result of cardiopulmonary resuscitation [8,9]. Most of these were, however, stents in the descending aorta or stented aortic valves. Owing to risk of caudal displacement, a balloon was thus inflated in the obstructed bronchus. The aim was that the stent would thus mold itself around the bronchus ensuring that patency of the bronchus would be maintained after deformation. This case also shows that one should always carefully deliberate all the indications for an intervention before proceeding and be careful of opportunism. Growth is a long and slow process and we could perhaps have waited longer before expanding the stent. As an additional preventive measure, a guidewire (and balloon) can be positioned in the LPA to manage overcompression of the stent. This would also allow molding of the stent in the event of marked deformation as the compression forces in the chest are unpredictable and difficult to control. We did not anticipate the compression to be so successful, highlighting the importance of additional safeguards.

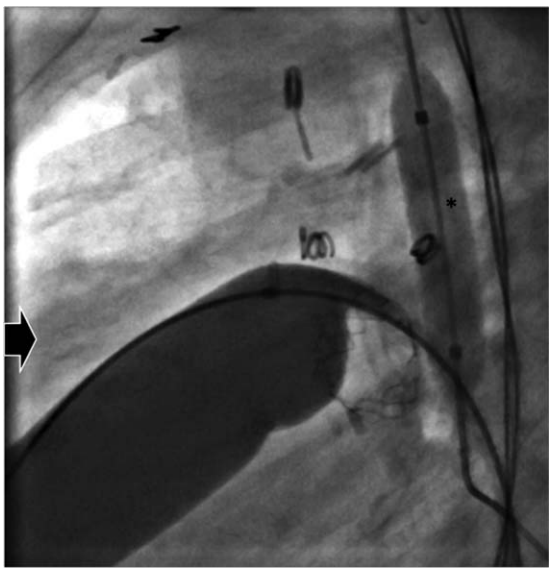

Fig. 3. Technique of compression of stent. Large Tyshak balloon in aorta, smaller balloon (*) in left main bronchus. Arrow demonstrates direction of external chest compression force.

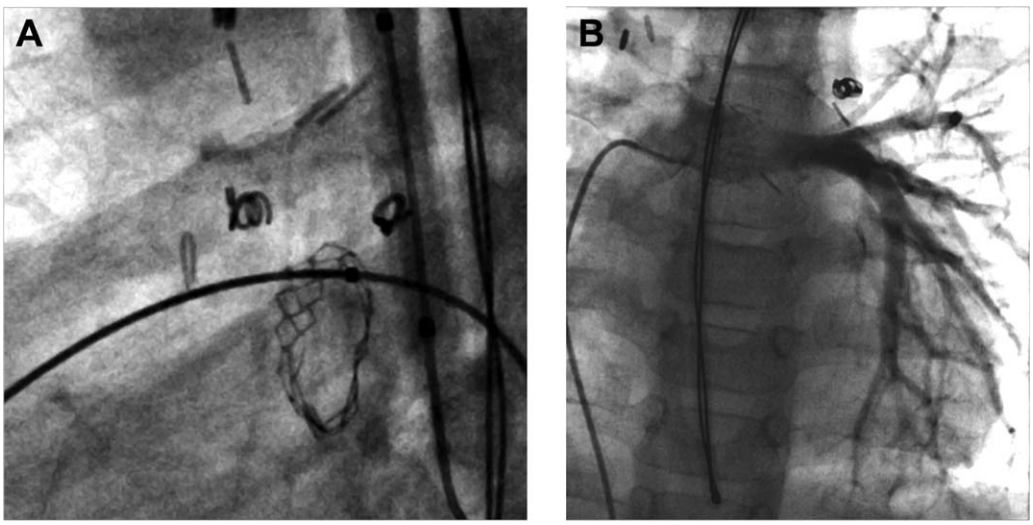

Fig. 4. Deformed stent in LPA. (A) Lateral projection during fluoroscopy shows compressed and deformed stent. (B) Pulmonary angiography shows good perfusion left lung (anteroposterior projection).

CONCLUSION

This report shows that by thinking "out of the box," bearing in mind spatial relationships within the chest, it is possible to diminish the anterior–posterior diameters of a stent. This intervention proved to be successful with relief of the left bronchial compression and resolution of chronic airway infection.

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